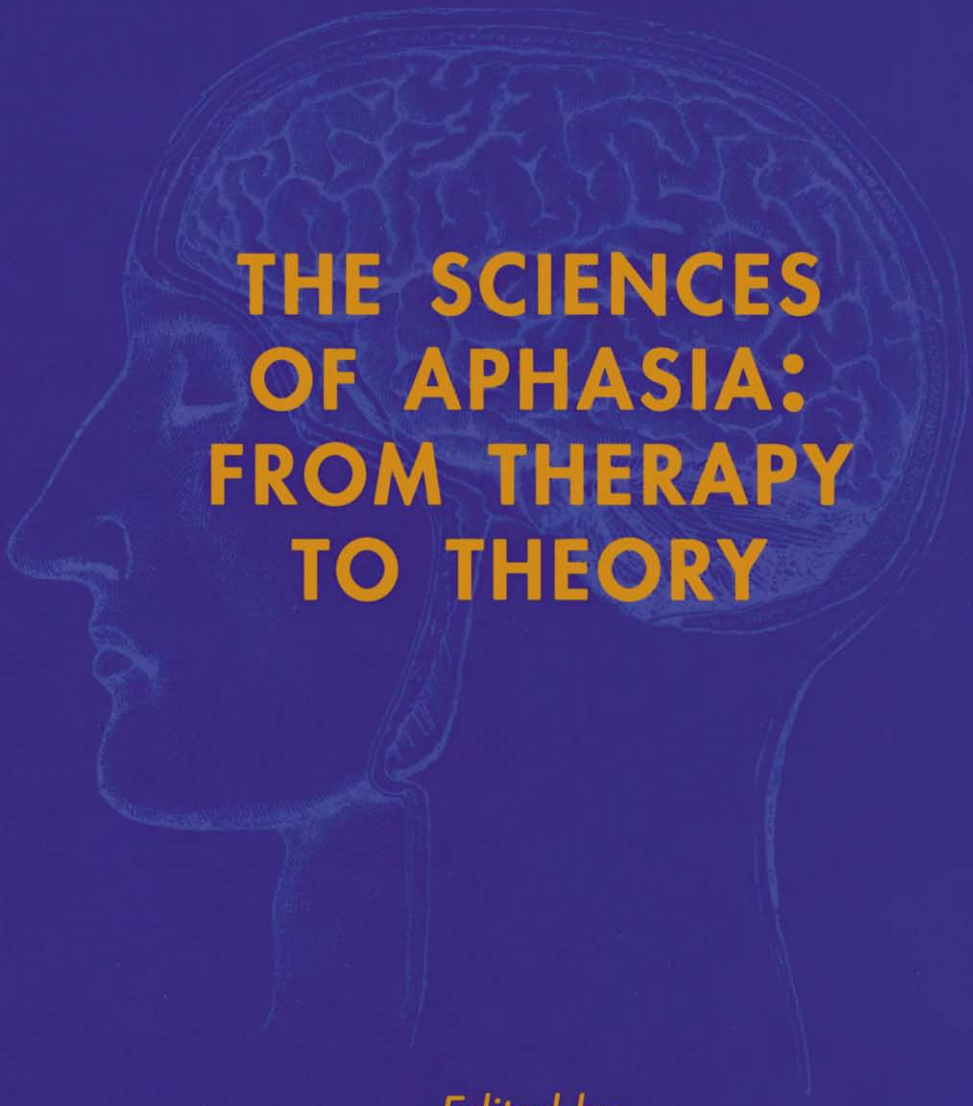

THE SCIENCES OF APHASIA: FROM THERAPY TO THEORY

Edited by
Ilias Papathanasiou
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Edited by

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PREFACE

APHASIA THERAPY: PAST, PRESENT AND FUTURE

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For many years, aphasia therapy has been the poor relative of studies in the neuropsychology of language. Like neurology itself (until quite recently), aphasiology has been strong on diagnosis but weak on effective remediation. I write "effective" remediation because speech and language therapists practiced their trade for many years without fully realizing the necessity to submit objective evidence of efficacy.

Consider the situation in the early 70s of the last century. Darley (1972) summarized the state of the art as follows: "Studies of the effect of therapy on the course of recovery from aphasia yield inconsistent results and permit no generalization to the population of aphasic patients". A few years later, Marshall, Holmes and Newcombe (1975) noted that some victims of aphasia recover a reasonable measure of communicative competence and others do not, an undoubted fact that raised two basic questions: "(1) Can the variables which account for the existence of different levels of residual disability be found? (2) Can therapies be devised that will speed rate of recovery and reduce the degree and scope of persistent impairment?" Although the questions were reasonably clear, the answers at that time were anything but.

Nonetheless, the 1970s and 80s did eventually see the beginning of a concerted effort to produce rational therapies and (even more important) rational evaluations of efficacy. Albert, Sparks and Helm (1973) had devised melodic intonation therapy (MIT), a technique that drew upon the (musical) intonation capacities of the intact right hemisphere to improve verbal expression after left hemisphere stroke. For the most part, the results obtained in non-fluent aphasia were highly encouraging (see, for example, Laughlin *et al.*, 1979); Goldfarb and Bader, (1979). On a more general note, Enderby and David (1976) proposed a serious randomized trial of speech therapy for aphasia. The final outcome (David, Enderby, and Bainton, 1982) was somewhat mixed. Patients seen by professional speech therapists and

untrained volunteers seemed to recover at much the same rate; patients who started treatment late made as much progress as those who started earlier. The authors concluded that "the improvement in communication which occurred during treatment may be due both to the appropriate stimulation which was based on detailed and accurate speech therapy assessment, and to the regular support and encouragement provided within the therapeutic relationship."

An unkind critic might suggest that therapy had been shown to be a placebo effect. A related study (albeit without full randomization and with a very large non-random drop out rate) was conducted by Basso, Capitani and Vignolo (1979). They found that time between onset and the first examination *was* negatively correlated with improvement. But like David *et al.* (1982) they reported that delay in obtaining language therapy after the onset of language disorder did not reduce the efficacy of rehabilitation.. Nonetheless, Basso *et al.* (1979) wrote, somewhat alarmingly, that with respect to language rehabilitation, "the relationship of type of aphasia to improvement was not significant". Unless the particular type of therapy was closely matched to the specific deficits shown by the patients, this latter finding is susceptible to a less encouraging interpretation than that which the authors wish to uphold.

The issue of specificity was finally seen to be of crucial significance in the 1980s and 90s. Under the influence of the somewhat earlier cognitive revolution in neuropsychology, a major reconsideration of both the structure of language and speech rehabilitation itself, and of how to evaluate the results thereof, was at last undertaken. In brief, it became clear that aphasia therapies must be explicitly tailored to the pattern of impaired and preserved performance in the individual patient. This single-case study approach (Coldheart, 1983) drew heavily upon arguments that had previously been advanced for why the description and theoretical interpretation of neuropsychological symptoms should, for the most part, be based upon the performance of individual patients and case-series (Marshall and Newcombe, 1984; Newcombe and Marshall, 1988). Group studies based upon the polytypic syndromes of traditional aphasia taxonomies are unlikely to be either theoretically revealing or practically useful.

One consequence of this emphasis upon the individual is that randomized controlled trials of aphasia therapy are not the best way to evaluate efficacy (Howard, 1986; Pring, 1986), although such large-scale trials could no doubt be improved by consideration of effect size (as assessed by meta-analysis) rather than statistical significance per se (Fitz-Gibbon, 1986). More importantly, longitudinal single-subject experimental designs are required that can evaluate the efficacy of treatment in the individual. A number of such protocols are now available including reversal and withdrawal designs, multiple baseline designs, and cross-over

treatment designs. Excellent discussion of these issues can be found in Willmes and Deloche (1997) and Franklin (1997). Two key books that outline some preliminary results from the new approach are Seron and Deloche (1989) and Berndt and Mitchum (1995). In order to know what the future may hold for best practice, I recommend the articles that follow, based upon the Euroconference 2000: The Sciences of Aphasia: From Therapy to Theory.

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Chapter 1

PROSPECTS IN THE STUDY OF APHASIA: THE NATURE OF THE SYMPTOM AND ITS RELEVANCE FOR FUTURE RESEARCH

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INTRODUCTION

There are several problems in aphasia research and neuropsychology that one could term fundamental. One of these is the nature of the image, for example, the verbal image of inner speech or the visual image of hallucination. The image is fundamental because the relation of the image to the perception goes to the heart of the transition from private to public space. Paul Schilder is one of the few prior to imaging research who have studied this problem, for example, finding that he could induce vertigo with cold water calorics in the hallucinatory images of cases of belladonna intoxication. Another such problem is the nature of the present moment. There are few investigations of the duration of the present since the days of William James. My own studies and those of Ernst Pöppel are exceptions (also Kinsbourne, 2000; Dennett and Kinsbourne, 1992). The duration of the present is important for many reasons, not the least of which is that it spans instants of physical passage and is a fundamental obstacle to a naïve mind-brain reduction.

Still another and perhaps even more fundamental problem is the origin of the error or symptom. It is fundamental because a symptom is a fragment of a behavior that points to a state of the brain. If the fragment points to a disrupted phase in the mind/brain state, behavior more generally is a symptom or expression of the brain state as a whole. This implies that the relation of the symptom to the defective segment is a nucleus of the relation of any behavior to the mind/brain state that generates it. The symptom is only pathological when it is deviant.

In neuropsychology, a symptom is a fragment of the unexpected in an otherwise normal behavior. An aphasic error is an example of an unexpected (deviant) performance.

I would hope that all of us accept that the study of symptoms is instructive with regard to the normal; otherwise clinical study would be a waste of time. Yet, after so many years of describing clinical symptoms, we still do not have a theory as to the nature of a symptom, how it arises or what it refers to. Until we do, clinical work, which depends on symptom description, will be considered unscientific discourse and relegated to the status of anecdote. In fact, this is what has happened over the past 30 years of cognitivist studies, which by and large have taken the view that symptoms are subjective, variable, qualitative, fleeting, non-reproducible, the static or noise in the damaged computer-brain. The cognitivist accepts that a symptom may be useful as a guide to an experiment, but beyond this has little scientific value.

Whereas process theory looks at continuities and transitions, cognitivism looks at dissociations and slices of behavior averaged or summed over samples. There is an emphasis on deficits, failures to perform or omissions. These have the virtue of being more or less repeatable and quantifiable, and thus conform to the requirements of scientific theory. It is argued that one can infer the damaged mechanism from the failure in performance. Thus, a score of 9/10 on one test implies an intact mechanism, and a score of 2/10 implies that the mechanism is damaged, or that connections to or from it are damaged. In single case studies, the same method is used to explore the fine structure of a process. The quantitative approach is simply re-asserted at progressively finer levels.

While soft cognitivists do utilize symptoms as confirmations of experimental findings, the tendency is to assert a finding on the basis of theory – modularity, quantity, dissociation, interaction – and then select a symptom that supports it. To illustrate a local model of function by an unusual case study is a fraudulent use of the clinical material which, in my opinion, should be taken as a whole or not at all. The one thing we learn from the clinical material is the transition from one symptom to another, the qualitative shift from moment to moment, i.e. the dynamic nature of the symptom. An isolated error is as meaningless as an isolated deficit. The Emperor's Clothes of cognitivist research are that it *ad hocs* mechanisms from nothing, from the failure to do something. As a result, the components and connections that are postulated to fill in the gaps in theoretical models driving the research proliferate beyond all correspondence to brain study.

I think this emphasis on quantity at the expense of quality is completely backwards. The symptom is not a mere decoration to a scientific psychology, but points to a pre-processing phase in language or cognition, while the omission tells us little or nothing about brain

process, though it does provide a constraint on theory construction based on the analysis of symptoms. The nature of the symptom is a central problem but what would constitute a scientific theory of the symptom?

THE SYMPTOM

Many years ago during a stay at the laboratory of A R Luria in Moscow, he told me that for him a theory of the symptom was the most important goal in neuropsychology. Luria did not have a definitive theory but rather different perspectives depending on the problem under analysis. In aphasia, he borrowed some of Pavlov's ideas, for example, with respect to paraphasia he proposed an equalization of associative strengths, so that one word was as likely to occur as another. At times he seems to have endorsed Wernicke's view that paraphasia developed when the posterior lesion disinhibited Broca's area and allowed it to run on chaotically. But paraphasias are not random misses or guesses. Such accounts do not explain their regularity or lawfulness. Luria also explained errors in relation to Vygotsky's hierarchic theory and the concept of regression, for example in the role of the automatic and the volitional in restitution or verbal regulation. The symptom was also linked to the notion of the functional system. But there was no unified theory. Kurt Goldstein, whose paper on the symptom Luria admired, thought that the aphasias reflected a progression from thought to speech, from a phase of abstract or categorical thinking in amnestic aphasia, to defects at the thought-speech transition in conduction aphasia, to motor speech impairments in Broca's aphasia. He was close to microgenetic and gestalt theorists such as Pick, Lotmar, Bay and Conrad, who interpreted the aphasias as disruptions in a process of gestalt-formation or in the actualization of speech out of thought.

My own work has centered on the concept of the aphasias as disruptions at successive moments or phase transitions in the realization of an utterance over planes of evolutionary growth in the forebrain (see review in Brown, 2001). The aphasic symptom represents a snapshot of a concealed, submerged or, better, transformed pre-processing phase that, for the moment, actualizes as a final form. From this standpoint, the symptom is a link from the pathological to the normal, as a piece of preliminary language that becomes, for that moment, a terminal product. There is no arrest of language production, no blockage or obstruction; the derailment is carried through into subsequent phases. For example, a lexical-semantic error that dates to an earlier phase in the realization of an utterance undergoes normal phonological processing that dates to a later phase. On this model, a paraphasia is a brief attenuation of normal language that is carried through normally to a distal termination.

Take, for example, the process of naming and its disruption in posterior aphasia (see Brown, 1986; 1994). An aphasic confronted with a chair may produce various types of errors. With lesions of left infero-lateral temporal cortex, often bilateral in younger patients, errors show wide semantic distance, e.g. wheelbase for chair. When pervasive, this is semantic jargon. There are similarities to schizophasia, and to the so-called "non-aphasic misnaming" mis-described by Weinstein and Keller (1964) and more carefully studied by Rochford and Williams (1962). With left temporo-parietal lesions, errors tend to conform to the object category, e.g. table for chair, or the word may be semantically constrained but not evoked, as in a TOT state. This is anomic aphasia. With focal left superior temporal lesions, the item is available but does not achieve adequate phonemic encoding, e.g. predisent for president. This is conduction aphasia, which is just fluent phonemic paraphasia apart from the repetition disorder, which is uniform in such cases. In most patients the combination of semantic and phonemic errors gives neologism, though there are other sources, such as blending.

In this model, there is progressive zeroing in on the target over growth planes in brain evolution. The fundamental process is a whole-part or context-item transform. We see this not only in the specification of lexical items but also in phonological realization, as targets actualize from wide to narrow feature distance. The model is evolutionary in more than its anatomical substrates; processing is uni-directional, like growth, and like growth it is obligatory and cyclical, or recurrent. The cycle of birth and death, waking and sleeping, is replicated each moment in the arising, perishing and replacement of the mental state. There is no multiplicity of possible routes or open-ended concatenation. Lesions do not knock out boxes or sever the arrows between them. Rather, the lesion displays phases in a transitional sequence from depth to surface. And like evolution and maturation, phylogeny and ontogeny, the progression in the mental state is from potential to actual, from past to present, from unity to diversity, not the other way around. Phylogeny delivers a brain, ontogeny fine-tunes it, microgeny deposits a cognition. The millions of years of evolution and the history of the individual are collapsed in a fraction of a second. This is an iterative process like the heartbeat that recurs billions of times over the life span.

For a long time I struggled to integrate the ontogenetic dimension with phyletic and microgenetic concepts in search of a deeper theory that might explain how symptoms arise. I wanted to better understand the attenuation effect with subsequent normal processing that would resolve the microgenetic model with the heuristic of double-dissociation. Isolated lexical-semantic and phonological defects imply separate components. One reason for the difficulty was the demise of the regression hypothesis, associated in aphasia study with Hughlings Jackson and Roman Jakobson, the idea that pathology unpeels the onion-skin of development. The hypothesis has been refuted many times over, but I think there is something

in it worth preserving that just needs a fresh interpretation. Ontogeny was also problematic from the anatomical standpoint since the brain matures more or less as a whole making it difficult to tease apart the anatomy of distributed systems with the specificity necessary to map the systems to pathological and normal cognition. In contrast, the long history of phyletic growth settles the brain into distinct anatomical formations - limbic, neocortical and so on - that can more readily be correlated with phases in language, perception and action. The symptom stands at the crossroads of all of these difficulties and ontogeny, far from being the obstacle to a coherent genetic model, now is the key to its solution.

MORPHOGENESIS AND SYMPTOM-FORMATION

I want to approach the symptom and its relation to normal function by way of a detour into ontogeny, specifically the topic of morphogenesis, the process through which the brain develops. Morphogenesis is the growth process in the fetal brain that mediates the translation from the genetic code to brain structure. We know there is no simple correspondence between the code and brain morphology. Even if 50% of the genome is devoted to the human brain, this cannot possibly account in a 1:1 manner for the trillions of cells and their connections. That is why developmental biologists focus on the correspondence rules or algorithms that govern the translation of code to structure. At the molecular level these are complex events involving timing, rate and contextual, even anticipatory, effects of gene combination, as well as a variety of epigenetic factors. In neurology, growth and morphology are usually conceived as different problems than those of function or behavior. We tend to think that developmental process lays down brain structure which then outputs function. Cognitivists assume that stages in evolutionary growth, like stages in the assembly of a computer, are unrelated to the operation of the mind or software. Surely this is an inadequate view of morphogenesis and of cognition.

The problem in the relation of brain structure to function is that we think of function as active and dynamic and of structure as fixed and stable. The result is that mental activity is artificially stabilized in compartments that can be localized, when what is needed is an enlivened concept of structure in terms of process. If we consider morphology and behavior in relation to growth, with growth a population dynamic and the cognitive process the configured properties of populations of cells, it might be possible to map the one to the other, i.e. to correlate the cognitive process with the growth process.

The first step in re-thinking the concept of structure is to consider the development of the brain not as the precursor of its final morphology but as morphology itself, i.e. morphology is

an artificial slice through development with behavior its four-dimensional structure. Put differently, morphogenetic process lays down form in the form of morphology early in life, and lays down form in the form of behavior later in life. On this view, the growth process becomes the cognitive process or, rather, a single process that initially deposits the structure of the brain continues or is reiterated to lay down language and cognition. The argument is that behavior is four-dimensional morphology, or structure over time. This means that if we can identify some features of the initial growth process that deposit a relatively mature brain, the same process would be responsible for depositing behavior.

There have been prior speculations along these lines. Karl Pribram (1991) in *Brain and Perception* alludes to this possibility, and mentions older work in embryology that indicates that patterns or lines of development in embryogenesis might persist as force lines that determine the processing direction or the characteristics of processing in mature perception. Others have commented on the possibility that ontogenetic sculpting relates to learning and information representation in the brain. But little more has been written on the topic. I want to consider two mechanisms involved in morphogenesis in the normal fetus, and in errors of development, which help to refine the regression hypothesis and provide a first pass at a scientific theory of the symptom. These mechanisms are parcellation and heterochrony.

PARCELLATION

Parcellation is the idea that early in fetal brain growth there is exuberant proliferation and migration of cells, initially an over-abundance and multiple overlapping connections, with a loss of both cells and connections through competitive interaction in order to achieve synaptic specificity. In almost every area of the brain that has been studied, exuberant cell growth is accompanied by cell loss as structure takes on definition. Initially, the elimination is for cells, then for the connectivity. For example, Changeux (1985) found in the juvenile mouse that the multiple climbing fibers innervating the Purkinje cells drop out as the cell arborizes leaving a single innervation. Perhaps the most dramatic example of elimination in development was reported by Rakic (1992) in macaque. Using electron microscopy, he found that by the age of sexual maturity there is a loss of over 2 trillion synapses in neocortex. The process of parcellation is innately driven but requires sensory input at birth to continue. Studies have shown that visually deprived animals do not develop the fine connectivity typical of the normal visual system. A more diffuse or redundant connectivity accompanies a more ambient mode of perception with a loss of fine discrimination. The process of specification by elimination is an innate characteristic of morphogenesis, but sensory experience drives the process into maturation without altering its fundamental properties.

At some point in morphogenesis, probably after most anatomical connections are established, the specification continues through the inhibition of an otherwise stable connectivity. That is, inhibition plays the role in function that elimination played in growth. Inhibition accomplishes more or less the same thing physiologically as elimination did anatomically. It is also less emphatic and reversible. There are many examples of this in normal development. Visual evoked potentials recorded over a wide area of cortex in juveniles gradually zero in on visual cortex in the adult and in pathology re-generalize. Coghill (1964) described successive inhibition of spinal reflexes in the differentiation of motility. In pathology, there is re-generalization with mass reflexes. Progressive inhibition may account for the concept of diffuse right and focal left hemisphere function (Semmes, 1968), as well as progressive specification of the left language areas. Inhibition occurs in the development of action in newborns, which goes from a global movement of the hands or face (the cherubic face of the infant) to one that is more finely individuated. From another perspective, Wall (1988) noted that cortical cells with wide receptive fields in juveniles undergo progressive inhibition as a means of establishing specificity. The fields re-generalize through disinhibition of latent synapses in pathology. This might be one mechanism of compensation through mirror brain areas.

The equivalent of parcellation in growth, or surround inhibition in physiology, is the whole-to-part or context-to-item transformation in cognition, the specification of parts out of wholes by way of constraints on developing form. The intuition that such a process is essential in language and cognition is confirmed by the ubiquity in the literature of such mechanisms as individuation, differentiation, specification, gestalt-formation, holistic-analytic and frame-content theory. These are different ways of describing the whole to part transform, where wholes are not sums but configured antecedents, and parts are realizations, not constituents. I think this transform is the basic pattern of the system, whether it is achieved by elimination, inhibition or context-item specification.

The sculpting by way of constraints at successive phases in a transitional sequence is the driving force in microgenetic process. A single process guides evolution, development and cognition, not multiple processes acting at different points. The argument is that parcellation by elimination, specification by inhibition and the elicitation of items from contexts by constraints at successive phases in cognition, are all instances of a common process in which growth lays down patterns that are derived into the patterns of cognitive activity. The concept that cognitive form is trimmed by the elimination of maladaptive routes of actualization in the mind/brain state is consistent with the evolutionary principle that unfit forms of life are pruned by the environment.

HETEROCHRONY

As parcellation is the pattern of process in development and behavior, heterochrony is the rate or timing of this process. Heterochrony is the idea that in brain development or evolution, different organ systems can develop at different rates, and that this difference in the timing of development can lead to shifts in evolutionary outcomes, including adaptations, errors and severe aberrations. *Neoteny* is a selective retardation or prolongation of a juvenile stage in development. The selective slowing of a stage can be the springboard of a new burst in evolutionary growth. An example of neoteny would be the lengthening of the period of sexual maturity over the primate series leading to man, from two years in the lemur, seven in the great apes to about twelve in humans. Delayed closure of the cranial sutures is a neotenous feature that permits the expansion of a similarly neotenous brain. A prolongation of juvenile forms leads to evolutionary change. There is an emergence of novel form from the prolongation of earlier phases in development. This is an illustration of the evolutionary principle that new growth occurs from antecedent stages, not by terminal addition. Put differently, innovation arises from earlier stages of potential, not endpoints of specification. As Steven Gould (1982) has put it, evolution is a branching bush, not a ladder of progress.

First, let us consider the adaptive aspects of neoteny, then the maladaptive ones. To do this I would like to mention a little noticed theory of dominance that I published many years ago that I believe offers an alternative to the "bigger is better" concept that is its main anatomic rival. We know that in pre-human primates, there is relative symmetry of function. One can rephrase this by saying there is bilateral representation of many functions and contralateral representation of others, for example, of motor function. There is little or no asymmetry or lateral representation as occurs in humans. The microgenetic theory of dominance maintains that the Wernicke and Broca zones are phonological processing planes that individuate out of a background association or integration neocortex. This individuation creates a phase in cognition intermediate between the integration and primary cortices. Lateral representation results from a subtraction of the functional connectivity mediating bilateral representation.
Nothing is added. Rather, new growth occurs from penultimate phases by way of elimination as an effect of parcellation. At the same time, the development of the Wernicke and Broca zones occurs as a juvenile phase in growth is prolonged. Here, parcellation and neoteny combine to give lateral asymmetry.

This is an example of a positive adaptation. Evolution and development are double-edged swords. The same process that gives a beneficial adaptation can account for one that is harmful. This is where the symptom comes in. A symptom is a kind of negative adaptation. In order to understand the symptom, it is necessary to turn to the role of parcellation and neoteny

in errors of development. Serres (1860, cited in Gould) described the neotenous origin of anomalies when certain parts lag behind in development and retain at birth the characteristics of earlier stages. He noted that the anomaly pointed to the stage in development that was unduly prolonged.

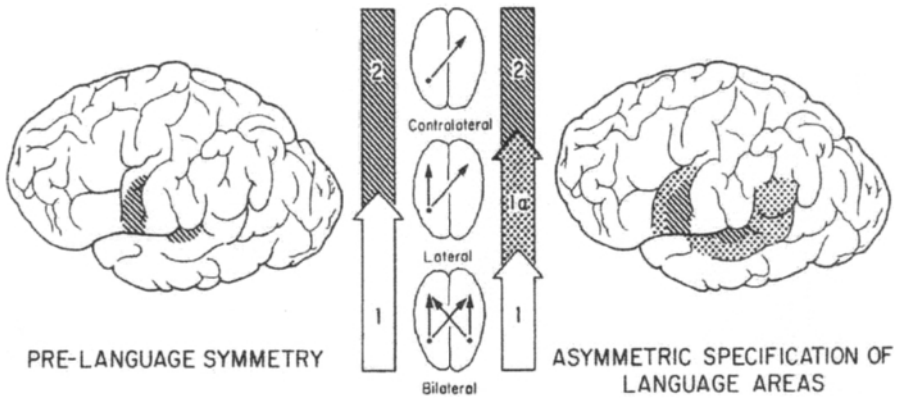

Figure 1. The emergence of Wernicke and Broca zones within "integration" neocortex is a neotenous expansion in brain development corresponding to the development of cerebral asymmetry through parcellation. Neoteny and parcellation combine to specify asymmetric planes in the actualization of language interposed between bilateral and contralateral modes of representation

To take an example, a neoteny at a stage in limb bud development may explain why the digits do not fully individuate and an infant is born with a fist-like hand. The local process stagnates but the overall development proceeds unabated. The defect points to the phase in development where individuation should have occurred. When foreshortened digits appear with syndactyly there is normal nail bed formation. Presumably, there is a disruption at the stage where the digits individuate. Yet, the ensuing phase takes place normally on an earlier one that is defective. Compare this with the cranial sutures where the retardation of closure allows for the expansion of the brain. Closure does occur but at a later stage in development. Such anomalies indicate that a slowing of development does not obstruct the process but is a marker of that phase in development where the alteration occurs. The error is a signature of an upstream segment in growth.

The principle effect of a brain lesion is to retard process, not destroy function. The lesion induces a change in a configuration that is a type of traveling wave. If an act of cognition is conceived in relation to evolutionary and maturational growth, we can conceive of a symptom

as a kind of developmental error. The lesion exposes pre-processing phases. The effect of the lesion is like that of a rock in a stream, it delays or perturbs the flow but does not block it. The relation of symptoms to eddy currents and whirlpools has been explored mathematically by Ralph Hoffman (1987) in Hopfield simulations. The nature of the error is determined by the location of the disruption in language, perception or action. A disparity in the timing of a process that advances like a wave front leads to a local delay that is "out of synch" with concurrent parallel streams. Consider the effects of such a lesion on an error of naming.

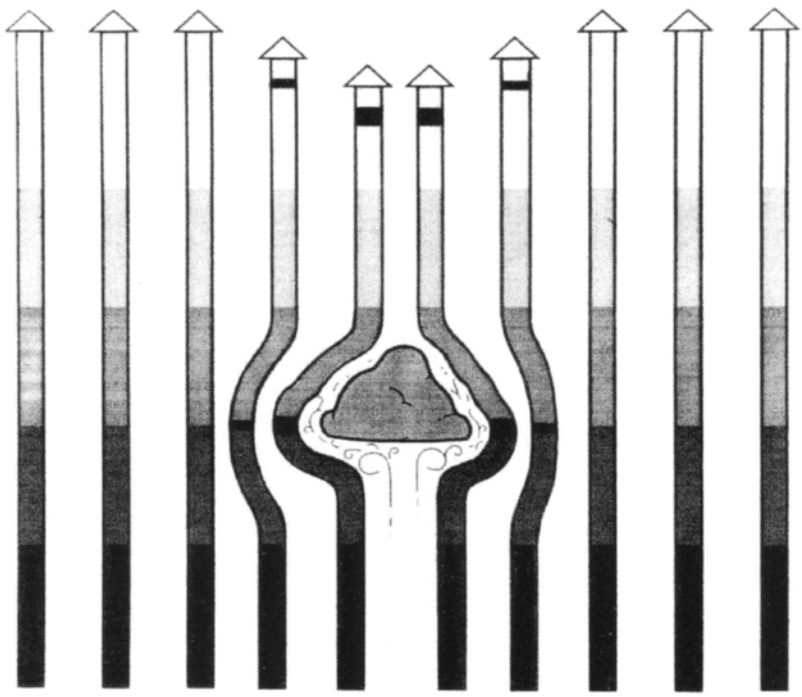

Figure 2. The lesion delays a segment of process (neoteny). The delay accompanies an incomplete specification (parcellation) of the lexical or phonological item. The neotenous form continues to a normal endpoint. The incomplete individuation of a lexical item undergoes normal subsequent phonological processing. A dissociation of lexical and phonological disruption can occur in a serial processing model

Take a patient with a left temporo-parietal lesion who calls a chair a table. How is this explained? I would say the process of lexical realization or the specification of word meaning develops to the object category, say that of furniture, where the potential to specify the word chair is equivalent to that for the word table. There is a momentary retardation at this phase that can be conceived as a brief neoteny. The result is an incomplete specification of the emerging content. The fractionation to the target item is prolonged at a phase just prior to full selection. If the word table, or the neural configuration corresponding to the word table, is selected, it then passes to an ensuing phase of phonological realization. The wrong word comes up but undergoes normal phonological encoding. Phases in phonological realization operate on a deviant semantic form. However, the word is not really deviant, it points to a phase where either chair or table might have been selected. Even the "correct" word, *chair,* might be considered an error. A holophrastic noun does not achieve the referential or denotative specificity of a fully individuated lexical item. I reported this years ago in studies on "fuzzy" semantic boundaries in posterior aphasics. The aphasic error, table for chair, results from a focal retardation in process (neoteny) and an incomplete specification (parcellation) of the lexical item. The fact that the derailment undergoes normal subsequent processing shows that double dissociation can occur in a serial model.

A brain lesion differs from a rock in a river in that unlike a river, where the current moves on, cognition is recurrent, more like a fountain, so that the disruption is encountered on each new traversal. This accounts for the variability of errors, as well as their regularity or clustering within distinct categories. The aphasic error samples the context at the phase of disruption. With errors of language development, the context is impoverished so errors tend to be simplifications that are more predictable. Contextual cues are less effective. A 3 years old is said to be at the stage of a normal 18 months old. *The difference between developmental errors in children and acquired errors in adults is neoteny in the initial formation of a system as opposed to a segmental delay in the reinstantiation of a system that is already formed.*

Given this concept of the error as a link between specification and timing, or the pattern of process and its rate, the regression hypothesis can be reformulated as follows. Microgeny does not retrace ontogeny, nor does pathology recapitulate stages in acquisition. It is not the stages that recur but the process leading to the stages. *The recapitulation is for the process, not the actualized elements it deposits.*

RECOVERY

Finally, a word on recovery in relation to this concept of the symptom. The first point is that

studies of linguistic change in restitution are vital to the documentation of the model, which predicts that symptoms will undergo a coherent rather than piecemeal transition. The finding that repetition in conduction aphasia improves in relation to comprehension defect and production pattern is an example of an observation that supports this formulation, one that is inconsistent with modular or mosaicist interpretations.

Second, if the symptom refers to a segment of preliminary process, one strategy in treatment might be to concentrate efforts on the just prior phase, or on constraining the damaged one, so as to facilitate a traversal of the disrupted segment. I think that hemiplegic writing, or noun activation by proximal motility, is an example of this effect (Papathanasiou, 2002). Lastly, those of you engaged in treating aphasics are in a position to test many of the postulates of microgenetic theory, for errors in therapy sessions are fragments of phases postulated in the pre-processing sequence. Some aspects of the theory can provide a motivation for research and a strategy for treatment. For example, the concept of a phase transition assumes generalization across neighboring, i.e. level-specific, linguistic or cognitive domains.

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SECTION 1

THE NEUROSCIENCES OF APHASIA

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Chapter 2

THE NEUROLOGY OF RECOVERY FROM STROKE

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INTRODUCTION

It is a fact that patients suffering from a stroke undergo some degree of functional recovery (Twitchell, 1951), the extent of which can up to a point be predicted. Variation occurs, implying that the outcome is influenced by many variables, and it is natural for the clinician to attempt to influence these variables to produce the best possible outcome. To date the cornerstone of restorative neurology in the field of stroke medicine has been good general medical care and more specific therapy (physiotherapy, occupational therapy, speech and language therapy), and this is of course still the case. However clinicians are starting to become aware of advances made in the basic sciences, which will add a new dimension to the treatment of functional deficits in patients suffering single insult brain injury.

EARLY RECOVERY: THE PRINCIPLES OF ACUTE STROKE TREATMENT

Physicians interested in stroke have an active approach to management aimed at reversing any remedial causes of stroke, preventing secondary damage to the brain and enhancing recovery (Brown, 2002). The concept of neuroprotection plays an important part in this approach to stroke. A large number of neuroprotective drugs have been tested in randomised trials in man but despite major benefits in animal models of stroke, none have proved effective in human stroke to date. However, thrombolysis using tissue plasminogen activator is beneficial in acute ischaemic stroke when given within 3 hours of stroke to suitable patients in specialised units (Lees, 2000). Unfortunately, only a small proportion of patients are eligible for

thrombolysis. Although we do not yet have any neuroprotective drugs that have been shown to be effective in acute ischaemic stroke, active management of acute stroke includes a number of measures to protect the brain from further ischaemia and extension of infarction. For example, systolic blood pressure can be maintained within a given range, antipyretic drugs given for fever and blood glucose maintained within the normal range. Nevertheless, despite these measures many patients with stroke continue to have significant disability after the acute period. Hence, enhancing recovery using modern rehabilitation techniques continues to play an essential part in stroke treatment.

Randomised clinical trials have provided good evidence that the best way to enhance recovery from stroke is to admit the patient to a stroke unit. These trials show that organised stroke unit care can produce significant benefits to patients in comparison with routine care on a general medical ward. Part of the benefit is due to a reduction in mortality from good quality care, but there is also a significant reduction in disability and handicap. The meta-analysis of the stroke unit trials has shown that for every 100 patients receiving stroke unit care, 3 additional patients survive, 3 avoid long term institutional care and an additional 6 return home compared to routine medical care (Stroke Unit Trialists' Collaboration, 2000). The benefits of stroke unit care are very similar in male and female patients and whatever the age of the patient or the severity of the stroke. These benefits are achieved not simply by gathering the patients together in a single location labeled "stroke unit", but by the development of a specialised multidisciplinary team. The characteristics of stroke unit care contributing to the benefit include nurses interested in stroke, a physician interested in stroke, multidisciplinary team meetings, regular staff training, the involvement of carers and the early onset of rehabilitation. Specialised nursing is particularly important, because nurses can practice the principles of active rehabilitation 24 hours of the day. Most of the benefits of stroke units accrue during the first 2 weeks after the onset of stroke, and it is likely that a number of measures contribute to the benefit e.g. the medical measures discussed above. Specialist assessment e.g. swallow assessment by a speech and language therapist, and prevention of complications of stroke, are particularly important. After the acute stage, it is likely that rehabilitation input from specialist therapists has an important role in improving recovery in the later stages.

LATE RECOVERY: ENHANCEMENT OF NEURONAL PLASTICITY?

Until recently it was felt that there was no evidence to support the effectiveness of physiotherapy for stroke patients in the post acute phase. A meta-analysis combining results of seven randomised controlled trials on the effects of different intensities of therapy after

stroke reported small but significant reductions in mortality and significant improvements in activities of daily living (ADL) scores as a result of higher intensity therapy (Langhorne *et al.*, 1996). More recently Kwakkel *et al.*, (1997) examined nine trials of physiotherapy in stroke patients involving 1051 patients, and concluded that there was a small but significant correlation between intensity of therapy and outcome. Furthermore, greater intensity of leg rehabilitation improves functional recovery and health-related functional status, whereas greater intensity of arm rehabilitation results in small improvements in hand dexterity (Kwakkel *et al.*, 1999). It is accepted that stroke units are beneficial in the management of stroke patients, but it is not clear which aspect of the service is beneficial. When considering physiotherapy, failure to demonstrate a beneficial outcome in the past is likely to be related to the fact that assessments were often made using global ADL scales, rather than parameters relating directly to the form of training being given. When considering recovery of motor function for example, a study involving 148 hemiparetic patients undergoing an intensive training period, demonstrated that gait symmetry (a principle goal of many physiotherapy programs) was unaffected. However, specific measurements of stance duration, weight acceptance, and push off of both legs improved significantly (Hesse *et al.*, 1994). Furthermore recovery of gait function in the severely hemiplegic patient using treadmill training, functional electrical stimulation, individually or in combination with each other, have been demonstrated to improve specific aspects of gait, indicating a significant task specific training effect (Hesse *et al.*, 1995).

It might be argued that the only truly relevant measurements are global functional assessment in the home environment. If however one wishes to investigate the mechanisms of recovery, it is important to know exactly what each component of the so called "black box" of a stroke unit is contributing to recovery. It is important to know, for example, what effects the motor and sensory experience of physiotherapy has on the motor system, as well as to know whether the patient becomes independent. These are two different questions, the former being concerned with the relationship between cortical plasticity and recovery. To focus on ADL's alone can also mask relevant information as in the case of recovery of hand function in a hemiplegic patient. We know that motor learning studies have demonstrated reduced skillfulness in the recovered hemiplegic patients, despite regaining the ability to solve simple spatial motor problems (Platz *et al.*, 1994). To begin to understand mechanisms of recovery in these patients, and potentially to be able to manipulate these mechanisms, one must look beyond using ADL's as an assessment. This is an important lesson in assessing any form of rehabilitative strategy, including treatment of aphasia, if we are to avoid discarding potentially useful treatments as a result of selecting poor or inappropriate outcome measures.

PLASTICITY IN THE NORMAL BRAIN

When considering mechanisms of recovery after focal brain injury, the term 'plasticity' is often used. In 1947, Hebb postulated that increments in synaptic efficacy occur during learning when firing of one neuron repeatedly produces firing in another neuron to which it is connected (Hebb, 1947). Hebb is suggesting that plasticity is a matter of synaptic efficacy, but it could just as well be brought about by changes in the number of synaptic connections. The key elements in a definition include a change in structure over time with a consequent change in function (Kolb, 1995). The cortex, with its myriad synaptic connections is the ideal site for this plasticity to take place (Sanes and Donoghue, 2000), and can occur in a number of ways. Firstly, it has been repeatedly demonstrated that enriched environments and motor learning in adult animals is associated with morphological changes in the cortex suggesting increases in the number of synaptic connections. These changes include growth of dendrites, increases in dendritic spines and synaptogenesis (Ivanco and Greenough, 2000; Bury *et al.*, 2000). Secondly, long term potentiation (LTP) and long term depression (LTD) have long been known about as mechanisms of changing synaptic efficacy, particularly in the hippocampus (Collingridge and Bliss, 1995). More recently there is evidence that these processes can occur in the neocortex if certain conditions, such as a concurrent ascending input, are in place (Hess *et al.*, 1996). Thirdly, the influence of one cortical neuron on another can be altered by factors other than environment or practice. Jacobs and Donoghue (1991) performed an experiment in which bicuculline (GABA antagonist) was applied to forelimb area of rat motor cortex. Stimulation of the adjacent cortex (normally representing the vibrissa) then lead to forelimb movements, suggesting cortical maps are maintained at least in part by GABA, and can be altered by manipulated pharmacology.

In the human brain very different techniques are used to study alterations in cortical function and neural networks. Studies using transcranial magnetic stimulation (TMS) have demonstrated changes in cortical function in response to sensory input (Hamdy *et al.*, 1998), motor imagery (Hashimoto and Rothwell, 1999), and motor practice (Pascual-Leone *et al.*, 2000), and functional imaging has been used to demonstrate changes in the organisation of neural networks during motor learning (Toni *et al.*, 1998; Karni *et al.*, 1995).

Work in both animals and humans suggests that the brain is not hardwired, and that plasticity, as previously defined, does occur, and that the cortex is the most likely site of such change. We need to consider whether plasticity also occurs in the damaged brain, and whether this process in any way contributes to functional recovery.

PLASTICITY IN THE LESIONED BRAIN

There is much evidence now to support the notion that the lesioned brain has an increased capacity for plastic change. Developmental proteins not normally expressed in the adult brain re-emerge in the hours and days following focal brain injury. These proteins are involved in changes in the extra-cellular matrix, structure of glial support cells, neuronal growth, apoptosis, angiogenesis and cellular differentiation (Cramer and Chopp, 2001). Structural changes have also been observed, with evidence of neurogenesis (Gould *et al.*, 1999), increased dendritic branching (Jones and Schallert, 1992), and synaptogenesis (Jones *et al.*, 1996). The correlation between some of these changes and behavioural recovery becomes clearer when considering that the magnitude and temporal course of cellular events often parallels this recovery (Cramer and Chopp, 2001). It is also interesting to consider the spatial distribution of such changes, with synaptogenesis (Stroemer *et al.*, 1998) and axonal outgrowth (Kawamata *et al.*, 1998) seen in perilesional tissue in rats, and evidence of dendritic branching in homotopic cortex in the non-lesioned hemisphere (Kozlowski *et al.*, 1996). The changes in cortical structure and function that might mediate recovery, may therefore occur at sites distant from the lesion.

The idea that intact areas of the brain become functionally and metabolically inactive because they are disconnected from the site of focal lesions (a phenomenon known as diaschisis), and that this might have an impact on not only the clinical presentation but also on recovery, was first discussed at the beginning of the last century by Von Monakow (Von Monakow, 1914). In a human PET study, survival of the metabolically active cortex surrounding an infarct correlated with neurological recovery in acute stroke patients (Furlan *et al.*, 1996), although the evidence that reversal of diaschisis (rather than recovery of stunned cells in the ischaemic penumbra) contributes to functional recovery is conflicting. Resolution of ipsilateral thalamocortical diaschisis seems to correlate with improvements in cognition and neglect (Baron *et al.*, 1992), but does not seem clearly related to motor outcome. Bowler *et al.*, (1995) performed SPECT scans in 50 unselected patients with cerebral infarcts at the time of infarct and 3 months later. They could not demonstrate that diaschisis independently added to the clinical deficit after stroke, and they found no correlation between recovery and reduction in diaschisis.

Widespread areas of hyperexcitability in cortex, distinct from the areas of metabolic depression, have been observed in several studies, both ipsilateral and contralateral to the lesion in animal models (Witte and Stoll, 1997). In animal models at least, the extent and time course of recovery of these areas of hyperexcitability does not correlate with the changes in metabolic diaschisis, so they are presumably distinct processes. It has been proposed that it is

these areas of hyperexcitability that develop in the first five days and partially reverse over months, that may be the substrate for use dependent plasticity (Witte, 1998). Hagemann *et al.*, (1998) demonstrated that the induction of long term potentiation (LTP) is facilitated in the surround of focal cortical infarcts in rats in vitro, associated with and probably mediated by reduced GABAergic inhibition and increased NMDA receptor mediated glutamate response. In this hyperexcitable cortex, inputs from neighbouring cortical neurones may become more efficacious, shifting the cortical map, but to take advantage of this hyperexcitable state and induce LTP is likely to require task related activation by training. That this is the mechanism of recovery of behavioural function is not established, but the link is tantalising.

There is also evidence of changes in the organisation of cortical networks following focal brain lesions in humans. Enlargement of cortical motor maps demonstrated with TMS correlates with functional improvement in stroke patients (Traversa *et al.*, 1997), and changes in activation maps post stroke have been demonstrated in both motor (Weiller *et al.*, 1993, Cramer *et al.*, 1997), and language studies (Warburton *et al.*, 1999). The controversies surrounding functional imaging studies in aphasics are discussed fully in chapter 4.

PROMOTING FUNCTIONAL RECOVERY AFTER STROKE

Given that the brain has the capacity for plastic change, and that this seems to increase in the lesioned brain as a consequence of expression of trophic factors and other changes previously discussed, how might we take advantage of such changes to influence outcome? A number of studies have demonstrated that these changes are dependent not only on the lesion, but on experiential demand (Schallert *et al.*, 2001). For example, in rats subjected to unilateral sensorimotor cortex damage, restraining of the impaired forelimb leads to reduced dendritic arborization in surrounding cortical tissue. This is not seen if the impaired limb continues to be used (Jones and Schallert, 1992). Animal studies have been supportive of the idea of task specific training effects in cortically injured subjects, and have begun to shed light on the underlying mechanisms. Nudo *et al.*, (1996), in an important study, trained Squirrel monkeys in the execution of a complex motor task using a hand. Focal infarcts of a small portion of the hand representation in cortex were induced and five days later intensive retraining identical to that previously applied was undertaken. This continued until pre-infarct levels of performance were attained. Using intracortical microstimulation techniques, they found that spared hand areas had either been preserved or had expanded into regions previously occupied by elbow and shoulder representations. Animals in whom retraining had not been attempted and in whom recovery had been less marked had lost remaining hand representation in the cortex. Rehabilitative training would therefore seem to have had an effect on reorganization of intact

cortex, as measured by changes in cortical maps, with a consequent beneficial effect on motor recovery. Exposure of animals to an enriched environment enhances dendritic growth and synapse formation (Schallert *et al.*, 2001), and has also been demonstrated to enhance post brain injury recovery (Ohlsson and Johansson, 1995). This effect is likely to be related to experience and consequent cognitive processing, as physical exercise on its own does not produce such significant results on post injury motor recovery (Gentile *et al.*, 1987). Parallels certainly exist in human studies, as has already been mentioned. In particular, improvements in motor performance in the chronic setting have been demonstrated with constraint induced therapy (CIT), based on overcoming learned non-use of the affected limb (Taub, 1993), and have been accompanied by increases in cortical representation of the affected limb (Liepert *et al.*, 2000). Other theoretically derived techniques are similarly under investigation (Whitall *et al.*, 2000). 'Constraint induced aphasia therapy', a technique in which chronic aphasia patients were constrained to systematically practice speech acts with which they had difficulty, has also shown some benefits. From the point of view of motor rehabilitation, there has been a reluctance to use these forced therapy approaches in the acute setting, because of evidence that such an approach could lead to exacerbation of cortical damage (Kozlowski *et al.*, 1996). This overuse-dependent exaggeration of injury can be blocked by administering MK-801 (NMDA antagonist), suggesting that glutaminergic mechanisms are involved. The question of timing of an intervention is clearly critical, as there appear to be definite change in the molecular and cellular environment at a certain time point after injury. For example, it has been hypothesized that during early development new synapses with a NMDA:AMPA receptor ratio are formed. This ratio rapidly becomes similar to that seen in adults. In any period where new synapses are forming, such as early development, new learning, or post focal brain damage (Stroemer *et al.*, 1998), use dependent plasticity is likely to occur in the new synapses with high NMDA:AMPA receptor ratios, which may explain why NMDA antagonists, although thought to be protective in acute ischaemia, may slow or prevent plastic changes occurring in perilesional tissue, with subsequent impairment of functional recovery (Barth *et al.*, 1990). Understanding of these processes is crucial if we are to use interventions correctly.

PHARMACOLOGICAL MANIPULATION OF RECOVERY AFTER STROKE

There has been recent interest in whether the processes collectively described as neuronal or synaptic plasticity can be influenced by pharmacological manipulation. As early as 1942, investigators concluded that the cholinergic drugs, strychnine and thiamine could enhance the rate and degree of recovery from motor cortex damage in monkeys (Ward and Kennard, 1942). However, by the 1950's it was felt that pharmacological stimulation of the reticular

activating system was the key to facilitating recovery. Amphetamine was first used in 1946 by Maling and Acheson (1946), who demonstrated that this drug transiently restored righting reflexes in low decerebrate cats, and subsequently Meyer *et al.*, (1963) temporarily restored the placing reflex in decorticate cats by the administration of amphetamine one year after they had originally undergone surgery. Faugier-Grimaud *et al.*, (1978) also demonstrated that a deficit could be temporarily reinduced after recovery had taken place. They performed either unilateral or bilateral parietal lesions in Java monkeys, inducing deficits in visually guided reaching tasks, after which spontaneous recovery took 2 weeks. One year after surgery the monkeys were given a small dose of the general anaesthetic, ketamine, which resulted in an immediate but temporary return of the deficit, with the deficit being unilateral or bilateral depending on the number of lesions performed one year previously.

Feeney and co-workers revisited amphetamine as a possible neuromodulator of functional recovery (Feeney *et al.*, 1982). A series of experiments were carried out on rats that had undergone suction ablation of the sensorimotor cortex. Rats are known to recover from such unilateral damage to the motor cortex in approximately two weeks (Maier, 1935). In one such experiment (Feeney *et al.*, 1982), 111 rats were operated on in this way, after they had been trained to run on a beam to avoid white noise and bright light. 24 hours post operatively, the animals each received intraperitoneal saline, amphetamine or haloperidol. Trials on the beam were then carried out at hourly intervals for the first six hours and then at 12 and 24 hours after drug administration. Trials were carried out on alternate days for a further 15 days or until animals had recovered their agility. Comparison was made with a similar group of rats in whom no practice trials were made. Animals given amphetamine (2 mg/kg) and practice made a recovery at 24 hours similar to that seen in the control rats at 1-2 weeks. Practice contributed to this effect only in the context of amphetamine administration, and vice versa. Haloperidol given with amphetamine blocked the effect, and given alone retarded recovery, suggesting a role for dopamine (DA) neurotransmission.

Further observations have implicated the cerebellum in this effect. Firstly, both amphetamine and haloperidol worsen beam walking recovery in rats with cerebellar injury (Boyeson and Feeney, 1991), and secondly microinfusions of noradrenaline (NA) into the cerebellum contralateral but not ipsilateral to the site of a cortical injury, mimics the systemic effects of drugs on beam walking recovery in rats (Boyeson and Krobert, 1992).

Although the effect of haloperidol in Feeney's initial experiments suggested that the effect was mediated by DA, further evidence implicates NA. Lesions to the contralateral but not ipsilateral dorsal noradrenergic bundle (projecting from the locus coeruleus to the cerebral cortex), impair motor recovery after subsequent cortical lesion (Goldstein and Bullman,

1997). Alpha-2 antagonists (increasing noradrenergic effect) such as yohimbine and idazoxan, have also been found to facilitate motor recovery in a single dose (Sutton and Feeney, 1992). Conversely drugs that decrease NA release in the CNS, or block post-synaptic effects (i.e. alpha-1 antagonists/alpha-2 agonists), are therefore likely to be harmful to recovery in the above model, and in fact clonidine (alpha-2 agonist) (Goldstein and Davis, 1990) impairs beam walking recovery, in the same way as haloperidol. In an echo of the work done by Faugier-Grimaud *et al* in 1978, it was noted that these drugs not only impair recovery, but if given to a rat that has made a spontaneous recovery, will reinstate the deficit temporarily, to a degree proportional to the original deficit. This effect has been seen with several drugs (clonidine, prazosin, phenoxybenzamine) and across species (Feeney, 1997).

Speculation as to the mechanism of action of these effects is fascinating. Noradrenergic induced changes in local metabolism or enhancement of LTP may allow motor experience to induce permanent changes. We have already discussed the evidence pointing towards how this may occur in relation to LTP (Hagemann *et al.*, 1998). It is also fascinating to note that the effects of neurotransmitters (and drugs that affect these neurotransmitters) on LTP induction, correlates strongly with their effects on recovery of function after sensorimotor cortex injury in animals (Goldstein,1990). More recently Stroemer and co-workers (1998) induced unilateral cortical ischaemia in a group of rats, and then treated one group with amphetamine and one with saline. Levels of GAP-43 and synaptophysin, as markers of neurite growth and synaptogenesis respectively, were measured in peri-infarct tissue at different intervals after the lesion was induced. Behavioural recovery was measured at the same time intervals. Levels of GAP-43 and synaptophysin were found to be significantly elevated and the degree of elevation correlated with behavioural recovery in a temporal fashion. It is tempting therefore to suggest that the increase in expression of these proteins promotes recovery directly, but it is known that these proteins are associated also with release of NA and DA (Dekker *et al.*, 1989), and possibly with LTP (Iwata *et al.*, 1997), both of which may be important factors themselves in functional recovery.

The fact that the recovery of function is so susceptible to reversal by pharmacological agents (e.g. ketamine, prazosin, clonidine), suggests that changes are not purely anatomical and that a change in the neurochemical balance of interacting systems has been effected. Meyer (1972) suggested that lesions in these animal models cause a suppression of retrieval of motor engrams, which have been formed in the brain as a result of previous learning and experience. Perhaps these motor engrams are not destroyed by cortical lesions but become inaccessible, and noradrenergic enhancement allows them to be accessed once more. This is of course speculation.

There is also evidence that modulation of other neurotransmitter systems can have similar effects. Dopamine has already been mentioned in relation to Feeney's work with haloperidol. Apomorphine reduces the severity of experimentally induced neglect from prefrontal injury, and spiroperidol reinstates this neglect (Feeney, 1997). GABA infused intracortically impairs beam walking recovery in rats (Brailowsky *et al.*, 1986), and diazepam impairs recovery of sensory asymmetry caused by unilateral damage to the anteromedial cortex in rats (Schallert *et al.*, 1986). Acetylcholine antagonist scopolamine interferes with recovery after motor cortex infarction in rats (De Ryck *et al.*, 1990), and in monkeys, cholinergic drugs increase the rate of recovery in animals with motor cortex lesions (Ward and Kennard, 1945). NMDA antagonist MK-801, has been found to be detrimental if given during the recovery period in rats (Barth *et al.*, 1990), which is in contrast to its proposed neuroprotective effect if given immediately after an infarct.

More recently, a greater understanding of the molecular and cellular events occurring post injury, has lead to attempts to manipulate them for the purposes of promoting functional recovery. Candidate compounds include osteogenic protein-1 (Ren *et al.*, 2000), brain-derived neurotrophic factor (Rossi *et al.*, 1999), fibroblast growth factor-2 (Kawamata *et al.*, 1998) and stem cell treatment (Kolb *et al.*, 1998).

In summary therefore, experiments in animals point most strongly towards the influence of neurotransmitter systems on functional recovery after focal brain injury. The effect is dose dependent, and the timing of administration is crucial, with the effect being dependent on close temporal linkage to behavioural experience.

What of attempts to translate these findings into promotion of functional recovery in humans? Much of the early work in this field has been done by Alexander Luria and colleagues in soldiers with head injuries sustained during the second world war (Luria, 1963; Luria *et al.*, 1963). They proposed two types of functional disturbance as a result of focal brain lesions. Firstly cell death, and secondly functional inhibition of intact neurons. They suggested that patients in whom the latter predominated might benefit from "removal of the diaschisis, restoration of synaptic conduction or to use another term, de-blocking". It was proposed that this could be achieved by the combination of two approaches. Firstly the administration of a pharmacological agent "capable of removing inhibition, modifying mediator metabolism, and restoring disturbed synaptic conduction", and secondly by methods of training which promote 'de-blocking', the essence of which is "that by means of various methods the level of excitability in certain functional systems is raised and the corresponding functions are 'de-inhibited'". The main de-blocking agents used by these investigators were anticholinesterases. In one such experiment, neostigmine was administered to a patient with a non penetrating

wound of the premotor area. Rhythmic tapping movements were recorded before and after neostigmine was given, and improvements of 'dynamic co-ordination' were noted after the drug, when none had been obtained with repeated training attempts prior to this experiment (Luria, 1963). Luria also made claims that the rate of recovery from aphasia could be increased, as long as the lesion was not in what he described as the 'primary speech areas', using galanthamine, (a specific, competitive, and reversible acetylcholinesterase inhibitor now under investigation for the treatment of dementia, was the drug of choice) (Luria, 1963).

It is surprising that despite this large body of work, further studies of pharmacological enhancement of recovery were not pursued further until nearly 40 years after Luria's original work was published. The first trial of noradrenergic enhancement coupled with physical therapy in human subjects was published in 1988 (Crisostomo *et al.*, 1988). In this experiment patients who had suffered from hemiplegic stroke, with no evidence of previous strokes, in whom the neurological status had been stable for at least 72 hours, were assessed over two days prior to treatment, using the Fugl-Meyer test of motor function. Patients were then randomized to receive either 10 mg amphetamine or placebo, 45 minutes prior to physiotherapy. Follow up assessment 24 hours after treatment indicated a 40% improvement from baseline scores compared to placebo. Small numbers of patients in this study (8 in total), make interpretation difficult, as the authors stated. A subsequent failure to replicate these findings by Borucki *et al.* (1992) were attributed to different experimental design, in particular a failure to schedule physiotherapy immediately after amphetamine treatment. Perhaps also significant, in view of the possible therapeutic window, was that treatment was not started until over a month after the stroke. More recent studies with d-amphetamine have had conflicting results (Walker-Batson *et al.*, 1995; Sonde *et al.*, 2001), but positive results have been published for daily doses of l-dopa (100mg) used in conjunction with physiotherapy (Scheidtmann *et al.*, 2001). Studies have initially focussed on motor recovery, most likely because of obvious parallels with animal studies, but a similar study looking at recovery in aphasia post stroke have been performed (Walker-Batson *et al.*, 2001). A comprehensive account of the pharmacotherapy of aphasia is given in chapter 3.

Following on from the idea that exogenously administered drugs may alter the balance of extracellular concentrations of various neurotransmitters, and that this might in turn have an effect on functional outcome following focal brain injury via an as yet undetermined mechanism, Goldstein *et al.* (1990) performed a retrospective analysis of the effect of drugs, (predicted from animal models to have a deleterious effect on motor outcome following focal brain injury) on outcome in stroke patients. Patients receiving phenytoin, benzodiazepines, or alpha-adrenergic antihypertensives at the time of stroke or shortly afterwards had poorer outcomes than the controls who did not receive any of these drugs. This was found to be the

case for a number of outcome measures, covering both activities of daily living and specific motor function, and 30 day recovery rates. These findings were replicated by the Acute Stroke Study Investigators (Goldstein, 1995), using a group of patients who were themselves the control group in a prospective acute interventional trial. 40% of these patients received one or a combination of drugs predicted to impair recovery, and were found to have poorer recovery as measured by a variety of measures. Similar findings have been published relating to the adverse effects of certain antihypertensives (Porch *et al.*, 1986). These analyses were retrospective and could not exclude the possibility that patients were given these drugs for medical reasons that themselves would predict poorer outcome, but this area of research clearly warrants further investigation.

Evidence therefore exists that certain drugs influence behavioural recovery in humans following focal brain injury. This has implications in as much as it suggests that more could be done to enhance recovery in these patients, and also that certain drugs are probably best avoided in these circumstances.

SUMMARY

Until recently it has been on the basis of intuition rather than evidence that physicians have recommended post-acute therapy for stroke patients. Evidence now exists that stroke units work, and that specific retraining techniques have measurable benefits. A wealth of evidence has been produced from work in animal models that the lesioned brain changes at a molecular, cellular and systems level, in a way that promotes experientially driven changes in synaptic structure and function. There is clearly a bias towards motor studies in animal models, but the principle of a post lesional plastic brain applies to any cortical function. The study of aphasia therapy will be equally invigorated and stimulated by these findings. The challenge is now to advance our understanding of the science of recovery after brain damage in humans, and crucially how we can use this information to promote recovery.

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Chapter 3

EVIDENCE FROM BASIC NEUROSCIENCE AND HUMAN STUDIES OF PHARMACOLOGIC THERAPY

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Aphasia is a major symptom of stroke and shares common neurophysiological and neurochemical features with other stroke consequences. Currently, there is considerable interest in applications from animal models for the development of science based approaches to stroke rehabilitation. Over the past 30 years there have been significant advances in knowledge regarding central nervous system plasticity and recovery of function in the basic science laboratory, yet there has been little application of this knowledge to rehabilitation practice. Consideration of physiologic events surrounding brain injury, manipulation of neurotransmitter systems and a fuller understanding of mechanisms of learning may in the future be combined to define a brain based approach to rehabilitation including aphasia.

Several laboratories are exploring post-stroke manipulations of brain plasticity following experimental injury in terms of the timing and type of intervention. This includes manipulation of central nervous system effects using pharmacologic agents to modify or accelerate recovery. This chapter will review: theories of neuroplasticity thought to underlie behavioral recovery, animal recovery studies which may have importance in the design of rehabilitation practice, and data from humans on pharmacologic therapy in the treatment of hemiplegia and aphasia.

CHANGING CONCEPTS OF RECOVERY OF FUNCTION

Contemporary theories of recovery of function after brain injury reflect changing concepts of

neural plasticity and specificity (Rauschecker, 1997; Juliano, 1998). From their work in neural plasticity Finger and Stein (1982) have proposed that recruitment of latent synapses or the release of previously suppressed pathways after damage to the dominant pathway may account for some of the recovery after central nervous system injury. Others refer to this concept as 'unmasking' (Wall and Egger, 1971; Merrill and Wall, 1972) or 'disinhibition' (Calford and Tweedal, 1988). Wall has suggested that neural plasticity is achieved by existing afferents becoming more effective rather than the development of novel synapses. The release of existing afferents is achieved by manipulation of the excitatory-inhibitory balance of cells. One of the mechanisms suggested by Wall to unmask existing cells is by the chemical transport of substances that change the excitation of central structures (Wall, 1988). These processes can take place from days to weeks post-injury.

THE USE DEPENDENT/LEARNING DEPENDENT MODEL

In a systematic series of experiments, Merzenich and his colleagues (Merzenich *et al.*, 1983, 1984, Jenkins *et al.*, 1990) have observed critical aspects of input during recovery which have strong implications for rehabilitation. Merzenich defines a type of reorganization following median nerve section and digit amputation in the monkey which is a dynamic self-organizing process resulting from use-dependent alteration of the neo-cortical field. An important aspect of this work is that it has shown that the type of input affects reorganization. Competitive advantage is given to those behaviours that arrive first to the to-be reorganized territory. Reorganization arises from alteration in synaptic effectiveness with intensive use over time. The time course of reorganization as defined by Merzenich occurs days to months post-injury. Other groups have suggested the term learning-dependence (Plautz *et al.*, 1995; Nudo *et al.*, 1997) for the activity dependent changes in brain reorganization following injury. Nudo and colleagues (1997) observed that motor maps are altered by motor skill acquisition not by repetitive use alone. Topographic plasticity coincided with the acquisition of new motor skills in intact animals or the reacquisition of motor skills in lesioned animals.

PHARMACOLOGIC MODULATION IN ANIMALS

Following stroke, catecholomines throughout the brain, brainstem, cerebellum and cerebrospinal fluid have been found to be reduced. Reductions in norepinephine are most prominent on the side of the lesion but are also reduced diffusely throughout the brain (Reding, 1998). The decreased availability of norepinephrine has led to a number of therapeutic studies in animals.

After experimental cortical lesions, administration of dextroamphetamine (which blocks re-uptake and enhances release of norepinephrine) produces significant improvements in motor recovery (Feeney *et al.*, 1982; Boyeson and Feeney, 1990) sensory-motor integration tasks (Hurwitz *et al.*, 1991) and binocular depth perception (Feeney and Hovda, 1985) in rats and cats. Importantly, the dextroamphetamine facilitated functional recovery has been found to be greater when drug treatment is paired with practice or training during the drug action period as compared with drug administration alone (Feeney *et al.*, 1982; Goldstein and Davis, 1990). The importance of norepinephrine mediation of recovery is supported by the fact that drugs which act as norepinephrine antagonists have reinstated motor deficits in animals (Boyeson *et al.*, 1993) and hindered recovery from aphasia in humans (Porch, Wyckes and Feeney, 1985).

CRITICAL TIMING WINDOWS/PHYSIOLOGICAL LIMITS

The critical time window after brain injury for initiation of use dependent practice or pharmacologic modulation is not known. In a small number of animals the time period for effective pharmacologic modulation of visual deficits did not extend beyond 90 days post injury (Feeney and Hovda, 1985). Recent motor recovery studies in animals without pharmacologic treatment suggest that very early intensive treatment (< 7 days post stroke) may be detrimental increasing both the deficit and the size of the lesion (Kowalski *et al.*, 1996; Risedal *et al.*, 1999). How this extends to humans is not clear.

Physiologic events following brain injury complicate the timing for administration of various agents. Drugs that are effective in the very acute or subacute period following injury may be ineffective or even detrimental at later recovery periods (Goldstein, 1998a). Theoretically, brain plasticity, once modulated pharmacologically or through use-dependent practice, may be increased and/or extended; however there are definite limits depending on the time post injury as well as the amount and site of tissue loss.

SUMMARY

Animal studies of recovery subsequent to experimental stroke provide a beginning science base which may influence rehabilitation practice. A number of studies suggests that the type of input may effect neural reorganization and that the timing of retraining or rehabilitation may be very important. This suggests treatment approaches can be either adaptive or maladaptive. Additionally, a number of studies provide evidence for efficacy of pharmacologic modulation to increase rate of recovery from a variety of deficits in cats and

rats. This model however, does not extend to all behaviours (Schmanke *et al.* 1996) or sites of brain damage (Mintz and Toner, 1986; Boyeson and Feeney, 1991). While acknowledging the limitations of animal models, there is a beginning recognition that the development of an adjunctive pharmacologic therapy to enhance recovery from stroke related deficits may have clinical application (Goldstein, 1998b; Gladstone and Black, 2000).

PHARMACOLOGIC TREATMENT IN HUMANS

Pharmacological treatment as an adjunct to traditional approaches to the treatment of stroke is not a new idea. There are clinical reports dating back over 60 years that suggest the use of various agents in the treatment of both hemiplegia and aphasia (Scicoulouff, 1934; Luria, 1969).

More recently, enhanced recovery from hemiplegia after occlusive stroke has been reported using noradrenergic agonists paired with physical therapy. Small pilot studies have explored drugs such as amphetamine (Crisostomo *et al.*, 1988; Walker-Batson *et al.*, 1995), methylphenidate (Grade *et al.*, 1999) and L-DOPA (Scheidtmann *et al.*, 2001).

Previous reports exploring the use of various drugs in the treatment of aphasia include a variety of agents. One early study included stroke patients treated with acetylcholine (Sciclounoff, 1934). Seventy hemiplegic/aphasic patients received intravenous acetycholine and were compared to 151 patients who received no drug, with treatment beginning 10 days or less of stroke. Patients showed improvement in both hemiplegia and aphasia. The aphasia aspect of this study does not provide methodological details or indicate how language change was measured. Sodium amytol was used in an early report by Linn (1947) with two patients. One patient showed improvement in language performance after the drug, which was ascribed to improved attention and energy. The second patient had markedly improved responses during drug infusion which were not maintained after the drug stopped. In a controlled clinical trial, Bergman and Green (1951) tried to replicate Linn's findings. A detailed battery of language and cognitive tests was administered to 27 individuals with aphasia before and after intravenous sodium amytal administration. Although the individuals with aphasia felt that their speech was more fluent during the drug infusion period, formal testing demonstrated no objective improvement. In a brief report, Benson (1970) described the use of amphetamine in the treatment of aphasia. This placebo-controlled study compared early (2-3 months after stroke) and late (> 6 months after stroke) treatment with 5 subjects in each group. Patients were given two daily doses of amphetamine (10mg and 5mg) for 2 months. Patients received intensive speech and language therapy 5 days per week during the study

period. A facilitative effect of amphetamine was found in the early but not late-treated patients. Darley (1977) and his associates reported a double-blind crossover study of 14 individuals with aphasia that compared methyphenidate with a benzodiazepine and placebo. The Porch Index of Communicative Abilities (PICA) was the dependent language measure and was administered 1 hour after drug or placebo administration. No direct speech/language therapy was provided in conjunction with drug administration and the time post onset of stroke was not specified. No drug-related effects on language were found.

Early efforts in pharmacologic modulation in the treatment of aphasia are difficult to interpret because of lack of adequate rationales for the use of specific drugs and the anecdotal nature of many reports. Additionally, many studies did not pair speech/language treatment during the active period of the drug action nor did they provide long-term follow-up with reliable measures of language change.

More recent efforts have explored dopamine agonists such as bromocriptine (Albert *et al*, 1988; MacLennon *et al.*, 1990; Gupta and Miloch, 1992; Gupta *et al.*, 1995; Sabe *et al.*, 1992, 1995) and nootrophic agents such Piracetam (Huber *et al.*, 1997; Kessler *et al.*, 2000). The results of the bromocriptine studies have been mixed. The primary limitations of all of the bromocriptine studies are that they were conducted in the chronic recovery period (from 1 to 4 years) and that language therapy was not tightly coupled with drug administration. The two placebo-controlled studies, pairing Piracetam with language therapy in the subacute recovery period, report positive results. However, in at least one of these studies (Huber *et al.*, 1997), the effects were not maintained after drug treatment was withdrawn.

Our group has explored the long-term effects of the noradrenergic agonist, dextroamphetamine in the treatment of aphasia. Single case (Walker-Batson *et al.*, 1990) and open label pilot studies (Walker-Batson *et al.*, 1992; Walker-Batson, 1998) were initiated to determine dosing and safety. The protocol we developed (adapted from an animal model, Hovda and Feeney, 1984) specifies that subjects receive 10 mg of dextroamphetamine 30 minutes before speech/language therapy alternating every third/fourth day for 10 sessions over a 5 week period. The Porch Index of Communicative Ability [PICA] (Porch, 1982) was used as the dependent language measure. A 15-percentile point shift on the PICA Overall score was set as a significant clinical difference to determine clinical change at the 1-week-off-drug assessment.

In a prospective, double-blind study of 21 aphasic patients we found a significant difference between groups after conclusion drug treatment phase with the greater gain in the amphetamine group (Walker-Batson *et al.*, 2001a). This difference was significant when

corrected for initial aphasia severity and age and was maintained at 6 months (Figure 1). By 1-week after drug treatment ended 83% of the amphetamine treated subjects compared to 22% of the placebo treated subjects had achieved the 15 percentile point clinical change that we had specified (Table 1).

Other studies, published in abstract form only, include use of this protocol in the chronic recovery period (> 60 days post stroke) (Walker-Batson, 1999) and extended dosing of amphetamine over 4 successive sessions for 10 weeks using physiological monitoring with fMRI (Walker-Batson *et al.*, 2001b). It is noteworthy that patients enrolled into this amphetamine protocol have suffered no adverse reactions and no differences in blood pressure and heart rate when compared to placebo patients (Smith *et al.*, 1999; Unwin and Walker-Batson, 2000).

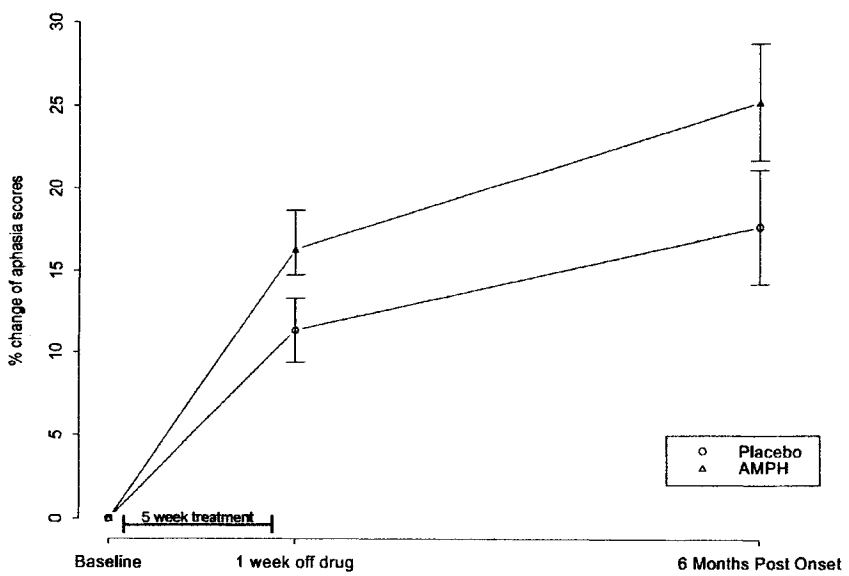

Figure 1. Mean ± S.E.M. percentile change in Overall PICA scores at 1 week off drug and 6 months post stroke onset for (amphetamine + ST) and (placebo + ST) groups. The one week off drug data point was taken one week after the last of 10 treatment sessions given at 3 or 4 day intervals over a 5 week period. Treatment was initiated between 16 and 45 days post stroke onset. (Walker-Batson *et al.*, 2001a)

Table 1. Main Results

	Placebo	AMPH	
PICA Overall mean gain score 1 week off drug	11.3	16.7	p=0.0153*
% achieved PICA 15 percentile point change 1 week off drug	22%	83%	p=0.0092**
PICA Overall mean gain score 6 month follow-up	17.7	25.2	p=0.0482* ns
Mean Speech/Language Therapy Hours 1 week off drug	27.22	33.45	p=0.3331*

* Two-sample t-test [Bonferroni Adjustment (alpha=0.025)], ** Fisher's Exact Test (Walker-Batson *et al.*, 2001a)

Our results thus far suggest that low-dose amphetamine accelerates the rate and in some patients the extent of aphasia recovery when entered in the subacute but not the chronic recovery period independent of initial language severity, age or lesion size (Walker-Batson *et al.*, 2001a). Our experience suggests that the timing post stroke of drug administration is very important. Unanswered questions include: How far post stroke can amphetamine be administered and have an effect? What is the dosage and the number of drug administrations needed to provide optimum recovery? Since dextroamphetamine is a potent releaser of neurotransmitters, it is possible that continuous administration would deplete the store of the neurotransmitter system (in particular norepinephrine) that needs to be stimulated to influence recovery rate. Thus, continuous drug administration could in fact have a diminishing return on therapeutic efficacy. Also unknown is the amount of use-dependent practice or re-training that must be paired with the pharmacologic intervention for optimal recovery.

In the future, pharmacologic modulation may prove to be an important adjunct when paired with behavioral therapy in the treatment of aphasia. Because of changing models of health care delivery, acceleration of rate of recovery from aphasia would have wide ranging application and clinical utility.

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Chapter 4

NEUROANATOMICAL SUBSTRATES OF RECOVERY OF FUNCTION IN APHASIA: TECHNIQUES AND EVIDENCE FROM NEUROPHYSIOLOGY

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In the last two decades, the description of language in aphasia has been predominantly based on the information processing models of normal language. However, these models are limited in describing the recovery mechanism involved. Recent advances in basic neurosciences and in neurophysiological techniques used to study brain structure and function have provided evidence how these recovery mechanisms operate. However, researchers are still setting the foundations to find the link between these mechanisms and therapy induced plasticity and subsequently recovery. This chapter aims to describe: a) the functional neuroanatomical mechanisms of recovery, b) the techniques available to study brain structure and brain function, and c) the current evidence from neurophysiological studies which supports these mechanisms of recovery.

WHICH ARE THE FUNCTIONAL NEUROANATOMICAL MECHANISMS?

In aphasia, evidence of plasticity is available not only from examination of the function of the synapse but also from examination of the brain's structure as a whole (Keefe, 1995). The physiological mechanisms refer to what happens at the cellular level to repair affected connections. (Papathanasiou & Whurr, 2000). The various theories describing functional neuroanatomical mechanisms of neocortex have been developed to provide an explanation for the reappearance of a "lost" function following brain lesion (Blomert, 1998). However today,

neuroscientists have established that the brain responds to damage by triggering complex processes which take place over long periods of time and that produce changes throughout the entire nervous system (Stein & Glazier, 1992). But the early theoretical accounts still provide a framework for the current research. These theories are described as:

- The resolution or regression of diachisis (Powell, 1981; Cappa, 1998): The initial diaschisis which is considered as an initial protective mechanism of the brain to the injury, like any other form of inhibition, tends to disappear, allowing for the brain to have several of its centres able to function again. According to Luria (1963), there is an established pattern to recovery from diachisis with physical and functional thresholds changing first, followed by thresholds of subjective sensation.
- Mass action theory also known as equipotentiality (Powell, 1981; Leonard, 1998): This theory states that any part of the brain is capable of subserving any function and that most functions are represented all over the cortex. This suggests that a function lost secondary to damage to a specific region of the nervous system can be mediated by a surviving structure or pathway. This view is not fully accepted at present.
- Redundancy (Powell, 1981; Blomert, 1998): The concept of redundancy can be referred to either at the level of neural processing or at a task level. At the neural processing level, redundancy theory assumes that uninjured neurons in the damaged area function as spare systems that can compensate for those that are non-functional. At a task level, it is assumed that a task might carry multiple cues, which are transmitted neurally. In case some of the cues become redundant, the task might still be completed by using compensatory strategies.
- Vicarious function theory (Blomert, 1998; Leonard, 1998): This theory suggests that another area of the brain, not previously involved in a particular function, takes over the function of the damaged areas. This might involve synapses that originally had a very weak input into the system, which after damage becomes the strongest available remaining input.
- Substitution theory (Powell, 1981; Leonard, 1998): This proposes that, behaviour can be performed by a different mechanism than that which originally controlled the behaviour. In other words, the end is achieved by different means, served by regions which were not involved before and now become active.
- Functional 'take-over' (Cappa, 1998): In the case of lateralized functions such as language, this theory suggests that functions can be taken over by the undamaged contralateral hemisphere. This is considered to be the 'unmasking' of a pre-existent functional commitment, inhibited by the left hemisphere (Moscovitch, 1977). Another possibility is that the functional takeover is the result of reorganization in the same hemisphere, as it is described in the vicarious function theory.

The role of the mechanisms proposed by these theories in the recovery of function is still controversial, as the mechanisms of recovery do not relate to these theories (Stein and Glazier, 1992). The gap between theories and mechanisms may be diminished by recent technological advances such as functional imaging and transcranial magnetic stimulation (TMS). Before we provide the evidence studies using these methods, the neurophysiological techniques available today are going to be described.

WHICH ARE THE TECHNIQUES AVAILABLE TO STUDY BRAIN STRUCTURE AND FUNCTION?

Today, the development of neurophysiological techniques has enabled scientists to study brains of live humans with a variety of different techniques rather than be restricted to studying the brains of live animals or deceased humans. The techniques available to use can be described under two major categories: the techniques used to study brain structure and the techniques used to study brain function.

Brain Structure

The structure of the brain can be studied by several imaging technologies. Mlcoch & Metter (2001) suggest that two types of neuroimaging methods have emerged to enable scientists to obtain an image of the brain: those that measure the transmission of energy through tissues such as x-rays and computed transmission tomography (CT), and those that produce images from natural or introduced energy sources, such as magnetic resonance imaging (MRI), positron emission tomography (PET), and single photon emission tomography (SPECT).

The X-rays are low wavelength rays in the light spectrum, which are invisible and are attenuated to different degrees by different kinds of tissue. The unabsorbed or transmitted through the brain x-rays are recorded by sensitivite film or video images devices (fluoroscopy). This provides a planar or two-dimensional image who has excellent spacial but poor contrast resolution (ie. the ability to distinguish between the gray matter and white matter).

The CT scans are high density X-rays in "slices" so can build up 3D picture of tissue eg brain. Within the brain, CT scan can distinguish structure, pathology and anatomy. Contrasts between structures depends on the amount of x-rays absorbed and the thickness, density, and atomic number of the structures. CT is capable of differentiating tissues with small absorption differences such as white and gray matter, cerebrospinal fluid and air passages.

Emission Tomographies produce images utilizing data from internal energy source. PET and SPECT are based on the technology of detecting gamma ray emissions from intravenously injected radioisotopes. Both these technologies are designed to measure functional changes such as cerebral blood flow (rCBF), metabolism and neurotransmitters. These methods are distinguished in the types of radiopharmaceuticals and equipment each employs. In PET the internal energy is radioactive isotopes with short half-lives in a substance such as water, which can cross the blood brain barrier (BBB). The radioactively labeled water is injected into a vein, then it travels in the blood to the brain where it crosses the BBB. In the brain, the O15 breaks down to the stable form, O12, emitting a positron. The positron collides with an electron and is annihilated, producing two gamma photons which send gamma rays in opposite directions. Then these are detected by an array of crystals surrounding the head. Where there is more blood flow, there are more positrons and hence more gamma rays are detected, thereby showing distribution of blood throughout the brain. The positrons inside the brain are located with excellent accuracy. In SPECT the isotopes used are injected also intravenously, but they differ from the ones used in PET as they have relatively long half lives and emit one gamma photon. The site which the photon was emitted is located by using a collimator. In SPECT, spacial resolution is less than any other imaging method.

Magnetic Resonance Imaging (MRI) is the most recent development in brain imaging. It does not utilize radioactive substances but examines the response of selected elements in response to a large magnetic field. It utilizes the behaviour of the nuclei as dipoles with very weak magnets. During this technique, the subjects head is placed in a scanner with a strong magnetic field. All the protons in each cell's nucleus will align to this field. A brief radiofrequency wave is applied which transiently shifts the magnetic field and hence the alignment of the protons. As the protons are realigned to the original magnetic field, they emit energy, which is used to construct a 3-D image of the brain. Different tissues emit different energies, so MRI provides a very sharp contrast between white and gray brain matter, making it particularly good for visualizing lesions.

Brain function

Brain function experiments may be designed to look at the activation corresponding to something, which consists of more than one psychological process. The theory of localisation of function approach makes it tempting to search for a relation of one area to one function relationship. However, even though there are areas specialized of particular types of processing, the brain works as a highly integrated system.

The techniques available to study brain function during action are based on different approaches. These approaches are:

- imaging brain during a "task" by looking at the blood flow
- recording the electric or magnetic activity of the brain during performance of a task
- stimulating brain neurons and examine the effects on behaviour.

Imaging Brain Function

PET and fMRI are task-dependent functional imaging techniques, which offer the potential to reveal areas of brain that are active during a task. The findings of studies using these techniques promise to expand our knowledge of the extent of areas involved in language processing and to allow further investigation of their roles in normal and impaired language processing. These techniques are offering improved spatial information about the underlying substrate of normal human cognition.

PET and fMRI differ in the specific requirements for experimental design and data analysis and have different spatial and temporal properties. However, both of them involve imaging that correlates neural activity during performance of a cognitive task in order to observe the particular areas of the brain that play the largest role in that cognitive task. The principle they are based on is that when a part of the brain is more active, its metabolism increases, and there is greater blood flow, to satisfy the increased demand for oxygen. Changes in blood flow/blood oxygenation reflect changes in neural activity over a population of neurons. Neurons may be excitatory or inhibitory and a population of neurons may contain some of each. Activation may therefore be increased activity in excitatory neurons or reduced activity in inhibitory ones. Or no activation may mean equal amounts of excitatory and inhibitory activity.

PET can give us a picture of the brains blood supply when the subject is at rest (not performing any particular task) versus when the subject performs a task of interest (eg reading). The patterns of positrons emitted are represented as a matrix of points called voxels. In order to see which voxels (and the corresponding points within the brain) are active during a task, we can compare activity within each particular voxel when the subject is at rest or is engaged in a task. However, one possible problem with comparing PET images of rest versus an active task such as listening to words is that the rest condition may reflect areas which are involved eg in internal thoughts that the subject is experiencing whilst at rest. Different experimental designs can be used to study human cognition using this technique. These are: a) a subtraction design, when in PET images are compared with a control condition to subtract

areas involved specifically to that task, b) parametric designs, when PET images compared when one task variable is altered, look for correlations of brain activity with this variable, and c) conjunction designs, when PET images compared across similar but different tasks, so areas which are activated in all suggest involvement of these areas in the aspect common to all the tasks.

The fMRI similar to structural MRI, is based on the notion that different tissues emit different energies when they realign to the magnetic field after the radiofrequency pulse has been applied. In the blood flow studies using fMRI, the same principle applies to different forms of blood: blood containing mainly oxyhaemoglobin (oxygenated form of blood which "gives up" oxygen on reaching the tissues) can be discriminated from blood containing mainly deoxyhaemoglobin (deoxygenated form of blood after oxygen has been extracted). Subsequently, regions of the brain in which the neurons are more active will receive an increased supply of oxygenated blood (containing oxyhemoglobin). The oxyhaemoglobin dilutes the deoxyhaemoglobin in the blood vessels surrounding the active brain region. The relative change in energy emitted from the active region due to the increased ratio of oxyhaemoglobin versus deoxyhaemoglobin (blood oxygen level dependent measure) is measured which is usually referred as the BOLD response. Like in PET imaging, BOLD is a relative measure, i.e patterns of BOLD response can be compared as subjects perform different tasks within the scanner. The experimental design and analysis in fMRI are similar to PET although a recent development in design is to use "event-related" rather than "blocked" design. In the former one, the task and the control task are interspersed, so it prevents the subject habituating to performance on a given task, which may result in brain activation effects being minimized.

Even PET and MRI imaging techniques are very popular in studies of brain function, they have some advantages and disadvantages. PET imaging is less susceptible to artifact during the course of imaging as subjects can talk in scanner and make motor responses without compromising the data and there are no restrictions on the equipment that can be used. In contrary fMRI is noisy and very claustrophobic for subjects, while the magnetic field imposes some restrictions in the equipment, which can be used. Also subjects with metals in their bodies such as pacemakers, artificial limbs, metal eyes etc cannot be used. Due to radioctivity involved in PET scanning, it can be used only with adults and for limited time, while no such limitations exist with fMRI scanning and the same person can be scanned several times. Also PET scanning involves an injection and has a limited block design, while in fMRI no injection is involved and you can perform even related studies and within subject designs. Furthermore the fMRI provides very good spatial resolution so can pinpoint brain regions more accurately.

Recording Electrical or Magnetic Activity

While PET and fMRI imaging techniques have a good spatial resolution and can indicate areas of brain that are active during, they cannot provide information how these areas act or interact over time, as their temporal resolution is very low. The temporal aspects are important when considering the speed with which language processing take place. Methods that are able to register the brain's activity in milliseconds domain are the electroencephalography (EEG) and magnetoencephalography (MEG). Both of these techniques are measuring the signal carried by neurons. EEG picks up the electrical potentials of this activity at the scalp, where MEG registers is magnetic field. The difference between these two techniques lies in the fact that the scalp recorded EEG signal can be affected by different structures through which the electrical potential has to travel through before it is picked up by the electrodes, whereas the scalp recorded magnetic field is unaffected by the intervening structures (Friederici, 2001). However, the distribution of activity measured at the surface of the scalp does not allow direct conclusions about the loci of the generator. So EEG cannot be directly used for localization studies, even some indirect conclusion regarding the loci of the activity can be drawn. In contrast, since magnetic signals are not distorted by structures surrounding the brain tissue, identification of the source of the signal with MEG is more straightforward.

Patterns of electrical activity time-locked to particular events, which reflect the summation of synchronous post-synaptic activity of large populations of neurons, are known as event-related brain potentials (ERPs). ERPs can be used with more complex experimental designs, similar to those used with PET or fMRI i.e alternate a cognitive task such as word reading with a control task and compare patterns of electrical activity associated with each. The patterns of brain activity recorded by ERPs are analyzed along four dimensions. The ERP can vary as a function of a particular cognitive process in its latency, its polarity, its size of the electrical response (amplitude), and its topological distribution. As it is an invasive technique it can be used with small children and even with newborn babies.

Stimulating the Brain

The neurophysiological techniques, which involve stimulation of the brain, are those of direct cortical electrical stimulation, where electrical interference is applied through electrodes resting directly on the cortex, and the transcranial magnetic stimulation (TMS) which operates on the principles of the electromagnetic induction. In TMS, electrical current is discharged into a stimulating coil, which is positioned on the scalp, over the cortical region of interest. Due to a process of electromagnetic induction, the current is converted into a magnetic field,

which in turn, induces a change in the electrical activity of the neurons at the surface of the cortex underlying the coil.

According to Gordon *et al.*, (2001), there are now two general methods for direct cortical electrical stimulation: the intraoperative and the extraoperative. Intraoperative stimulation is done at the time of the surgery, allowing the direct visualization of the sites of stimulation and the location of the electrodes. The limitations of this method are that the subjects might not be most alert for the testing and the areas that can be stimulated are limited to those exposed with the craniotomy. In the extraoperative method, electrodes are implanted via a craniotomy and patients can be tested following the recovery from the procedure using these implanted electrodes while awake. However, this method does not allow independent movement of the electrodes sites.

Transcranial Magnetic Stimulation is a technique developed in the 1980's for non-invasively stimulating the cortex in neurologically normal subjects. During the stimulation magnetic currents pass unattenuated through scalp and skull so can be used without exposing the cortical surface and since the current is not converted to an electrical one until it has passed through the skull, pain receptors are not stimulated in the scalp. The application of TMS to the skull has the effect of stimulating neuronal activity, and thus of disorganizing the functioning of the underlying cortex. For example, when used over primary visual cortex, can induce phosphenes (brief flashes of light in the absence of any external stimulus) while over the motor cortex it causes activation of specific muscle groups. Two modes of stimulation are widely used to study brain function: single pulse TMS, in which a brief pulse usually of large amplitude is applied at a particular time during a performance of a task, and repetitive pulse TMS, in which small magnetic fields may be applied over a period of several hundred milliseconds at a rate of up to 50 Hz (Walsh, 1998). Repetitive TMS can be applied for up to a second while the subject performs the cognitive task of interest. Behavioural effects (reaction time, error rates) can be compared when the subject receives TMS to when the subject receives no TMS or TMS over a cortical area which is not thought to be involved in the task. In this mode, it can be viewed as a "lesion" technique since it is causing synchronous activity of a population of neurons, which would otherwise be firing in a highly specific and non-synchronous way. Hence, it is a "lesion" technique because you are preventing normal neuronal activity. All these temporal "lesions" are transient and reversible. Finally, TMS also provides cortical maps of neuronal function and can highlight specific pathways and connectivities within a functional system (Pascual-Leone & Meador, 1998).

WHAT IS THE EVIDENCE FOR FUNCTIONAL NEUROANATOMICAL MECHANISMS INVOLVED IN RECOVERY OF FUNCTION?

Today, data from animal studies, brain imaging techniques, and neurophysiological techniques such as TMS provide some empirical evidence for functional neuroanatomical mechanisms involved in the recovery of function.

Animal studies

Keefe (1995) gives an account of animal studies that indicate changes in the structure of the nervous system. These changes are described as an increase in the dendritic branching and functional reorganisation of the cortical maps. First, dendritic branching is influenced by rich environmental stimulating tasks, which indicates that learning or environment-induced changes in cortical structure are highly associated with the specific task demands. Also, dendritic branching of neurons is influenced by central system damage. In animal studies, the functional reorganisation of cortical maps is observed following behavioural intervention and is associated with changes in both the ipsilateral and contralateral areas adjacent to the lesioned brain regions. However, these changes in nervous system networks could be limited by the nature and extent of the existing sensory connections and their association to primary and secondary cortical areas. Similar mechanisms of functional reorganisation are observed under the influence of stimulating environmental tasks (Keefe 1995). Similar cortical reorganisation in animals, as reflected in alterations in cortical maps, is seen following cortical injury. In the site of the lesion, the size of the receptive field of remaining neurons become larger and the lesion affects more distant cortical areas, and in particular, the secondary areas of representation which are related to long term recovery of function. The parameters determining the nature of this aspect of reorganisation are not known.

Functional imaging studies

The brain imaging studies provide functional anatomical information by using techniques such as positron emission tomography (PET) and functional magnetic resonance imaging (fMRI). Cappa (1998, 2000) gives an account of the studies of the mechanism of diaschisis using PET. He suggests that the presence of diaschisis has been related to the severity of the clinical picture following the acute period after the lesion. In a series of studies, Metter *et al.*, (1992) assessed regional cerebral metabolic abnormalities in structurally affected and unaffected areas in aphasic patients. They found a significant positive correlation between changes in left and right temporo-parietal glucose metabolism and changes in auditory

comprehension. Heis *et al.*, (1993) reported that the level of glucose metabolism in the left hemisphere outside the infarcted area in the acute stage was the best predictor for recovery of auditory comprehension after 4 months, suggesting the importance of intrahemispheric diaschisis in the recovery of function. Also in the same study, they reported that in a subgroup of mild aphasic patients, metabolic values in the infarcted temporo-parietal area, left Broca's area and contralateral mirror to the lesion area were predictive of the recovery of auditory comprehension, which indicates the possibility of extensive bilateral contribution of brain areas to the recovery of function. Cappa *et al.*, (1997) examined the changes in functional deactivation during the recovery of the lesion in the cortical language areas by measuring regional glucose metabolism. The study showed that even 6 months post-onset, metabolic depression, which correlates with functional deactivation, was present in structurally unaffected areas both ipsilateral and contralateral to the lesion site. In addition, an association was found between the deactivation in the unaffected areas connected to the lesion site and the recovery observed. Recently, Price *et al.*, (2001) used functional neuroimaging to investigate how lesions to the Broca's area impair neuronal responses in remote undamaged cortical areas. In this study four patients with speech output problems but relatively intact comprehension, were scanned while viewing words relative to consonant letter strings. They suggested that the presence of dynamic diaschisis, the anatomically remote and context-sensitive effects of focal brain lesions, reveals abnormalities of functional integration that may have implications for neuropsychological interference and cognitive rehabilitation.

Cortical functional reorganisation in both hemispheres, has been reported in a number of functional imaging studies investigating regional cerebral activation in recovered and non-recovered aphasics while engaged in linguistic and non linguistic tasks which allows more direct investigation of functional recovery after acute damage. Assuming that the damaged area can not support function, there are three potential locations of recovery: peri-infarct, in a distal brain region, and in the homologous contra hemispheric region. However, the exact nature of this will vary with the size and location of an infarct.

Regarding the potential role of the peri-infarct regions, some studies have shown peri-infarct activation in recovered aphasics - evidence that brain regions around the edge of the infarct have taken over the function. In their study of six aphasic patients, Warburton *et al.*, (1999) found that recovery of function was related to peri-infarct activation with no activation of the right hemisphere suggesting that recovery of language depends on the viability of peri-infarct tissue and the premordid lateralisation of language. The amount of peri-infarct activation has been claimed to be a predictor of amount of recovery (Karbe *et al.*, 1998), but this might reflect differing initial damage. Peri-infarct activation can be due to some surviving tissue, rather than actual re-organisation.

Mummery *et al.*, (1999) demonstrated that the right superior temporal gyrus (STG) and superior temporal sulcus (STS) are involved in 'normal' passive speech perception. They studied neural activity during speech perception in six normal subjects and two aphasic patients with left temporal infarction. In this study the patients (EJ and SS), performed well on single word comprehension tasks, though EJ was distinctly better than SS who could understand single words. Both showed extensive right temporal lobe activations when listening to speech, no peri-infarct activation in EJ and only a small residual region of the medial left dorsal temporal lobe responded in SS. Since both aphasic patients had mature lesions, the assumption was that vascular reactivity around the infarct had returned: that is, peri-infarct activation would have been observed if it had been present. Furthermore, in SS activation was observed in the most medial part of the left dorsal temporal lobe, the only residue of superior temporal cortex left after his stroke, and this was directly adjacent to the infarct. This confirms a role of the homologous in recovery from aphasic stroke regions, which in this study is the right STG/STS. In the same study, in addition to extensive right STG/STS activation to speech, EJ also showed right prefrontal activation when listening to speech. This was not seen in the controls or in SS. Strikingly, EJ's recovery is much better than SS's, and she seems to have recruited some prefrontal, 'executive' brain regions in comprehending speech. The involvement of anterior brain regions in recovery from stroke and brain injury is becoming clearer. The role of the prefrontal cortex is generally in aspects of controlled behaviour that involves decisions, attention and the use of rehabilitation strategies.

Weiller *et al.*, (1995) investigated changes in the organisation of the brain after recovery from aphasia by measuring increases in regional cerebral blood flow (rCBF) during repetition of pseudowords and during verb generation. Six right-handed patients who had recovered from Wernicke's aphasia caused by an infarction destroying the left posterior perisylvian language zone were compared with 6 healthy, right-handed volunteers. In the control subjects, strong rCBF increases were found in the left hemisphere in the posterior part of the superior and middle temporal gyrus (around Wernicke's area) in both tasks. During the generation task activation was greater in lateral prefrontal cortex (LPFC) and in inferior frontal gyrus (Broca's area). There were some weak right hemisphere increases in superior temporal gyrus and inferior premotor cortex. In the patients, rCBF increases were preserved in the frontal areas. There was clear right hemisphere activation in superior temporal gyrus and inferior premotor and lateral prefrontal cortices, homotopic to the left hemisphere language zones. Increased left frontal and right perisylvian activity in patients with persisting destruction of Wernicke's area emphasizes redistribution of activity within the framework of a pre-existing, parallel processing and bilateral network as the central mechanism in functional reorganisation of the language system after stroke. This study shows that, with a more complex task, the sensory processing of the input has possibly transferred to the right hemisphere (or has been taken

over entirely by the right STG/STS). However the more frontal regions involved in the generation of language, which are distal to the stroke and thus not directly compromised, do not re-lateralise. Similarly, Buckner *et al.*, (1996) also reported a right lateralised prefrontal response in aphasic patients during a word stem completion task, suggesting recruitment of a brain compensatory pathway. Frackowiak (1997) reported two further studies of language recovery in aphasic patients compared with normals, one investigating non-word repetition and verb generation and the other reading. He observed bilateral language activation not only in their recovered subjects but also in the controls when they were viewing words, which indicates the contribution of the right hemisphere in some language functions.

Small *et al.*, (1998) in a study of a patient's reading before and after therapy with fMRI reported similar intrahemispheric reorganisation, which supports this notion of altering brain physiology following rehabilitation therapy. Recently another two studies (Calvert *et al.*, 2000; Gold and Kertesz, 2000), which examined patterns of the brain reorganisation in aphasia, suggest that the areas of activation depend both on the different processing strategies the patients are recruiting within the damaged hemisphere as well as the unmasking of functions in homologous areas in the other hemisphere. Gonzalez Rothi (2001) distinguishes these two patterns of reorganisation as two different processes of recovery. She suggests that the recovery of function accomplished by reconstruction involving portions of its formal structure is restitutive recovery, while recovery accomplished in a manner different to its former functional structures is substitutive recovery. Overall, these findings suggest considerable evidence, that there are long lasting changes in the patterns of cerebral activation, which are related to clinically observable behavioural changes and vice versa.

Further clues about possible mechanisms of brain plasticity after stroke are provided by Chollet *et al.*, (1992). They studied patients who had recovered from striatocapsular infarcts with PET, and found that movements with the recovered arm were associated with bilateral increases of regional cerebral blood flow in premotor areas, parietal and prefrontal cortices. The bilateral activation suggests that recruitment of ipsilateral pathways in addition to contralateral pathways may play a role in recovery of function after stroke.

TMS studies

Further evidence of the mechanisms of recovery and plasticity have started to emerge from the increased use of non-invasive neurophysiological techniques not only in experimental but also in clinical studies. Transcranial Magnetic Stimulation (TMS) is a technique, which can provide insight into areas that are essential for performance and the timing of information

processing within a neuronal system. Cohen *et al.*, (1998) described the different ways in which TMS can be used to identify patterns of reorganisation of function, the mechanisms involved in cortical plasticity and the relevance of these patterns to behavioural changes. Pascuel-Leone *et al.*, (1993) used TMS to study plasticity in motor cortex in blind subjects. They reported that the motor cortical representation corresponding to the reading finger of blind subjects proficient in reading Braille is larger than the representation of the same finger in the non reading hand. Interestingly, this effect was more marked at the end of a complete day of reading than following a day during which subjects did not read Braille. They suggested practice can result in plasticity related changes in the motor cortex but this plasticity is use- and disuse- dependent. In other studies, TMS has been used to investigate the plasticity associated with implicit and explicit knowledge (Pascuel-Leone *et al.*, 1994), skill acquisition (Pascuel-Leone *et al.*, 1995) and practice (Classen *et al.*, 1998).

Turton *et al.*, (1996) used TMS to investigate hand motor function recovery in stroke patients. They found that TMS over the undamaged hemisphere was more able to elicit responses in the muscles of the ipsilateral hand in the patients that recovered poorly after the stroke than in the patients that managed to recover their hand functions to a much higher level. Moreover, in the same group of poorly recovered patients, they were able to elicit responses in the distal muscles more easily than it has been previously shown to be the case in the healthy people (Carr *et al.*, 1994). These findings further suggest that the easier elicitation of responses in the distal muscles following ipsilateral stimulation might relate to the importance of ipsilateral pathways for any attempts for recovery of hand function in the poorly recovered chronic stroke patients. Liepert *et al.*, (2001) reported that cortical reorganization can happen following appropriate rehabilitation therapy. In this study, patients showed an increased cortical representation area of the affected hand muscle in the contralateral side following a period of constraint induced movement therapy, where they were encouraged to use the paretic limb. This increased cortical excitability was accompanied by a significant improvement in dexterity. This approach has been developed over the years from a combination of basic neurosciences and small clinical studies (Ostendorf and Wolf, 1981; Taub *et al.*, 1993; Liebert *et al.*, 1998; Blanton and Wolf, 1999; Kopp *et al.*, 1999; Liepert *et al.*, 2000).

Epstein (1998), in an overview of the current experimental use of TMS in language function, reports that TMS is used to interfere with language expression, comprehension and verbal recall by creating temporarily reversible 'lesions'. It can also occasionally facilitate language-related functions and motor aspects of language. Topper *et al.*, (1998) used TMS to study the effect of the proximal muscle area of the motor cortex and the Wernicke's area on picture naming latencies in normal subjects. They reported that TMS over Wernicke's area decreases

picture-naming latencies when TMS presented picture presentation by 500 or 100ms. They suggest that focal magnetic stimulation can facilitate lexical processes due to a general pre-activation of language related neuronal networks when delivered over Wernicke's area. Tokimura *et al.*, (1996) demonstrated lateralised speech effects on the excitability of cortical arm areas in healthy subjects. They reported that reading aloud produced a significant increase in the size of responses evoked in the distal FDI muscle of the dominant hand but had no or a smaller effect on the non-dominant hand. Papathanasiou *et al.*, (in preparation) have used TMS to study the recovery patterns in aphasic agraphic hemiplegic patients following writing therapy with a writing device. They reported that therapy induced recruitment of ipsilateral pathways, which were related to the improved hand function. The mechanisms of reorganization and functional take-over were considered responsible for the plasticity observed.

CONCLUSION

In recent years, the available neurophysiological techniques have enabled scientists to study the mechanisms of recovery in aphasia. However, the use of these techniques in therapy lead recovery studies has been very limited. Future therapy studies should use these techniques, as they will enable us to understand the neuroscientific basis of therapies used in clinical practice. This will lead to delivery of therapy, which will induce plastic changes in the brain, and subsequently, it will facilitate better behavioural recovery, which will improve the quality of life of the patients.

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Chapter 5

SUBCORTICAL APHASIA: EVIDENCE FROM STEREOTACTIC SURGICAL LESIONS

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INTRODUCTION

Although the cerebral cortex has traditionally been considered the neural substrate of language, over the past two decades this traditional view has been challenged by the findings of a proliferating number of clinico-neurological correlation studies that have noted the occurrence of language disorders in association with apparently subcortical lesions. In particular, the introduction in recent decades of new neuroradiological methods for lesion localization *in vivo*, including computed tomography (CT) and more recently magnetic resonance imaging (MRI), has led to an increasing number of reports in the literature of aphasia following apparently purely subcortical lesions involving the striato-capsular region and/or thalamus. Consequently, in recent years, there has been growing acceptance of a role for subcortical structures such as the globus pallidus and thalamus in language processing.

One outcome of the reported clinico-neuroradiological correlation studies has been the development of a number of theories regarding the function of subcortical structures in language. These theories, largely developed on the basis of speech and language data collected from subjects who have had cerebrovascular accidents involving the thalamus or striatocapsular region, have been expressed as neuroanatomically-based language models – two of the most influential models developed include those developed by Crosson (1985) and Wallesch and Papagno (1988).

Crosson (1985) proposed that subcortical structures such as the globus pallidus and thalamus

participate in language via a cortico-striato-pallido-thalamo-cortical loop. In essence, according to his model, language formulated in the anterior language centre is relayed to the posterior language area, via the thalamus, for verification of semantic content. Once language segments have been verified for semantic accuracy, the temporoparietal cortex releases the caudate nucleus from inhibition. The caudate nucleus then serves to weaken inhibitory pallidal regulation of thalamic excitatory outputs in the anterior language centre, which in turn, arouses the cortex to enable the generation of motor programs for semantically verified language segments. Crosson (1985) hypothesised that lesions of the globus pallidus would result in disinhibition of the thalamus and hence, hyperarousal of the anterior language centre, leading to the production of extraneous verbal output, including semantic paraphasias. On the other hand, lesions in the thalamus would lead to disruption of the arousal of the cortex and thereby disturb the process of pre-verbal semantic monitoring. It was suggested that the loss of spontaneous speech noted after thalamic lesions may be due to the interruption of excitatory input from the thalamus to the cortex. Further, thalamic lesions would, according to this model, interrupt the transfer of information between the anterior and posterior language centres, disrupting preverbal semantic monitoring leading to poor monitoring of the semantic content of language, characterised by the production of semantic paraphasias.

The original conception of the response-release mechanism proposed by Crosson (1985) has since been revised and elaborated in terms of the neural substrates involved (Crosson 1982a, b). These revisions were based on the observation that excitatory fibres from anterior and posterior cortical regions project onto adjacent areas of the head of the caudate nucleus, which in turn project to the same dendritic fields in the globus pallidus via inhibitory fibres. Further, the striatum was assumed to convert patterned neural input from the cortex into quantitative output, based on evidence from stimulation and lesions of the cortex and striatum. The actual response-release mechanism resembles the original conception, however, the route for this release is altered. The formulation of a language segment causes frontal excitation of the caudate which increases inhibition of specific fields within the globus pallidus, however, this level of inhibition alone is not sufficient to alter pallidal output to the thalamus. An increase in posterior language cortex excitation of the caudate, which occurs once a language segment has been semantically verified posteriorly, provides a boost to the inhibition of the pallidum. The pallidal summation of this anterior and posterior inhibitory input allows for the release of the ventral anterior thalamus from inhibition by the globus pallidus, causing the thalamic excitation of the frontal language cortex required to trigger the release of the language segment for motor programming.

According to this revised model, lesions of the dominant caudate nucleus would lead to nonfluent aphasia as the globus pallidus is released from inhibition, leading to chronic

thalamic inhibition. Lesions of the globus pallidus itself would cause semantic paraphasias and fluent speech as the ventral anterior thalamus would be released from pallidal inhibition, allowing the release of language segments prior to semantic monitoring. The revised model provides an integrated account of how the basal ganglia might influence language output through a neuroregulatory mechanism which is consistent with knowledge of cortical-subcortical neurotransmitter systems and structural features. It is also compatible with the results of electrical stimulation studies, and considers the issue of patterned versus quantitative neural output. One of the major drawbacks of this model is determining how it may be verified or refuted empirically. Although nonfluent output and semantic paraphasias have been documented following nervous system lesions, these features have not always been strictly associated with caudate nucleus and globus pallidus lesions respectively and claims that there is no consistent syndrome of language deficits in this population are even more difficult to reconcile with this theory. The use of data from nervous system vascular lesions to test the cogency of this model appears somewhat inappropriate, however, given that such lesions typically involve multiple structures and may compromise the input or output of given nuclei, depending on lesion location, with the present model predicting different behavioural patterns on the basis of these fine distinctions.

Wallesch and Papagno's (1988) model also proposed that subcortical structures participate in language processes via a cortico-striato-pallido-thalamo-loop. They postulated that the subcortical components of the aforementioned loop constituted a "frontal lobe system" comprised of parallel modules with integrative and decision making capabilities rather than the regulatory function proposed in Crosson's (1985) model. Specifically, the basal ganglia system and thalamus were hypothesised to process situational as well as goal-directed constraints and lexical information from the frontal cortex and posterior language area, and to subsequently participate in the process of determining the most appropriate lexical item, from a range of alternatives, for verbal production. The most appropriate lexical alternative is then released by the thalamus for processing by the frontal cortex and programming as speech. Cortical processing of selected lexical alternatives is made possible by inhibitory influences of the globus pallidus upon a thalamic gating mechanism. The most appropriate lexical alternative has an inhibitory effect on the thalamus, promoting closure of the thalamic gate, resulting in activation of the cerebral cortex and the production of the desired response. Cortical processing of subordinate alternatives is suppressed as a consequence of pallidal disinhibition of the thalamus, and the inhibition of cortical activity. Wallesch and Papagno (1988) hypothesised that lesions of the globus pallidus would result in characteristics of non-fluent language pathology (eg. difficulty initiating speech). They suggested that, in the case of such lesions, the thalamus is disinhibited so that the thalamic gate is opened, the cortex is inhibited, and no language (or more difficulty initiating language) is produced. Indeed, there

have been several reports of transcortical motor-type aphasia subsequent to pallidal lesions (Brunner *et al.*, 1982; Damasio *et al.*, 1982), although this is not always the case. Further, they suggested that lesions in the thalamus may lead to the release of inappropriate, poorly monitored responses by the frontal cortex (due to disinhibition of the cortex). In such a lesion, the thalamus may receive inhibitory input from the globus pallidus in order to produce a response, but due to the thalamic lesion, all gates in the thalamus are permanently closed. The cortex is disinhibited and all parallel circuits may arrive at the cortex for a response. Unfortunately, the first response completing the circuit and reaching the cortex will be produced (possible semantic paraphasia) rather than the correct response.

The observation that mainly lexical functions which may be considered to involve a large number of degrees of freedom (eg. lexical selection in spontaneous speech) are often disturbed in this population, while syntax and repetition is often spared, lends some credence to Wallesch and Papagno's (1988) model. Finally, it has been suggested that a more marked and persistent impairment in language function might be expected following subcortical lesions if the basal ganglia did play such an intrinsic role in information processing related to lexical production (Crosson *et al.*, 1997). Wallesch and Papagno (1988) did, however, emphasise the possibility that lexical gating might not be carried out exclusively by the cortico-striato-pallido-thalamo-cortical loop, allowing for the possibility that other neural systems could subsume this role in the event of disruption by vascular or degenerative damage.

More recently, Wallesch (1997) cautioned that the original unidirectional loop which underpinned this theory may have been oversimplified. It is acknowledged that cortical projections probably do not directly interact at the striatal level, but instead remain as segregated "miniloops" which are subcortically modulated (Parent and Hazrati, 1993, 1995). The suggested segregated nature of these circuits casts doubt upon the hypothesised integrative function of the striatum in bringing together external and internal constraints from diverse cortical regions to influence the selection of lexical items. Wallesch (1999) argued that the basal ganglia may still influence cortical language processing, possibly through the time domain synchronizing of linked cortical outputs. The basal ganglia may be involved in the parallel processing of different miniloops which subserve cortical modules possibly through some modulatory function involving the reconfiguration of cortical activation. Like the prefrontal lobes, this subcortical system would still be involved in situations involving many degrees of freedom, and damage to this system may result in either 1) erroneous responses (ie. semantic paraphasias), 2) an increase in the level of activation required for response release (ie. transcortical motor-type aphasia), or 3) an increased dependency upon external constraints (ie. propositional aphasia) (Wallesch, 1997). It is not clear at present

whether these revisions constitute a fundamental shift from an information processing role to a neuroregulatory or neuromodulatory role for the basal ganglia in language functioning.

Although several theories of subcortical language function have been postulated, our understanding of the role of structures such as the globus pallidus and thalamus in language remains poor. From this perspective, Kinnear Wilson's (1928) original characterization of the basal ganglia as the dark basements of the brain still rings true today. The principal reason for the limited progress in this area is the presence of deficiencies and limitations in the research techniques employed to date to investigate subcortical participation in language. Contemporary theories of the role of subcortical structures in language are largely based on examination of subjects with language pathology associated with poorly defined lesions of vascular origin. These lesions rarely involve a unitary subcortical structure and in many cases the neuroradiological techniques used to identify the location of the lesion have been inadequate. The majority of the evidence linking subcortical lesions to the occurrence of language disorder reported in the literature so far has come from studies based on CT localization of the lesion site. Unfortunately, the limitations of CT in defining the extent of vascular lesions is well-documented. Although there is one earlier reported study which has noted language changes following stereotactic surgery involving subcortical structures (Svennilson *et al.*, 1960), the surgical techniques used at that time were associated with some complications and only relatively primitive neuroradiological procedures for confirming the exact location of the lesion were available. Consequently, an inability to strictly define vascular neuropathology and the use of rudimentary pallidotomy techniques supported by only emergent neuroimaging technology, have largely limited opportunities to validate contemporary models of subcortical participation in language, until now.

In recent years, a number of factors including, enhanced understanding of the pathophysiology underlying movement disorders, advances in neuroimaging and neurosurgical techniques and a degree of disillusionment with drug treatments for movement disorders, has led to a revival of stereotactic surgical procedures such as pallidotomy and thalamotamy in the treatment of Parkinson's disease (PD). The availability of discrete circumscribed lesion sites therefore provides an unprecedented opportunity to empirically test models that implicated the globus pallidus and thalamus in language. Further, although there is a plethora of literature documenting the benefits of stereotactic surgery in ameliorating the motor symptoms associated with PD, there is a paucity of information pertaining to the effects of this procedure on cognitive functioning. Recent studies that have evaluated the effect of stereotactic lesions on cognitive function, including language, have been largely neuropsychologically-based. Reports have consistently described performance on tasks of attention, memory, concentration, visuospatial ability, executive functioning and, in relation

to language, confrontation naming, categorical and phonemic fluency. Therefore, the aims of the research to be presented were to: 1) Investigate the effect of surgically induced lesions of the globus pallidus on language functioning, utilising a comprehensive linguistic assessment battery; 2) discuss post-operative language profiles in relation to operative theoretical models of subcortical participation in language; and 3) validate or nullify the hypothesis of a cortico-striato-pallido-thalamo-cortical loop subserving language, which promotes functionally distinct roles for the globus pallidus and thalamus. These aims will be addressed by reference to the pre- and post-surgery language abilities of two subjects, one of whom underwent pallidotomy, and the other thalamotomy. Prior to examination of these cases, however, it is necessary to briefly summarize the surgical procedures involved.

PALLIDOTOMY AND THALAMOTOMY

There are four major reasons why the past decade has witnessed a renaissance in the surgical treatment of movement disorders associated with Parkinson's disease. First, medications such as L-dopa have shortcomings and there continues to be a large number of patients who are disabled despite optimal medical therapy. Second, advances in neurosurgery, in imaging the brain and in neurophysiological monitoring make functional procedures more accurate and safer. Third, advances in the understanding of basal ganglia circuitry, function and pathophysiology, based largely on experiments conducted on the MPTP (1-methyl-4-phenyl-1,2,3,6 tetra-hydropyridine) primate model of Parkinsonism now provide a solid scientific rationale for intervening at the level of the basal ganglia. Finally, recent preliminary clinical results of pallidotomy suggest that this procedure can provide striking benefits.

It appears that certain Parkinsonian features may respond to surgery better than others. Features that respond to surgery are tremor, rigidity, bradykinesia, postural and gait disturbances, and drug induced involuntary movements. Features that are resistant to surgery include cognitive deficits, psychiatric disorders and autonomic disturbances. For patients with PD in whom tremor is the dominant feature, the ventral intermediate (VIM) nucleus of the thalamus or its afferent axonial projections is still the most commonly chosen surgical target. Patients with tremor-dominant PD, however, constitute a small minority. More common are patients with akinetic/rigid PD with fluctuating on/off periods and drug-induced dyskinesias. For this larger group of patients, the internal segment of the globus pallidus (GP) is a more appropriate surgical target. The VIM thalamic target is largely ineffective for the non-tremor components.

Surgical Techniques

The surgical technique is carried out by the consulting neurosurgeon. First, a pre-operative T-1 weighted Magnetic Resonance Image (MRI) scan is acquired. Just before the operation and under general anaesthesia the Cosman-Roberts-Wells (CRW™) head ring is fixed to the patient's head low enough to acquire a CT scan of the entire skull. We prefer to use a general anaesthetic for this stage to avoid patient movement and for patient comfort. A stereotactic Computed Tomography (CT) scan of the entire skull is acquired using 3mm contiguous slices. Next the MRI and stereotactic CT are transferred to the StereoPlan workstation. Using the Image Fusion software, the MRI is aligned to the stereotactic CT with at least three anatomic landmarks, we use the orbital fissures, lateral ventricle horns, the lenses of the eyes, and the pineal gland. Once alignment is complete the ImageFusion software volumetrically correlates the MRI image set to the stereotactic CT, independently scaling X, Y, Z and all rotational axes. Upon completion of the complete correlation of the MRI to the stereotactic CT the StereoPlan software is employed to pre-operatively plan a surgical trajectory for electrode placement. The spatially corrected and volumetrically correlated MRI is used for anatomic localisation of the Gpi that is clearly seen on the MRI images. Additional anatomic verification is performed with the AtlasPlan module of StereoPlan. The Schaltenbrand and Wahren Atlas (used in the software with permission from Thieme) is co-registered with patient anatomy by the AC and PC points as well as a known lateral landmark (we compare the left and then right putamino-pallidal boundary of the patient's scan, depending on the initial target, to that in the atlas). The AtlasPlan module then shows the placement of the target on the Schaltenbrand and Wahren Atlas images. The planned trajectory can be evaluated with the StereoPlan software once the arc angles are set to enter from an entry point 1.0cm anterior to the coronal suture and 5-6.0cm lateral to the midline with the CRW arc system in place.

Whilst planning the procedure, the patient is woken up after insertion of an arterial line to monitor blood pressure. The patient's BP is maintained at 20mm HG below their normal systolic pressure using intravenous hydralazine or sodium nitroprusside. We do not routinely shave the patient's hair but clean it with aqueous and alcoholic chlorhexidine and then shave a few hairs at the point of entry. A dermal skin punch is then used to incise the skin and a 4mm twist drill made along the planned trajectory, the dura is then punctured with a biopsy cannula. This prevents any significant CSF leakage that could cause brain sag. For posteroventral pallidotomy (PVP), the entry point chosen is 1.0cm anterior to the coronal suture and 5-6.0cm lateral to the midline. This approach avoids the internal capsule and allows for lesions to be placed from medial to lateral in a single pass of the electrode.

Having determined the co-ordinates using ImageFusion and StereoPlan, the electrode (2mm exposed tip, 1.88 diameter) is passed to the target. We chose the initial target to be in the medial pallidum just beyond the Gpe and Gpi border and all lesions are placed ventromedially. This avoids encroaching onto the lateral pallidum which may have untoward effects. Despite the large numbers of cases published, there is still no precise target as such that one could aim for and obtain reliably good results. We feel that in the anter-posterior

direction, the posterior third of the medial pallidum should be the target region. In the coronal plane the lesions are localized to the medial pallidum without a great deal of encroachment into the ansa lenticularis.

Impedance using this electrode is 500-700 ohms in the pallidum. Stimulation at 100Hz beginning at 2.0mm above the calculated target is then begun with the attending neurologist examining the patient. The electrode is advanced in 1.0mm steps and testing repeated. The optimal site is that at which rigidity, bradykinesia and, if present, tremor are alleviated at 0.5-1.0V at 100Hz. Very occasionally, patients may show exacerbation rather than alleviation of symptoms. We feel that it is important to examine the patient during the first pass of the electrode as impedance is measured since the stun effect of passing the electrode to the anticipated end target may render subsequent neurological examination unhelpful. It may be that this is why many groups do not find macrostimulation and examination helpful. Certainly multiple passes to plot single cell activity will cause a significant stun effect. In addition, with the macrostimulator left on, a brief cognitive screen including tests of memory, orientation, and language functioning is administered.

The electrode is then advanced until the impedance begins to rise to about 800-1100 ohms, which indicates that the bottom of the medial pallidum has been reached and no further advancement is made. If capsular motor responses are not obtained at less than 2.0V and 2Hz, and there are no visual phosphenes on stimulation at 3.5-5.0V, it is safe to begin lesioning. Another confirmatory sign that we find is one of bobbing of the patient's tongue at 2Hz and 2.0V. The first is a temporary lesion at 45C for 60 seconds and, if there are no side effects, the first lesion is placed at 80C for 60 seconds, the electrode is then withdrawn 2.0mm and a second lesion made at 80C for 90 seconds and on withdrawing another 2.0mm a final lesion is made at 75C for 60 seconds if bilateral lesions are made and 90 seconds if not. The largest middle lesion is chosen as the site where the minimal voltage on macrostimulation alleviated the neurological signs. It is not unusual to observe what we call release dyskinesias in completing the lesions. These tend to herald a good clinical outcome and vary in duration from a few minutes to a few days. The lesions thus obtained are 100-150mm in volume overall, with bilateral PVP we aim to make the lesions assymetrical with a smaller lesion

(~100mm^3) in the dominant hemisphere and the larger (~150mm^3) in the non-dominant side. The total awake time to perform a pallidotomy is, on average, 40mins for each side

CASE REPORTS

Case Report 1 – Pallidotomy

LM, a 73-year-old, right-handed female with idiopathic PD (12 years post-diagnosis) and goitre served as the subject for this case study. The subject reported that she had achieved ten years of formal education, and that English was her first and only language. No previous history of head injury, cerebrovascular accident, cerebral tumour or abscess, co-existing neurological disease, substance abuse, psychiatric disorder, developmental language disorder or speech and/ or language disturbance prior to the onset of PD was reported. LM underwent a bilateral ventromedial pallidotomy to alleviate dyskinesias, dystonia and unpredictable on/off states associated with PD.

Prior to this procedure, a neurological evaluation was conducted by a qualified neurologist. LM achieved a total score of 51/176 on the Unified Parkinson's Disease Rating Scale (UPDRS) (Fahn and Elton, 1987). This score was achieved by summating component scores of sections 1-4 of the aforementioned scale, in the defined 'off' state (ie. no medications overnight or 1 hour after waking). This rating was indicative of bilateral pathology with mild to moderate reduction in activities of daily living and mild to moderately reduced motor skills. In addition, LM achieved an overall rating of 3 on the Hoehn and Yahr Staging of Parkinson's Disease scale (H&Y) (Hoehn and Yahr, 1967), indicative of a moderate to severe generalised dysfunction including a significant slowing of body movements and impaired balance on walking and standing. Pre-operative MRI noted no brain abnormalities. LM's drug regime prior to surgery consisted of the following: Parlodel 5mg, Sinemet 100/25mg, Sinemet 250/25 mg, Sinemet CR 200/50mg, Inderal 40mg and Thyroxine 100mg. A mild dysarthria was evident at both the pre- and post- surgery assessment phases as perceived by a speech pathologist, however, this speech disturbance was not considered to adversely affect functional speech intelligibility on language measures requiring verbal responses

An evaluation of cognitive functioning was also conducted by a neuropsychologist prior to surgery. An estimate of premorbid level of intellectual functioning placed LM within at least the average range of performance. Average to above average performance was reported on assessments of cognitive functioning with the exception of angular judgement (mildly impaired ranked at the 22nd percentile) and facial recognition (moderately impaired ranked at

<5th percentile) tasks. Executive functioning was reported as largely intact with no evidence of co-morbid dementing illness or indicators of anxiety or depression documented.

For measures of high-level language functioning where no age appropriate normative data was available, the experimental subject's performance was compared to that of a group of seven non-neurologically impaired individuals. One standard deviation above or below the control group mean was established as the criterion for performance outside the range of normal. A conservative criterion was selected in order to account for any subtle changes in linguistic functioning post-pallidotomy. Control subjects were native speakers of English with no previous or existing history of head injury, cerebrovascular accident, cerebral tumour or abscess, substance abuse, psychiatric disorder, developmental language disorder or speech and/or language disturbance. The control subjects presented with perceptually normal speech as judged by a speech pathologist and scored within the range of normal cognitive functioning

(ie. score between 139.9-144) (Troster *et al*, 1989) on the Mattis Dementia Rating Scale (Mattis, 1988). The mean age of the control group was 63.86 years (range 50-78) and the mean level of formal education was 11.57 years (range 9-19).

Procedures

Language assessments. LM was administered a comprehensive battery of language assessments considered to be sensitive measures of both gross and high-level linguistic functioning. The assessments utilised were then subdivided into two separate test batteries for analysis purposes. LM was administered the test battery 1 month prior to bilateral pallidotomy and 3 months after surgery, within perceived "on" periods (ie. when optimally medicated). Testing was undertaken in a quiet distraction free environment according to standardised instructions and was conducted over two 2-hour sessions, in order to compensate for fatigue. The assessment battery incorporated tasks that were primarily dependent on oral language abilities, however, tasks dependent on written expression and reading comprehension skills were also included.

Battery 1 incorporated measures of gross language function including the Neurosensory Centre Comprehensive Examination of Aphasia (NCCEA) (Spreen and Benton, 1969) and the Boston Naming Test (Kaplan *et al.*, 1983) including an analysis of error types derived from those methods utilised by LeDorze and Nespoulous (1989), Smith *et al.*, (1989), LaBarge *et al.*, (1992), and Chenery *et al.*, 1996). Battery 2 contained measures of high-level linguistic functioning including the Test of Language Competence Expanded-Edition (TLC-E) (Wiig and Secord, 1989), The Word Test-Revised (TWT-R) (Huisingh *et al.*, 1990), a subtest of the Test of Word Knowledge (TOWK) (Wiig and Secord, 1992), the Wiig-Semel Test of

Linguistic Concepts (WSTLC) (Wiig and Semel, 1974), and the animal fluency subtest of the Western Aphasia Battery (WAB) (Kertez, 1982).

Results and Discussion – Case 1

Post-operative neurological and neuropsychological profiles. A post-operative neurological evaluation, conducted by a neurologist, revealed a total score of 31/176 on the UPDRS. This score was indicative of mild improvements across a number of activities of daily living and general motor skills. Despite these overall improvements, a mild decline in LM's swallowing function and a moderate to severe decline in speech intelligibility and legibility of handwriting were reported. LM's overall H&Y rating remained the same following pallidotomy. A post-operative MRI, taken 1 day following surgery, reported bilateral pallidotomy lesions measuring 1 cm in diameter in addition to possible small bilateral areas of associated haemorrhage and oedema extending into the posterior rim of the internal capsule. LM's post-operative drug regime included the following: Solprin 300mg, Inderal 40mg, Pravochol 40mg, Probanthine 15mg, Parlodel 2.5mg, Sinemet 250/25mg, Sinemet CR 200/50mg, Madopar 200/50mg. Post-operative neuropsychological assessment conducted by a neuropsychologist indicated that executive functioning remained largely unaltered subsequent to pallidotomy, however, fluctuations in performance were observed across a number measures of cognitive functioning. Mild declines on tasks of non-verbal reasoning and abstraction (50-75th percentile), focused attention (16th percentile), working memory and delayed recognition of verbal information were documented. In addition, a moderate decline in angular judgement skills (4th percentile) and a severe decline in the ability to retain verbal information, was noted. Psychomotor speed, was reportedly unchanged following pallidotomy, however, mild improvements in immediate attention span were documented. It was reported that post- operative declines in performance may have been partially attributable to demonstrated fatigue during follow-up assessment.

The results of this study support the hypothesis of a cortico-striato-pallido-thalamo-cortical loop subserving language function (Crosson, 1985; Wallesch and Papagno, 1988), and indeed are consistent with the suggestion that the globus pallidus has a role to play in mediating linguistic processes. The results revealed that subsequent to pallidotomy, gross language skills remained relatively intact, however, fluctuations in performance across a number of high-level linguistic tasks were observed. When compared to a control group, the experimental subject also demonstrated reduced accuracy in conducting lexical decisions pertaining to legal non-words and words with few meanings following pallidotomy, in addition to overall longer mean reaction times for all stimuli. The post-operative changes in language profiles observed

were attributed to disruption of the aforementioned cortico-subcortical-cortical loop, subsequent to the surgical generation of discrete lesions within the globus pallidus.

Battery 1: Gross language function. LM's performance on gross measures of language function was relatively intact following pallidotomy, with the exception of scores achieved on subtests Phonemic Fluency (PF) and Sentence Construction (SC) of the NCCEA. Of note, pre-operative scores achieved for each of these subtests were also below average, suggesting that pre-existing subcortical pathology may have negatively influenced performance on these tasks. Reduced scores on the SC task were attributed to the use of time restrictions in the administration of this subtest. Slow response initiation was observed during LM's pre-operative performance on this task and may have represented a form of linguistic akinesia. Indeed, Crosson (1992a) hypothesised that a form of language akinesia may mirror manifestations of physical akinetic symptoms. This phenomenon was claimed to be the result of impaired language release mechanisms, presenting as a deficit in initiating and maintaining the production of language (Crosson, 1985). Reduced post-operative performance was attributed to an inability to accurately and efficiently combine specified word forms into a syntactic structure. Despite the production of syntactically correct sentences post-operatively, the subject demonstrated a tendency to utilise word forms that were a close approximation to the target stimulus *(eg. drove versus drive, walked versus walk)*. This behaviour may be best interpreted in terms of Wallesch and Papagno's (1988) model, as an inability to efficiently integrate specified linguistic information and to apply and monitor the application of goal-directed rules in the production of verbal output. Lesions of the globus pallidus and the subsequent disruption of subcortical linguistic integration and rule application processes, may manifest as sentences which contain linguistic units that deviate from the intended target, in terms of morphosyntactic structure, and remain uncorrected unless prompted. Despite the inclusion of a PF subtest within the NCCEA, this subtest has been routinely incorporated into cognitive assessment batteries which aim to evaluate high-level executive functions of the frontal lobe. As such, these results are discussed further below, relative to high level-linguistic abilities.

LM demonstrated a post-operative decline in confrontation naming abilities, despite pre-and post-operative scores both falling within the range of normal. This decline in performance was associated with an increase in the number of semantic errors produced, supporting Crosson's (1985) theory that lesions of the globus pallidus would serve to disinhibit the thalamus, thereby increasing cortical activity to produce extraneous verbal output, such as semantic paraphasias. Significant changes in post-operative confrontation naming abilities have not been previously reported (Masterman *et al.*, 1998; Scott *et al.*, 1998), yet among these reports detailed error analyses of responses produced have not been conducted. As such,

previous studies have failed to delineate any changes in lexical retrieval mechanisms subsequent to pallidotomy, which is an issue worthy of further consideration.

Battery 2: High-level linguistic functioning. The TWT-R, TLC-E and WSTLC, in addition to letter-cued fluency (PF), represented the measures of high-level linguistic functioning most sensitive to changes in complex language following pallidotomy. Average to above average pre-operative performances on the Multiple Definitions (MD) and Definitions (DF) subtests of the TWT-R, declined post-operatively to below the range of normal, indicating that lesions of the globus pallidus may impact upon the ability to formulate complex divergent language. Post-operatively, a number of LM's responses on the DF subtest were lacking essential semantic elements. For example, in defining the verb *scribble*, requiring the essential elements of write/ colour/ draw + without order/ meaning, LM responded *used to take notes*. These results suggested a deficit in recognising and describing key semantic attributes of a target word (Huisingh *et al.*, 1990). Semantically inadequate definitions in terms of critical attributes, such as circumlocutory responses, support the hypothesis of Crosson (1985), that lesions of the globus pallidus will result in extraneous verbal output, as a consequence thalamic disinhibition and hyperactivation of the anterior language area. In addition, post-operative errors produced on the MD task involved stimulus-bound responses (Huisingh *et al.*, 1990) and definition references not indexed within the scoring criteria. Stimulus-bound responses (eg. for the word *park*, LM provided the definitions *parking a car* and *to park yourself somewhere*) fulfilled only one definition reference criterion, *to leave temporarily*, as opposed to a minimum of at least two semantically distinct criteria (including, *land* or *car gear*). Inflexibility in the production of definition references on this task may suggest a reduced capacity to accurately select or gate multiple competing definitions for expression following pallidotomy. As a result, the same definition reference is selected more than once for production. These results are in line with Wallesch and Papagno's (1988) model, that the basal ganglia system and thalamus integrate cortically generated situational and goal-directed constraints in additional to lexical information, and determine the most appropriate lexical alternative from a range of alternatives for production as speech. Lesions of the globus pallidus may disrupt thalamic gating mechanisms, resulting in disinhibited thalamic excitation of inhibitory cortical interneurons, preventing the processing of appropriate competing alternatives. It has been observed that lesions of the basal ganglia system typically impair performance on tasks where degrees of freedom, in relation to possible responses, are large (Wallesch and Papagno, 1988). On such tasks, syntax is preserved yet semantic content may be inadequate (Wallesch and Papagno, 1988), such as the replication of previously selected definition references on a multiple meanings task. The production of unindexed definition references (eg. for the word *down*, one of LM's responses included hair on your face) also

supports the hypothesis of disturbed lexical selection mechanisms following pallidotomy, whereby subordinate alternatives are gated for verbal production.

LM's performance on verbal fluency subtests revealed post-operative declines on both phonemic and semantic fluency tasks. Pre-operative performance on the PF subtest was below normal, LM demonstrating a further decline post-operatively. In contrast, LM's semantic fluency score was within the range of normal both pre- and post-operatively, however, a decline in performance was also evident following pallidotomy. These results suggested a post-operative impairment at the level of the phonological output lexicon and perhaps a reduction in semantic drive (Caplan, 1994). Murdoch (1996), in line with Wallesch and Papagno (1988), stated that lexical content in the production of responses on verbal fluency tasks would also involve many degrees of freedom, where a large number of appropriate responses generated by the cortex, compete with each other for selection by the striatum and subsequent release to the anterior language centre via thalamic gating mechanisms. Consistent with the results achieved on the DF and MD subtests in the present study, where task responses were unrestricted, this postulate would explain the post-operative reduction in performance on both PF and Semantic Fluency (SF) tasks as an impaired or functionally reduced lexical selection mechanism, especially for words specified by a particular phonological form. Pallidotomy may serve to disrupt thalamic gating mechanisms resulting in the inefficient release of competing alternatives for frontal processing, and the inhibition of cortical activity. The results of the current study also suggest that a deficit in the ability to retrieve phonologically constrained word forms was evident prior to pallidotomy in this subject. Previous studies have reported variable results in relation to the performance of people with PD on PF tasks. A number of studies have reported reduced PF scores in people with PD (Epker *et al.*, 1999; Beatty and Monson, 1989), while others report performances within the normal range (Beatty and Monson, 1989; Caltagirone *et al.*, 1989; Raskin *et al.*, 1992; Auriacombe *et al.*, 1993; Lewis *et al.*, 1998). The variation in results reported may be attributed to the possible existence of heterogeneous cognitive profiles across subject groups and the premise that as cognition declines in PD, linguistic deficits may manifest across a wide range of language tasks (Lewis *et al.*, 1998). Indeed, Beatty and Monson's (1989) study identified PD subgroups relative to performance on the BNT. PD subjects with impaired naming abilities performed below normal on a PF task relative to controls, however, PD subjects with normal naming abilities performed within the range of normal on the same task. In addition, a number of distinct criterion levels were utilised to classify normal performance across studies, perhaps contributing to the variable results reported. Furthermore, surgically induced lesioning of the globus pallidus contributed to an exacerbation of the aforementioned lexical selection deficit for phonologically constrained word forms, and also a decline in performance on a task of category cued fluency.

In relation to other studies, reductions in PF and SF scores following pallidotomy have been reported. Declines in PF have been reported following unilateral pallidotomy of the left hemisphere (Lombardi *et al.*, 2000; Trepanier *et al.*, 1988;) in addition to bilateral pallidotomy (Scott *et al.*, 1998), but not subsequent to unilateral pallidal lesions within the right hemisphere (Lombardi *et al.* , 2000). Declines in semantic fluency scores have also been reported following unilateral (Masterman *et al.*, 1998; Scott *et al.*, 1998) and bilateral (Scott *et al.*, 1998) posteroventral pallidotomy, with no identifiable interhemispheric effects (Lombardi *et al.*, 2000). Of note, however, more anteromedially placed lesions were documented to produce declines in performance in the production of words that were related categorically, and posterolaterally positioned lesions, to effect an increase in performance on this task (Lombardi *et al.*, 2000).

In contrast, post-operative improvements in performance were observed on the Figurative Language (FL) and Remembering Word Pairs (RWP) subtests of the TLC-E, the Associations (ASS) subtest of the TWT-R and on the SP component of the WSTLC. An increase in the total score achieved for RWP may have been closely related to the mild post-operative improvement in immediate attention span reported in the post-surgical neuropsychological evaluation, rather than an enhancement in linguistic functioning. The increase in scores achieved on the FL subtest of the TLC-E and the ASS subtest of the TWT-R, however, was highly inconsistent with the results achieved on other complex expressive language tasks within the present study. LM was expected to perform below the control group mean on these tasks, consistent with her performance on other tasks where the degrees of freedom for lexical content pertaining to potential responses was great. The FL and ASS subtests involved complex divergent expressive language tasks, requiring the interpretation of metaphorical speech and the explanation of differences between category members in terms of critical semantic features, where degrees of freedom in relation to possible responses were unrestricted. LM, however, achieved normal post-operative scores on these tasks, her performance improving from below the range of normal prior to pallidotomy to within normal following surgery. In addition, required responses on the WSTLC (yes/no) involved a restricted number of degrees of freedom and consequently, reduced competition for selection between potential lexical alternatives. As such, performance was not expected to change significantly on this task between pre- and post-operative test phases. Despite this prediction, however, LM demonstrated an increase in total score achieved on the WSTLC following surgery.

LM's performance on the Semantic Absurdities (SA) subtest of the TWT-R and the Passive (PS) subtest of the WSTLC remained stable across pre- and post- operative test phases. Pre- and post-operative scores on the SA subtest that were stable yet reduced, relative to normal

controls, lent support to both Crosson's (1985) and Wallesch and Papagno's (1988) models of subcortical participation in language. Despite recognising semantic incongruities within absurd statements, LM demonstrated an inability to consistently repair incongruities by evaluating and contrasting the critical semantic features of discordant words (Huisingh *et al.*, 1990). Explanatory and negated responses lacking critical semantic features were observed inconsistently. These extraneous responses may have been the result of an impaired response-release mechanism for semantically verified language (Crosson, 1985) or a defective lexical selection mechanism that resulted in the thalamic gating of semantically subordinate or rudimentary responses for frontal processing, on tasks where potential responses entailed large degrees of freedom (Wallesch and Papagno, 1988). Reports of PD performance on semantic absurdity tasks have not been previously reported in studies of high-level linguistic functioning in this population (Lewis *et al.*, 1998). Despite an inability to draw comparisons from previous research, the results achieved in the current study may represent the existence of dysfunctional cortico-subcortical-cortical circuitry in populations with pre-existing subcortical neuropathology that remains similarly impaired following pallidotomy. In addition, stable, yet enhanced performance on the PS subtest was also acceptable in relation to contemporary models of subcortical participation in language. Required responses for this task involved limited degrees of freedom and as such, were not hypothesised to change significantly subsequent to pallidotomy. The variation in post-operative performance observed across high-level language tasks, indicates that disruption to the hypothesised cortico-striato-pallido-thalamo-cortical loop alters the way in which complex language is understood and formulated, lending support to models of subcortical participation in language (Crosson, 1985; Wallesch and Papagno,1988). Despite the documented changes in high-level linguistic performance following pallidotomy, the inconsistencies observed complicate the delineation of the mechanism underlying these post-operative alterations in function. Scott *et al.*, (1998) also reported significant individual variation in post-pallidotomy cognitive gains and deficits, despite minimal post-operative alterations in cognitive functioning within a group of 20 pallidotomy patients.

Post-operative fluctuations in performance within the current study may have been indicative of normal variability between assessment phases, possible improvements in motivation and attention subsequent to pallidotomy (Scott *et al.*, 1998), cyclical alterations in medication levels throughout assessment sessions, which served to influence cognitive functioning (Gotham *et al.*, 1988; Saint-Cyr *et al.*, 1993) or may in fact negate the hypothesis of a specific linguistic function for the globus pallidus. In the event of the globus pallidus undertaking a specialised linguistic function, similar levels of impairment were predicted following pallidotomy for task responses that potentially involved large degrees of freedom, such as complex divergent expressive language tasks. The post-operative inconsistencies in

performance (ie. decrements and improvements) observed on comparable linguistic tasks in this study may be more accurately interpreted as the result of disruption to, or the disorganisation of the regulation of language processes along reciprocal cortical- subcortical loops (Crosson, 1985; Wallesch and Papagno, 1988). The precise mechanism underlying the subcortical-cortical integration and regulation of lexical information, however, remains largely misunderstood, further complicating the interpretation of results. The capacity for lexical alternatives to also be gated via thalamo-frontal and fronto-thalamic circuits which bypass the basal ganglia system, in addition to the assumption of functional plasticity within a disordered neural network (Wallesch and Papagno, 1988), may explain in part, some of the post-operative inconsistencies in performance observed in a subject with long-standing basal ganglia pathology. In order to further validate these findings future research requires the compilation of linguistic profiles for larger subject numbers. Despite the above limitations, however, defective or reduced lexical selection mechanisms were identified in this single case, following pallidotomy.

Conclusion – Case 1

Based on the results of this single case study, it may be concluded that fluctuations in cognitive functioning following pallidotomy extend into the domain of high-level linguistics and semantic processing. The results of this study revealed evidence of deficient lexical selection mechanisms on complex divergent expressive language tasks subsequent to pallidotomy, in addition to inefficient lexical access in the semantic processing of orthographic word forms. These post-operative linguistic impairments lend support to a hypothesised role for the globus pallidus in language, which is in all probability performed via participation in a cortico-striato-pallido-thalamo-cortical loop (Crosson, 1985; Wallesch and Papagno, 1988). Despite these endeavours, however, the results of the present study will remain unsubstantiated until comprehensive pre- and post-operative linguistic profiles are compiled for larger subject numbers.

Case Report 2 - Thalamotomy

TR, a 75-year-old, right-handed male with idiopathic, tremor dominant PD (10 years post-diagnosis) served as the subject for this case study. The subject reported 10 years of formal schooling, and that English was his first and only language. No previous history of head injury, cerebrovascular accident, cerebral tumour or abscess, co-existing neurological disease, substance abuse, psychiatric disorder, developmental language disorder or speech and/ or

language disturbance prior to the onset of PD was reported. TR underwent a left ventral intermediate (VIM) thalamotomy to relieve a chronic tremor of the right upper limb.

Prior to this procedure, a neurological evaluation was conducted by a qualified neurologist. TR achieved an overall rating of 3 on the Hoehn and Yahr Staging of Parkinson's Disease Scale indicative of a moderate to severe generalised dysfunction including a significant slowing of body movements and impaired balance on walking and standing. Unified Parkinson's Disease Rating Scale data was unavailable for this subject. Pre-operative MRI reported no brain abnormalities. TR's drug regime prior to surgery consisted of Artane 20mg, taken three times daily. A mild dysarthria was evident at both the pre- and post- surgery assessments as perceived by a speech pathologist, however, this speech disturbance was not considered to adversely affect functional speech intelligibility on language measures requiring verbal responses.

An evaluation of cognitive functioning was also conducted by a neuropsychologist prior to surgery. No evidence of co-morbid dementing illness or indicators of anxiety or depression were documented. In addition, executive functioning was reported to be largely intact.

A group of 16 adults with no neurological disease served as a control group (mean age = 61.94 years; mean level of education = 12.63 years). One standard deviation above or below the control group mean was established as the criterion for performance outside the range of normal. A conservative criterion was selected in order to account for any subtle changes in linguistic functioning subsequent to thalamotomy. Control subjects were also native speakers of English with no previous or existing history of head injury, cerebrovascular accident, cerebral tumour or abscess, substance abuse, psychiatric disorder, developmental language disorder or speech and/ or language disturbance. Control subjects presented with perceptually normal speech as judged by a speech pathologist and scored within the range of normal cognitive functioning (ie. score between 139.9′-144) (Troster *et al.*, 1989) on the Mattis Dementia Rating Scale (Mattis, 1988).

Procedures

Language assessments. TR underwent comprehensive language assessments aimed at evaluating gross and high-level linguistic functioning in addition to on-line semantic processing, 1 month prior to and 3 months following a surgically induced lesion of the left VIM thalamus. All assessments were conducted during perceived "on" periods (ie. when optimally medicated). Testing was undertaken in a quiet distraction free environment according to standardised instructions and was conducted over two 2-hour sessions, in order to compensate for fatigue. The assessment battery incorporated tasks that were primarily

dependent on oral language abilities, however, tasks dependent on written expression and reading comprehension skills were also included.

Battery 1 incorporated measures of gross language function including the Neurosensory Centre Comprehensive Examination of Aphasia (NCCEA) (Spreen and Benton, 1969) and the Boston Naming Test (Kaplan *et al.*, 1983). Battery 2 contained measures of high-level linguistic functioning including the Test of Language Competence Expanded-Edition (TLC-E) (Wiig and Secord, 1989), The Word Test-Revised (TWT-R) (Huisingh *et al.*, 1990), the Conjunctions and Transitions subtest of the Test of Word Knowledge (TOWK) (Wiig and Secord, 1992), the Wiig-Semel Test of Linguistic Concepts (WSTLC) (Wiig and Semel, 1974), in addition to semantic (animal and tool) fluency tasks. Battery 3 consisted of a lexical decision task incorporating legal non-words and real word stimuli classified by number of meanings and meaning relatedness (Azuma and Van Orden, 1997).

Results and Discussion – Case 2

Post-operative neurological and neuropsychological profiles. A post-operative neurological evaluation, conducted by a neurologist, revealed a Hoehn and Yahr rating of 2, indicating a mild improvement in motor functioning and the alleviation of tremor, despite persistent bilateral symptoms involving posture and gait. Post-operative neuropsychological and MRI data were not available for this subject. TR's post-operative drug regime was unchanged.

Language profile. A myriad of previous studies support evidence of a laterality effect with respect to linguistic performance, subsequent to surgically induced lesions of the thalamus. Indeed, Samra *et al.* (1969) reported language impairments in general, to be more commonly associated with left as opposed to right thalamic lesions. Specifically, post-operative deterioration in verbal expression has been well documented (Almgren *et al.*, 1976) in addition to receptive deficits (Vilkki and Laitinen, 1974; Wester and Hugdahl, 1997; Hugdahl *et al.*, 1990). It is difficult, however, to compare the results of the current study with previous findings, given the paucity of earlier research entailing high-level linguistic analyses. An exception to this observation were the findings of Darley *et al.* (1975) who reported high-level linguistic deficits such as reduced completeness and accuracy of verbal language formulation and expression, subsequent to left thalamotomy.

Battery 1: Gross measures of language function. TR failed to demonstrate significant post-operative changes in performance when comparisons of total test scores for measures of gross language functioning were made. Non-significant post-operative declines in performance were observed for the NCCEA and BNT, however, both pre- and post-operative total test scores fell within the range of normal.

Battery 2: Measures of high-level linguistic functioning. Significant post-operative declines in performance were evident across the majority of total test scores pertaining to measures of high-level linguistic functioning, including: TLC-E, TWT-R, TOWK and semantic fluency (animals and tools).

TLC-E. TR demonstrated significant post-operative declines on the Recreating Sentences (RS), Figurative Language (FL) and Remembering Word Pairs (RWP) subtests of the TLC-E.

With regard to the RS subtest, TR demonstrated a decline in performance to 6.80 SD below the control group mean. His post-operative performance indicated an inability to consistently and efficiently integrate specified linguistic units into semantically, syntactically and pragmatically appropriate sentences. For example, in constructing a sentence with the specified lexical units *neither, week, were*, in the defined context of *at the supermarket*, TR responded *" The shoppers....were not...."*. In relation to subcortical language models, left thalamic lesions may indeed serve to: 1) disrupt the efficient and appropriate organization, integration and production of specified lexical-semantic content within contextually bound syntactic structures (Wallesch and Papagno, 1988); 2) disconnect cortical nets responsible for concept generation, resulting in verbal output which is lacking in detail and well defined conceptual markers (Nadeau and Crosson, 1997); or 3) impair spontaneous speech (ie. impair fluency or efficiency of verbal output) and disorganise language as a direct consequence of cortical hypoarousal and/ or redundant semantic monitoring mechanisms (Crosson, 1985).

Post-operative performance on the FL and RWP subtests fell 1.40 and 1.21 SD respectively, below the control group mean. The FL task required subjects to interpret figurative expressions within contextually defined boundaries, and to identify additional metaphorical expressions with fundamentally parallel meanings (Wiig and Secord, 1989). TR demonstrated post-operative deterioration in both skill components of this task. For example, when explaining the meaning of the expression *" It's still up in the air"* within the context *'A student talking to his friend about a trip'*, TR responded *"It means I'm still on top of the world"*. This response reflected an inability to independently formulate an explanation which contained conceptually and semantically appropriate content, despite the ability to identify the fundamental meaning of the expression when given a choice of 4 additional figurative expressions. In relation to operative subcortical language models, this result supports the hypothesis that thalamic lesions 1) disrupt lexical selection mechanisms (Wallesch and Papagno, 1988) or 2) semantic monitoring mechanisms (Crosson, 1985), resulting in the inclusion of inappropriate semantic content in verbal output. In addition, thalamocortical disconnection may result in concept generation and lexical-semantic deficits during expressive speech tasks (Nadeau and Crosson, 1997). Furthermore, TR also demonstrated an inability to correctly identify figurative expressions with similar meanings in a consistent

manner. For example, the expression *"That team couldn't win even with new blood"* was correctly interpreted as *"They couldn't win even with new players"*. TR was, however, unable to correctly identify which among several additional competing figurative expressions contained a fundamentally parallel meaning (eg. TR selected the literal interpretation *'That team couldn't win even with a blood transfusion' as opposed to the correct response 'Even some new faces couldn't make that team win'*). This performance profile indicated that thalamic lesions may also impair comprehension abilities via cortical disconnection mechanisms (Nadeau and Crosson, 1997) and that competition of lexical material for cortical processing (Wallesch and Papagno, 1988) may also apply to receptive language, with more literal concepts having a processing advantage subsequent to thalamic lesions.

Reduced post-operative recall of word pairs on the RWP subtest was consistent with the findings of Almgren *et al.* (1969). From the above results, it was hypothesised that left thalamic lesions may impair the processing of verbal language and the ability to retrieve words from short term memory. This postulate was consistent with the models of: 1) Crosson (1985) based on the work of Ojemann (1975; 1976) that the thalamus may influence attentional mechanisms which gate the storage and retrieval of items within short and long term memory; and 2) Nadaeu and Crosson (1997) who theorised that thalamotomy may prohibit the selective engagement of cortical nets responsible for the storage and retrieval of verbal language.

TWT-R. Overall post-operative reduction on the TWT-R was attributed to significant declines on the antonym generation and multiple definition formulation subtests. In relation to performance on the antonym generation task, TR's post-operative score fell to 2.29 SD below the control group mean. Errors consisted of semantically inappropriate responses (eg. opposite of *worst = better; first = second*). These results can be explained in terms of subcortical models of language production, in that semantically inappropriate responses result from disruption to thalamic gating mechanisms as a consequence of left thalamotomy (Wallesch and Papagno, 1988) and/ or thalamic lesions impair semantic monitoring mechanisms resulting in semantic paraphasias/ extraneous content within verbal output (Crosson, 1985). In addition, thalamic lesions may disconnect cortical nets (Nadeau and Crosson, 1997) dedicated to the storage and retrieval of semantically coded material (eg. antonyms versus synonyms).

Post-operative performance on the multiple definition formulation task fell to 2.48 SD below the control group mean. Two error types in responses reflecting vocabulary inflexibility (Huisingh *et al.*,1990) were observed, including 1) stimulus bound responses (Huisingh *et al.*, 1990) where the same essential meaning was produced for both definitions (eg. show = a) *an exhibit and b) a musical*), 2) inability to formulate more than one meaning for target words

(eg. fall = *a) a waterfall; b) don't know*). This performance profile was supported by the postulates of Wallesch and Papagno (1988) and Crosson (1985), respectively, in that 1) stimulus bound responses indicate a reduced capacity to select or gate multiple competing definition concepts for expression, subsequent to thalamic lesion and 2) the thalamus may serve to promote linguistic flexibility (Riklan *et al.*, 1969) by mediating the efficient release of semantically monitored information for verbal production, via cortical activation and organization mechanisms.

TOWK. TR demonstrated a significantly reduced post-operative ability to identify logical semantic relationships in the presence of defined contexts, on the conjunctions and transitions subtest of the TOWK. On this task, the subject was required to select one of four written conjunction or transition words to define the logical semantic relationship existing between a contextually defined and an incomplete sentence (Wiig and Secord, 1992). This performance was best interpreted by Nadeau and Crosson's (1997) model of subcortical participation in language, in that thalamic lesions may disrupt mechanisms underlying the activation of cortical nets dedicated to the comprehension of semantic information. In addition, an extrapolation of Wallesch and Papagno's (1988) model may also support these findings, if competition between lexical alternatives is considered with regard to input channels as well as verbal production mechanisms.

Verbal fluency (animals and tools). TR demonstrated a significant post-operative decline in semantic fluency scores (ie. animals and tools), with the greatest decrement observed for the animals category. These results were in contrast to those of Fukuda *et al.* (2000) who reported no significant reduction in animal fluency scores 4 weeks following left thalamotomy. This disparity may be attributable to incongruities in the duration of post-operative assessments (ie. 4 weeks as opposed to 3 months). Our research group promotes post-operative follow-up assessments to be conducted at 3 months, in order to obtain a neurologically stable performance.

Additional research, however, has also reported reductions in semantic fluency scores subsequent to left thalamotomy (Petrovici, 1980; Riklan and Levita, 1970; Riklan *et al.*, 1969; Vilkki and Laitinen, 1974; 1976). Of note, Riklan *et al.* (1969) hypothesised that verbal fluency tasks require creative and flexible sorting and searching for linguistic information. As such, the observed post-operative profiles may reflect a disturbance to activation mechanisms underlying linguistic fluency and flexibility, as a consequence of thalamic lesions of the dominant hemisphere. Furthermore, Vilkki and Laitinen (1974) reported post-operative deficits in tasks requiring continuous and rapid production of appropriate verbal responses, including semantic fluency.

The semantic fluency results obtained in the current study support operative theoretical models of subcortical participation in language. Specifically, the thalamus may serve to promote linguistic flexibility and fluency by mediating the efficient temporal release of semantic information via cortical activation and organization mechanisms (Crosson, 1985; Riklan *et al.*, 1969) and/ or thalamic lesions may impair lexical selection mechanisms causing inefficient thalamic release of competing alternatives for cortical processing (Wallesch and Papagno, 1988). Given that PET studies have revealed the temporal lobe to be active during semantic fluency tasks (Boivin *et al.*, 1992), thalamic lesions may actually disconnect areas of the temporal cortex dedicated to the storage of category specific semantic knowledge (Nadeau and Crosson, 1997), resulting in impoverished performance on semantic fluency tasks. The observation of a greater post-operative decline in the number of animal names generated as compared to tools, indicated that further studies of verbal fluency performance utilising categories controlled for frequency or familiarity, may shed further light on the way in which semantic knowledge is stored or classified within neural systems. Indeed, heightened thalamic neuronal activity has been observed, in relation to verbal responses containing specific semantic categories (Gogolitsin and Kropotov, 1981).

Battery 3: Semantic processing. Post-operative accuracy of lexical decisions remained within normal limits, however, TR demonstrated significantly longer pre- and post-operative mean reaction time (RT) scores when compared to the control group. A general post-operative improvement in RT was observed, however, mean scores remained significantly above the range of normal. In addition, a post-operative RT advantage was demonstrated for few meaning words, whereby fastest responses were observed for few/ high (FH) and few/ low (FL) words, respectively. Faster post-operative RT's were expected given an anticipated improvement in overall motor functioning. In harmony with this postulate, TR achieved a post-operative Hoehn and Yahr score of 2, indicative of a mild improvement in motor abilities. Concurrently, Vilkki and Laitinen (1976) reported persistently prolonged motor RT's on the Purdue pegboard test subsequent to unilateral thalamotomy of either the dominant or non-dominant hemisphere. Despite strong correlations, however, between verbal and sensorimotor impairments in the above study, it was reported that delayed post-operative RT's could not be held entirely accountable for any cognitive deficits observed.

Indeed, TR demonstrated an atypical profile with respect to RT's for few/ meaning words, possibly indicating that thalamic lesions may alter word processing mechanisms. Spreading activation theory dictates that words with many meanings are processed faster as a consequence of probabilistically mediated nodal activation (Rubenstein *et al.*, 1970). The unexpected results achieved in the current study may be attributed to a number of factors and must be interpreted with caution. Of note, it has been reported that number of meanings

(NOM) and relatedness effects are challenging to replicate (Azuma and Van Orden, 1997), as such, our results may simply reflect this phenomenon. In the event of future findings which support our results, however, thalamic lesions may in fact serve to interrupt word processing mechanisms. Specifically, left thalamotomy may impact upon the efficiency of information transfer via nodal networks. Given the premise that word recognition occurs when the orthographic, semantic and phonological representations of a word cohere (Azuma and Van Orden, 1997), thalamic lesions may influence coherence mechanisms to become dependant upon the number of nodes shared, with an efficiency advantage for minimal nodal distribution. As such, reduced processing demands (ie. NOM) result in faster RT's.

Conclusion – Case 2

The results of this study support the hypothesis of a cortico-striato-pallido-thalamo-cortical loop subserving language function (Crosson, 1985; Wallesch and Papagno, 1988; Nadeau and Crosson, 1997), and indeed further support the suggestion that the dominant thalamus has a role to play in mediating linguistic processes. The exact mechanisms underlying thalamic participation in language remain unclear, however, our results revealed that subsequent to left VIM thalamotomy, gross language skills remain relatively intact, with a marked deterioration in performance evident across a number of high-level linguistic measures and semantic processing abilities. In particular, post-operative deterioration in performance was observed on tasks of verbal memory, construction of syntax within contextually constrained boundaries, comprehension of semantic relationships, interpreting figurative language, antonym generation, multiple meaning explanation and temporal processing of words coded for NOM and meaning relatedness. The post-operative changes in language profile observed were attributed to disruption of the aforementioned cortico-subcortical-cortical loop, subsequent to the surgical generation of discrete lesions within the dominant thalamus. The collection of data for larger subject numbers is required, however, before these results can be substantiated.

SUMMARY

Contemporary theories of subcortical participation in language are based largely on data collected from subjects with language pathologies associated with poorly defined brain lesions of vascular origin. Consequently, due to limitations in the methodologies employed, to date these theories have not been empirically tested. The recent renaissance of stereotactic neurosurgical procedures such as pallidotomy and thalamotomy, however, has made available a group of subjects with discrete, circumscribed lesions involving the globus pallidus and thalamus and provides the opportunity to empirically test theories of subcortical

participation. The preliminary findings reported in relation to the two cases described in the present chapter lend support to the suggestion that examination of the pre- and post-surgical language abilities of subjects with stereotactic neurosurgically induced lesion in the globus pallidus and thalamus has the potential to further inform debate regarding the hypothesised roles of subcortical structures in language.

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SECTION 2

COGNITIVE AND PSYCHOLINGUISTIC APPROACHES TO APHASIA THERAPY

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Chapter 6

COGNITIVE NEUROPSYCHOLOGICAL APPROACHES TO APHASIA THERAPY: AN OVERVIEW

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APPROACHES TO APHASIA THERAPY

The three most widely applied aphasia therapy approaches are the communicative approach, the so-called linguistic approach and the cognitive approach. These correspond to the classification of language impairments proposed by Schwartz and Whyte (1992) following the general I(mpairment) D(isability) H(andicap)-scheme of the World Health Organisation (WHO, 1980) into **I**mpairments of cognitive language functions, linguistic **D**isabilities, and communicative or psychosocial **H**andicaps.

Cognitive impairments are described with respect to explicit psycho- and neurolinguistic models. For example, agrammatic language production has been characterised in some models as a lexical impairment with respect to function words (Bradley *et al.*, 1980), in others as an impairment of syntactic processing (cf. Grodzinsky, 1984). The so-called linguistic disabilities, on the other hand, are not defined with reference to specific theoretical systems but in pretheoretical terms with respect to some language performances, especially those which are relevant for daily life such as spontaneous speech or comprehension. Communicative handicaps are defined in terms of the psychosocial consequences of the language disorder such as reduced functioning in employment or in the family.

In the major aphasia therapy approaches, different IDH-levels are emphasised. The aim of the communicative approach is to optimise the patient's communicative potential rather than

improve his linguistic disabilities (cf. Blomert, 1990; Davis and Wilcox, 1985; Holland, 1991). Patients have to learn to use the remaining verbal and nonverbal means of expression in a flexible way in order to achieve efficient interaction with the communication partner. As a consequence, linguistically incorrect reactions are not necessarily corrected by the therapist as long as they do not influence interaction.

The so-called linguistic approach (LA) and the cognitive approach (CA) both address the language impairment directly. However, they differ in important respects. Whereas LA-methods focus on surface symptoms of the language disorder, CA emphasizes hypotheses about the functional origin of impairment. The difference between LA and CA can be illustrated with a patient who has an impairment in writing to dictation. Intervention within LA would treat writing to dictation. Intervention within CA would first analyse the possible causes underlying the impairment. The patient's performance pattern may be obtained by using different tasks and various stimulus materials based on a model of normal processing. Following this detailed deficit analysis, the functional impairment can then be localised in the model and training procedures can be developed which either attempt to reactivate the impaired subprocedures or to circumvent them by means of alternative processing routes (cf. Howard and Patterson, 1989). For example, if it is found that the impairment in writing to dictation is due to dysfunctioning auditory processes, the treatment will focus on auditory processing such as auditory discrimination of minimal pairs (rat/mat) in order to reduce the problems in writing to dictation as well as in all other tasks relying on auditory processing.

Therapy studies within CA thus refer to psycho- and neurolinguistic models of language processing in the design and execution of therapy and the development of therapy evaluation measures. A basic assumption of these models is their modularity, i.e., information models of language processing contain dedicated subsystems which are interconnected. Another basic notion is that of transparency between the normal models and impairments, postulating that acquired language disorders can be explained as functional impairments of one or more cognitive subsystems in the normal model, and that the basic structure of the normal system is retained in the impaired one. A third essential ingredient is the detection and distinction between dissociated and associated impairments, as the models predict multiple qualitatively different unimodal impairments which do not correspond to the association of symptoms in the taxonomy of aphasic syndromes.

THE EFFICACY OF APHASIA THERAPY

As in neuropsychology in general, there has been a major shift of paradigm in the last twenty years in the study of the efficacy of aphasia therapy. This shift was associated with the

general change of paradigm from group studies to single case studies. Traditional approaches to aphasia therapy research were largely based on clinical studies using the group study paradigm. A central issue within this approach was the study of the efficacy of language intervention using the sampling method. A sample of aphasic patients was treated with a certain intensity of therapy for a particular time, and language performances were measured before and after the therapy as a dependent variable. In order to reduce the interfering influence of spontaneous recovery, the sample was usually restricted to chronic patients (e.g. Poeck *et al.*, 1989).

Other studies within this paradigm attempted to approach the standards of medico-clinical research, in which randomized matched samples are used (e.g. Mazzoni *et al.*, 1995). They tried to establish two samples of aphasic patients which were as comparable as possible in terms of type and severity of aphasic symptoms. Patients who underwent treatment constituted the experimental group, patients who did not functioned as a control group. If there was better recovery in the experimental group than in the control group, this was seen as evidence of the efficacy of language intervention.

However, there were strong ethical reasons for rejecting that control groups be established by therapy denial. An ethically more viable alternative was the use of control groups consisting of patients who did not receive therapy for "natural" reasons, e.g. due to the unavailability of speech therapy in the geographical region (Basso *et al.*, 1979). This method was criticised on methodological grounds (cf. Wertz, 1992), since comparability of these patients to the experimental group was unlikely.

Studies on the efficacy of language intervention conducted within this paradigm were equivocal in several respects. There were studies which demonstrated positive results and showed that the recovery process can be influenced by therapeutic intervention (e.g. Poeck *et al.*, 1989; Wertz *et al.*, 1986), whereas other studies were unable to find a positive influence of language therapy on recovery (e.g. Lincoln *et al.*, 1984). In addition to the empirical (e.g. David *et al.*, 1982) and methodological (e.g. Schoonen, 1991) weaknesses of studies with a positive as well as a negative outcome, it remains unclear whether the results were contradictory because certain patients or impairments respond better to language therapy, or because certain therapy methods or therapeutic processes are more efficient than others. Given that there was no theoretical foundation nor experimental control of the contents and design of the therapy measures, these questions cannot be answered. Clearly, with the heterogeneity of impairments inherent in patient groups, there must have been a large variety of individual therapy measures.

Some of the studies also documented a lack of maintenance effects (cf. Howard *et al.*, 1985a), so that clinically relevant generalisations could not be made. Therapy effects which were statistically significant in structured therapy situations and could be demonstrated during and after therapy had already vanished within four weeks post-therapy. Thus, they were of little use for the communicative daily requirements of the patient (cf. Fratalli, 1998). Several authors therefore required that an evaluation of the "clinical significance" of therapy effects (cf. Barlow and Hersen, 1984) should involve the demonstration of a widespread and stable generalisation of learning effects on untreated performances and materials (e.g. Kearns, 1992; Thompson, 1989).

These weaknesses favoured a shift from the group study approach to the single case approach and a change of focus on treatment from so-called linguistic or communicative disabilities to that of impaired cognitive language functions. Such cognitive approaches attempt to reliably demonstrate for each single case a causal relation between particular therapy procedures for particular impairments and the subsequent improvement of linguistic performances. In addition, their aim is to provide a theoretical explanation of the relation between cause and effect in order to derive generalisable conclusions on the cognitive learning mechanisms which can be triggered by language therapeutic intervention (cf. Kearns, 1992; Thompson, 1989).

COGNITIVE APPROACHES TO LANGUAGE IMPAIRMENTS

A central issue of cognitive therapy research is whether it is possible to determine which subsystems of a normal processing model can be supported by a specific training procedure if a deficit has been identified. A final goal is to make predictions on the success or failure of specific treatment methods for a particular deficit from treated patients to others, allowing inter-individual generalisations. In order to answer such questions on the specificity and generalisability of treatment effects, at least the following requirements should be met by the therapy design (cf. Kearns, 1992; Poling and Grossett, 1986).

The behaviour to be changed, i.e., the dependent variable, has to be clearly defined and evaluable in order to provide an operational definition of the target behaviour. This requires the determination of a stable baseline, so that the occurrence of the target behaviour in the baseline can be used as a predictor for the expected frequency of the behaviour without intervention. One way to reduce the effect of naturally occurring fluctuations in the target behaviour is to take continued baseline measures during an intervention-free observation stage (Stage A).

The therapy method used in the subsequent intervention stage (Stage B) should be applied consistently and according to a preplanned therapy protocol in order to allow an evaluation of its efficacy. The protocol contains at least an explicit description of the sequence and contents of the procedures and the therapy conditions in order to reduce variability and subjectivity and to allow replication and systematic modification.

The experimental design should make it possible to trace changes in the dependent variables to a specific impact of the therapy methods and control the influence of potential intervening variables. It is therefore advisable to use logically consistent intervention steps and keep them to a minimum. For example, in a naming therapy, phonemic and semantic cues should preferably not be mixed. Although ABA-designs are sufficient to examine the specificity of therapy effects, ABABA-designs have the advantage that intra-individual replicability can be examined (cf. Coltheart, 1983).

A distinction which is crucial in the cognitive approach is that between theoretically expected and unexpected intra-individual generalisations, which requires multiple baseline designs (cf. Barlow and Hersen, 1984). They have the advantage that therapy-free observation stages become superfluous. Theoretically expected generalisation effects are defined with respect to a specific model. In line with the architecture of the cognitive model, direct intervention of a particular function should indirectly influence untreated related behaviours but not functionally unrelated ones. In multiple baseline designs, the frequency of the target behaviour and of theoretically related and unrelated behaviours is examined before the intervention. If following intervention there are greater changes for intended and related parameters than for others, it may be concluded that positive effects are a direct result of the applied intervention.

An example based on dual route models of word processing can illustrate this point. In such models (cf. Morton and Patterson, 1980), oral naming and reading aloud via the lexical route both require activation of word forms in the phonological output lexicon. Functional impairments of this subsystem will therefore influence reading aloud as well as oral naming.
An expected generalisation effect would be that training oral naming may not only specifically influence naming but also reading aloud by reactivation of the word forms in the phonological output lexicon. There would be an unexpected generalisation effect, on the other hand, if training oral naming would specifically influence reading aloud nonwords or written naming, which do not rely on lexical phonology.

For an analysis of these generalisation effects, multiple baselines are obtained during the preintervention deficit analysis (Stage A) to assess the state of oral and written naming,

reading aloud words and nonwords. This is followed by the cognitive intervention (Stage B), for which therapy of oral naming could be designed, e.g. presenting a fixed sequence of different phonological cues when the response is incorrect. In the post-intervention deficit analysis (Stage A'), the state of oral and written naming, reading aloud words and nonwords is again determined with multiple baselines. An analysis of the specificity of the therapy effects can then not only establish task specific training effects (oral naming), but also expected (word reading) and unexpected (nonword reading, written naming) generalisation effects.

To compare the adequacy of different intervention methods, cross-over designs may be used, which contain components of the ABA-design and the multiple baseline approach (cf. Coltheart, 1983; Howard, 1986; Pring, 1986). A comparison of methods is sensible in the case of a single impairment in a patient to examine the differential efficacy of the intervention procedures. It may also be used in patients if they have multiple impairments which are due to breakdowns in at least partly different subsystems. In this case, the specificity and generalisation of theoretically independent, consecutively applied intervention measures can be examined. Method A, for example, is theoretically expected to be relevant for the treatment of performance A but not B, whereas method B is designed specifically for treating performance B but not A. If it turns out that method A indeed leads to improvement of performance A, in accordance with the theory, but not to improvement of performance B, whereas the reverse happens with method B, this is evidence for the specific efficacy of the therapy procedures.

The use of cross-over-designs may also help to identify the exact locus of impairment if baseline analysis has not been able to exclude some alternatives. For example, if a patient has difficulties in reading aloud words, this may be due to problems with the graphemic input lexicon or the phonological output lexicon. To decide between these alternatives, two tasks may be developed, reading aloud words (task 1) and graphemic word to picture matching (task 2). Within the dual route models of word processing, word reading is assumed to rely on cognitive subsystems A (the graphemic input lexicon), B (the semantic system) and C (the phonological output lexicon). Graphemic word to picture matching uses subsystems A (the graphemic input lexicon) and B (the semantic system) but not C (the phonological output lexicon). If therapy effects specifically occur on intervention using task 1, reading, but not task 2, word-to-picture matching, training of reading must have caused a change in C, and there was obviously an impairment in the phonological output lexicon rather than in the semantic system per se.

The final goal of therapy is that the improvement of language abilities is relevant to daily activities. In its applied component, cognitive therapy research also deals with the issue whether the therapy effect is goal-oriented with respect to its practical utility. It is of course impossible to completely evaluate the global question whether particular therapy measures give rise to particular beneficial effects for the patient's social skills and his quality of life for the entire life span, so that it is necessary to establish hierarchies of partial goals (cf. Wottawa and Thierau, 1990) to evaluate the applicability of a language therapy measure. Relevant questions here concern intra-individual generalisations, transfer and maintenance of the effect and the general efficiency of the intervention measure (cf. Fratalli, 1998; Holland *et al.*, 1996).

With respect to intra-individual generalisation effects (Seron, 1997; Thompson and Kearns, 1991), a distinction is made between across-item-generalisation and across-task generalisation. If there is across-item generalisation, there is not merely improved access to item-specific knowledge resources but a reactivation may be postulated of the cognitive processes and strategies which the patient can independently apply to new stimulus material.
If there is across-task-generalisation, this shows that there is not merely improved performance on the treated task but in the functional component subserving this task and thus also of other, untreated tasks relying on the functioning of this component.

The presence of transfer effects signal the applicability of the reacquired processing routes not only in the highly structured therapy situation but also in ecologically valid tasks ("functional outcome measures") . If possible, they should refer to theoretical systems (cf. Grosz and Snyder, 1986), so that there is not only a valid analysis of the results but clinically successful and unsuccessful therapy procedures also receive a theoretical explanation..

Maintenance effect can only be observed if there is repetition of the baseline-assessment over larger time distances after finishing the intervention.

Whether a cognitive intervention is efficient or not depends on whether the clinical benefits are adequately related to the required burden, e.g. the duration and intensity of the therapy, the stress on the patient, etc. This requirement is best met by studies in which methods are compared to examine the relative benefit of two approaches with a comparable burden on the therapist and the patient.

There may be several reasons for the limited scope or the lack of generalisation and transfer effects of a particular intervention. If there are underlying associated deficits and the cause of the communication disorder is multifactorial (cf. Schwartz and Whyte, 1992), the successful

treatment of one specific deficit may not be generalisable. Consequently, as many deficits as possible should be assessed and specifically treated, if possible successively, and the effect of each new intervention should also be examined at the level of communicative abilities.

Although no single cognitive therapy study has met all of these requirements, a number of studies have contributed to the development of quite rigid standards in the field. This is illustrated by the therapy studies on deficits in segmental written language processing discussed below.

EXAMPLES OF COGNITIVE THERAPY RESEARCH

Some of the earliest cognitive interventions have dealt with impairments of central dyslexias and dysgraphias. De Partz (1986), for example, described a French speaking patient SP, who was more severely impaired in reading nonwords than words. His reading of words was further characterized by the presence of a part of speech effect (content words: 51% correct, function words: 11% correct) and the absence of a regularity effect (67% correct for regular GPC-words, 74% for irregulars). Thus, the results of a model-based deficit analysis revealed severe impairment of Grapheme Phoneme Conversion (GPC), a partial impairment of lexical processing in reading aloud, and it could be concluded that the patient's reading was lexically guided.

Accordingly, the therapy goals and procedures of the dyslexia therapy aimed at a reactivation of GPC by using the better preserved lexical reading route. The patient should learn to use his lexical knowledge in order to support segmental processing. In a first step, SP had to associate a relay word to each grapheme (e.g. <A> → "allo") and then produce the target phoneme in isolation relying on the word onset. After the grapheme-phoneme-correspondences had been stabilised in this way, SP had to learn in a next step to blend the phonemes into simple words and nonwords. In a third step, the same method was used to retrain context-dependent suprasegmental GPC-rules (e.g. <au> → "eau").

After an intensive training stage of approximately nine months, SP showed significant improvements in reading words of different parts of speech and in nonword reading. An evaluation of the specific effect of the therapy showed task-specific generalisation effects and a reactivation of GPC. Reading (of words and nonwords) before and after therapy resulted in 72% and 14% errors, respectively. The difference between words and nonwords, which was significant before therapy with a word superiority effect (55% versus 7% correct), had become non-significant. The pretherapeutic concreteness effect had disappeared, and a regularity effect was visible only post-therapeutically (regular GPC words correct, irregulars

20% errors). The patient now showed more frequent regularisation errors than before therapy in reading irregular words (e.g. <c> in <celle> was read as <k> as in <carole>), which usually characterises patients with surface dyslexia.

The successful reactivation of the segmental route apparently had led to a partial neglect of the lexical reading routes and thus to increasing errors in words with irregular GPC. However, the large number of correct responses in reading irregular GPC-words (80%) showed that the lexical route was not blocked but that both lexical and non-lexical routes were now often used adequately and in parallel.

In a subsequent study (Bachy-Langedock and de Partz, 1989), SP's deficit in oral naming was treated by relying on his improved reading skills. The deficit analysis had shown an impairment in oral naming (48% correct) with significantly better performance in written naming, i.e. there was impaired access to the phonological output lexicon.

The therapy goals and procedures in the naming therapy were a reduction of the activation threshold of word forms in the phonological output lexicon. This reactivation relied on the partially recovered reading by progressing from internal written naming to reading aloud the so generated internal graphemic form and thus producing a phonological output name.

The therapy procedure developed by de Partz was used with an English patient TC by Nickels (1992). This patient also demonstrated severe impairments in reading nonwords (0% correct), indicating a deficit of the segmental reading route. However, his word reading was also severely impaired and the presence of an imageability effect pointed toward deep dyslexia (28% correct for words of high imageability, 0% correct for words of low imageability). Although it was possible to retrain the GPC-rules by means of relay words with this patient, he was unable to learn to blend the single segments, so that reading nonwords could not be improved in the five months of therapy. However, there was a significant improvement of both oral naming and word reading following therapy. These generalisation effects can be explained if one assumes that the patient was successful in self-cueing. Due to the relay word training for single segments, TC was now able by himself to find the phoneme corresponding to the onset of the graphemic word, thus facilitating lexical access in reading. The improvement of oral naming may be explained by assuming that TC spontaneously developed the mechanism used for explicit training by Bachy-Languedoc and de Partz (1989). Like SP, TC's written naming was better than his oral naming (pre-therapy oral naming: 12% correct, written naming 65%). After therapy, he may have relied on internal written naming and then used phonemic self-cueing to produce the oral name.

It is not clear why the therapy results for patients TC and SP were different even though their deficits were similar. The problem of generalisability across patients is highlighted in another important study on the cognitive therapy of graphemic processing by Hillis and Caramazza (1994), in which the potentials and limits of cognitive therapy research become obvious. By using intra- and interindividual comparisons of methods, the authors clearly demonstrated that there may be different therapy effects although similar deficits are treated with the same method.

The study included two english speaking dyslexic patients, HW and PM, who had a similar impairment. HW had deep dyslexia, and the deficit was analysed as impaired access to the phonological output lexicon and impaired GPC. She was unable to read nonwords and had a part of speech effect in word reading (nouns: 38% correct, verbs: 18% correct). Furthermore, HW was unable to write nonwords. PM was also unable to read nonwords but words were read correctly in 76% of cases. However, there was a strong effect of part of speech, so that rare words and non-nouns were only read correctly in 10% of cases. Both patients thus had a deficit of the segmental reading route with associated impairments to the lexical route. This was localised at the level of the phonological output lexicon, since both patients had preserved auditory word comprehension.

The patients were treated with the same method and intensity. The intervention relied on a version of the relay method applied in the studies by de Partz (1986) and Nickels (1992). HW did not benefit from this therapy method. Unlike TC (Nickels, 1992), who could relearn GPC-rules, yet was unable to blend the segments and to read nonwords after this training, HW could often not derive the GPC from the relay word. HW also did not show a lexical self-cueing effect which could have been evoked by at least partially successful GPC. Remarkably, a different training consisting of reading aloud words for several weeks led to a significant across-task generalisation in oral naming, at least for the trained items.

Patient PM, whose reading pattern was similar to that of HW, showed important improvement with the relay method including phonemic self-cueing. In reading untrained high frequent nouns, her performance improved from 76% correct pre-treatment to 89% post-treatment

Carlomagno and Parlato (1989) conducted a dysgraphia therapy with the relay method similar to the reading therapies described above. Clinically, their Italian patient had a moderate impairment of reading and a severe writing impairment without accompanying aphasia. The reading assessment showed partial impairments of both the sublexical and the lexical routes, but the word superiority in reading showed that the segmental route was relatively more severely impaired. However, lower performance on words with irregular word stress revealed

the lexical impairment. Partial preservation of the segmental route was demonstrated by the correct realisation of context-sensitive GPC-rules and complex graphemes, which pointed toward some residual sublexical knowledge at the syllable level.

Writing was impaired in all modalities, writing to dictation, written naming, spontaneous writing and, except for an effect of length, there were no specific variables affecting writing. The authors analysed the deficit as a partial impairment of both the lexical and sublexical writing routes.

Based on the deficit analysis and taking into account the high transparency of the PGC-rules in Italian, the authors considered a reactivation of segmental processing the most meaningful from a therapeutic point of view. This was supported by lexical processing. Given that there was some evidence for sensitivity to the graphemic syllable in reading, the patient was trained to render CV-syllables graphemically by associating them with the onset of an Italian city used as relay word (e.g. "Pa-Palermo"). The choice of these relay words was based on the patient's profession in the Italian railway system. Even though GPC and PGC-mechanisms are functionally independent in the dual route model, the authors assumed that the patient might monitor his written realisations of CV-syllables by reading aloud and then correcting them.

First, 30 CV-combinations spoken by the examiner had to be matched to their associated relay words, then the patient had to write the syllables to dictation. After a few sessions, he reached criterion (95% correct), and he then had to write 2- to 3-syllabic nonwords to dictation. Criterion (95% correct) was reached within one month. In the following four months, further neologistic stimuli were trained with CV-combinations as well as with consonant clusters and diphthongs. In stimuli with consonant clusters, the patient was asked to process them segmentally rather than by the syllabic strategy. However, he refused to do this, segmented them syllabically and looked himself for appropriate relay words (e.g. "spa" →"Spagna").

There were significant improvements in spontaneous writing, from 20 of 45 words incorrect pre-therapy to only 3 of 71 after the therapy in comparable writing samples. After about one month, there were also significant generalisations to word reading (from 50% correct pre-therapy to 80% correct), probably due to a spontaneous transfer of the relay strategy to reading.

Hillis and Caramazza (1994) reported on two english-speaking patients who underwent segmental writing therapy. One patient, SJD showed a pattern of deep dysgraphia. There was

impaired access to the graphemic output lexicon and a deficit of PGC. SJD was also unable to read nonwords, reflecting a deficit of GPC as well.

The aim of the dysgraphia therapy was to use the partially preserved lexical writing abilities to retrain sublexical writing, in order to improve writing in general. Again, the relay word strategy was used, but in this case with single phonemes, given that English is less transparent than Italian. Within 12 sessions, SJD not only learned 30 PGC-conversions, but her lexical access in reading words also improved.

Patient PM, whose reading had successfully improved by using the relay method for treating segmental reading, was also trained by this method to improve her writing. After 5 sessions, PM's writing had improved for trained items but not for untrained ones, so the authors discontinued the therapy.

Even though PM and SJD had similar deficits in writing, the segmental writing therapy which functioned for SJD and even generalised to reading did not function for PM's writing, although a similar therapy for PM's reading disorder had been successful and had generalised to untrained items. The same segmental reading therapy applied to patient HW with a reading deficit similar to PM's was without effect, but a lexical therapy led to an item-specific generalisation in HW's oral naming.

DISCUSSION: POTENTIALS AND LIMITS OF COGNITIVE APPROACHES

An important question raised in the cognitive studies reviewed here is why certain therapeutic procedures but not others lead to the desired results with certain patients, or why some patients but not others benefit from a particular intervention procedure. At present, there are still serious problems with the definition of the comparability of cognitive deficits. It is still unclear for which aspects of the individual performance profile and to what extent two patients should be similar in order to justify expectations on the generalisation of an appropriate therapy method.

For instance, the two patients HW and PM in the study of Caramazza and Hillis (1994) had deficits which seemed to be comparable from a model-theoretic view. They had an impairment of the segmental reading route and associated impairments of the phonological output lexicon. Although they were treated with the same relay-word therapy method and with comparable intensity, treatment with a lexically supported segmental reading strategy had no effect for HW, whereas PM made "dramatic" improvement. Interestingly, PM, who also had an impairment of the segmental writing route, did not benefit from a similar therapy

to ammeliorate segmental writing to dictation. However, SJD, who had a similar impairment of PGC, did benefit from a lexical relay word method and writing to dictation improved significantly.

It is hard to explain on the basis of the model why there are such interindividual differences and why there are intraindividual differences, for example in the case of PM, who benefited from a relay word method for segmental reading but not for writing. A conceptual limit of cognitive models of language processing or the deficit analyses based on them is that they give a simplified presentation of the actual processing procedures, and the assessment of qualitatively different impairments within a component is highly unsatisfactory. In addition, they do not yet allow a well-motivated selection between alternative therapy methods nor do they formulate theoretically based therapy goals and procedures. It can only be speculated that differential therapy effects are due to different degrees of impairment, or to different subcomponents of PGC which are impaired. Alternatively, the particular type of cues may use certain cognitive resources which are available for one patient but not the other, or different non-cognitive factors such as general psychological condition, supporting measures from friends and family, kind and extent of the brain lesion may play a determining role.

A basic weakness is that cognitive models are quite sophisticated at outlining normal and pathological processing procedures. However, in order to explain the efficiency of cognitive training on language processing skills, one would need models containing those cognitive and neuronal mechanisms responsible for the compensation and/or reacquisition of impaired performances. In order to examine such questions more coherently, several authors advocate that new models on cognitive learning mechanisms must be developed in addition to the existing cognitive models of language (e.g. Hillis and Caramazza, 1994; Schwartz and Whyte, 1992; Wilson and Patterson, 1990).

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Chapter 7

LURIAN APPROACH TO APHASIA THERAPY - A REVIEW

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INTRODUCTION

Luria's extensive work in the field of human cognition influenced many researchers, clinical aphasiologists not only in Europe but also professionals in the rest of the word. In the present review I would like to stress the unique feature of his work in clinical aphasiology, namely, an approach with logical link between diagnosis of aphasia and its treatment. It is obvious that the time period when Luria developed his theory (first publication appeared before 1940) strongly influenced his view on processes such as language, cognition, etc. From the point of view of contemporary linguistics and cognitive (neuro)psychology some concepts and terminology would sound unusual or simplified. In this review, however, I did not change the original terminology used in Luria's publications.

When reading Luria's publications, we should remember, that his theoretical framework was developed during four decades and Luria himself recognized the importance of permanent reflection on new linguistic and psychological theories into his work. When reading his books and other publications it is obvious, that he was able to modify concepts when necessary and apply new ideas to his own work. Caplan (1987, p.132) points out that "Luria provided the first reasonably detailed model of language processing related to aphasia and to the brain. These models became more complex as linguists and psychologists began to study aphasia in depth".

The aim of this review is to show how Luria's theoretical paradigm was implemented into its

clinical application, especially to treatment for aphasia. I would like to highlight some similarities between Luria's concept to aphasia therapy and contemporary cognitive-neuropsychological approach. Of course, those similarities are more on a general level, than in detail. Both approaches have provided a ground for understanding the mechanisms of aphasic disturbances using a theoretical model of cognitive mechanism.

Almost in every survey on aphasia, there is at least a comment on Luria's contribution to this field. On the other hand, we must agree with A. Holland who writes in the foreword to "An Introduction to Luria's Aphasiology", written by Kagan and Saling (1992, p. xiii), "that many speech and language pathologists who are interested in aphasiology all recognize the importance of A. R. Luria and his monumental contributions to aphasiology, but only a few of them truly understand what those contributions are and how to apply those ideas to their own work". It should be included, that this may be true for professionals in the North American continent since there has been a more wide application of Luria's treatment procedures into clinical practice especially in countries of Eastern and Central Europe and Scandinavia. However, well designed controlled experimental studies aiming the efficacy of those procedures are rather rare.

Mitchum (1994, p. 22) states, that surprisingly "that approach with greatest potential to influence both diagnosis and treatment (i.e. Luria) has had minimal impact in either area". Of course, this statement is not completely true, at least for the current Russian neuropsychology, represented by T. Akhutina, E.D. Khomskaya, L. S. Tsvekova, J. M. Glozman, etc.

The possible explanation for the limited application in clinical settings is that, Luria and his coworkers did not prepare a "simplistic manual for aphasia therapy". In a sense it is not possible. To apply Luria's ideas into clinical practice in our opinion means that, (1) the clinician must be able to appreciate the relationship between the brain and higher cortical functions; (2) in assessing individuals following brain damage the clinician must go beyond the description of symptoms and has to identify the possible underlying mechanism of the deficit (or deficits) and, (3) in the treatment the role of the therapist is to structure the therapy process by implementing therapy techniques appropriate to the detected internal structure of the deficit and not only to the symptoms.

In the Lurian approach *the framework for therapy* is stressed more strongly, than the description of a particular therapy technique. Kagan and Saling (1992) states that a wide array of current clinical techniques developed since Luria can be incorporated into a therapy program along the line he suggests. One of the appealing aspects of his work is that it does not

preclude the use of techniques developed by others but often enhances their use by placing them in a meaningful context.

The recent review is based on two of Luria's basic publications: "The human brain and psychological functions" originally published in 1963 and "Fundamentals of neuropsychology", published in 1973. Information about treatment strategies in detail was described by Luria's close coworker L. S. Tsvetkova (1989).

THEORETICAL FRAMEWORK

Luria's approach to cognitive functions was founded upon the basic concept of (1) function, (2) localization and (3) symptom. Luria revised the classical view of neuroanatomic localization of cognitive functions. This revision was based on Vygotsky's premises of social determination of higher psychological functions and the thesis of the interiorization during the individual development.

Luria defined *function* as a complex and dynamic functional system, which is constituted from integrated basic components. Even the functional system outwardly looks as a unitary function; the inner structure of the functional system is complex. This complexity is created by many different components of the particular functional system. Impairment of any component can lead to the disintegration of the whole functional system or all systems, which contain this component. For instance, the process of writing can be disintegrated differently when different parts of the brain are damaged. In a case of the lesion in the acoustic analyzators (the upper part of left temporal lobe), copying and writing of highly automatized words are preserved, since it does not require acoustic analysis of words, but writing on dictation will be strongly affected, since in this process acoustic analysis of phonemes plays an important role. The structure of the deficit will be different when lower parts of the left premotor area are involved. Perseverations in writing will occur, because of the problem in sequencing elements (impaired "kinetic melody"), such as graphemes or syllables, but writing of isolated letter will be preserved.

The functional system is dynamic, which means that the invariant task (i.e. reading) can be realized in many different ways (reading written words, but also "reading with fingers" in Braille). Functional system has a "polyreceptoric character" in a sense that it receives information from different parts of the brain. This means, that complex cognitive processes (such as language) are not localized in narrow circumscribed areas of the brain. The

components of the functional system are spatially independent, and are formed in a course of the development. Functional system has "chronogen" *localization* in the brain and the construction of the functional system is strongly influenced by the process of learning and training (i.e. there is a difference between involved components of the functional system of reading in the young child who only starts to read and a skilled adult reader). During development the inner structure of the cognitive function is changing, some components are less involved than in the earlier stages, which also means, that the localization of that functional system differs.

From the above mentioned it is clear that the symptom is only an isolated manifestation of the disintegrated functional system. An understanding of the structure of the functional system - *an analysis of the syndrome* - is crucial for detecting the primary problem (a breakdown in a general underlying mechanism). Primary problem, in Luria's framework, can result externally heterogeneous but internally interconnected symptoms. For example the structure of processes such as space orientation, calculation and understanding of logic-grammatical structures involve common component in their structure, and the breakdown of this element - in this case a lesion in lower parietal and occipital areas - leads to the disintegration of all those processes.

It is logical then, that the *classification of different types of aphasia is related to the primary problem* (the internal structure of the deficit), which is a result of the focal damage of the brain. For instance, damage of secondary zones of frontal lobe results in disintegration of skilled sequential movement, also called a "kinetic melody", which outwardly is manifested as a problem in initiation and perseveration and it is labeled as "efferent motor aphasia". For this type of aphasia there is problem in sequencing elements, such as syllables or words, which is observed as perseveration of those elements at different linguistic levels. However, for patients with efferent motor aphasia articulation of isolated sounds is in most cases preserved.

This theoretical construct of human cognition guides the *assessment.* Briefly, the goal of the diagnostic procedure is to identify the primary problem, the internal structure of the deficit. To achieve this goal, one has to compare different cognitive processes such as language, gnosis and praxis, memory, intellectual functions, etc. This comprehensive assessment is required, since according to Luria; non-linguistic functions are also accomplished by those areas of the brain, which play an important role in language functions. Luria believed that the primary problem can result in not only language deficits, but can cause also apraxia, agnosia, acalculia, etc. For example, if the brain damage results in the disintegration of sequential movements (breaking kinetic melody), clinically it will manifest not only as efferent motor aphasia (with

typical symptoms such as perseveration) but also as kinetic apraxia and agraphia and also motor perseverations.

The assessment includes introductory conversation with patients, in which the clinician formulates "working hypothesis" about the possible mechanism of the deficit(s). The following, very comprehensive testing contains "probes", in which the clinician analyzes patients accomplishment in different tasks such as speech, receptive and expressive language, reading and writing, praxis, gnosis, memory, intellectual functions. Syndrome analysis is closely related to task analysis, and reveals the psychological structure of each task. This type of analysis not only permits to understand why the patients was poor or unable to perform a given task, but also to see what tasks with similar cognitive structure will present difficulties for this individual (Glozman, Tupper, 1995). As Mitchum (1994) points out, the complexity of this kind of assessment, which requires great experience in using process oriented diagnostics, may be responsible for the limited influence of his approach in contemporary clinical aphasiology. Unfortunately we have to agree with her that the published formalization of Luria's assessment methodology, developed by Christensen (1975) remained outside the repertoire of most speech and language pathologists.

At this point, I would like to stress some similarities between Luria's approach and the contemporary cognitive neuropsychological (CN) approach. Detailed and systematic analysis of single cases is a basic requirement for both approaches. The aim of assessment in both approaches is to identify impaired and intact components within the framework of cognitive processing models. In Luria's methodology it means to identify the primary problem (deficit), which was in the background of impairment of different cognitive function. On the basis of this syndrome analysis the therapist builds up a treatment strategy. Hillis (1998, p. 659) formulates that "a detailed analysis of language performance, with the goal of identifying which cognitive mechanism underlying a specific language task are impaired and which are spared, is a productive way focus therapeutic intervention." Contrary to Luria's theoretical paradigm, where syndrome analysis leads not only to the detailed description of structure of the impaired psychological function but also to identification of the proposed lesion in the brain, in the CN approach, the interest is placed on the identification and location of functional lesion within the information processing model. Mitchum (1994) points out, that in attempting to identify the locus of deviation from normal event, a model of normal processing may be consulted at any time. This kind of "consultation" is apparent in Lurian diagnostic procedure, when the clinician analyzes the internal structure of a particular function. Both assessment approaches are qualitative in essence, and are similar in regard, that the clinician hypothesis about the impaired component (factor) is refined in the course of the complex assessment. Cognitive

dysfunction is revealed in the form of performance errors in different functions (such as naming, reading, writing, etc.). Both approaches offer opportunity to individualize assessment. Mitchum (1994, p.32) points out that the greatest advantage of the CN approach to aphasia is it potential to reveal the underlying source of the impairment. The goal to go beyond verification *that* symptom exists and identify *why* a symptom exists aligns the CN approach with the analytical approach of Luria. This qualitative assessment creates then a basis for therapy. The identification of the primary problem is significant for formulation of the goal of remediation.

LURIAN APPROACH TO APHASIA THERAPY

In the following I would like to summarize Luria's concept on aphasia treatment in describing methodological and psychological principles of remediation, besides depicting principles and requirements on concrete methods.

Luria strongly emphasized that remediation must always be directed toward the whole person, not on his/her isolated abilities. Activities are influenced by motivation, and as such, character of the motivation significantly stimulates the effectiveness of any activities. Communication is induced and directed by motives. According to Glozman (1981) a study of emotions and motives of individuals with aphasia are very important to understand many phenomena of aphasic disturbances of verbal communication as well as to find adequate ways for its treatment.

Patients personality is taken into account during the whole process of treatment since in the course of the individual development people acquire different social experiences, many of them remain also after the brain damage. Impaired abilities have to be remediated in the framework of patient's practical activities. Luria and his coworkers from the very beginning stressed the importance of group therapy for these patients. All methods used in individual and group therapy were designed to restore "communicative function of speech", which is according to Luria the central deficit in aphasia and creates a barrier between patients and his/her environment. This aspect of the therapy is very similar to the functional approach in aphasia therapy.

In formulating general psychological principles, Luria underlines the role of programming of the process of remediation. This means, that the therapist in certain phases of the therapy

process divides the tasks into special steps, which can be realized first with the help of the therapist but then also independently.

There are five principles of remedial teaching, which Luria and his colleagues called methodological principles (Tsvetkova, 1989):

(1) Identification of the primary problem, which is a basic task before planning any treatment strategy. The damage of different parts of the brain can result in disintegration of the same functional system, but the mechanism of this deficit will be different and will depend on the localization of lesion in the brain.

(2) Inclusion of intact systems of analyzators (afferentations); this means, that in the process of reorganization of the impaired function (i.e. reading) a new link is incorporated into the functional system (i.e. tactile-kinesthetic analysis). This is an example for intersystemic reorganization of the functional system.

(3) Transfer of the function to hierarchically higher (voluntary, intentional) or lower (involuntary, automatic) level of realization. This vertical shift within the functional system is called as intrasystemic reorganization.

(4) Inclusion of intact cognitive processes, when language is impaired (i.e. memory, intellectual function, etc.). In the course of development all higher psychological functions are interrelated. The role of the language is important in many of them.

(5) Including control and feedback into therapy. This principle is based on the thesis of afferentation, which means controlling the realized activity with the original intention and making correction when needed (tape- and video-recording, praise, etc.).

Requirements on the methods are that they have to be appropriate to the mechanism of the deficit, not to a symptom. Techniques used at least at the beginning have to be indirect. For instance, when phonemic hearing is impaired - which leads to an inability to differentiate between similar sounds (b-d, p-b) and consecutively to impaired comprehension, spontaneous speaking, writing and reading -, the therapist does not address acoustic analysis, but tactile-kinesthetic and visual analyzators. Simultaneously, the focus is placed to the meaning of the target word.

Publications after Luria's death (e.g. Tsvetkova, 1988) implemented those principles and strategies into a comprehensive system. A detailed description of that therapy system is beyond the scope of this chapter.

I would like to illustrate the complexity of treatment procedure on the case of the treatment procedures for afferent *motor aphasia*, described in detail by Tsvetkova (1989). All the above

mentioned principles are incorporated into the therapy program for individuals with this specific impairment of spoken production. According to Luria, the primary problem (the impaired tactile-kinesthetic feedback) is manifested in production as incorrect position of the articulators. Individuals with afferent motor aphasia have mainly difficulties with production of phonemes that are similar in articulation. Patients with mild impairment usually do not have difficulties with automatic production of word or even sentence. When the focus of his/her attention is shifted to another cognitive task, for example sorting pictures into two categories, the patient usually spontaneously produces correct word and sentence. The problem is much more apparent when patients are focusing on their articulation (i.e. when he/she is asked to repeat). The above mentioned symptoms are present also in reading and writing.

Techniques utilized in the following procedure are indirect (any task is realized in connection with other activities), they have to move the patient's attention from articulation to acoustic and semantic components of his language and they have to facilitate spontaneous speech production.

In the first stage of the therapy the "primary problem" is not addressed directly, so the stimulus is always a meaningful unit (word or sentence) and not an isolated sound. The patient is asked to listen to the words or sentences and is not forced to imitate them. However, any spontaneous imitation is accepted. The therapist uses pictures of objects and actions, or objects and actions themselves. At the beginning only a limited number of high frequency words are used as a stimulus material when the therapist speaks about these objects. This is similar to the "semantic feature analysis", when the therapist discusses different aspects of the objects or actions. When doing so he/she is emphasizing the prosodic component of the word, in the sentence the target word is exaggerated. The purpose of this task is to stimulate the patient with a small group of words that he/she already knows but for which he/she may not have a consistent acoustic representation.

Afterwards, more active participation is encouraged from patients. First, he has to finish sentences like "The girl has a ball, but the boy does not have a..." and the picture simultaneously presented to him. Another type of task is one in which the patient has to name the object when the clinician describes it to him. Sometimes he is also asked to close his eyes and imagine the object and only then is asked to name it. When real objects are used, the patient is asked to take it, to include tactile-kinesthetic perception of objects. The aim of these tasks is to increase the number of words, which are correctly produced by a patient.

The next step is to teach the patient to analyze phonemes and the syllabic structure of the target word. Following this, programmed instructions are used: (1) imagine the structure of the word, (2) tell the sounds of the word, (3) match the plastic letter to the word, (4) copy or write the word form of the plastic letter, (5) write the word from memory, (6) read the word silently, (7) read the word aloud.

With the help of reading and writing the patient develops kinesthetic analysis of the word. Developing correct articulation using kinesthetic feedback is the aim of the following tasks. Now, the patient has to analyze similar phonemes. Written stimuli are used in this procedure, and the main goal is the identification and production of those phonemes (target phonemes are presented in different positions in the words). Later on only pictures are used to stimulate the production of the target phoneme.

A comprehensive and updated description of all strategies used in this kind of syndrome-specific therapy was done by Luria's close coworker Tsvetkova (1988, 1989). Methods delineated in her books are based on Luria's original ideas but also further developed on the basis of extensive clinical experiences.

Besides individual treatment, group therapy is employed from the beginning at the rehabilitation process (Tsvetkova, Glozman, Kalita, Maksimenko, Tsyganok, 1980). The theoretical framework and many therapy techniques described in that "small booklet" are in essence similar to those in contemporary functional approaches to aphasia therapy.

Clinical application of the Lurian approach in different aphasia syndromes was described not only by Luria's co-workers, but also outside of his laboratory. For instance, Gielewski's (1989) paper deals with the application of remediation programme based on Luria's work in-patients with sensory aphasia. Kagan and Saling (1992) outline a therapy plan for a patient with multiple primary problems (efferent and afferent organization of movements, simultaneous synthesis and audioverbal difficulties). From a methodological point of view, it should be noted that these studies are mostly illustrative case studies aiming to describe in detail patients' language profiles, identifying their major problems (primary deficits) and report on strategies successfully used in a treatment process.

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Chapter 8

THERAPY FOR LEXICAL DISORDERS

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THE STRUCTURE OF THE LEXICON

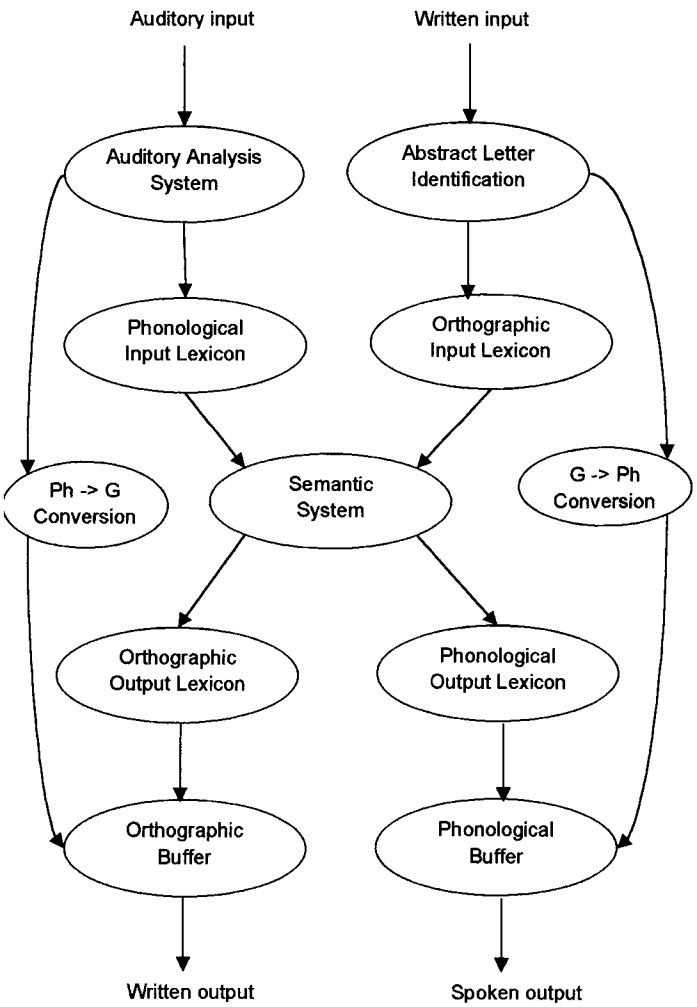
Before discussing therapy for lexical disorders it is important to agree on what is meant by lexical disorders. There are at least two possible meanings, one which refers to the form of the disorder -- lexical errors -- and one that refers to the origin of the errors -- damage to the lexical system. Although lexical errors and errors originating in the lexicon generally coincide it is important to keep this distinction in mind because therapy can be theoretically motivated only in case the origin of the error is taken into consideration and not only its form. I shall therefore briefly sketch a model of the structure of the lexical system which has been used as a reference point in analyzing the patients' functional damage.

One of the basic assumptions of cognitive neuropsychology is that a cognitive process can be characterized as a set of sub-processes that are sequentially computed in the course of cognitive performance. Figure 1 depicts a dual-route model of the lexical system.

A first distinction must be drawn between the meaning of a lexical item and its form. There are patients who have preserved meaning of concepts (they know what a "cat" or a "hammer" is) but they have lost information about the form (phonological and/or orthographic) of the words *cat* and *hammer*. Other patients show the opposite pattern: they have lost the meaning of concepts (maybe knowing that a cat is an animal but not which kind of animal it is) but have not lost the phonological and/or orthographic representations of words. A second generally accepted distinction is that between input and output lexicons. Allport and Funnell (1981), however, argued that a single phonological lexicon is used for the recognition and the production of spoken words (and a single orthographic lexicon is used for the recognition and production of written words). But a single phonological lexicon, according to Ellis and Young (1988), cannot explain performance of those patients, so called deep dysphasics, who produce

semantic paraphasias in repetition. Mediation from the phonological input to the phonological output lexicon through the semantic system easily explains semantic paraphasias in repetition. On a one-lexicon model the same representations in the single lexicon should be used for comprehension and production and it is not possible to explain semantic paraphasias.

Figure 1. A dual-route model of the lexical system.



A third distinction is that between orthographic and phonological information. Until rather recently orthographic representations have been argued to depend on phonological representations. The *phonological mediation hypothesis* states that an orthographic representation cannot be accessed without previous access to the corresponding phonological representation. In fact, a common introspective experience is that when we have to write a word we first conjure up the phonological form and then write it down. However, there is by now sufficient experimental evidence that the written and the spoken forms of words can be separately affected by brain damage.

Thus far the proposed architecture includes separate semantic, phonological and orthographic representations; moreover, orthographic and phonological representations are separate for input and output. Besides the lexical and the semantic components, figure 1 depicts a mechanism for the recognition of heard phonemes and one for the recognition of written graphemes on the input side; on the output side it depicts a phonological and an orthographic buffer. The buffers are short-term working memory systems that temporarily store representations in preparation for subsequent processes. In order to produce a word, for example, one has to retrieve its phonological form in the phonological output lexicon; the actual production of the word requires a given time during which the abstract representation of the word must be continually reactivated for production to take place. The same happens when one has to write a word; the abstract orthographic representation must be reactivated as long as it has been written. The output buffers perform just that.

On the input side, processes in the auditory analysis system abstract from the details of the acoustic stimulus and compute an auditory representation, which serves as input to the phonological lexicon. When graphemes instead of phonemes are presented, the abstract letter identity system recognizes letters independently of their font and letter case and holds them for subsequent processing.

Finally, a distinction must be drawn between lexical and sublexical processes. A normal literate adult is able to read and write known and novel words. Known words can be read (or written) through the lexicon: the component phonemes (or graphemes) are identified in the auditory analysis (or abstract letter identity) system, the word is then recognized in the phonological input lexicon (or orthographic input lexicon) and is given meaning in the semantic system, which in turn addresses the phonological output lexicon (or orthographic output lexicon) for production. This lexical route is the only one that allows one to read (and write) "irregular" words, such as *yacht* or *pint*, which cannot be read (or written) by applying the language conversion rules. A novel word cannot be read through this route because by definition its representation is not present in the input and output lexicons. Normal literate

adults can give a phonologically (or orthographically) plausible rendition of the novel word by applying the language conversion rules. Figure one depicts the nonlexical routes for reading and writing.

It has frequently been maintained that to be theoretically justified aphasia therapy must be based on a functional model of normal processing and that to be effective treatment methods must be founded in a rational analysis of the patient's problems (e.g., Howard and Hatfield, 1987). The above described model of the lexical system has frequently been taken as a starting point in evaluating patients and planning therapy.

"COGNITIVE" REHABILITATION

I shall now review some reportedly "cognitive" rehabilitation of naming disorders and see in what way the cognitive approach differs from more traditional clinical approaches.

I shall review studies reported in two books (*Cognitive Approaches in Neuropsychological Rehabilitation* edited by Seron and Deloche (1989) and *Cognitive Neuropsychology and Cognitive Rehabilitation* edited by Riddoch and Humphreys (1994)) and three special issues of three journals (*Aphasiology*, vol. 7, no. 1, 1993; *Neuropsychological Rehabilitation*, vol. 5, no. 1-2, 1995, and *Brain and Language*, vol. 52, no.1, 1996). Four studies deal with naming disorders. The reference model in all these studies is the same or very similar to the one just described.

Raymer *et al.* (1993) treated four chronic Broca aphasics with severe anomia. The patients had different lexical disorders but they all had severely impaired written naming and failure of the lexical-semantic information to access the phonological representations. The investigators chose to implement a phonologically based treatment for all patients. Two sets of 30 monosyllabic picturable nouns were selected; ten were target words, ten rhymed with the targets and ten were semantically related. Treatment consisted in presenting the patient the ten target pictures one by one. He or she was asked to name the picture; in case of failure the patient was given a phonological cue -- first a rhyming word, then the initial phoneme of the target word, and finally the whole word. At whatever level the patient produced the correct word he or she was asked to repeat it five times and then naming was re-attempted. All subjects improved oral naming of the target words; generalization to untreated words and untreated tasks (reading and written naming) varied across subjects.

Greenwald *et al.* (1995) treated two patients with multiple loci of deficit in lexical processing. Both SS and MR had anomic aphasia with severe alexia and agraphia. Memory disorders and

impaired visual object processing were also present in SS, and oral and limb apraxia in MR. Extensive testing revealed an impairment in activation of lexical phonology from semantics and an impairment in activation of semantics from viewed objects in both patients. A phonological cueing hierarchy was used for the phonological disorder which was treated first. Forty picturable nouns (20 experimental and 20 controls) were selected and the patients were asked to name to definition. When they failed a phonological cueing hierarchy was used for the experimental stimuli: first phoneme, first two phonemes, whole word. At the end of treatment both patients showed improved naming of the experimental pictures (and SS also some generalization to the control pictures). A second treatment targeted the semantic impairment and a visual-semantic cueing hierarchy was used whenever the patients failed to name a target picture. The cueing consisted in first saying the semantic category of the stimulus, and then naming and description of a visual characteristic of the stimulus pointed to by the examiner. After each question the examiner summarized what had been said and asked the patient to name the picture. For both patients treatment two resulted in improved naming of the trained items and no generalization to untreated items. To control for the effect of simple rehearsal on improvement, an oral repetition treatment for picture naming was administered to both patients but only MR showed a statistically significant improvement. The investigators argue that their results provide evidence that treatments targeted at two specific impairments – phonological cueing for lexical retrieval damage and semantic cueing for visual-semantic processing – are more successful than an unspecific control treatment – repetition.

Le Dorze and Pitts (1995) implemented three experimental treatments and two control conditions with patient RT, a severely anomic woman with impaired comprehension. Further testing suggested problems in accessing semantic information and word-form information in naming task. The first treatment simultaneously targeted the semantic and the word-form disorder. For the semantic disorder word-picture matching tasks with semantically related distractors was chosen. For the word-form disorder RT was asked to read the target word and try to remember it; she was then given the first letter and asked to name the picture. The second treatment only used the semantic technique, and the third only the word-form technique. In the two control conditions the patient was offered repeated opportunities to name without being given any help. In each treatment and control condition the patient was asked to name five pictures. RT reached criterion, namely four of five correct naming, in two consecutive sessions within six sessions for the semantic-phonological technique, and the experiment was terminated. Treatments two and three, but not the control conditions, also produced some improvement. According to the investigators the experiment demonstrated that "substantial improvement occurred when therapy was oriented towards the patient's disorders" (Le Dorze and Pitts, 1995).

Finally, Miceli *et al.* (1996) described rehabilitation of patients RBO and GMA with unimpaired comprehension and damage to phonological output representations. For both patients therapy required retrieval of the phonological forms of the target words. Two treatments were implemented for RBO: naming and repetition of 30 experimental written words. Each treatment lasted five days and the experimental words were presented ten times each. After treatment completion, RBO named the experimental words better than the 30 control words. GMA underwent three treatments: in treatment one he was presented the picture and the written word of one of the 20 experimental words and was asked to read it; in treatment two only the written word was presented, and in treatment three only the picture and he was asked to name it. In case of failure he was given the first phoneme of the word. Each treatment lasted seven days and GMA showed improvement after each treatment. Neither of the patients showed generalization to untreated control words. These results were predictable because according to the model there is a one-to-one relationship between a lexical-semantic representation and its corresponding phonologic form. The investigators therefore conclude that to start from an explicit and detailed model of the normal function is useful in guiding treatment.

The two books and the three journal special issues reviewed also report case studies of rehabilitation of reading and writing disorders. Three studies treated rehabilitation of conversion rules (Bachy-Langedoch and De Partz, 1989; Berndt and Mitchum, 1994; Carlomagno and Parlato, 1989), one treated direct learning of irregular words in a surface dyslexic patient (Coltheart and Byng, 1989). Two studies targeted treatment for the output orthographic buffer (Aliminosa *et al.*, 1993; De Partz, 1995) and one targeted treatment for letter-by-letter reading disorders (Behrmann and McLeod, 1995). They will not be reviewed here and the interested reader is referred to the original papers.

What can we conclude from this review of therapy for lexical disorders? I think three main considerations can be drawn. Firstly, in the papers considered, which hopefully are sufficiently representative of the literature, no treatment has been reported for rehabilitation of comprehension. Secondly, no really new technique for naming disorders has been reported (rehabilitation of reading and writing disorders is somewhat more accurate and specific than it used to be in classic rehabilitation). Raymer *et al.* (1993) used a cueing hierarchy for the treatment of 10 words, Greenwald *et al.* (1995) a phonological and a visual-semantic cueing for the treatment of 20 words, Le Dorze and Pitts (1995) a semantic and a phonological cueing technique for the treatment of 5 words, and Miceli *et al.* (1996) repetition and naming of the written name for the treatment of 30 and 20 words. Phonological and semantic cueing, reading and repetition techniques are part of the equipment of any experienced speech therapist.

Thirdly, these studies represent investigations about which techniques could be efficacious but they do not represent clinical treatments. The number of stimuli used in each research (from a minimum of five to a maximum of thirty) is very low and one cannot take it for granted that the same method would be efficacious with a much larger number of stimuli. The treatments described are useful suggestions about how to treat patients in clinical settings but do not have any impact on the patients' everyday life.

A PROPOSAL

It has often been argued that we lack a theory of aphasia rehabilitation (e.g., Caramazza, 1989). A theory of aphasia rehabilitation must not only be able to specify "how to do what to whom" but, more importantly, the theoretical underpinnings of the theory. I certainly do not have the pretence to have any detailed proposal, but I think that it is possible to advance a suggestion. As long as we do not know more about how recovery from aphasia occurs, data about learning in normal subjects -- both about adults who enlarge their knowledge and about children who are acquiring a new capacity -- should be taken into consideration.

Models of the acquisition of reading and spelling in normal children are rare and not very detailed. Frith's (1985) model describes only the sequence of the different stages of the learning process without any further specification. In her model, the first stage is the logographic one in which the child acquires a small reading sight vocabulary and the capacity to write some of these words by a whole-word strategy. At stage two the child learns an alphabetic strategy, and at stage three the whole-word strategy is resumed, first for reading and then for spelling. According to Frith, acquisition of the representations for spelling is based on "transfer" to spelling processes of stabilized input orthographic representations for reading. In other words, well-acquired representations in the input lexicon are automatically transferred to the output lexicon. The model does not specify how this transfer occurs.

Basso *et al.* (1999) argued that knowing whether and how in normal subjects input orthographic representations are transferred into the output lexicon for spelling could be used in the rehabilitation of patients with damage to the output orthographic lexicon. Twenty healthy Italian controls with previous knowledge of French participated in the study by Basso and co-workers (1999). Twelve picturable French words (e.g., *seau* (bucket) and *feuille* (leaf)) which could not be written by applying Italian orthographic rules were selected. The 12 pictures were presented to each subject individually three times in pseudorandom order while the examiner clearly said the corresponding French word. Participants were then asked to point to the picture named by the examiner; errors were corrected by the examiner who also

repeated the correct word. The procedure was continued until the subject correctly pointed to the 12 pictures three times consecutively, showing acquisition of the input phonological representations. Subjects were then asked to write the name of the pictures, which were presented one at a time (baseline). The written words were then presented and the same procedure was followed and continued until subjects correctly pointed to the 12 pictures three times consecutively demonstrating acquisition of the orthographic forms of the words for reading. After a filled delay of 10 min the subjects were asked to write the name of the pictures (testing) and once again a week later, at follow-up. Results indicated that number of words correctly written at testing and at follow-up significantly differed from number of words written at baseline, and that number of words correctly written at testing was significantly higher than number of words written at follow-up. The investigators take these results as indicating that knowledge of orthographic representations for reading can support correct spelling of irregular words the first time they are spelled. Correct writing could not have taken place by retrieving the representation in the output orthographic lexicon because, by definition, it only contains known words. A possible explanation is that people can conjure up the mental visual image of the written word and copy it from the visual buffer. Correct writing at follow-up could be explained in a similar way but it is also possible that output orthographic representations had been acquired and that words were lexically spelled. It is suggested that this strategy could advantageously be used in the rehabilitation of patients with damage to the orthographic output lexicon.

A second investigation with normal subjects was carried out by Basso and co-workers (2001). They aimed at evaluating the best strategy for normal adults to learn new words in the belief that aphasic patients with word-finding difficulties could benefit most from such a technique.

Briefly, three learning methods were compared in normal adults: repetition, reading aloud, and orthographic cueing. Thirty subjects (ten in each condition) were asked to learn 30 legal nonword arbitrarily assigned to 30 different pictures; thirty further nonwords were used as controls. All subjects learned the thirty words but number of trials to criterion was significantly lower for the orthographic cueing method. In addition, at follow-up a week later, number of remembered words was significantly higher for the same method. No difference was found either in learning or at follow-up between repetition and reading aloud.

The same experimental design with only minor changes was applied to two aphasic patients and they too showed significantly better recovery of words with the orthographic cueing method, supporting the idea that the best strategy for normal controls can also be the best strategy for brain-damaged aphasic patients.

CONCLUSION

Many different methods for the rehabilitation of naming disorders have been shown to be efficacious, even when the same method has been applied to patients with different disorder (e.g., Raymer *et al.*, 1993).

The future of aphasia therapy can follow the same lines as in the past. Skilled and intuitive speech therapists will continue to devise new methods that will prove successful for some patients and slowly become part of each therapist equipment. The link between the treated disorder and the method used, however, will not always be clearly stated and our theoretical knowledge will not be implemented.

Otherwise, a cooperative effort could pose the foundation of a shared theoretical basis. Some basic questions can be agreed on and evidence for a response can be slowly accumulated, permitting to reach more rapid answers to our questions about aphasia therapy implementation and effectiveness.

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Chapter 9

VERB RETRIEVAL PROBLEMS AT THE WORD AND SENTENCE LEVEL: LOCALISATION OF THE FUNCTIONAL IMPAIRMENTS AND CLINICAL IMPLICATIONS

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INTRODUCTION

All aphasic patients suffer from word finding difficulties. The severity and nature of this deficit may differ, depending on where in the production process the functional impairment arises. Most studies to word finding problems focus on nouns, but over the last fifteen years it has been demonstrated that most aphasic patients encounter problems in verb retrieval as well, even more than in noun retrieval (Williams and Canter, 1987; Kohn *et al.*, 1989; Jonkers, 1998). Since verbs play a central role in sentence building, it is more complicated to find the locus of the functional deficit in the case of verbs, because more factors play a role in verb than in noun retrieval.

This chapter deals with verb retrieval. Verb retrieval deficits in Broca's and anomic aphasia will be discussed, with a focus on both the word and the sentence level. It will be argued that the impairments in verb retrieval in Broca's and anomic aphasia arise at different levels. At the end of the chapter, the implications for assessment and therapy will be discussed. For describing the verb retrieval problem, Levelt's model for speech production (Levelt, 1989) will be used. For explaining the deficits found at the sentence level, Chomsky's Government and Binding (GB) theory will be the theoretical framework.

First, a short description of Levelt's model will be given, followed by an overview of the factors that play a role in verb retrieval. This will result in a hypothesis about the level of breakdown in verb retrieval in Broca's and anomic aphasia at the word level. For Broca's aphasia this functional localisation will be further refined using the results from two Dutch tests for verb retrieval at the sentence level.

LEVELT'S SPEECH PRODUCTION MODEL

Levelt (1989) introduces a speech production model for the sentence production in non-brain-damaged speakers. This model can be very helpful in describing aphasic behaviour, although it is not originally meant for this purpose. In order for a word to be retrieved from the lexicon, several stages need to be processed, starting with the activation of a concept. This concept, in turn activates a lemma in the lexicon. Lemmas contain semantic and syntactic information. For example, the semantic information belonging to a verb specifies its meaning and its thematic roles, whereas the syntactic information is on word class and subcategorisation frame. Lemma information is used by the Grammatical Encoder to construct a sentence. The lemmas activate the so-called *lexemes*, the underlying word forms that contain information about the phonological and prosodic form of the word. During Phonological Encoding, these lexemes, which are still quite abstract, are inserted into the sentence frame built by the Grammatical Encoder. After phonological encoding, several phonetic processes take place, but these are beyond the scope of this chapter and will further be ignored. In Figure 1, a graphical representation based on Levelt's model is given.

This can be illustrated by an example. Suppose someone wants to produce the verb 'to milk'. The concept activates the lemma TO MILK, including the information that it is a verb meaning something like 'to get milk out of a cow' and that it has two thematic roles -- an agent and a theme -- and that it is a transitive verb, subcategorized for a subject and an object. The lemma subsequently activates the lexeme /tu mIlk/.

When this representation of a verb is compared with that of a noun, we see the following: the semantic lemma information for a noun contains its meaning, but usually not information about thematic roles, as nouns usually do not have thematic roles. The syntactic information says that it is a noun, but there is no information about subcategorisation, since nouns usually do not have subcategorisation frames.

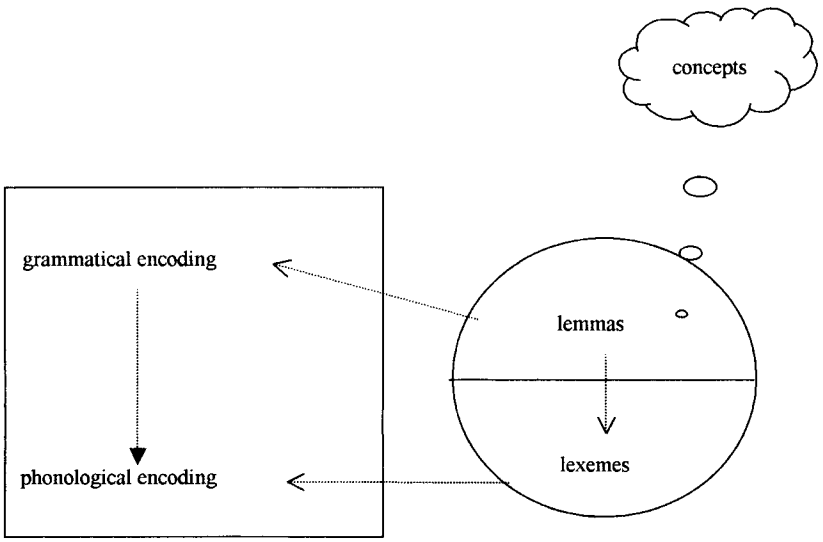


Figure 1. A simple representation of Levelt's (1989) speech production model

VERB RETRIEVAL AT THE WORD LEVEL IN APHASIA

Many studies have compared verb and noun retrieval in aphasia. With respect to Broca's aphasia, there is general agreement that verb retrieval is more impaired than noun retrieval (e.g. Bates *et al.*, 1991; Miceli *et al.*, 1984; Kohn *et al.*, 1989; Zingeser and Berndt, 1990, Jonkers, 1998; Kauske and De Bleser, 2000;). With respect to anomic aphasia, the results are less equivocal. According to some authors, nouns are more impaired than verbs (Miceli *et al.*, 1984; Zingeser and Berndt, 1989), but others claim that verbs and nouns are equally impaired in anomic aphasia (Basso *et al.*, 1990); there is also some evidence that verbs are more impaired than nouns in anomic aphasia, just as in Broca's aphasia (Williams and Canter, 1987; Kohn *et al.*, 1989; Jonkers, 1998). Some of these studies show that verb retrievability is influenced by several factors. These factors can be divided into four linguistic levels: lexical-semantic factors, factors related to argument structure and thematic roles, morphological factors, and syntactic factors.

Lexical-Semantic Factors

There are several factors which are assumed to play a role at the lexical level, for example, word frequency, imageability, instrumentality and name-relation to a noun. Most studies that compare verb and noun retrieval have controlled their items for word frequency. This is often considered to be a very important factor in noun retrieval, but so far there is no evidence that it plays a role in the retrievability of verbs (Jonkers, 1998; Kemmerer and Tranel, 2000).

Currently, a lively debate is going on in *Brain and Language* on the influence of the factor *imageability*. Bird *et al.*, (2000) attribute the often-reported difference between retrievability of verbs and nouns to the difference in imageability between the two word classes: verbs are less imageable than nouns and are therefore more difficult to retrieve from the lexicon. This conclusion is challenged by Shapiro and Caramazza (2001), but a study of Luzzatti (2001) confirms that imageability of nouns and verbs plays a crucial role in verb retrieval.

Jonkers (1998) has developed a test in which instrumental and non-instrumental verbs are included. An instrumental verbs is defined as referring to an action for which a man-made instrument is required, for example, *to drill, to cut*. Jonkers' results show that instrumental verbs are easier to retrieve than non-instrumental verbs for anomic speakers but not for Broca's aphasics. The influence of instrumentality on the facility with which verbs are produced has been confirmed by Kemmerer and Tranel (2000) in their study of a patient group that was not selected for aphasia type.

Jonkers (1998) also made a distinction between instrumental verbs that are name-related to their instrument (*to drill*) and those that are not (*to cut*) and he compared their retrievability with that of the accompanying nouns (*a drill, a knife*). The data reveal that verbs related in name to their instrument are as easy to retrieve as the names of the instruments, whereas verbs that are not-related in name are significantly more difficult to retrieve than the name of their instruments.

Factors related to argument structure and thematic roles

One of the characteristics of verbs is their argument structure, that is the number of arguments that they are subcategorized for. The arguments fulfill thematic roles. Action verbs, the verbs that are usually used on action naming tasks, have an external argument, the agent, and often one or two internal arguments. Intransitive verbs have one (external) argument (such as *to swim*) transitive verbs have one external argument and one (such as *to repair*), two (such as *to*

put), or even three (such as *to exchange*) internal arguments. Examples are given in (1); external arguments are **bold**:

(1) intransitive (one external argument)
***John** is swimming*
transitive (one external, one internal argument)
***John** repairs the house*
transitive (one external, two internal arguments)
***John** puts the book on the table*
transitive (one external, three internal arguments)
***John** exchanges his book for a CD with Mary*

Broca's aphasics are sensitive to the number of arguments that belong to a verb. Both Kim & Thompson (2000) and Kiss (2000) show that the more arguments belong to a verb, the more difficult it is to retrieve for Broca's aphasics. So far, no similar effects have been found for anomic aphasia. However, it is not only the number of arguments. Kiss (2000) also demonstrates that the kind of argument may influence retrievability: verbs with a locative argument (e.g. *to sit, to put*) are more difficult than other two place (for example *to throw*) and three place (for example *to send*) verbs. Kim and Thompson (2000) took the nature of the arguments, that is, whether or not they are obligatory, into account. Transitive verbs may be subdivided into those that require phonetic realisation of the internal argument (such as *to fix, to repair; *John is repairing* is an ungrammatical sentence) and the so-called *pseudo-transitives* verbs for which realisation of the internal argument is optional (both *the woman knits the sweater* and *the woman is knitting* are grammatical sentences). This means that verbs such as *to repair* have only one possible argument structure and verbs such as *to knit* have two possible argument structures. Kim and Thompson (2000) convincingly argue that verbs with one possible argument structure are easier for Broca's aphasics than those that have two or three. This has not yet been tested on anomic aphasics or any other fluent aphasia type.

From these studies, we can conclude that argument structure and thematic roles play an important role in the retrievability of verbs in Broca's aphasia; as to date, we do not know what the influence of these factors is in anomic aphasia.

Morphological factors

Since morphology plays a minor role in verb retrieval at the word level, this point will mainly be addressed when verb production at the sentence level is discussed. One interesting with

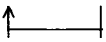
respect to morphology at the word level, however, is Kiss's (2000). She tested Hungarian Broca's aphasics and used verbs with increasing morphological complexity. For example, she compared verbs such as *alszik* (to sleep) with *fésülködik* (to comb one's own hair) and *gitározik* (to play the guitar) and found that the more morphologically complex the verb is, the more difficult it is to retrieve for Broca's aphasics.

Syntactic factors

A factor that has repeatedly been claimed to influence naming in Broca's aphasia is *transitivity*. Intransitive verbs are supposed to be subcategorized for a subject, whereas transitive verbs are subcategorized for a subject and an object. The results of the studies that controled for the factor 'transitivity' are not unequivocal. For English (Thompson *et al.*, 1997), German (Kauske and De Bleser, 2000) and Italian (Luzzatti, 2001), it has been argued that transitive verbs are easier than intransitive verbs, but this could not be confirmed for Dutch (Jonkers, 1997; Bastiaanse, 2001).

This suggests that comparing transitive and intransitive verbs is not easy. This is not surprising, since within each class, different categories can be distinguished. As mentioned above, within the category of transitive verbs, real transitives and pseudotransitives can be distinguished and we know from the Kim and Thompson study (2000) that this affects the retrievability of verbs at the word level in Broca's aphasia. The studies to German, Italian and Dutch do not allow for pseudotransitivity.

Within the class of intransitive verbs, another distinction can be made and two recent studies demonstrate that this is an extremely important distinction. Both Thompson (in press) and Luzzatti (2001) tested Broca's aphasics on two types of intransitive verbs, unergatives and unaccusatives. The difference between these two types is in their underlying structure. Unergative verbs, such as *to sleep* and *to swim*, are subcategorized for a subject and this subject is supposed to be the agent. Unaccusatives, such as *to fall* and *to sink*, however, are also subcategorized for a subject, but this is supposed to be the theme. This theme should be 'moved' to the subject position, as shown in (2).

(2) the man swims
the man$_i$ falls i


The 'i' indicates the connection between the original and the surface position of the subject. *The man* is co-indexed with its original position. This movement of the theme to the subject position is a syntactic operation.

Both Thompson (in press) and Luzzatti (2001) show that unaccusatives[1] are significantly more difficult to retrieve than unergatives in Broca's aphasia, suggesting that the syntactic operation complicates verb retrieval. Luzzatti (2001) also tested speakers with anomic and Wernicke's aphasia; in these aphasic subgroups, no significant differences in the retrievability of unergatives and unaccusatives were found.

CONCLUSIONS

Summarizing, speakers with anomic aphasia and speakers with Broca's aphasia encounter problems with verbs. More interesting, however, are the differences in retrievability within the class of verbs. In Broca's aphasia, argument structure, optionality of arguments, morphological factors and syntactic factors play a role. When a verb is more complicated in terms of any of these factors, retrievability diminishes. Interestingly, these are all factors that are related to the lemma. Factors related to lexical semantics, such as word frequency, instrumentality and name relation with a noun do not influence the ability of Broca's aphasics to retrieve the verb.

When we look at anomic aphasics, we see that such lexical-semantic factors do affect verb retrieval: instrumental verbs are easier than non-instrumental verbs and verbs related in name to a noun, a relationship at the lexeme level, are easier than those that do not have a name relation. At the same time, from some of the factors that play a role at the lemma level, such as ergativity, we know that they do not influence the ability of anomic aphasics to retrieve a verb. This suggests that the problems that both aphasic subgroups encounter with verbs arise at different levels: in Broca's aphasia the verb retrieval deficits seem to arise at the lemma level, in anomic aphasia at the lexeme level. The latter finding is not really surprising, since it has repeatedly been shown that the noun retrieval problems in anomic aphasia arise at this level, although usually a different model is used (e.g. Ellis and Young, 1988).

To assume that the problems in Broca's aphasia arise at the lemma level, raises the question whether the lemmas themselves are impaired or whether it is retrievability of the lemmas that is affected. There is ample evidence that the lemmas are intact, since comprehension of verbs

[1] Luzatti's refers to these unaccusative verbs as *ergatives*.

is unaffected in Broca's aphasia (Jonkers, 1997; Kim and Thompson, 2000). Apart from that, Shapiro and Levine (1990) have demonstrated that during online processing, Broca's aphasics show the same reaction patterns with respect to verb argument structures as non-brain damaged speakers: the more arguments a verb takes, the longer it takes to make a lexical decision; similarly, the more possible argument structures a verb has, the longer it takes to make a lexical decision, both for Broca's aphasics and for non-brain-damaged subjects. This means that at the input level no verb problems exist, whereas retrievability is impaired.

The next question, then, is whether lemma selection itself is impaired or whether processing of lemma information, that is grammatical encoding, is affected. In the following section, two Dutch studies will be presented to answer this question

VERB RETRIEVAL AT THE SENTENCE LEVEL IN BROCA'S APHASIA

The grammatical encoder uses the lemma information that has been retrieved from the lexicon to build syntactic frames. In the previous section, Levelt's (1989) model was used as a theoretical framework for word retrieval, but unfortunately, Levelt's theory on grammatical encoding is not sufficiently detailed to explain the results found in the two studies that will be presented in this section. Therefore, Chomsky's (1986) Government and Binding (GB) theory is used, in which so-called *movement* of constituents plays a role.

Two studies will be discussed in which verb retrieval in sentence context has been tested. Both studies have two conditions in which the same verbs are used. This enables us to find out whether it is lemma selection that is deficient, in which case one expects the same problems in both conditions, or whether it is grammatical encoding, in which case one expects the grammatically more complex construction to be more difficult.

Verb movement in Dutch

Linguistic background

Dutch has been analyzed as an SOV-language, meaning that the base-generated position of the verb is after the object (Koster, 1975). In the Dutch declarative matrix sentence, the finite verb has to be moved to second position. This movement is known as *Verb Second*. If the main verb clusters with a modal verb or auxiliary, the main verb remains *in situ* and the modal verb or auxiliary is moved to the Verb Second position. In embedded clauses, the finite verb remains in

its base-generated position. This is illustrated in (3-5), where *t* designates the canonical verb position, which is co-indexed with the Verb Second position.

(3) matrix sentence without modal verb/auxiliary

de jongen	koopt$_i$	een fiets t$_i$
the boy	buys	a bike

(4) matrix sentence with modal verb

de jongen	wil$_i$	een fiets	kopen	t$_i$
the boy	wants	a bike	(to) buy	

(the boy wants to buy a bike)

(5) embedded clause without modal verb/auxiliary

(ik denk)	dat	de jongen	een fiets	koopt
(I think)	that	the boy	a bike	buys

(I think that the boy buys a bike)

Broca's aphasia and verb movement

If it is lemma selection that is impaired, then one expects patients to score similarly on both conditions, since exactly the same lemmas have to be retrieved from the lexicon. If it is grammatical encoding, one expects completion of the matrix clause to be more difficult, since this contains a moved finite verb and thus requires an extra syntactic operation. Notice that the more frequent matrix clause is the more complex construction, because the finite verb has been moved to the second position. A third possibility is that it is just superficial grammar that is affected in Broca's aphasia, and theoretically founded operations such as verb movement have no influence on that. In that case, one expects completion of an embedded clause to be more difficult, since embedded clauses (which can only occur in combination with a matrix clause) are generally more complex than matrix clauses. This is what one would expect when one adopts Levelt's theory of grammatical encoding, which assumes that we speak 'from left to right'.

The following test was used. Two pictures were shown to the patient on which the same person was performing the same action with different objects. An example is given in figure 2.

The experimenter read a sentence to the patient, who was asked to complete it. There were two conditions: the matrix and the embedded condition. The sentences given as prompt for the picture in figure 1 for the two conditions are given in (6-7).

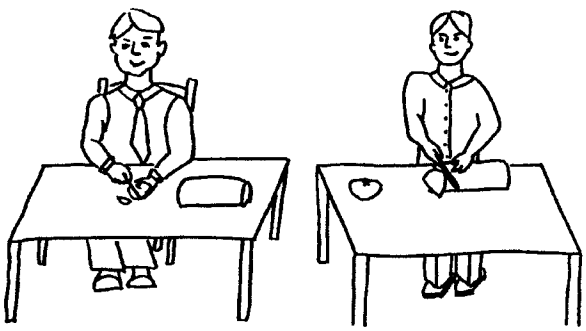


Figure 2. An example of the pictures used in the test for the production of finite verbs

(6) matrix clause
Dit is de jongen die de tomaat snijdt en dit is de jongen die het brood snijdt. Dus deze jongen snijdt de tomaat en deze jongen .....
Patient: *'snijdt het brood'*
(This is the boy that the tomato cuts and this is the boy that the bread cuts. So, this boy cuts the tomato and this boy .....
Patient: '*cuts the bread'*)

(7) embedded clause
Deze jongen snijdt de tomaat en deze jongen snijdt het brood. Dus dit is de jongen die de tomaat snijdt en dit is de jongen die .....
Patient: *'het brood snijdt'*
(This boy cuts the tomato and this boy cuts the bread. So, this is the boy that the tomato cuts and this is the boy that .....
Patient: *'the bread cuts'*)

Nine Dutch agrammatic Broca's aphasics were tested. All patients were right-handed and in all patients the aphasia was due to a single stroke in the left hemisphere and at least three months post-onset. All patients were Broca's aphasics in the classic way: they spoke in telegraphic speech and their comprehension was relatively intact. None of the patients had articulation disorders to an extent that might have influenced the test results.

The results are shown in Figure 3: the production of a (moved) finite verb followed by an object in the matrix clause is significantly more difficult than the production of an object and a finite verb *in situ* in the embedded clause (Wilcoxon: z=-2.37, p=0.018).

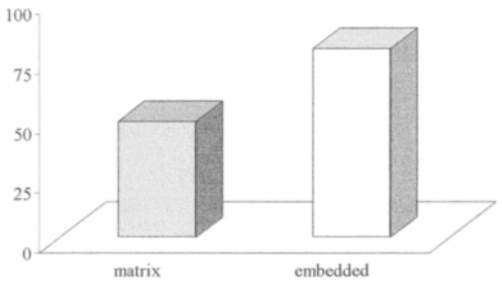


Figure 3. Percentages correct on the test for the production of finite verbs in the matrix and the embedded clause

These findings indicate that the production of the same verb is more difficult when an extra syntactic operation (in this case verb movement) is involved. This suggests that it is not lemma selection (the lemmas are the same in both conditions), but grammatical encoding that limits the production of verbs in Broca's aphasia. However, it is not grammatical encoding in the sense that Levelt (1989) suggests: the matrix clause condition, in which the verb has been moved, is the most difficult one, even though a matrix clause is much more frequent and superficially simpler than an embedded clause.

The production of verbs with alternating transitivity

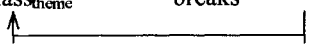
Theoretical background

Above, two studies have been mentioned, Luzzatti (2001) and Thompson (in press), that evaluated the retrieval of unaccusatives (or ergatives) and unergatives at the word level. There is, however, a class of verbs (the so-called *verbs of alternating transitivity*, Levin, 1993) that allow a transitive as well as an intransitive construction, for example, *to burn, to ring, to break*. In their transitive reading the agent is the grammatical subject and the theme the object, but in the intransitive reading, the theme is in subject position, just as in the unaccusative verbs mentioned above. Examples are given in (8-9).

(8) to break

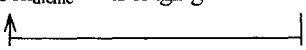
the $\text{woman}_{\text{agent}}$ breaks the $\text{glass}_{\text{theme}}$

the $\text{glass}_{\text{theme}}$ breaks



(9) the $\text{priest}_{\text{agent}}$ is ringing the $\text{bell}_{\text{theme}}$

the $\text{bell}_{\text{theme}}$ is ringing



It is assumed, as mentioned above, that in the second sentences, the theme has been moved from its original position to the subject position.

Alternating transitivity and Broca's aphasia

If grammatical encoding is indeed the affected component in speech production in Broca's aphasia, as suggested by the previous experiment, then we expect that producing a sentence with the intransitive version of the verb will be more difficult than producing a sentence with the transitive version. Notice that the construction with the transitive version is hypothesised to be easier, although it is superficially more complex since it has a subject and an object whereas in the intransitive reading only a subject needs to be produced. When the underlying syntax is taken into account, however, the latter requires an extra syntactic operation, movement of the theme to subject position, which is thus hypothesised to be complicated for speakers with Broca's aphasia.

In order to test the subjects' ability to construct sentences with verbs in the transitive and intransitive reading, a task was developed to elicit sentences. Underneath the picture of an action, the verb was printed in its infinitive form. Notice that in Dutch, the verb is marked for its infinitive form (stem + en: *dans* + *en* 'to dance'). Two examples of the test (one of the transitive and one of the intransitive condition) are given in Figure 4.

Figure 4. Two examples of the test for verbs with alternating transitivity. Left a picture for the transitive condition, right for the intransitive condition

burn

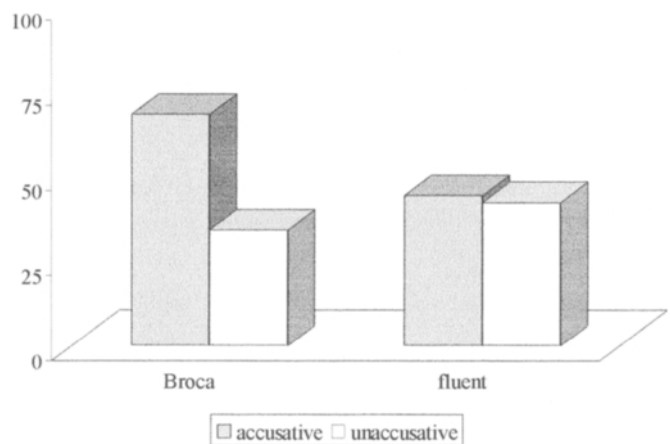
burn

This is a study in progress and so far, seven speakers with Broca's aphasia and five speakers with fluent (both anomic and mild Wernicke) aphasia have been tested. Above, it was hypothesised that these speakers with fluent aphasia have problems with retrieval of lexemes, not of lemmas. This means that the discrepancy that we expect to find for Broca's aphasia is not expected to arise in the group of fluent aphasic speakers. All subjects were aphasic due to a single stroke in the left hemisphere and at least three months post-onset.

The results are shown in Figure 5: for speakers with Broca's aphasia, producing a sentence with an intransitive verb is more difficult than producing a sentence with the same transitive verb (the difference approaches significance: Wilcoxon: $z=-1.94$, $p=0.053$), although the latter sentence is superficially more complex, as both the subject and the object should be realized. This difference is not found for the speakers with fluent aphasia (Wilcoxon: $z=-0.52$, $p=0.600$).

These findings support the hypothesis that it is not so much lemma selection as grammatical encoding that is the problem in Broca's aphasia. So far, it seems that grammatical encoding is intact in fluent aphasia, but for both aphasia groups more subjects need to be tested before we can make any definitive statements.

Figure 5. Percentages correct on the test for the production of verbs with alternating transitivity



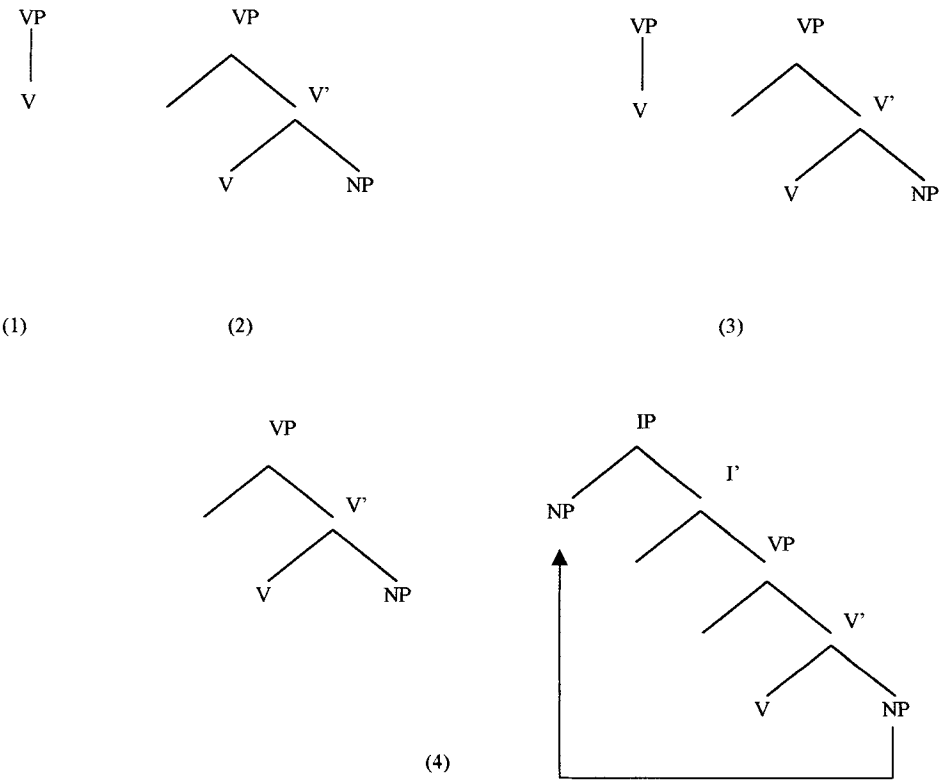
CONCLUSION

The main question of this chapter was why verbs are so difficult to retrieve for aphasic speakers. It has been shown that for fluent aphasics, lexical aspects, such as instrumentality and name-relatedness to nouns, influence retrievability. This suggests that selection of the lexemes is compromised, as these aspects seem to play a role at the word-form level. In Broca's aphasia, however, morphological, syntactic and argument structural aspects influence retrieval, suggesting that the problem is related to the lemmas. It has been argued on the basis of comprehension and online-processing studies, that the lemma themselves are intact. Two recent studies show that lemma selection as such is not impaired, but that Broca's aphasics encounter problems with the grammatical encoding of the lemma information. The more information needs to be encoded, the more errors are made.

The results of several studies to the retrieval of verbs as single words showed an effect for the amount of information contained in the lemma: the more complex this information is (for example, the more complex argument or morphological structure is), the more difficult it is for Broca's aphasics to name an action. This implies that even verbs as single words need to be grammatically encoded, although no sentence is built. An impression of such verb lemmas is given in Figure 6.

Notice that it is a matter of debate whether this information is indeed part of the lemma, or whether the lexicon contains merely abstract, unspecified representations, the work of the grammatical encoder being to encode the lemma (see, for example, Hale & Keyser, 1993). For our argument here, this does not make a difference, since the locus of impairment in Broca's aphasia is assumed to be in the grammatical encoder. The conclusion of the findings on verb retrieval in Broca's aphasia so far is that verbs are more difficult than nouns, because more information needs to be grammatically encoded. On top of that, the more verb information needs to be encoded, the more difficult it is to retrieve (cf. the studies of Thompson and Kiss). At the sentence level, the production of verbs and their arguments is, again, influenced by the information that needs to be grammatically encoded: the more complex the structure of the sentence is, the harder it is to produce the required construction (cf. the studies of the production of finite verbs and alternating transitives). This means that the functional locus of the impairment in Broca's aphasia is assumed to be to the process of grammatical encoding. Since the verb retrieval problems in patients with anomic aphasia are influenced by lexical rather than by morphosyntactic factors, it is suggested that their deficits are due to a word-form, or lexeme, retrieval problem.

Figure 6. Lemma representations of verbs: (1) intransitive verb, e.g. *to sleep*; (2) transitive verb with one possible argument structure, e.g. *to fix*; (3) transitive verb with two possible argument structures, e.g. *to read*; (4) alternating transitive verb, e.g. *to break*.



Clinical implications

The conclusion that the functional loci of verb production deficits are different in anomic and Broca's aphasia has the clinical implication that the problems should be treated differently. Unfortunately, there are not many treatment programmes or methods for the mediation of verb deficits, nor are there many devices to diagnose the level of verb retrieval deficits. One of the goals of our research group is to translate the findings of scientific research to clinical tools. For the production of verbs and sentences, this has resulted in a test for comprehension of verbs and sentences in Dutch (Bastiaanse *et al.*, 2000) and English (Bastiaanse *et al.*, 2002)

and a treatment programme for verb production in Dutch (Bastiaanse *et al.*, 1997) and German (Bastiaanse *et al.*, in press[a]). Both in the test and in the treatment programme, verb production is addressed at the word and the sentence level. At the sentence level a distinction is made between the production of infinitives and finite verbs.

In the treatment programme, two different approaches are possible: the therapist can either decide to work on *restoration* of language function, in which case he/she will train the production of sentences with finite verbs to avoid overloading of the grammatical encoder, or work on *compensation*, in which the aphasic speakers is trained to circumvent the problem with finite verbs by using modal verbs and auxiliary constructions learnt by heart (for e.g. *the man wants to read a book* instead of *the man reads a book*).

Training of verb production is, in this view, an essential part of language therapy: verbs are the core of the sentence and without a verb, no sentence frame can be built. Apart from that, many function words are dependent on the verb: the production of determiners, pronouns, and many prepositions is only possible when a verb is produced (see, e.g. Bastiaanse *et al.*, in press[b]).

CONCLUSION

Verbs play a central role in the sentence production process: they form the core of the sentence, on which the use of thematic roles, tense, determiners, pronouns, prepositions and many other linguistic aspects depends. Verb production is compromised in many aphasic individuals, but the level of breakdown varies. Nowadays, there are some tools to identify the level of breakdown and to train the production of sentences by focusing on the role of the verb. The idea behind these tools is that once verb retrieval improves, sentence production is facilitated. This will result in an increase of communicative content, because verbs are not only central in the sentence construction process, but also contain much semantic information.

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Chapter 10

REDUCED SYNTAX THERAPY (REST) – A COMPENSATORY APPROACH TO AGRAMMATISM

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INTRODUCTION

The ultimate aim for therapy-oriented research is to determine which therapy is most appropriate in dealing with a particular type of disorder and degree of severity, also taking account of the cognitive and pragmatic resources available and individual goals of the patients. However, despite numerous approaches to treatment of agrammatism (see reviews of Byng and Lesser, 1993; Van de Sandt-Kondeman and Bonta, 1998; Mitchum *et al.*, 2000), discussion about the application and efficacy of the specific approaches remains controversial.

This paper deals with a compensatory approach to the treatment of severe agrammatism. The main goal of this approach is to reduce the demands of morpho-syntactic processing in on-line communication by reducing utterances to simple non-finite syntactic fragments. For this reason this approach is called Reduced Syntax Therapy (REST) and has been developed and modified in recent years (Schlenck *et al.*,1995; Springer *et al.*, 2000). The main results of the outcome study and further application options in Computer-Mediated-Communication (CMC) are reported.

There is a consensus that expressive agrammatism is a combination of symptoms with different underlying disorders. Severe chronic agrammatism is characterized by the following main surface features (cf. e.g. Caplan, 1987; Huber *et al.*, 1997):

- incomplete sentence structures consisting of individual content words which are linearly grouped together on semantic grounds rather than morphology,

- Limited repertoire of frozen expressions, mainly emotional expression, such as the German "Oh Gott!" (*Oh, God!*) or stereotypical phrases, such as "Ich weiss nicht" (*I don't know*), and highly overlearned structures, such as "Ich möchte..." (*I'd like to...*) or "Das ist..." (*That is...*), which may be the outcome of pattern drill.
- reduced repertoire of content words exhibiting little lexical variability, mainly restricted to concrete nouns of high frequency,
- absence of morphology that is governed by demands of syntactic agreement, such as personal endings to verbs, or case markings to nouns and adjectives,
- highly limited repertoire of so-called function words consisting at most of co-ordinating conjunctions or adverbs (like German "und" (*and*), "dann" (*then*)), or prepositions which carry content (e.g. German "in" (*in*) "auf" (*on*)).

However, these symptom combinations prove to be non-homogenous, varying from patient to patient, and also varying in a given individual patient. Attempts at explanation in terms of linguistics which assume a basic (morpho-)syntactic deficit do not explain this phenomenon sufficiently.

According to the deficit hypothesis, these symptoms are always related to the normal language system. The symptoms directly reflect an impairment of morphosyntactic knowledge and/or a specific restriction of grammatical processing capacities (Bastiaanse, 1995; Menn and Obler, 1990; Paradis, 2001). As an alternative to a morphosyntactic deficit, some authors postulate an impairment of lexical/semantic knowledge and/ or processing (cf. the discussion by van Lancker, 2001). A strong deficit assumption was put forward by generative syntacticians, who assumed that the basic property of agrammatic speech, as compared to normal speech, is that the structural representation of sentences lacks functional categories/projections (e.g. Inflectional Phrase: Tense Phrase versus Agreement Phrase). Several studies revealed that there is no general loss of functional categories, but these categories can be selectively impaired (cf. Friedmann and Grodzinsky 1997, Hagiwara, 1995). Besides the individual variation of symptoms, however, other clinical findings exist which cannot really be covered by the strong deficit approach and therefore require alternative explanation.

These findings relate to the late occurrence of agrammatic symptoms during the course of recovery. It seems plausible to assume that compensatory language learning involving right-hemisphere areas and executive functions of the frontal become operative in these patients. A more detailed discussion of the implications of acute and chronic agrammatism are provided by Huber *et al.*, 1997 and Springer *et al.*, 2000.

Unlike the deficit hypotheses, the adaptation theory provides an alternative explanation for agrammatic symptoms. The basic assumption is that aphasic symptoms can reflect either the underlying deficit or the attempt to compensate this deficit (cf. Hughlings Jackson, 1879; Kolk, and Heeschen, 1990). Chronic agrammatism represents adaptation on the different levels of neurophysiological reorganisation as well as on the cognitive and interactive processes. The adaptation hypothesis especially favours therapy planning, because not only cognitive but also communicative aspects of the language processes can be explained.

THERAPY OF AGRAMMATISM

The chances of achieving transfer into spontaneous language may, of course, depend on the treatment method. The starting point of planning therapy is to specify the deficit in a model of normal language processing. According to sentence production models (Garrett 1984; Bock and Levelt 1994), two types of treatment must be distinguished, i.e. either focusing on the positional or on the functional level of normal sentence processing.

In the *positional level* approach it is assumed that surface structures are specifically affected in agrammatism. Therefore, linguistic parameters such as word order, syntactic functors (realised by closed class words) as well as morphosyntactic marking are the objective of therapy. These units and regularities are introduced along hierarchies of complexity and practiced across major linguistic modalities (cf. HELPSS developed by Helm-Estabrooks *et al., 1986;* symptom-specific training, Huber *et al.*, 1993; Springer *et al.*, 1993).

The *functional level approach* focuses treatment on predicate-argument structures or thematic roles of verbs with increasing number of arguments. Several variants of treatment were developed under the hypothesis of a mapping deficit, i.e. the projection of functional onto positional level information (and vice versa) is assumed to be specifically impaired in agrammatism, whereas information on either level remains relatively accessible (cf. Jones, 1986; Le Dorze *et al.*, 1991; Schwartz *et al.*, 1994, Mitchum *et al.*, 2000). The impaired mapping processes are treated primarily by metalinguistic tasks. The patients learn to identify thematic roles irrespective of word order. Subsequently, sentence production is expected to be similarly improved, even without explicit training. However, a positive transfer of this kind did not always occur (Nickels *et al.*, 1991).

In either approach, functional or positional, direct practice effects seem to be achieved easily. After training on the functional level, practice effects appear to be greater than after training on

the positional level. It is not clear, however, to what extent this is simply a consequence of differences in underlying mechanisms of agrammatism being present in the patients. But a transfer into spontaneous language is again difficult to achieve and is rarely investigated.

In recent years, we have developed a treatment method, the so-called Reduced Syntax Therapy (REST; Schlenck *et al.*, 1995; Springer *et al.*, 2000) deliberately encouraging rather than preventing the production of a telegraphic style.

REDUCED SYNTAX THERAPY (REST)

Goals and Concepts

The first goal is to enable the patient to generate reduced sentence structures in on-line communication as fluently as possible. The following linguistic criteria must be met. First, the starting point of each utterance is a main verb and its obligatory complements. Second, the verb phrase has to be non-finite, i.e. no inflected endings for person or tense and no auxiliary verbs. Third, the position of obligatory complements has to respect the basic word order of the target language, e.g. as a SOV language, the order in German has to be complement before verb. Fourth, syntactic morphology is neglected, i.e. training focuses on main categories (verb, noun, adjective, predicating preposition), functors such as determiners and pronouns, as well as verb agreement and case marking, are excluded.

Severe chronic agrammatism is primarily seen as a compensatory response of intact right hemisphere functions to a complete loss of the syntactic capacities of the left hemisphere. This is why the REST approach aims primarily at stimulating and expanding basic strategies of content word activation and concatenation. From this starting point basic syntactic parameters of the target language are introduced, consisting of content words or fixed word combinations.

In chronic agrammatism with good recovery residual capacities of the left hemispere may be reactivated. Whereas normal processing involve the whole redundant perisylvian system, agrammatic patients may perform language tasks by activation parts of the bilateral language net-work under intensive therapy/practice. Typically, these patients show a broad task-specific variation ranging from complete simple sentences to one-word utterances. Why performances are highly prone to disorders might be from only partially activating the language network. In contrast to severe agrammatism, the REST approach offers for these patients the option to choose a simple register as a compensatory strategy when it is pragmatically applicable. The

REST approach enables a reduction in processing capacity for complete sentence planning and the patient is likely to benefit from free capacity to cope with other aspects of the production process, such as message planning, word activation and motor speech planning. Thus, the utterances are produced faster, and remain comprehensible despite their reduced sentence.

A further justification for focusing on reduced sentence structures of this kind is, that various studies show overlap of similar structures found in mild forms of agrammatic speech and normal colloquial style; however the syntactic repertoire is reduced in agrammatic patients. The hypothesis that agrammatic speakers (over) use normal constructions has been confirmed and research shows that both in normal and aphasic speech a compensatory strategy for adaptation to an elliptic style can be found (Kolk and Hofstede, 1994). Typically, in German and Dutch these contextual ellipsis lack finiteness e.g. either the verb appears in a non-finite form or is omitted (Klein, 1993; Kolk, 1998).

In comparison to mapping therapy (Schwartz *et al.*, 1994), the focus of REST is to stimulate the production of fixed phrases and to expand syntactic frames rather than improving the processing of thematic roles. But it is a secondary aim of REST to practice variable thematic roles by changing the strict subcategorization frame of the verbs. In contrast to the sentence generation program of Thompson (1998), REST systematically leaves out morphosyntactic structures, thus focusing on base structures alone.

Treatment Methodology

The REST-approach consists of five levels as exemplified for both German and English:

LEVEL 1	**2-word utterances: VP with direct object**	
• THEME	**WHAT / HOW DOING**	
• STRUCTURE	**N - V**	**V - N**
	A - V	**V - A**
• EXAMPLES	Kaffee trinken	drinking coffee
	schnell laufen	running fast

LEVEL 2	**2^{+1}-word utterances: VP with prep phrase**	
• THEME	**WHERE / WHERE TO - WHAT DOING / DONE**	
• STRUCTURE	**PP - V**	**V - PP**
• EXAMPLES	nach Wien gefahren	gone to Vienna

LEVEL 3	**3-word utterances: VP plus subject**	
• THEME	**WHO - WHAT DOING / DONE**	
• STRUCTURE	**N - N - V**	**N - V - N**
• EXAMPLES	Katie Bein gebrochen	Katy broken leg

LEVEL 4	**3/4-word utterances: VP plus S-adverb**	
• THEME	**WHEN - (WHO) - WHAT DOING / DONE**	
• STRUCTURE	**A - (N) - N - V**	**(N) - V - N - A**
• EXAMPLES	gestern Brief geschrieben	written letter yesterday

LEVEL 5	**3/4-word utterances: VP plus indirect object**	
• THEME	**(WHO) - WHOM - WHAT DOING / DONE**	
• STRUCTURE	**(N) - N - N- V**	**(N) - V - N - N**
• EXAMPLES	(Lisa) Leo Brief geschrieben	(Lisa) written letter to Leo

In severe chronic agrammatism the ceiling performance is usually reached in level 2. In recovering chronic agrammatism the treatment can go on up to level 5, requiring repeated periods of intensive therapy. In acute agrammatism and mild forms of chronic agrammatism the training of level 3 often facilitates immediately complex syntactic processing. The treatment methods should then be changed to a treatment of those morphosyntactic features which are still disturbed.

Level 1: Initially, only two-word utterances always consisting of a non-finite verb and a noun functioning as direct object are practised (cf. German *Hände waschen, Kaffee trinken,* or English *wash(ing) hands, drink(ing) coffee*). We thus start with object-verb phrases have a more or less fixed collocation. A collocation is defined here as the joint occurrence of two or more words in the same utterance. This could be an idiomatic expression or highly overlearned phrase (e.g. in English *shake hands* or *wash dishes* etc.).

The two-word structures (N-V in German or V-N in English) are systematically practised in tasks that allow the patient to comprehend and produce elliptic answers to cueing questions from the therapist. The linguistic context may vary, for example

- naming of multiple choice sets of pictures/written stimuli with minimal semantic contrast in agent, object or activity,
- completion of brief and long stories with and without parallel depiction.

Irrespective of the context, for cueing the therapist uses always the same type of question, which is orally put forward in morphosyntactically complete forms, e.g.

- Therapist: *Was macht/machen X? Was hat X gemacht?*
  (What is X doing? What has X done?)

The spoken cues are supported by written and graphic cues which for the patient model the structure of the elliptic answer. The graphic cues introduce minimal metalinguistic information marking both word category (noun, verb) and word order.

In level 1 of REST, the structure of VPs should be changed according to the strict subcategorization parameters of the target language. In German, we introduce intransitive verbs together with morphologically unmarked adverbs, e.g. *gut schlafen, lange bleiben, viel arbeiten* (cf. *sleeping well, staying long, working hard*).

Level 2: On this level of REST therapy, prepositional phrases are introduced which express spatial relationships (location and direction), e.g. *im Bett liegen, nach Köln fahren* (cf. *lying in bed, driving to Cologne*). It might appear inappropriate to use prepositions at all in a reduced syntax approach, since prepositions are often considered to be function words like articles and pronouns. However, in logical semantics they are treated like predicates, and it was empirically found that they are relatively better preserved in agrammatism when they carry meaning of their own (Friederici, 1985).

The REST structures are again stipulated by questions of the format *Was macht/machen X? (X what is/are X doing?*). If the patient's response contains only the verb, the missing locational or directional complement is stimulated in a second step by introducing the corresponding interrogative pronouns *Wo/Wohin? (Where/Where to?*).

The following example (translated from the original German) illustrates stimulating of REST-structures:

- Stimulating context: action picture cards and written stimuli with object-verb-phrases demonstrating a sequence of every day actions: Going to the dentist, sitting in the

waiting room, reading newspaper, making a date, opening mouth, rinsing mouth, saying Goodbye

- Starting practice:

Th. *How did the story start ?*
P. feel pain . . go to dentist
Th. *Yeah, what happened next ?*
P. I have ...
Th. *Imagine you're already at the dentist's . . what are you doing in the waiting room?*
P. newspaper . .
Th. *I dont know what are you doing . . buying or reading newspaper? (pointing to the written card: What doing)*
P. reading newspaper . . (groans)
Th. *OK .. reading newspaper (pointing to the written cards and pictures) now you're being called in and you're going in to the dentist . . . what are you doing?*
P. but here (points to head). . I know it . .
Th. *You know it but you can't say it . .*
P. rinse mouth
Th. *and ?*
P. I don't know (laughs) . . goodbye
Th. *If you didn't have a new appointment . .*
P. ahh . appointment
Th. *I have to guess what you are doing . . please give me the full information . . more than one word . . What are you doing?*
P. make an appointment
TH. *OK . . now it is comprehensible . . what you are doing*

Level 3:

The REST structures being practised in level 3 consist of three constituents: subject - complement - nonfinite verb, e.g. Frau Zeitung lesen, Mann zum Krankenhaus gehen (cf. lady reading paper, man going to hospital, etc.). These structures are rarely found in elliptic utterances of normal speakers.

Why do we nevertheless introduce and practice them in level 3? Obviously, subject-verb agreement, one of the most demanding morphosyntactic operations is not required, just as case assignment was not required in level 1 and 2. From a functional perspective, however, these

structures allow to convey thematic relations and are consequently comprehensible for the communicative partner.

Level 4 and 5:
On level 4, free adverbial expressions are introduced primarily for temporal and local modification. They are again stipulated by an interrogative pronoun (English: When and Where (German: Wann and Wo). In a step-wise fashion, these free adverbials are combined with all REST structures practised earlier in level 1 to 3. Examples are: yesterday got flowers, next year going to Europe, tonight president speaking on TV.

On level 5, one finally proceeds to verbs with the most complex subcategorization frames. These are transitive verbs being complemented by both a direct and an indirect object. The corresponding thematic roles are always inanimate object and animate experiencer as in taken (away) bag (from) granny (cf. German: Oma Tasche weggenommen).

On all levels, it is necessary to support the formulation of REST structures by pictorial material, especially when new structures are introduced. The selection of the linguistic material should be adapted to the personal interests, linguistic skills and communicative needs of the patients as long as the overall principles are observed, namely systematic expansion of sentence fragments.

Outcome of the therapy study

Here we report the main results of the therapy study. Further data and the detailed discussion are provided by Springer *et al.* (2000). The outcome of the REST approach was examined in a group of eleven right-handed patients having chronic agrammatism. Six of the treated patients were male, five were female. The median age was 46 years (range: 31-73). The aetiology was vascular in all cases with extensive infarcts of the left perisylvian lesion.

Each patient received a total of 30 full hour treatments exclusively according to the principles of Reduced Syntax Therapy. After treatment, in eight of these patients the spontaneous language showed significantly more constituents per utterance, more non-finite verbs and in four cases even more morphosyntactic elements. As REST focuses specifically on verbs, we also assessed changes in their relative occurrence. Five of the eight patients with expanded clause structures indeed produced more verbs.

From follow-up data in four of the patients one can conclude that the improvements obtained from a relatively short training period may be long-lasting. Even if the linguistic outcome appears to be limited, it is important to note that transfer into spontaneous language was achieved. This improvement took place despite the fact that the mean duration of aphasia was over three years, during which time several attempts were made to stimulate the relearning of grammatically correct sentences. None of these earlier attempts had led to a stable transfer into spontaneous speech.

DISCUSSION

In short, the REST approach fits into a number of assumptions of the adaptation theory. REST tries to motivate and stimulate patients to choose a "simpler language" in conversation. In acute agrammatism, this may facilitate a restoration of normal syntactic processing. In chronic agrammatism, this may optimise the reduced language capacities being left and in some cases even stipulate long lasting relearning of complex syntax and morphology. Future neurolinguistic research has to specify what this "simpler language" relies on. Even after extensive left hemisphere damage, the ability to produce normal elliptic speech might remain relatively undisturbed, with the advantage of demanding less computational capacity than morphosyntactically complete speech. Alternatively, agrammatic language as it evolves during the natural course of aphasia reflects bilateral protolanguage capacities, which can be facilitated in the intact nondominant hemisphere. In fact, these two hypotheses could even be seen as compatible: the remaining ability to construct the skeleton of elliptical utterances and to gradually spell them out morphologically could be a bilateral "protolanguage" function. In either case, the REST approach would seem appropriate and extend the capacities still available in conditions of aphasia.

Preliminary results of our research open further perspectives of integrating REST structures into Computer-Mediated-Communication (CMC). Through visible, permanent and simultaneously presented language signs, the media platform of the PC supports the working memory and allows for repeated processes of self-repair and interactive repair. Even in standard CMC, mean length of utterances is short and non-finite structure occur frequently. Furthermore, the tolerance of the partner in relation to graphemic or grammatical errors is generally greater. Thus, the task of therapeutic research must be to analyze both the structural features of these media and the new conventions of communication, with the aim of achieving more targeted use of the resources of agrammatic subjects. The REST approach provides even patients with impairments of sentence production possibilities for remote communication.

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SECTION 3

FUNCTIONAL, PRAGMATIC AND PSYCHOSOCIAL APPROACHES TO APHASIA THERAPY

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Chapter 11

FUNCTIONAL AND PRAGMATIC DIRECTIONS IN APHASIA THERAPY

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HISTORY

The year 1975 was pivotal. Egypt reopened the Suez Canal eight years after it was closed during the Arab-Israeli War. Fighting occurred between rival rightist Christians and leftist Moslems in Beirut. Saigon, Vietnam was taken over by Communist forces and the United States presidency was besmirched by the Watergate scandal. In the UK, Margaret Thatcher became leader of the British Conservative Party, succeeding Edward Heath. An animal encephalitis outbreak raged in 16 U.S. states. Mrs. Junko Tabei, a Japanese woman, became the first female to climb Mt. Everest, and the Anglican Church of Canada approved ordaining women to the priesthood. Charlie Chaplin and P.G. Wodehouse were knighted by Queen Elizabeth II. One month later Wodehouse died. We watched *Jaws*, read *Watership Down*, and sang *Lucy in the Sky with Diamonds*. Some events ring familiar harmonics through the pages of history.

In the science of aphasia a shift was evident as well. The emphasis on the characterization of language solely in terms of semantics and syntax began to lean toward language use and language in context. This cool breeze of language use or "pragmatics" began to influence thinking about child language and eventually was refreshingly transferred to acquired adult language disorders. In the United States, Audrey Holland (1982) and Bloom and Lahey (1978) interpreted the speech act theory of John Austin and John Searle (Austin, 1962) as a viable and important area of clinical focus. Traditionally, language analysis, particularly in aphasia, had been largely word and sentence based. Increasingly, glimmers of interest began emerging on such seemingly "unique" language uses as conversation, discourse styles, social

interaction, and even participation in language events in the real word context of negotiating a purchase of toothpaste, arguing a football decision, or calling a Chihuahua.

Speech act theory first colored our assessment devices. Holland (1980, 1999) directed us to assessment strategies that were based on simulated and actual communicative activities of daily living and eventually the use of narratives, participation in life communication events, and the use of conversational partners crept into our approaches to therapy. In addition, the time and payment constraints imposed on the delivery of health care services of the 1990's forced attention to modes and models of aphasia therapy that were now directed more to accepting the notion that aphasia in most cases is a chronic condition and the resurrection of the popularity of group treatment. Slowly, a transformed model of aphasia began to appear and questions were posed about chronicity, adjustment, and accommodation to aphasia within the context of the World Health Organization models of activity and life participation. In an important and fascinating document on the history of speech pathology in America, Duchan (2001) traces what she calls the Pragmatics Revolution 1975-2000. Consulting this history is reminiscent of the lessons learned by passing under the engraved quotation above the entrance of the Norlin Library at the University of Colorado-Boulder that reminded fledgling scholars "Who knows only his own generation remains always a child."

SOCIAL MODELS OF APHASIA

Models of aphasia that incorporate social elements as vital signs of a living, growing scientific and clinical discipline are increasingly apparent. The medical model of the assessment, diagnosis, and treatment of the acute phase of aphasia is no longer as appropriate as earlier translated. Certainly aphasia is associated with certain unalienable medical conditions and constructs, and these should neither be ignored nor abandoned. But aphasia is also social. It is based on communicating, interacting with others, and living in a societal context. The isolation and imposed withdrawal seen in so many people with aphasia may well be iatrogenic. That is, caused by the intervention. The relevance of knee-to-knee individual sessions of drill on word retrieval and phonologic precision ("No, no, no. Get that little tonguey up on the gum ridge. That's 'tea' not coffee.") may well be approaching passé for all but a few process oriented conditions. In Scandinavia, Australia, South Africa, Canada, the UK, Japan, and other parts of the globe, international conferences on treatment approaches for aphasia are increasingly imbued with elements of social context and interaction needs of communicators.

Life Participation approaches

A confluence of elements continues to shape the flow of aphasia treatment. Fitting well into social models of aphasia is the recently articulated Life Participation Approach to Aphasia. The American Speech-Language-Hearing Association (ASHA) sponsored and encouraged a group of clinical aphasiologists and scholars in what has become known as the Life Participation Approach to Aphasia Project (LPAA) Project Group (2001). This group has articulated the values that grew from their discussions and proposed a philosophy of service delivery designed to confront the pressures on the delivery of aphasia services from professionals, providers, funding sources, and consumers. Participants in this LPAA Project group included Roberta Chapey, Judith F. Duchan, Roberta J. Elman, Linda J. Garcia, Aura Kagan, Jon Lyon, and Nina Simmons-Mackie. Although much of what will be reviewed in this chapter has emanated from this group, there are many others who have contributed to the worldwide movement to incorporating social models of aphasia into contemporary treatment modes. In the United Kingdom, Pound, Byng, and Parr (2000) three onomatopoetic names that are reminiscent of sounds heard at a construction site, have contributed mightily to advancing social models of aphasia. In Australia, Linda Worrall (2000) has studied and implemented pragmatic and social strategies. In the US, Fratalli *et al.*, (1995), Lyon (1995), Damico, *et. al.*, (1999), and LaPointe (2000) all have incorporated these values into their study and advocacy of aphasia intervention. This movement is becoming increasingly global and surely there are others who have been instrumental that are not mentioned here.

The statement issued by the LPAA Project group was intended to have an impact on assessment, intervention, policy making, advocacy, and research in aphasia. The life participation approach to aphasia was conceived foremost as a consumer-driven movement that was intended to broaden and focus the goals of aphasia treatment. The pivotal concept of the approach is re-engagement in life with real life goals generated from the idiosyncratic needs of each unique participant in aphasia intervention. This approach is like the seasons. Changes occur in the needs and goals of any person in the rehabilitation process and the LPAA adapts to the seasons of recovery. In the acute phase the emphasis might be on establishing effective communication with nursing staff, physicians, rehabilitation therapists and immediate family members. A new season, beyond the acute phase, might dictate focus on getting back to work at the tax office or candy shop or becoming reintegrated into community football booster club activities. No matter what the season of evolution of the condition might be, the emphasis is squarely placed on re-engagement and strengthening daily participation in activities of choice. Re-engagement is highly dependent on a richly supportive environment. This dictates a treatment model that goes beyond the word-retrieval skills of the person with aphasia and extends into the family system and the community. A

non-supportive or non-enlightened environment can affect the daily routines of even those with relatively mild aphasia. Those researchers or clinicians who subscribe to a life participation or social model of aphasia rehabilitation must be prepared to extend their efforts not only from the communication functions of transmitting and receiving messages but to the sowing and nourishing of social links. The quintessence of the life participation approach to aphasia can be realized by appreciation of the following core values:

- Life concerns and empowerment of the consumer are at the heart of clinical decision-making
- Life participation is the primary explicit goal
- Real-life goals that change over time must be sensitive to the evolution of aphasia
- Inclusion: all with aphasia are entitled to treatment
- Outcome measures should document life enhancement changes
- Personal and environmental issues are targets of intervention

In the United Kingdom strategies for addressing living with communication disability have received considerable attention as well. Pound, *et al.*, (2000) have outlined the philosophy and therapies developed at an aphasia center in London called Connect: The Communication Disability Network. This social model is based on British disability theory and allocates considerable attention to the barriers that exist to appropriate social participation. The Connect project advocates therapies based on clients' experiences with their disability. It expends considerable attention to eliminating environmental, instructional, and attitudinal barriers that hinder participation in social experiences.

Another thrust of the British social model of aphasia is to expend some effort of the establishment of a new, self-actualized social identity. This is an area, in my opinion, and in the opinion of others, of another crying need in the study of disability in general and specifically in aphasia. Very little research has been conducted on identity and aphasia. Some research on identity and self esteem in aphasia (Brumfitt, 1999a, 1999b) has contributed to the observation that all may not be right in the twisted identity world of people with aphasia. Brumfitt (1999a) continues her work on methods to assess identity issues with her Visual Analogue Self Esteem Scale (VASES) and Code *et al.*, (1999) and others have noted the need for work in this area.

The crucial nature of identity change in aphasia was first illuminated to me in the early 1980's when John Rosenbek and I were speaking at a conference and workshop on aphasia treatment sponsored by the parents of a young clinician who had been tragically killed in an automobile accident on her way to a home visit of a client with aphasia. At this Texas conference, Rosenbek and I presented the workshop portion and the celebrity keynote speaker was the

noted American playwright, Arthur Kopit. Kopit had recently completed his powerful play, *Wings*, which revolved around the altered life of Emily Stilson, a former aviatrix and wingwalker, who had become aphasic. Kopit based the play in part on his experiences with his father's aphasia and on his extensive observation of people with aphasia at a New York rehabilitation center. In a late night conversation with Mr. Kopit he enlightened us to some issues in aphasia of which we had given little previous thought. He said that one of his father's recurring questions was, "Arthur, when am I going to be me again? Is this me? Will the therapy return me to me?"

This revelation had a significant influence on our appreciation of the need for research on psychosocial aspects of aphasia, especially on the crying need for research on identity change and the perception of identity in stroke and aphasia.

CHRONICITY AND LIFE QUALITY

Pragmatic and life participation approaches to aphasia research and intervention are closely tied to the concepts of chronicity and quality of life. The psychosocial sequelae of stroke and aphasia are discussed by Code and by Byng in other chapters of this book. Families of those who live with aphasia appreciate these sequelae on a daily basis. The world is inverted, twisted, and sometimes as distorted as images in a carnival house of mirrors. Mood disorders and emotional alterations abound. Role change is necessary. Employment is lost or radically altered. Leisure activities change. Mobility and communication disability issues mingle to radically amend living arrangements and personal living space. Most importantly, interpersonal relationships with spouse, children, and friends are transformed. All of these factors are amenable to influence through life participation approaches to aphasia intervention. Yet, as influential as these factors might be, we have a long way to go to fully understand them or deal with them. For example, the issue of relative value placed on mobility versus communication is unclear. As professionals deeply interested in human communication and its disorders, we are quick to boost communication high up the hierarchy of activities precious to us. Little research has been conducted on perceived relative value of mobility versus communication. A recent study conducted by me and some of my Florida State University colleagues and graduate students revealed some surprising results (LaPointe, *et al.*, 2001). We entitled the project the "Walk the walk, talk the talk Project," and surveyed over 140 members of the general public with a series of questions designed to reveal the relative value they placed on regaining walking versus regaining talking after a stroke or brain injury. The hypothetical situations we described were, "If you were to have a stroke or other brain damage and loss the ability to walk and the ability to talk, which ability would you most

like to regain?" We posed the same question for spouse or significant other; for a child (if they had one); and for their parents. Interesting gender effects emerged. Female respondents were likely to favor the return of walking versus talking. Men, however, favored walking for themselves, for a spouse or caregiver, and for parents. They did value the return of talking more than walking for a child. These findings have illuminated a need for some fairly basic research on relative value of human activities that could have an impact on our models of rehabilitation and particularly on the allocation of limited resources among the therapies. We need to find out a lot more within the realm of aphasia sociology to better guide our services.

Storms and wrecks

In terms of communication rehabilitation, the late 1990's provided a rather stormy ride into the new century. In the United States the profession was rocked by policy decisions that decimated services for those with aphasia. Reimbursement caps were placed on services and the heavy hand of managed care (whom many thought was more managed than caring) created loss of jobs, reduction in rehabilitation services, and the relegation of people with aphasia to human warehouses or back home with no continuation of care. Fortunately the times have changed somewhat, but the clouds are still somewhat austere. Holland (2001) has commented on reasons for optimism about aphasia rehabilitation. She presented an invited lecture at the sesquicentennial of our university on the rise of group intervention as a viable modality of intervention and support. She highlighted as well the benefits inherent in shifting the focus of aphasia treatment from impairments to the new ICIDH-2 Beta 2 models that emphasize outcomes related to activity and participation. With the rise of group modes of intervention it becomes quite typical to hear participants with aphasia reiterate the issues of chronicity and adaptation that are echoes of all who have faced these storms. When the wreck of aphasia occurs, the big questions surface and recur. How does one accept a radically tilted life? How does a family adjust to a life that may be tenfold the challenge on Friday what it was on Monday? Is it possible to deal with chronicity and a prodigiously changed existence? Can relative happiness ever be attained or regained? Is there precedent for dealing with the illness experience? How does one cope with chronicity? How does one live with aphasia?

It isn't unusual for persons with aphasia to endure stages of chronicity that are not unlike stages of grief. These stages include realization, denial, mourning, and finally perhaps adaptation. Theories of adaptation to chronicity involve all of the systems that revolve around and within the individual, the family, the community, the culture, and society. Demographic variables also can have an influence on perceived wellness or life quality. Recent work by Blanchard, *et al.*, (2001) has highlighted age differences as well as circadian rhythm

influences on measured perceptions of life quality. These are more good reasons for increased attention to the sociology of aphasia. Researchers have studied personality characteristics of the individual that seem to be related to variability in adaptation to illness, chronicity, and aphasia. Concepts that emerge include the personality traits of temperament, hardiness, introspection, dispositional optimism, and perception of locus of control. These issues have been discussed in previous work on adaptation, coping with chronicity, and quality of life with aphasia (LaPointe, 1996; 1997; 1999; 2000).

Keys to successful aging and coping

In the United States some longitudinal research conducted at Harvard University has revealed some interesting lifestyle attributes or keys that are alleged to be associated with successful aging, wellness, and positive life quality. George E. Vaillant and his associates have conducted longitudinal studies with a duration of more than 30 years (Vaillant and Mukamal, 2001). Vaillant and several of his colleagues through the years followed three demographic groups (n = 824). One cohort was comprised of Harvard University graduates, one of inner city Boston men, and one of gifted California women. These groups were given psychological and medical tests, physical examinations, and interviews by psychiatrists and asked to evaluate their own lives and feelings. Some of these groups were followed and evaluated for up to 40 years. The evaluations and perceptions of a large group between the ages of 60 and 80 years were analyzed and three distinct groups emerged: a group characterized as the "happy well;" one labeled the "sad sick" (who had various ailments and decreased perception of enjoyment of life); and a group called the "prematurely dead" (a subset of participants who had died prior to the age of 60). The study is fraught with complexity and defies oversimplification, but some of the conclusions rendered by Vaillant and his associates are difficult to ignore. The personal attributes that characterized the happy well group (and qualities that were remarkably less prevalent in the other two groups) included:

- **Orientation toward the future** – this included the ability to anticipate, plan, and hope
- **Gratitude, forgiveness, and optimism** – these traits included the classic views of important life lessons during adversity and generally perceiving bottles, cups, and glasses and life events as half full rather than half empty
- **Empathy** – the ability to imagine the world as it seems to the other person (this is strikingly close to the concept of "theory of mind" that has attracted considerable research interest in persons with right hemisphere damage)
- **Reaching out** – this is the desire to do things with other people, not to do things to them; or the rumination that other people keep doing things to us

The Harvard studies also pointed out the positive aspects of support systems and of participation in societal and community activities. It is not difficult to see the links among all of these concepts that have been associated with positive and successful aging and coping to the chronicity of aphasia. Surely the functional, pragmatic, and life participation approaches reviewed and advocated in this chapter are vehicles for facilitating successful life with aphasia. It is sadly apparent, however, that very little empirical research has been conducted on many of these issues, particularly of effectiveness and efficacy of life participation approaches. Gigantic questions remain to be addressed by both qualitative and quantitative research. These include:

- Whose role is it to facilitate these attributes of successful coping?
- How do we identify, validate, and implement these attributes?
- What needs to be done to establish a research base of credible evidence on the value of coping and life participation?
- Can we establish a merger of qualitative and quantitative research approaches that coalesce and complement rather than being perceived as disharmonious?

Our challenge in clinical aphasiology, in my view, is to adapt to the changing landscape of aphasia intervention. Pragmatics, functionality, and life participation need to be clearly and acceptably defined. Issues in aphasia are medical, rehabilitative, economic, humanistic, sociological, and ethical. Attention to process and impairment need not be abandoned. But the complexities of aphasia and challenges of reintegration into the neighborhoods and communities of the planet need more. More than just the wisdom and prescription of the health care professionals who have been involved traditionally in the medical model of aphasiology. Participation and choice are golden. We hope for the rise of the consumer. We anticipate the power of the advocate. We call for the incorporation of relevance. Not only will this create a more holistic model and theory of aphasia and address the genuine plight of wives and fathers with aphasia but, as has been whispered before, it will better prepare those future clinicians destined to treat us.

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Chapter 12

CONVERSATION ANALYSIS AND APHASIA THERAPY

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INTRODUCTION

It has been a source of unease for aphasia therapists for many years that their and their patients' efforts at reducing the impairment of language, particularly in respect of word-finding and sentence construction, seem to have had little carry-over to everyday life. Success at picture-naming and picture description has often been regarded as an end in itself, even to the extent of resulting in discharge from therapy when a stable plateau has been achieved, without an assessment as to whether there has been improvement in the daily use of language to manage one's life. Improving picture-naming and description remains a favourite achievement in clinics. The sophistication achieved by the application of models from cognitive neuropsychology has led to a major increase in therapists' skills in diagnosing and treating facets of psycholinguistic impairment. Moreover there is burgeoning evidence through statistically valid experimental designs that such treatments are effective in improving results on these clinical tasks (e.g. Nickels and Best, 1996). Treatment of the *impairment* aspect of aphasia has thus developed a powerful momentum.

A reaction since the seventies to this unease at the emphasis on clinical tasks has been to consider enhancement of "functional communication" as a parallel duty of the aphasia therapist. This was spearheaded by assessments of everyday communication notably developed by Sarno (1969) and Holland (1980). Today, the World Health Organisation and hence the purse-holders have recognised that reduction of *handicap* (with increased *activity* and *participation* in social life) is more relevant than improvement in picture naming as an end in itself. Consequently the assessment of functional communication has become a growth industry (Worrall and Frattali, 2000). Less certain has been how to translate this into therapy,

although a number of approaches have been advocated. These range from the clinically based Promoting Aphasics' Communicative Effectiveness (PACE) (Davis and Wilcox, 1981), through Holland's Conversational Coaching (1991) to Lyon's (2000) vision of the aphasia therapist as a facilitating agent of change, guiding the aphasic client in matching existing skills with life challenges.

The problem for the therapist is that, whereas treatment of the impairment has become increasingly skilled and elaborate, the handicap approach hovers between the clinically prescriptive (as in PACE) and the nebulously well-intentioned. If the use of language as central to everyday life is the admitted ultimate target of intervention, what means do we have for examining it, for analysing its structure, and possibly for influencing it through therapy? For this aphasia therapists have turned to the techniques and principles of Conversation Analysis (CA).

CONVERSATION ANALYSIS

The beginnings of CA have been traced to Harvey Sacks' appointment in 1964 to the Suicide Prevention Center in Los Angeles. Here he had the opportunity to study recordings of telephone calls. This provided him with natural data on the management of two-way exchanges, through which to search for the structure and mechanics of conversation. CA has followed the same principles since then, of seeking a natural data base, without preconceived theoretical categories of analysis, though expanding its realm to face-to-face interactions and including non-audible communication, through the now generally accessible use of videorecording.

There are three central characteristics of CA, as it has been applied in aphasia research. First, all the data must be considered, including minimal utterances such as "uh" and "er", micro pauses, sound prolongations, coughs and laughter: all have communicative consequences depending on the sequential contexts in which they occur. This brings us to the second characteristic, that each utterance or non-verbal occurrence has a significance which is sequentially constructed by the conversational partners step by step. This means that utterance "tokens" cannot easily be grouped together out of context for quantification. "Yeah", for example, can function, depending on context, as an answer, a conversational continuant, a marker of a topic shift, an absorber of an overlap, or as a claim to take the floor. The third key characteristic is that the analyst interprets the exchange through the participants' own behaviours, and not by pre-defined categories or an observer's judgements of success. For a fuller account of CA as relevant to aphasia, see Lesser and Milroy (1993).

The traditional "functional communication" approach to aphasia, therefore, differs from that of CA. The functional approach applies a theory base, drawn variously from linguistic constructs such as speech acts, or appropriacy and normality. Assumptions of what is appropriate or normal have their risks, especially with a culturally diverse and older population (Garcia and Orange, 1996). Assessment is made through the observer's judgements and focuses on the aphasic individual's failures and successes. Recommendations for therapy, in contrast, are not essentially individualised but are based on general principles, such as advice to the relatives to "give time", "use simple sentences", "rehearse the vocabulary needed". In contrast CA applies structures drawn from the data, rather than based on invented samples, uses the participants themselves as judges of success, concerns itself with interactions rather than the one-sided contributions of the aphasic speaker, but draws implications for therapy which are inherently individualised to the particular dyad.

REPAIRS IN CONVERSATION

The aspects of "talk-in-action" which have been particularly productive in aphasia research so far relate to turn-taking and repairs of trouble sources. Sacks *et al.* (1974) analysis distinguishes turn-constructional units in conversation, as evidenced by the avoidance of overlaps and gaps at places where a turn transition to another speaker is relevant. If an overlap occurs, absorbers such as "you know" or "yeah" may be used, and turn beginnings may be recycled. Pairs of turns sometimes include preferred responses. For example, silence in response to an invitation is likely to be taken as a refusal. These are all aspects in which an aphasic person's linguistic restrictions may cause trouble. A major restriction is likely to be the ubiquitous word-finding difficulties of aphasia, leading to the need for repairs. Here CA recognises two possible initiations of the repair ("self-" and "other-initiation") and two possible completions ("self-" and "other-repair"). Self-initiated other-repair is relatively frequent in aphasia, as in the example below:

(i)

1. Mr A: and once I get those done I'll have to cut all the er trousers the the the
2. Mrs A: the trees
3. Mr A: the trees that's right

(Lesser and Perkins,1999, p 103)

Here Mr A recognises that he has produced the wrong word and hints that he needs help. Mrs A supplies the word, which is repeated and accepted by Mr A.

Repairs initiated by an aphasic speaker can sometimes result in long hint-and guess sequences, not always achieving either self- or other-repair, as in extract (ii) (where pause length is indicated in seconds in brackets and overlap by arrowheads):

(ii)

1. Mrs D: anyway I'll tell you where we supposed to be going erm (1.0) in a few
2. weeks er from (7.0) erm what do you call it [w ∂](2.0)
3. Mr D: work
4. Mrs D: no from the Tuesday
5. Mr D: g<roup>
6. Mrs D: <[w∂]> aha what do they call it
7. Mr D: er foun-? fou<n>?
8. Mrs D: <the> foun fountain (1.2) the fountain (3.0) ee I don't know I'm very
9. I'm not very sure
10. Mr D: OK well where are you going what are you
11. Mrs D: we're going (4.2) the (4.0) no (2.8) the (4.2) hhh in it's in (1.5) what do
12. they call it erm in Whickham
13. Mr D: Whickham?
14. Mrs D: Whickham (1.5) the (2.3) [k∂] the (2.6)
15. Mr D: the baths
16. Mrs D: no no no hehehehehe the [grei] not the <[grei]>
17. Mr D: <the> garden centre
18. Mrs D: the garden centre
19. Mr D: the garden centre

(Lesser and Perkins,1999, p 105-6)

This kind of lengthy collaborative sequence, characterised by self-initiated prompts for other-repairs, with further repairs of misunderstandings, is not atypical. It illustrates the collaborative nature of conversation, which is particularly noticeable in interactions with aphasic speakers (Milroy and Perkins, 1992). In this example Mr D frequently waits for quite long periods (up to 7 seconds in line 2) without offering an other-repair, giving his wife the opportunity to initiate an explicit repair request herself. He also uses a strategy of backtracking to the original theme (line 10), which seems to be successful in moving the conversation onward. Overlaps occur where each speaker is responding to a clue (lines 5, 8 and17), and are generally followed by repetition (lines 8, 18 and 19). In example (ii) Mrs D shows awareness of her word-retrieval failures through her initiations of repair, and her provision of phonemic cues (lines 2 and 16) and circumlocution (line 12).

More serious misunderstandings can occur in everyday conversations due to problematic sequential implicativeness (Schegloff, 1987). Idiomatic utterances may be taken as literal, serious as humorous, humorous as serious, statements as complaints. With aphasic speakers who do not monitor their semantic paraphasias, it can be the referent which is problematic, as in (iii).

(iii)

1. Mr. P. [chipsei set] what does Walter do with them
2. Walter: eh?
3. Mr. P. what's he gonna do what's he gonna
4. Walter: he's coming in about an hour he's pubbing isn't he
5. Mr. P: he's what?
6. Walter: he's coming to pick you up in about an hour
7. Mr. P: who's back by what's that for?
8. Walter: Douglas
9. Mr. P: oh he's coming is he ah
10. Walter: he's coming down here
11. Mr. P: ah

(Lesser and Perkins,1999, p 103)

In line 1 Mr P does not seem to be aware that he has produced what is evidently a semantic paraphasia (since it is Walter that he is speaking to). His indeterminate "them" also leads Walter to initiate a repair. From line 9, however, we learn that Walter's other-repair (based on the assumption that Mr P's paraphasia had been a reference to his son, Douglas) was a misunderstanding. The original topic has become derailed, and the original theme of "Walter" and "them" is abandoned.

CORRECTION AS THE INTERACTIONAL BUSINESS

It is not only in such sequences where the aphasic speaker does not appear to be self-monitoring that the conversation can become derailed, with exposed correction itself becoming the interactional business. Take example (iv) where Mr W is insisting on correct pronunciation of a phrase that his wife has at last achieved in finalising a shopping list.

(iv)

1. Mrs W: and I'll tell you what I want I need some (2.5) oh (2.5) it's it's a [d∂]D
2. Mr. W: D?
3. Mrs W: a (2.4)
4. Mr. W: to eat?

5. Mrs W: no it's a (3.0) it's not a D a [dð] a death no
6. Mr. W: a death you don't want a death
7. Mrs W: *{laughter}* shush I'm thinking it's a (2.0) death no ***heh***
8. Mr. W: er (1.0)
9. Mrs W: [desh]
10. Mr. W: dish?
11. Mrs W: dish thank you dish (1.5)
12. Mr. W: washer
13. Mrs W: wipe wipe
14. Mr. W: a dish wipe
15. Mrs W: wiper aha
16. Mr. W: a dish cloth
17. Mrs W.: mhm
18. Mr. W: a what?
19. Mrs W: er what?
20. Mr. W: dish cloth
21. Mrs W: dish (1.0)
22. Mr. W: cloth
23. Mrs W: cloth cloth
24. Mr. W: a dish cloth
25. Mrs W: dish cloth
26. Mr. W: fine
27. Mrs W: erm because Steven (1.5) I don't know what he's done with those ones
28. that I've got but they're (1.5) they're terrible

(Lesser and Perkins, 1999, p 108-9)

Here the correct production of "dish cloth" becomes the interactional business in line 18, although the collaborative sequence with the aphasic wife providing phonological and orthographic cues has achieved understanding. This kind of behaviour keeps the aphasic person's linguistic incompetence on the conversational surface. Lengthy correction is unusual in conversation between nonaphasic people, where corrections may be unobtrusively embedded through later repetition of the corrected word rather than exposed as the interactional business (Jefferson, 1987).

Another strategy adopted by some aphasic speakers with certain partners is to allow them to carry the main burden of the conversation, through relying on minimal turns to pass the conversational floor back, as in (v).

(v)

1. Cousin: Why you can't understand it when you're <2 syll.>
2. Mrs M: <well its its> its [te]
3. Cousin: I must be <thick> never mind Jean
4. Mrs M: <mhm> {*laughter*}<eee>
5. Cousin: <aye you cannot understand> half of them
6. Mrs M: aye (2.3)
7. Cousin: but er y'know we used to get a a rebate from Scotland
8. Mrs M: mhm
9. Cousin: y'know off the rent
10. Mrs M: mhm
11. Cousin: well that's been stopped 'cause we cannot get a rise on our pensions
12. Mrs M: mhm
13. Cousin: so I'm not allowed that anymore <so> that's stopped it's finished
14. Mrs M. <tsk> *{laughter}*

(Lesser and Perkins,1999, p 141)

This reliance by Mrs M on minimal turns was only observed with interactions with her cousin. With another speaker Mrs M successfully took the floor and produced major turns.

All the above examples show the importance of the role played by the nonaphasic interlocutor. Simmons-Mackie and Kagan (1999) have distinguished "good" and "poor" conversational partners. In this study the poor partners were unfamiliar with aphasia and with the individuals concerned. They used more words that highlighted the disability, and more disjunct markers such as "well". The good partners, on the other hand, used many acknowledgement tokens, congruent overlaps such as head nodding and laughter, face-saving clarification sequences and accommodated readily to the use of other media of interaction such as writing. Since relationships in everyday life are constructed and maintained through talk, the consequences of interactions with a habitually poor conversational partner can be severe. They can include repeated loss of "face", and a change of role associated with a reduction of self-esteem leading to social restrictions, depression, frustration and anger. The involvement of the aphasia therapist in facilitating conversation can therefore be significant.

IMPLICATIONS FOR INTERVENTION

It is evident from the examples above that facilitation of conversation is unlikely to be achieved through work with the aphasic individual on his or her own. Therapy needs to be

mediated through both contributors to the conversation, and, given the inherent limitations of one of the speakers, the main burden is likely to fall on the nonaphasic partner. The partner needs to be aware of what the linguistic impairments of the aphasic speaker are, and here the clinician's detailed assessment of psycholinguistic processing plays an important part in helping the partner to understand the nature of the individual's restrictions. Perkins has shown that the nature of the psycholinguistic impairment influences the time taken to resolve trouble sources (1995). The generalised advice to relatives given in the functional-communication approach may help or hinder conversation exchanges, depending on the type of processing which has been impaired. For example it may not be helpful to use multiple choices in other-repairs with someone who has a semantic disorder, or to rush in with an other-repair with someone with a phonological lexical disorder who frequently achieves word retrieval by self-repair after a delay. The conversation partner also needs to be sensitive to the strategies used by the aphasic speaker to gain or keep the conversational floor or to encourage the partner to carry more of the burden. Such strategies can include simplifying syntax, dislocation of noun phrases, reliance on pro-forms and direct reported speech, verbal deixis (e.g. "the second one"), re-use of the partner's word or phrase, and use of laughter as a conversational lubricant (Simmons-Mackie and Damico, 1997; Wilkinson, 1995, 1999; Leiwo and Klippi, 2000).

APPLYING CA IN THERAPY

Conversation has been the direct target of therapy for at least a decade, since Holland (1991) developed her system of Conversational Coaching. More recently Kagan (1998) has productively trained volunteers at an Aphasia Centre in Canada to support conversation. To date, however, these therapists have not reported the application of principles from CA.

An early attempt at CA informed therapy by Lesser and Algar (1995) used a personalised booklet given to two conversation partners of a 49 year old woman. Her persisting word-finding difficulties were attributed to impaired selection in the phonological lexicon, and this was illustrated in the booklet. Extracts from recordings of conversations made at home with her friends showed that she was able to self-repair given enough time, and preferred this to other-repair. Examples from the conversations were included in the booklets, with advice to the friends. The analysis of recordings made three months later showed an increase in successful outcomes to repair sequences, and a reduction in abandonment of repairs. Picture-naming on a clinical test showed no improvement. This study used minimal involvement of the therapist, apart from the labour-intensive transcription and analysis of the tapes.

Wilkinson *et al.* (1998) provided more therapy input with a 37 year old aphasic woman,

Connie, and her husband, in the form of four 2-hour sessions where the couple and the therapist discussed a videotape made at home and highlights from a transcription of it. It was clear from the videotape that the husband's prolonged other-repairs had caused distress to the wife. There were also sequences of up to 50 seconds in length where the wife's pronunciation became the topic (as in example (iv) above) and the conversation became derailed. A videotape made after the therapy sessions showed that the husband was now allowing his wife's phonemic mispronunciations to pass without initiating other-repair sequences.

The different reactions of Connie and Mrs W in example (iv) to other repairs of their pronunciation show the importance of judgements of conversational success being made by the participants rather than by the therapist. In an attempt to address this issue Whitworth *et al.* (1998) devised a Conversation Analysis Profile for People with Aphasia (CAPPA). This compares an aphasic person and carer's perceptions of their conversational abilities with an objective analysis of a sample of conversation audio-recorded at home by the two participants. The profile includes a structured questionnaire for each participant on their use of language, repairs, turn-taking and topic management. It asks them what they consider to be problems or not, investigates what they consider to be successful strategies and compares pre- and post-morbid communication styles, situations and contacts. CAPPA can be used to probe how insightful the speakers are into their conversational practices, and, as in the Wilkinson *et al* (1999) study, to discuss what topics could be used other than repair of pronunciation. CAPPA is both simpler and more explicitly focussed on conversation that the four-stage Communication Profiling System being developed by Simmons-Mackie and Damico (1996), which involves the therapist in data collection and introspection.

CAPPA provided the framework for another application of CA in aphasia therapy by Booth and Perkins (1999). The intervention in this study involved group training of four carers of different patients, comprising six 2-hour sessions discussing the patients' psycholinguistic abilities and CA transcripts of 10 minute recordings made at home using a radio-microphone. One dyad consisted of JB, with impaired comprehension and speech characterised by semantic and phonemic paraphasias, and his brother. It was noted that repair frequently became the interactional business, with topics becoming derailed, and 78% of major turns being involved in repair sequences. The brother would sometimes withhold collaboration after JB requested help. CAPPA revealed that the brother was unaware of JB's comprehension difficulties, but knew that few of his own strategies were successful. After intervention repair sequences were reduced to 29%, there were no examples of repair becoming the interactional business, the mean length of repair sequences decreased from 35 to 8 turns and the brother was checking on JB's understanding and seeking clarifications in collaborative repairs. JB himself was also now initiating clarification searches.

QUANTIFICATION

Studies of the effectiveness of intervention inevitably run into the question of quantification. The CA approach is inherently cautious about quantification, given the dependence of utterances on sequential context (Schegloff, 1993). Nevertheless if CA is to be shown to be of use in intervention, and to justify the time taken in transcription and interpretation, some measures of pre- and post-testing are essential. Hicklin *et al.* (2001) have tackled this by using three measures. One compares major and minor turns. Minor turns are defined as those which do not move the conversation on but merely signal acquiescence in its being continued. A second measure is the number of breakdowns and repairs. The third is a count of content words, including nouns as a subset, and various trouble indicating behaviours which show problems of lexical retrieval. Using this measure a strong relationship was found between picture-naming success and noun retrieval in conversation.

Perkins *et al.* (1999) have directly addressed the question of test/retest reliability in the use of CA. They recorded and analysed the conversations of eight aphasic people with their relatives on four consecutive weeks, five of the dyads video-recording themselves and three audio-recording themselves at home, each at a consistent time of day. The quantitative measure used was the proportion of major turns involved in collaborative repair. There was significant variation between conversations within the dyads, but the variation between the different dyads was much greater. Perkins et al stress the need for extended baselines for quantitative comparisons within dyads over time, but they also noted qualitative consistencies in interactional management across the four occasions for each dyad.

LIMITATIONS AND STRENGTHS

The problem of quantification is only one aspect of CA which currently limits its application in aphasia. The therapist's time taken to obtain the data may be minimal, given the increasing familiarity of many households with the making of recordings, but transcription, even of the five or ten minute samples considered adequate by Boles and Bombard (1998), remains laborious. Transcription from video-recordings takes longer but has the advantage of permitting the analysis of the use of gaze and other non-verbal means of interaction (Oelschlaeger, 1999). The task can be facilitated by the use of laboratory techniques such as VideoLab at University College London, where the recordings are transferred to compact discs (Ray Wilkinson, personal communication). Crockford and Lesser (1992) also showed that, although CA takes longer for transcription and analysis than two other standard means of assessing communication through role play or questionnaire, it provides considerably more

useful information for the therapist. The variability noted between partners of the same aphasic person (Lindsay and Wilkinson, 1999) also raises the question of whether therapy should involve the whole gamut of partners with whom the patient may interact.

CA, nevertheless, has considerable potential. The very variability between partners just noted can be used as an index of the patient's conversation resources under best conditions, and be used as a model. Its key strength is that it provides a structure for directly targeting the ultimate aim of therapy in improving quality of life. It is both collaborative and economically focussed on individuals' own circumstances and needs. With further development it promises to be a powerful addition to the aphasia therapist's resources.

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Chapter 13

SUPPORTED SELF-HELP GROUPS FOR APHASIC PEOPLE: DEVELOPMENT AND RESEARCH

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INTRODUCTION

Participation in self-help activities is seen as a useful approach to social reintegration for people with chronic conditions, contributing to the development of confidence and psychosocial reintegration (Jacobs and Goodman 1989; Coles and Eales 1999), empowerment and autonomy (Gay 1989). In this chapter we describe the development of supported self-help groups for people with aphasia and report the results of a study which aimed to determine the profile of the membership of the supported self-help groups for aphasic people in Britain.

SELF-HELP AND THE DISABILITY MOVEMENT

The concept of self-help as a collection of individuals coming together to cope with a shared problem has a long history. Self-help groups have been developed by people to improve housing and working conditions, enfranchise women and cope with the effects of drugs and alcohol. But it is in the second half of this century that self-help has come of age in the fields of health and disability. The development of the Disability Movement with its emphasis on self-determination has had a profound impact on people who want to take control of their own lives and also on the professionals who work with them (Finklestein and French, 1993). To a lesser extent service providers are also beginning to recognise the contribution that self-help can make to social and community reintegration (Wisdom, 1996).

Some of the largest associations in Britain representing disabled people were initially set up by relatives or parents to raise public awareness of a condition and to lobby for improved services. Many are now run on professional lines with accountants, fund-raisers and political lobbyists. Many employ large numbers of volunteers, contract out services and support research projects into the prevention or treatment of particular conditions.

For many years in Britain the British Stroke Association has run volunteer-led speech and communication groups and stroke clubs, based on an essentially philanthropic model. They provide support to survivors of stroke and their relatives through their Dysphasia Support and Family Support schemes within a sheltered environment. Professionals and volunteers determine the structure of conventional therapy-type activities, and people with aphasia are the generally passive recipients. The Dysphasia Support schemes receive statutory funding from the health authorities in the areas in which they operate.

It is perhaps because of this existing support that self-help in Britain has got off to a slower start than in some other European countries. In Germany and Belgium considerable statutory funding is provided and the rapid growth of self-help in parts of Europe may reflect the paucity of therapy for the majority of aphasic people. The European Commission has provided financial support for the establishment of the *Association Internationale Aphasie* as a focus for the exchange of information between countries, through its funding of a four year Helios project.

APHASIA THERAPY AND THE SOCIAL THEORY OF DISABILITY

In recent years a social model of disability has become more influential and we have seen some shift in the focus of aphasia rehabilitation from the traditional medical model to seeing aphasia as a disability rather than as an impairment. Aphasia results from impairment to cognitive processing underlying language, and therapy has been planned and executed to either restore or compensate for the impairments. But there has been an increasing recognition that language has an equally important social role, and language may have originally evolved through social interaction and for social purposes (Dunbar, 1996). Recognising the social relevance of language and communication requires that assessment and therapy involve analysis of the barriers and restrictions to successful communication (Togher *et al.*, 1996) and social interaction experienced by aphasic people, and working on challenging, overcoming and changing them (Jordan and Kaiser, 1996). There are indications that focusing education and training on those who come into contact most often with aphasic people (Kagen Forum), and the actual communicative act between interlocutors (Togher *et al.* 1996 and in press) can result in significantly more successful communicative interactions.

In Britain, therapy is still mainly provided by health and community services, and is mainly impairment based. It has been difficult to prove the efficacy and effectiveness of therapy for aphasia (see Code, 2000; Howard, 1986; Greener *et al.*, 1989; Robey, 1998; Robey & Schultz, 1998). Some of the problems of demonstrating effectiveness are probably due to a narrow focus on aphasic impairment and a failure to take full account of the importance of the social barriers faced by aphasic people. The lack of understanding of the significance of some of these barriers means that they fail to be included as important variables in successful therapeutic intervention. Acceptance of an altered self accompanying a disability and coming to terms with a new set of life constructs and circumstances is ultimately the most important task for the person with chronic aphasia. While many speech and language therapists understand and accept this, it has not been traditionally recognised as contributing to the goals of intervention and financial constraints have determined that clinical time is rarely invested in more socially relevant and authentic rehabilitation.

Membership of a self-help group for aphasic people can be seen to offer an important opportunity to develop empowerment and self-determination (Wisdom, 1996). Wann (1995) suggests that the benefits include the ending of isolation and the sharing of personal experiences of pain and anger and of finding practical solutions to problems. The atmosphere of openness and the development of trust and confidence also allows the airing of taboo subjects.

Members are all affected by the same condition and come into direct contact with each other. Activities are oriented directly to the needs of members. The groups are small, which facilitates face-to-face interaction and discussion. Contacts are unorganised, spontaneous and lively. Structure is frequently unstable, and often groups are often short lived (Matzat, 1989-1990).There may be significant overlap between the aims and activities of self-help groups, and other kinds of groups and organisations, as illustrated by Figure 1.

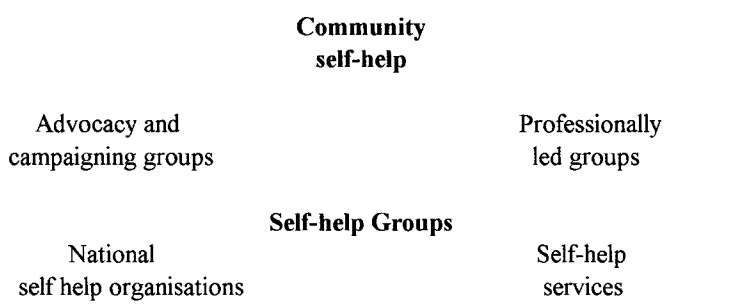

Figure 1. Overlap between self-help groups and other related groups and organisations (adapted from Wilson, 1995)

Rootes and Aanes (1992) identify seven criteria that define a self-help group, shown in Table 1.

Table 1. Rootes and Aanes (1992) seven criteria

1. A self-help group is supportive and educational.
2. Its leadership comes from within the group.
3. The group addresses a single major life disrupting event.
4. Group members participate voluntarily.
5. The group has no monetary interest or profit orientation.
6. The primary purpose of group membership is individual personal growth.
7. Membership is anonymous and confidential

THE ROLE OF THE APHASIA CLINICIAN IN SETTING UP A SELF-HELP GROUP

Self-help groups for aphasic people can offer an additional resource providing further opportunities to extend the network of long-term support outside a clinical setting. A self-help group can provide a natural opportunity for developing gains made in therapy and offer ways of exploring and developing skills and roles not promoted in formal therapy. Members can share emotional burdens and membership can help to enable and empower people to establish a new identity, where their own disability is recognised. The aphasic person can reach out to those with a shared experience and break away from being a 'victim' encouraged to refer to experts for help (Coles and Eales, 1999; Wann, 1995).

Typically, a self-help group starts with two or three interested people with aphasia and one of them will have sufficient language skills to take the lead. They are likely to need support with practical matters like find a meeting room, accessing transport and attracting other members. At first members may have difficulty simply in deciding issues such as where and when to meet. Time and patience are required to facilitate discussion and allow the time necessary for confidence to build. The clinician will need to commit time for maybe up to a year, supporting the group and helping it to understand the nature of self-help, to explore what they hope to

achieve and to develop aims for the group. The clinician will find that the important early issues are the practical ones of finding somewhere to meet and making sure that members can get to the meetings. Existing groups in the UK meet outside Health Service premises, in community halls, village halls and schools. Early meeting may take place in a member's home or a pub.

As a professional, the clinician will have held the power in the therapeutic relationship. Facilitation of a self-help group demands the development of new skills. The clinician/client relationship must give way to an equal partnership. As the group grows in confidence and assertiveness, members may decide that they do not wish to have a professional at their meetings, a development which should be celebrated by the clinician as it denotes an important stage in the development of the group, and the empowerment of its members. It may be difficult when the result of enabling people with aphasia is to empower them to be highly critical of the service they have received.

ISSUES FOR SELF-HELP GROUP MEMBERS.

Members should develop ownership of the group decision making and reduce dependence on the clinician. This will involve developing an understanding of the therapist's role as facilitator and enabler and they may take some time to learn to recognise the therapist as a resource on which the group can draw. They will also need to learn how to communicate with one another and understand each other's communication and to establish turn taking, and may develop ground rules for the conduct of meetings. There may be different benefits for members who take the most active roles in the group. Active participation is seen as taking on the role of Chair, Secretary or Treasurer of the group.

Different groups will develop very different identities. They groups identify different aims for themselves and achieve different goals. Members of one group may regularly speak at conferences and fora, work towards raising public and professional awareness of aphasia and educating professionals and service providers in their community. Some groups have sent letters to health care planners lobbying for improved services. One group has worked towards reducing barriers in the community by targeting local retail outlets and high street services, explaining aphasia to staff and awarding window stickers to enlightened participants. These are examples of the disabling barriers which the groups come to identify, for themselves, as confidence grows.

Group members comment: 'Setting up a self-help group is hard enough when you are able-bodied, but when you have disability which means you have a language disorder, its a [very] challenge' (Cressida Laywood, Nottingham group).

'My first meeting with the self-help group ..... was..... I didn't want to be part of it.....I was looking for people who were... my colleague types, who were able to communicate and outward going. But it's changed my life, meeting people at the self-help group, because they were bright, many of them very bright, successful people before their strokes, and their re-adapting again as I am to a totally new life, and being very positive.... the self-help group helped me a lot '(Margot Larkin, Central London Group,Personal Insights audiocassette, Speakability,1995).

Regional Development Advisers

The qualified professionals who work as Regional Development Advisers for Speakability in the UK, support a network of self-help groups, visiting them when requested, and providing help in times of difficulty or transition. They support and facilitate the early discussions and work to enable all members to express their views. They help in finding a meeting place and with organising transport to meetings and they assist in forming a committee and opening a bank account. They may help to make links with local radio and newspapers or help with press releases.

Liaison with Clinicians

It is important for the clinician to establish a good working relationship with the groups. The local clinician is a valuable link for the group and the most usual agency for referring new members. Some clinicians enjoy keeping in touch with a group and may become honorary members. Some groups will ask their local therapist to screen potential members, while others will accept direct contact from new members.

Networking opportunities for the Self-Help Groups

In the UK, Speakability encourages the sharing of information and experiences by sending representatives to national and regional Forum meetings and working parties set up to develop policy documents used to lobby decision makers. The assertiveness of established groups and the campaigning and awareness projects in which they are involved show up the hesitancy and anxieties of newer groups. Some members will need facilitators who can assist them with

issues they wish to raise.. Information from the groups is published in *Speaking Up,* Speakability's newsletter.

The Profile of Self-Help Groups for Aphasic People.

There has been very little research into the impact of self-help groups on the quality of life of aphasic people. We assume that active participation brings fundamental benefit impacting on the individual's psychosocial life. However, we have not tested this assumption.

In 2000 we conducted a study to determine the profile of the membership of *Speakability* self-help groups in England. We wanted to know what kinds of people become members and what are the factors associated with taking an active role in a group. We aimed to find out the age range of the membership, how severe their aphasia was, what parts of society they were drawn from, and the roles individuals played in the groups. We wanted to know their physical limitations, how long they had had aphasia, and examine the relationships between these factors. We also wanted to know something about how members got to their meeting.

Membership Profile. A survey was designed to collect data on the membership of the self-help groups affiliated to Speakability in England. Data were collected by Speakability's Regional Development Advisers. We gathered data on 135 aphasic members of 19 SHGs around England, which represents approximately 50% of the national self-help group membership at the time of the study. Table 2 shows that in terms of age the sample may be representative of the aphasic population (Davis and Holland 1981). There is a wide range of months post-onset (MPO) showing that most group members have a predominantly chronic aphasia with few having more acute forms. The mean distance of group meetings from members' homes was 6.1 miles (SD=6.7; Range=0.25-40 miles), with most members living within 5 miles of the meeting place with a few, from rural regions, living between 25-40 miles (see Figure 2).

Table 2. Age, months post-onset of aphasia (MPO) and the distance in miles members lived from the group meeting place

	Age	**MPO**	**Miles**
Mean	61.8	40.2	6.12
SD	11.2	36.2	6.75
Range	32-87	4-288	0.25-40

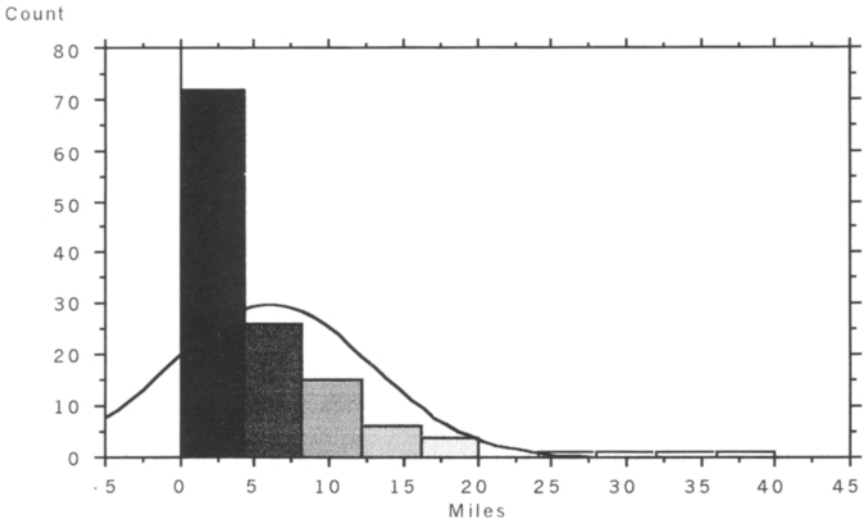

Figure 2. The distance in miles that participants live from the meeting place (numbers of participants on the Y axis)

Table 3 shows that over half of members either drive themselves to meetings or are driven by a relative in their own car, use public (N=20) or private transport (N=22), with few (N=9) needing to use community transport or volunteer transport. Seven members walk to meetings and one cycles

Table 3. How group members get to meetings

Transport to Meetings	**Numbers (%)**	
Own car	76	(56.3)
Public Transport	20	(14.8)
Lift	17	(12.6)
Community Service	9	(6.6)
Walks	7	(5.2)
Taxi	5	(3.7)
Bicycle	1	(0.7)
Total	135	(100)

Table 4 shows that members live predominantly with their spouse, their family or alone. Only one member lived in a residential home

Table 4. Group members' home circumstances

Circumstances	**Numbers**	**(%)**
With Family	56	(41.3)
With Partner/spouse	51	(38)
Alone	27	(20)
Residential Home	1	(0.7)
Total	134	(100)

Group Roles and Participation. The association between the role a member takes in the group and the severity of their aphasia is shown in Table 5. The likelihood of a member taking a participating role in the self-help group is significantly related to the severity of their aphasia (as determined by the Boston Diagnostic Aphasia Examination Severity Rating Scale, Goodglass and Kaplan, 1972)) (Monte-Carlo exact version of the Jonckheere-Terpstra Test for correlation: two-sided: JT=3.163; p=0.0017). Figure 3 shows a clear relationship between age and whether a member takes a group role or not, with older members participating less (ANOVA, F=3.0; $p<.02$). There are significant differences between the severity levels of those taking on the roles of Chair ($p<.01$) and Secretary ($p<.024$) compared to those taking no active role.

Table 5. Group role and severity of aphasia as determined on the Boston Aphasia Exam. Severity Rating Scale (Goodglass and Kaplan, 1972. 0=most severe, 5=least severe)

BDAE Severity	Chair	Secretary	Treasurer	Other Role	No Role	Totals
0	0	1	0	0	1	2
1	0	0	1	1	13	15
2	1	2	3	3	13	22
3	3	5	0	0	15	23
4	6	3	4	6	20	39
5	4	5	5	0	7	21
Totals	14	16	13	10	69	122

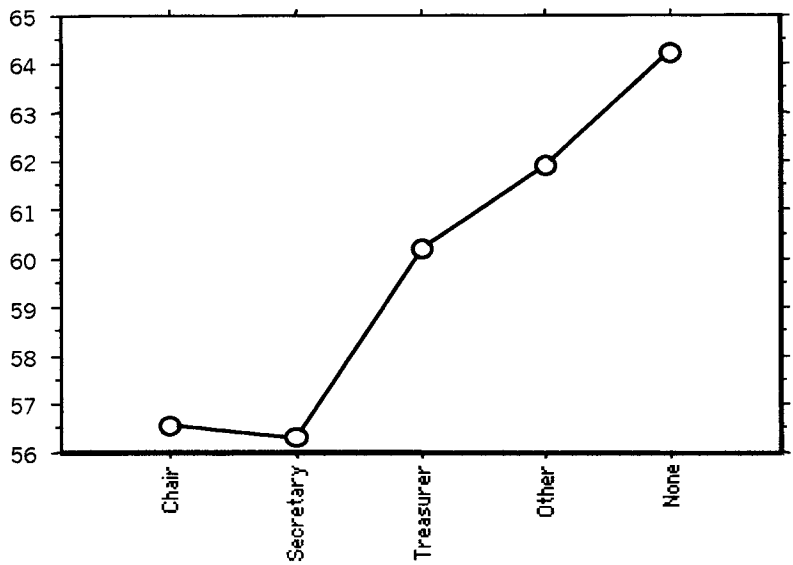

Figure 3. The relationship between age (Y axis) and group role

We examined the association between physical disability and the roles members take in self-help groups. Of the members taking a participatory role, 15 had a hemiplegia (Chi Sq=NS), suggesting that having a hemiplegia does not prevent members taking an active role. Of the 135 members who participated in this study, only 13 (10.4%) were non-ambulant. Of these 13, only 1 took a group role. Being non-ambulant does appear to influence whether members take on an active group role.

We examined too if socio-economic class, gender or age influences whether a member takes a role. Table 6 presents the distribution of socio-economic class (OPCS 1980) and group role in the 124 participants for whom we have this information, where socio-economic group I includes the higher earning professions (e.g., lawyer, physician, academics), group II the lower earning professionals (teachers, therapists, managers, administrators), group IIIN skilled non-manual workers, group IIIM skilled manual workers, group IV partly-skilled manual workers and group V unskilled manual workers. Table 6 shows a relatively unequal distribution with a significant association between socio-economic class and group role (Monte-Carlo exact version of the Jonckheere-Terpstra Test for correlation: two-sided:JT=3.867;p=0.0001). Those in the higher socio-economic classes are more likely to take group roles, particularly those in socio-economic group II, professional and managerial workers, like therapists, teachers, administrators and managers.

Table 6. Group role and socio-economic group (SEG).

SEG	Chair	Secretary	Treasurer	Other Role	No Role	Totals
I	2	3	2	1	1	9
II	9	9	9	4	22	53
IIIN	3	1	1	0	14	19
IIIM	1	2	2	3	15	23
IV	1	2	0	2	14	19
V	0	0	0	0	1	1
Totals	16	17	14	10	67	124

Table 7 shows that there are slightly more men (46.7%) than women (34.8%) who take an active role, but this difference is not significant (Chi Sq=NS)

Table 7. Group Role and Gender

	Chair	Secretary	Treasurer	Other Role	None	Totals
Male	11	12	12	8	49	92
Female	5	5	2	3	28	43
Totals	16	17	14	11	77	135

However, Table 8 suggests that there is a relationship between age and whether a member takes a group role ($p<0.01$), with older members participating less

Table 8. Age and group role

Age	Chair	Secretary	Treasurer	Other Role	None
Mean	56.5	56.2	60.2	61.9	64.2
SD	11.9	10.5	8.8	10.4	11.2

CONCLUSIONS AND IMPLICATIONS

The data seem clear. Most self-help group members have been aphasic a long time, are relatively less severely aphasic and relatively young. While hemiplegia is common, the large

majority of members are ambulant. Only one member did not live in their own home or with their own family.

Understandably, most groups are centred around densely populated urban areas. Groups that form in rural areas often have problems with members travelling large distances to get to meetings. Our study shows that members tend to live close to their meeting place and use their own transport or other private or public transport to get to meetings. Community or volunteer transport is used less often than public and private transport.

The role an individual takes is seen as an index of group participation. Members who take a full participating role are males and females and are more likely to be younger and less severely disabled, both communicatively and physically. They are also more likely to be drawn from the higher professional and managerial groups in the community.

Our data does not answer questions about whether active participation brings fundamental benefit impacting on the individual's psychosocial life (Jacobs and Goodman 1989), but it does suggest that membership comes from a relatively well defined section of the aphasic population. But there is some indication that belonging to a self-help group interacts significantly with the number of hours a person with chronic long standing aphasia spends out of the house engaged in social and community activities (Code, 2001).

Self-help in aphasia is still evolving and, like other human groups, things do not always go smoothly. Misunderstandings may arise. Arrangements for meetings and outings can be casualties of impaired communication. Members may have different expectations of their groups and some may go away disappointed. Some members may be unwilling or unable to recognise the need for facilitation and support. As in other groupings of human beings, leaders may be insensitive or over dominant which can impact on the development of autonomy in members.

In this chapter we have presented some data and related our experiences based on the development of over 50 self-help groups over a six year period. Despite the difficulties, and the lack of systematic research, it seems clear that Self-help provides many people with a valuable vehicle to make the changes needed in adapting to the disability of aphasia. For clinicians who embrace self-help and see the value of assisting people to dismantle the barriers in the community caused by aphasia, the work can be challenging and rewarding. Listening more closely to what people with aphasia themselves require should influence service planning and inform new initiatives in service delivery.

The goal for people with aphasia and for the professionals and voluntary organisations working with them, is to dismantle the barriers which frustrate and disable people. Self-help can make an effective contribution to the psychosocial reintegration of aphasic people, encouraging autonomy and empowerment.

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Chapter 14

THE SCIENCE OR SCIENCES OF APHASIA?

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INTRODUCTION

In this chapter we pick up our theme from the title of the conference on which this book is based - the Science of Aphasia. We suggest that there is a wide range of sciences that we draw upon to underpin our exploration of the causes and nature of aphasia, its impacts, and the issues faced by those who live with it. We discuss the methodologies required by those sciences. In particular we discuss the role, alongside quantitative methods, of qualitative research methods in exploring some aspects of aphasia. We discuss the issue of judging the quality and rigour of all research methods, and the implications of new, explicit standards for judging the quality of research. These standards include a requirement for researchers and the 'subjects' of research to collaborate much more closely than has previously been the case, with the subjects of research gaining more influence over the nature of the research agenda. We indicate that the methodologies used by researchers will need to respond to the research questions being prioritised by the consumers of that research. We go on to illustrate the use of qualitative research by demonstrating how the outcomes of qualitative research that we have undertaken have challenged our concepts of intervention in aphasia. We describe briefly how a man with aphasia used his therapy also to challenge those concepts of intervention, highlighting to us the critical relationship between identity, language and communication for people with aphasia.

SCIENCE AND SCIENTIFIC METHOD

Delegates and speakers attending the first Science of Aphasia conference represented a diverse range of nations and disciplines. The gathering provided an opportunity to hear linguists, cognitive neuropsychologists, speech and language therapists, neurologists, neurolinguists and others debating current research related to aphasia in their fields, and to reflect upon the relationship between sometimes very different representations of aphasia. The eclectic programming demonstrated that aphasia cannot be understood from the perspective of one discipline alone.

In keeping with the title of the conference, there was discussion of recent technological and scientific advances in the investigation and treatment of aphasia. Some of the investigations described (for example PET scans showing patterns of brain activation during language processing tasks) are clearly founded upon the culture and traditions of classical science. Procedures such as these involve empirical observation and induction; they adhere to experimental principles; they are objective, controlled and focused; they generate theories and predictions, and they define and develop themselves through these processes. Such methods accord with the assumptions underpinning traditional scientific method, identified by the physicist John Barrow (1988):

- There is an external world separable from our perception
- The world is rational: A is not equal to not A
- The world can be analysed locally: that is, one can examine a process without having to take into account all the events occurring elsewhere
- There are regularities in nature
- The world can be described by mathematics
- These presuppositions are universal

Similar rational and objective principles underpin the research of many other non-medical disciplines concerned with aphasia. Thus, linguists, psychologists and cognitive neuropsychologists design specific, controlled experimental studies, the results of which are used to develop models of language organisation and cognitive processing that can in turn be tested, refined and generalised. The conference demonstrated how, at present, aphasiology is largely mediated through controlled experiment, observation and scientific reasoning, within disciplines that model and define aphasia in diverse ways.

Although it is relatively easy to identify scientific method within these diverse disciplines,

science itself is notoriously difficult to define (Wolpert, 1992). This becomes particularly problematic in areas such as medicine and healthcare (and aphasiology), where different conceptual frameworks, including lay perceptions, start to converge. Although a strong scientific conceptual framework underpins biomedical practice, this is subject to increasing scrutiny. Previously unstated assumptions have been identified, articulated and questioned (Nettleton, 1995):

- The mind and body are separate entities
- The mechanical metaphor: the body is a machine
- Disease is a matter of biological change
- Biomedical practice is concerned with observation, hypothesis, control, experiment and outcome.

Greenhalgh (1999, p 248) comments:

> 'The medical profession has aspired to an empiricist paradigm for more than a century. This approach is based on the somewhat tenuous assumption that diagnostic decision follows an identical protocol to scientific enquiry- in other words that the discovery of 'facts' about a patient's illness is exactly equivalent to the discovery of new scientific truths about the universe. Until fairly recently, the empiricist framework was loosely applied and rarely questioned.'

Writers within academic and popular discourses have been developing critiques of the conceptualisation of biomedical science and the practice of medicine since the 1970s. Prominent examples would be the work of Foucault and the struggles of the womens' movement, with particular reference to the management of childbirth. Such analyses have called attention to cultural aspects of scientific biomedical practice. They argue that this practice currently takes insufficient account of the socio-environmental contexts of sickness and social inequalities in health. Nor does it consider how interpretations and experiences of health and illness are mediated through gender, race and class. This neglect of socio-cultural and contextual impacts can lead to the exercise of professional medical power in ways which users of healthcare construe as over simplifying and excluding (Nettleton, 1995). These critiques suggest that these issues may have been neglected in the pursuit of science. Although these analyses have been started in biomedicine, it is probably fair to say that the culture of aphasiology has not yet been subjected to such sustained and critical scrutiny.

QUALITATIVE METHODS IN MEDICINE

There has been an increasing application within healthcare research of methodologies that have their provenance in the social, rather than the natural sciences. In the UK the context in which these methodologies are developing is one in which there is increasing questioning of the power base and culture of medicine fuelled by a number of high profile cases of serious medical malpractice (c.f. Kennedy 2001). These cases are focusing attention on both the social relations of healthcare delivery and the sciences that underpin the evidence base for healthcare services.

Qualitative methods are particularly suited to capturing the subjective, social and changing aspects of illness and healthcare and the complexities of sickness in context. These are the aspects of illness that commonly lie beyond the gaze of biomedical science. They are concerned with social phenomena, they gather social evidence and thereby operate on different ontological and epistemological premises. Qualitative methods highlight the interpretative aspects of research, emphasising *reflexivity* rather than assuming objectivity. The relationship between researcher and researched in qualitative studies is also under scrutiny, the investigator generally acknowledging naivety and inexperience concerning the issues being investigated, while the subject of the research is considered the expert. Ethical issues raised by the research method are also central to the qualitative endeavour (Mason, 1996).

Qualitative methods are becoming more recognised within medical and health research, even though they appear to be founded upon conceptual principles that have been less familiar in biomedical sciences. The authors of a series of articles in the British Medical Journal focused on these qualitative methodologies within medical research, outlined their range and potential and argued that such methods, while different, can and should be as robust, rigorous, empirical and systematic as quantitative investigation (e.g. Mays and Pope, 1995). More recently, the same journal ran a series of papers on narrative-based medicine, in which the relationship between interpretative approaches and evidence-based practice was discussed. In one contribution to the debate, Greenhalgh (1999 p 251) argues that diagnostic and therapeutic judgements are always based on an integration of different sorts of evidence: the clinician's case-based experience, the patient's individual and cultural perspectives, and the results of clinical research trials and studies. Her reflections call into question the universal applicability of research findings in the practice of medicine:

> "The generalisable truth that we seek to glean from the research trials pertains to the sample's (and hopefully the population's) story, not the story of individual

> participants. There is a serious danger of reifying that population story - that is of applying what Whitehead (1925) called the fallacy of misplaced concreteness - and erroneously viewing statistics as hard realities. Misplaced concreteness is also an apt description of the dissonance we experience when we try to apply research evidence to clinical practice. Hence although there are certainly 'wrong' answers to particular clinical questions, it is often impossible to define a single 'right' one that can be applied in every context."

Alderson (2001) points out that quantitative confidence has been privileged over qualitative attention to complex meanings as a sound basis for planning healthcare. But the potential contribution of qualitative evidence alongside quantitative evidence is underlined. Qualitative methods have a variety of specific uses: probing little explored phenomena; providing detailed descriptions; reporting peoples' views and experiences in their own words; combining a range of methods such as observations, group discussion, interviews and diaries; adding new questions and theories; searching for examples that challenge and test emerging conclusions; including silenced and excluded groups and enabling them to be heard; and recording and analysing ambiguity, contradiction and gradual change.

It is important to stress here that although we are drawing attention to the potential contribution of qualitative research we are not arguing that qualitative research methods are any better than any other research methods. Rather the point that we are making is that there is a range of methodologies that can be employed and what is critical is to ensure that (i) the most appropriate methodology is used to address the research question being asked, and (ii) we do not constrain the research questions that we are asking to fit only with a certain type of methodology. As Alderson (2001, p 5) argues:

> "The 'gold standard' method is not any one particular method, but the one which is most appropriate and effective in answering the research question."

Quantitative and qualitative methods are increasingly being used within the same research studies to address different aspects of the same issue (e.g. Ritchie *et al.*, 1994), demonstrating the utility of drawing on a range of ways of thinking, in combination with one another, in order to create something larger than any one way of asking a question can achieve on its own. As Riessman (1993, p 70) suggests

> "Science cannot be spoken in a singular universal voice. Any methodological standpoint is, by definition, partial, incomplete and historically contingent."

The arrival of qualitative and narrative methods within the culture of health and medical research constitutes a timely response to some current trends in health policy. These reconfigure the patient as consumer, scrutinise the principles of practice, and urge increased user/consumer involvement and 'shared clinical decision making' within healthcare (Department of Health, 2000).

> "Pressures are now building to persuade clinicians that decision-making should take account of evidence on clinical effectiveness, cost-effectiveness and patients' and public preferences. To do so adequately requires some form of decision support, which could include clinical guidelines, patient information materials and formal techniques for decision analysis. It also requires the acknowledgement of uncertainties in medical care. It will depend on enhancing the skills of clinicians to facilitate knowledge transfer and sensitive determination of patients' values."(Coulter, 1997)

QUALITATIVE METHODOLOGIES WITHIN APHASIOLOGY

Qualitative methods are beginning to be applied to the study of aphasia. Damico *et al.* (1999) suggest that qualitative methods, designed to analyse interpretative data, arise from a well-accepted paradigm within social sciences that has a place within the discipline. They list the key features of qualitative methods as follows:

- focus on phenomena in natural, not experimental, settings
- highly systematic set of procedures
- open and flexible research designs
- acknowledgement of the researcher as a key instrument of data collection
- descriptive data
- narrow and in-depth focus
- concern for detail and process rather than product
- inclusive of the mundane
- focus on individual perspectives and experience.

Damico *et al.* describe and illustrate a range of qualitative methods that have been used in the study of aphasia. Perhaps Conversation Analysis is the most well-established and familiar method within the field, and Damico *et al.* (1999) explore this contribution in detail. Those unconvinced of the potential of Conversation Analysis are encouraged to read Goodwin's (1995) elegant account of Rob, a severely aphasic individual, whose interactions with his

carers enable co-construction of his precise meanings, despite the fact he only has three words: 'yes', 'no' and 'and'. Goodwin concludes by reminding his readers that aphasia is a social as much as a physiological or biological event, and one that must be understood systemically and not in isolation from the social context.

Simmons Mackie and Damico (1999) also describe the principles and application of ethnographic techniques in the study of aphasia. This eclectic, flexible method of studying social phenomena, incorporating observation, analysis of artefacts, interview and diary methods, enables insights into what Goodwin has termed 'the social life of aphasia.' Indeed, the social life of aphasia *therapy* does not escape scrutiny either. Simmons Mackie and Damico's (1998) ethnographic study of feedback in intervention sessions explores the relationship between therapist and client, and demonstrates that therapy is a complex social event that influences considerably more than language. Narrative analysis, another qualitative method that is gaining influence in healthcare research (Greenhalgh 1999), has as yet been little used within the domain of aphasiology (however see Pound *et al.* (2000) for practical applications of narrative method).

In-depth interviewing also enables the exploration of the phenomenon of aphasia from the perspective of the individuals who have it. This brings to mind Alderson's (2001) mention of 'silenced and excluded groups' whose experience can be accessed using qualitative methods. People with aphasia are often excluded from research, even when issues that seem to relate directly to communication are being explored, such as quality of life following stroke (Maclean *et al.*, 2000, Williams *et al.*, 1999). This is ostensibly because they struggle to talk and to explain themselves, and may have difficulty in comprehending and responding to questionnaires and surveys.

Our qualitative study 'Talking about Aphasia' (Parr *et al.*, 1997), in which fifty people with aphasia took part in in-depth interviews, was conceived with the intention of enabling the experiences, views and perspectives of people with aphasia to be heard. Such inclusion is possible if methods of supporting conversation (Kagan, 1998) are imported into interviewing techniques, although such techniques are by no means straightforward to apply.

Methods and findings from our study have been detailed elsewhere (Parr, 2001; Parr *et al.*, 1997). However, there are two points relating to the contribution of interviews that are worth emphasising in this context. The first is that they allow us to hear not only what people with aphasia are saying, but also how they say it. Transcribed interview material can be organised in such a way as to enable case-by- case analysis and conceptual or thematic analysis across respondents, and yet to retain the actual words used. Far from being a reason for excluding

their opinion, the fragmented, jumbled, often densely metaphoric language of people with aphasia strongly conveys the ideas expressed. Its very directness becomes an asset rather than a disadvantage, as these quotations from data collected in the study illustrate:

'Money, work worry. Worry me all around.'
'I express me I me painting'
'Mary touchpaper'

The second point concerns the nature of qualitative methods. Qualitative research is often perceived as woolly and anecdotal. However the process of qualitative data analysis includes a methodical and systematic process of transcribing, indexing, charting, summarising, interpreting and validating (e.g. Ritchie and Spencer 1994). Data are available for independent scrutiny, cross-checked, and the process of analysis is transparent and reflexive. In any scientific paradigm there are examples of good and poor implementation of the science. Evaluators of scientific enquiry need to be adept at identifying both good and poor methods.

Reflecting on the advent of qualitative methods within aphasiology, Kearns (1999 p.649) comments that the contributions of unfamiliar paradigms may not always be understood or appreciated:

> 'As we achieve success with our methods and traditions we may become intolerant of less familiar approaches to research. Tragically, our healthy scepticism can turn into cynicism and mistrust. This kind of conceptual myopia is not good for scientists and it is particularly bad for our science. We must be careful to avoid pre-judging an entire scientific tradition we know little about.'

Kearns takes pains to underline that adopting an open approach to qualitative methods does not necessarily mean suspending the criteria traditionally used for judging good science. Damico *et al.* (1999) explore these issues with particular reference to the reliability and validity of qualitative studies. Other commentators (for example Mason 1996, Alderson 2001) indicate that these criteria, just like science itself, are not set in stone, but can themselves be adapted in line with a different ontology and epistemology. Thus, criteria for the evaluation of qualitative research may include the following questions (British Sociological Association, 1996):

- Are the methods of the research appropriate to the question being asked?
- Is the selection of cases or participants theoretically justified?
- Are the data available for independent scrutiny?

- Has the relationship between field workers and subjects been considered?
- Is there evidence about how the research was presented and explained to its subjects?
- Was the data collection and record keeping systematic?
- Is there adequate discussion of evidence both for and against the researcher's arguments?
- Are the results credible?
- Have ethical issues been adequately considered?
- Is the author's position clearly stated?
- Has the meaning of their accounts been explored with the participants?

STANDARDS IN QUALITATIVE AND QUANTITATIVE RESEARCH

Such challenging questions addressing ethical issues, methods and systems set high standards for qualitative research. But it would be hard to argue that these questions are not relevant also to quantitatively based research studies. The challenges posed reflect good practice in the conduct of research of any form. The recent research governance directive issued by the UK government's Department of Health (Department of Health, 2000) reflects the growing pressure on **all** researchers working with 'patient' groups within any research methodology (as well as the funders and administrators of research), to adopt similarly high standards of research conduct. In future all UK health-related research will have to show that is has conformed to the standards set in the directive.

One of the interesting challenges contained in the questions posed by the British Sociological Association, and more latterly by the Department of Health, is that they require researchers to be more explicit about the social and political relations of research. All researchers need to engage with the subjects of their research by explaining and presenting the purpose of the research and exploring **together** the research findings and outcomes. This requirement assumes that the process of exploration and explanation is engaged in accessibly, in a form that ensures that the subjects of the research can follow (an interesting challenge when carrying out research with people with aphasia):

> 'Participants or their representatives should be involved wherever possible in the design, conduct, analysis and reporting of research ... Researchers are responsible for selecting appropriate means of communication to ensure that potential participants are fully informed before deciding whether or not to join a study.... Findings must be made available to those participating in the research... Reports need to be

> comprehensible and take language and other needs into account.' (Department of Health 2000)

This move towards including the subjects of the research in the process of implementing the research reflects the 'user/consumer involvement' movement in health care generally. The pressure to carry out research collaboratively across not only different scientific disciplines, but also with consumer group representatives, is mounting. Such representatives are not only joining research teams and advisory boards to carry out qualitative research studies but also, increasingly, to conduct large randomised controlled trials (for examples see www.hfht.org/Consumers in NHS Research). The National Health Service in the UK is also promoting the development of training courses to facilitate the involvement of user representatives (for examples see www.hfht.org/Consumers in NHS Research). Users are represented on the board of the UK National Institute for Clinical Effectiveness (NICE), a powerful government agency that evaluates evidence for the effectiveness of health care interventions before recommending them for implementation.

These developments open up new issues that must be taken into account by people engaged in research. Researchers are accustomed to being accountable to their funders and disseminating research findings to their scientific colleagues. But authentic and inclusive explanation and dissemination (let alone actual involvement in the research process) may require the development of new skills for researchers. These might include explanation in accessible lay language; an ability to communicate with the public; inclusive interaction and engagement with research subjects; an ability to listen and respond to the perceptions, questions and priorities of user groups.

Qualitative researchers have for many years been able to be explicit about the mutual roles of researcher and researched because of their acknowledgement of the reflexivity that is part of **all** research. They are used to challenging (and being challenged by) the dynamics of the research process, and attending to the attitudes that they bring to the endeavour. The relationship between researcher and researched reveals underlying, often unspoken, value systems (Byng *et al.*, in press). Increasingly, public attention to the ethical aspects of recent scientific and technological developments (such as embryo-related research or cloning) is making the articulation of assumptions and values an imperative for all researchers, not just those who use qualitative methods.

We anticipate increasing challenges to all researchers, emanating from the subjects of research. These concern making discussions of research accessible and meaningful, revealing the value systems of the research community and eliciting those of other stakeholders,

engaging with users in the generation of research questions, negotiating how these are prioritised and by whom, and dealing with incompatibility and conflict. The disability movement has long argued for control of the research agenda by disabled people (Barnes, Mercer and Shakespeare, 1999). The 'consumer involvement' movement is gaining in power and also has the potential to influence the research agenda in healthcare.

The impact that consumers exert over the nature of the research that is being carried out may well have implications for the range of methodologies with which researchers will need to be familiar. Methods have to be matched to the questions being asked, as we suggested above. If researchers are less in control of the research agenda, then they are also going to have to be more flexible in the methodologies they implement in order to be responsive to the questions. What will be critical in this process is constructive dialogue and negotiation between researchers from different traditions and consumers, to ensure that relevant research is conducted using good science.

LISTENING TO PEOPLE WITH APHASIA: REVISITING DEFINITIONS

The qualitative methodology used in the Talking about Aphasia study referred to earlier allowed us to access the views and experiences of people with aphasia. What they had to say about their interpretations of language impairment and their experience of services caused us to revisit our own assumptions as aphasiologists. We will use this study as a way of demonstrating the contribution of qualitative research to shaping thinking about aphasia and intervention. We will summarise the main points emerging from the study, then explore what issues these raised for our conceptualisation of aphasia, and our implementation and interpretation of therapy.

The interviews with fifty purposively sampled people with aphasia revealed some common aspects of the social experience of aphasia, offering insights into what it means to people who live with it. To summarise: aphasia is long-term, continually revealing itself as life events unfold. It is systemic, affecting not just individuals but entire social networks. It is wide-ranging, having a major impact on all aspects of a person's social experience: work, financial status, education, interactions, community and family life, citizenship, identity. These impacts are inter-related. Struggles with language mean struggles with identity, with one's sense of self. Difficulties returning to work bring financial losses, compounded by struggles to negotiate the maze of financial aid provided by government.

For those who live with it, aphasia is bewildering, invisible, difficult to conceptualise and understand. It is idiosyncratically interpreted and experienced by those who have it. As well as these direct impacts, aphasia compromises the very tools needed for negotiation, enquiry, understanding, and complaint. People with aphasia talked about its impact on their lives, but also their ideas concerning therapy and various health, social care and welfare services. To summarise: they valued services that were relevant to their concerns, timely and flexible, long-term, respectfully negotiated and clearly communicated. Their direct experiences of health, welfare and social services did not always provide them with examples of these qualities.

Documenting these reactions caused us to wonder what aphasia is. Professionals define it in terms that relate to the disciplines within which they have studied. People with aphasia define it using very different concepts and terms. Neither definition is wrong, but then nor is either right on its own. They are just different. If we are to study the nature of aphasia and the interventions relevant to it, we need scientific methods that allow us to explore all of these different meanings.

At the moment, there is a bias in the type of definitions employed professionally and in the type of methods used in research. As we discussed earlier, the western tradition of biomedicine currently provides the predominant research culture and language of exploration for aphasia. This tradition serves the study of some aspects of aphasia well, but not all. The importance of broadening our methods of scientific enquiry is underlined when we consider the impact of our definitions on the development of intervention.

RECONCEPTUALISING THE NATURE OF INTERVENTION

Listening to people with aphasia focused our attention both upon the nature of the impairment and upon the focus and process of therapeutic intervention. McNeil and Pratt (2001) argue that the clinical management of aphasic individuals depends critically on the assumptions of a scientific definition of the impairment. Our study precipitated for us a definition, or re-definition of aphasia, not simply as linguistic or cognitive deficit, but in social terms.

The study enabled us to see what is obvious to those who have it: aphasia is a biological event, which takes forms that can be observed and described and which has complex, long-standing, socially-related impacts. If this is what aphasia represents, broadly, for people who have it then what does that mean for the professionally generated 'scientific' definitions of aphasia? Doesn't a 'scientific' definition of an impairment need to be able to include or accord

with the perspective of the person who experiences the impairment? A scientific definition of impairment, which is generated by data observed through experimentation, one form of science, does not contain the fundamental 'truth'. It represents a perspective formulated from the application of one form of science.

For us, the need became paramount to develop a means of conceptualising both aphasia and aphasia therapy, in a way that related to the meanings expressed by people with aphasia, not just the scientific traditions provided in our training and education. We felt that previously we had perceived the experience of people with aphasia through a filter formed by the traditions of western biomedicine. Aspects that we had perceived as priorities or as important had reflected the importance our scientific disciplines attached to particular features of aphasia. Learning to listen reflectively to how people were describing their perceptions and experiences led us to formulate a new conceptual framework to underpin our intervention (Byng *et al.*, 2000).

In this reconceptualisation we acknowledged that we did not want to throw numerous babies out with bathwater. People with aphasia included in their interpretations issues that accorded with existing focus and methods of scientific enquiry. Many people talked about their loss of speech or difficulty talking and wanted to understand more about why it had come about, why they had the difficulties they did and what could be done about them. They wanted to know more about the stroke, why it had happened and how further strokes could be prevented. The current sciences of aphasia have a lot to offer in answer to these kinds of questions, and knowledge about these issues is growing and developing all the time, fed by current research. But people also raised many other, wider, issues about living with aphasia. When we turned to the scientific literature to explore these issues further there was a marked change in the volume of literature from which to draw: the wider impacts of living with aphasia raised by the people we were listening to have received scant attention from the scientific community (Sarno 1993). Neither did it seem as if these wider issues were being explicitly addressed in the literature on intervention. The full impact of aphasia was not being addressed. Or if it was, few people were writing about it.

Perhaps the reason behind this one-sidedness is related to the assertion made by McNeil and Pratt (2001), that the clinical management of aphasic individuals depends critically on the assumptions of a scientific definition of the impairment. If this is true, and if our 'scientific definition' of aphasia omits issues critical for the person with aphasia, then these issues will remain outside the focus of our clinical management. They will not be addressed explicitly (often tantamount to not addressing them at all). We would prefer to restate McNeil and Pratt's assertion as follows: 'clinical management' needs to depend on the person with

aphasia's definition of the *impact* of the impairment. In this case, clinical management would need to address explicitly a much wider set of issues and we would have to develop a range of methods to explore the nature of the impact of the aphasia. It seemed to us that what was missing from our intervention was an understanding and definition of *impact.*

IMPACT-LED INTERVENTION

Following the Talking about Aphasia study, we developed a framework to represent our changing understanding of (and aspirations for) intervention (Byng *et al.*, 2000). This framework (see figure 1) represents the person with aphasia within a number of social domains and identifies six areas for intervention. The representations arise directly from what was identified in the interviews. Although the areas for intervention are depicted as separate entities of course they are inter-connected. Just as for the people who experience them, the complex impacts of aphasia are integral to each other rather than discrete events. One impact articulates with and often illuminates another.

We will illustrate both the range and the inter-relatedness of the impacts of aphasia by considering briefly the experience of one person, Tony, reported in detail elsewhere (Cairns, in press). Tony's story exemplifies the interaction of issues critical to his learning to live with aphasia. In Tony's case, the relationship between his communication and his sense of himself - his identity - were impossible to tease apart - it was not possible to address his communication without dealing explicitly with issues related to his identity. In fact in Tony's case, despite his complete loss of speech and quite severely impaired ability to write, what seemed to be most important for him in his process of adapting to his new life was to focus on issues related to acknowledging and supporting his identity. If intervention with Tony had addressed only his communication then therapy for him would have missed the main point. In practice, Tony turned therapy to address communication into therapy addressing his identity.

Tony: impacts and interventions

Tony had a stroke eighteen months before the interventions highlighted here began. He was born in Switzerland but grew up as a child in Portugal. He lived in France and Brazil prior to taking up residence in the UK in 1959. Tony therefore has a rich language background, speaking both Portuguese and English fluently, but also having some competence in Spanish, French and German, of which he is proud. He now relies on his strong non-verbal

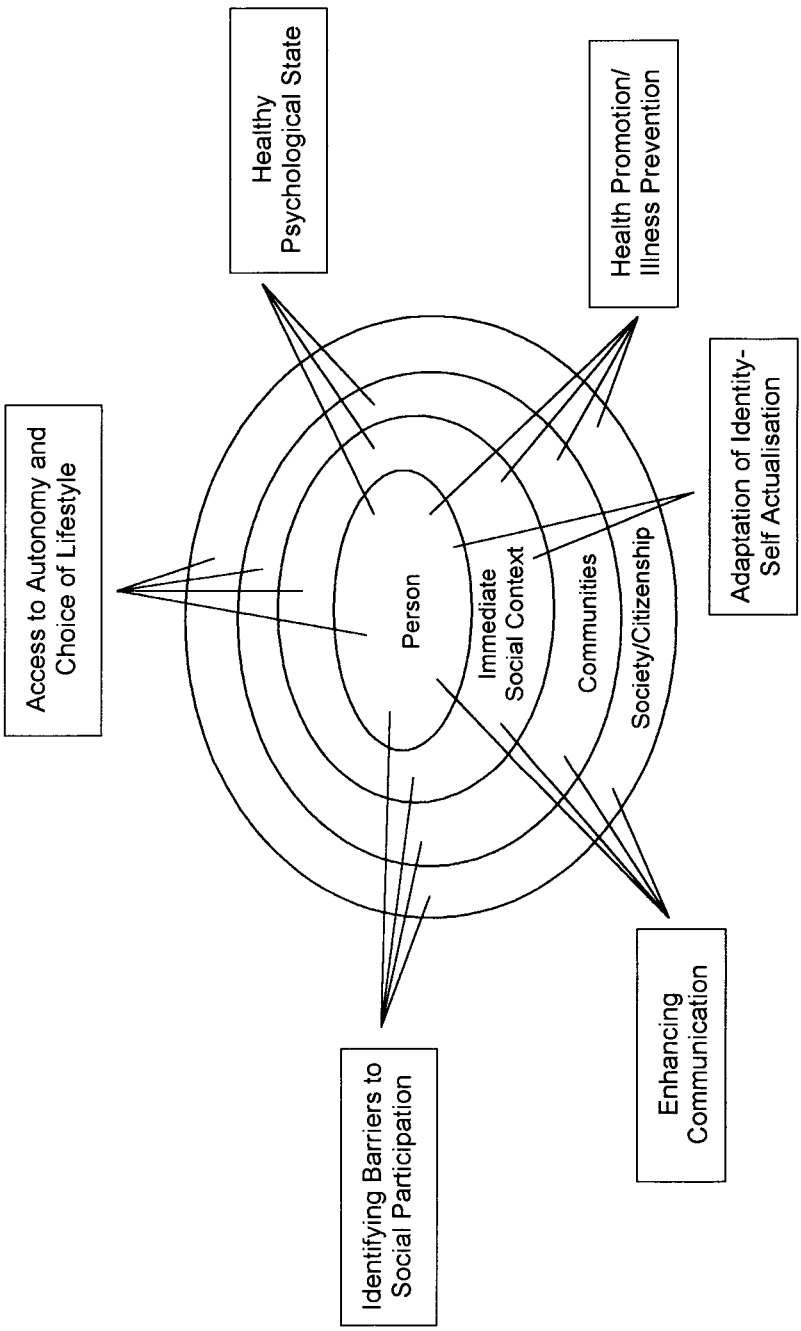

Figure 1. Living with Aphasia: Goals of Intervention

communication skills and some writing ability as his primary means of communication. Tony's writing often contains a mix of Portuguese and English. At the start of the therapy described here he had little awareness of this mixing or code-switching, and this often dramatically affected the success of his attempts to communicate. The mixing of languages was a source of frustration and distress to Tony, and led to frequent communication breakdowns with monolingual therapists and students and with other people with aphasia. Even with his wife, who was also a bilingual speaker of Portuguese and English, communication for Tony was often difficult and problematic.

Over a one and a half year period, Tony attended for group and individual therapy and counselling twice a week at a facility for people living with aphasia that is now called Connect. Table 1 summarises the type of activities that Tony engaged in.

These included working on developing a wide range of communication strategies in total communication, identifying the nature of the language impairments across a group of people with aphasia, developing a group video to describe participants' experiences of aphasia in their own terms and to educate relatives about ways of supporting communication, planning outings, individual counselling, and developing a personalised communication portfolio. (See Cairns, in press, for more information about these therapies.) These activities address the major areas of intervention set out in the framework shown in figure 1, as table 2 demonstrates.

The bilingualism project. One project in particular addressed Tony's bilingualism. This project focused on an investigation of Tony's retained writing ability in both English and Portuguese and drew specific attention to his code switching. It was conceived as a project that focused directly on Tony's language impairment. The process of investigating the phenomenon proved to be therapeutic in itself for Tony. Through it, Tony developed his awareness of when the code switching was happening, in what circumstances and contexts, and also developed skills in managing the situation with his interlocutor, even though he was unable to translate from one language to the other. The project thus served to enable Tony to exert more control over his communication. However it also offered a means of acknowledging his competence across languages, providing him with a new role in the group he was working with as a promoter of discussion about bilingualism - an issue relevant to a number of group members who spoke more than one language. This provided an opportunity for group members' expertise as controllers of several languages to be acknowledged, and linked their aphasia to their often international histories and their ongoing life stories. So, a project which was looking at specific aspects of language performance not only had a wider

Table 1. Tony's pathway through therapy

Term	Spring 1999	Summer 1999	Autumn 1999	Spring 2000	Summer 2000
Group Work	Total Communication Relaxation Planning outings	Video project Use of Telephone Cognitive Neuropsychology project	Video project Leaflet project Access to information	Video project Conversation Outing	Portfolios Conversation Transport Aphasia awareness
Individual Work	Extend personal communication book	Personal communication file / Portfolio	Use of Computer	Use of Computer + Dictionary/ Spellchecker to translate	Interaction with wife Writing
Counselling	→				→
Other activities		← Teaching on Volunteers and Assistants' Courses → ← Bilingualism project → ← User consultation initiatives at Connect → Advising the 'Severe Aphasia' research project Contributing to the Proper names research project			
Tony's wife's involvement	Counselling	← Relatives' Information & Support days →			Interaction work with Tony Relatives group Counselling

Table 2. Examples of therapy projects within a framework for therapy interventions (Byng et al, 2000)

	Enhancing communication	**Identifying barriers to social participation**	**Access to autonomy & choice of lifestyle**	**Healthy psychological state**	**Health promotion / illness prevention**	**Adaptation of identity**
Project	Total communication Computer work Interaction work	Access to information Conversation Outings	Access to information Transport	Counselling Conversation	Relaxation	Video Portfolios Aphasia awareness

communicative application, but also related to issues about Tony's, and other group members', identity and social role. For Tony it highlighted aspects of his identity that were of particular significance to him, and made them a focus of intervention.

The communication book project. A second project Tony engaged in related to the development of a communication book. The therapists conceived of this as providing an augmentative communication aid, but in the course of the project it became clear that Tony was developing other ideas. The sequence of events during which Tony changed the nature of the communication book is documented in the notes that were kept at the time. An early entry in the notes is as follows: "Tony is very keen to develop a communication book. This will provide him with the means to a useful communication strategy beyond Connect and his home environment. It is hoped that it will add to his independence, promoting his confidence in social situations and reducing his isolation." A little later Tony's student key worker wrote "It was decided to include a section with useful vocabulary for when Tony goes shopping, or to the gym, park, café or hospital. His week is organised in such a way that these are regular places he goes, where a communication book would enable him to communicate with staff".

A while later Tony made clear that the book as it stood was not serving a useful purpose: "Tony expressed that shopping is not a problem for him as he goes to the supermarket and picks things off the shelves. Similarly at the gym the manager and staff know him and he does not encounter any real communication difficulties. In view of Tony's ability to function adequately in these situations, it may be more useful for him to use his book for social reasons. Therefore we should consider taking more of a personal portfolio approach (see Pound *et al.*, 2000) with personal information about himself and what he does, and less

emphasis on functional needs and vocabulary." Under Tony's guidance the 'communication book' was steered towards becoming a collection of representations of himself and his life. What started out as a static tool became a dynamic resource through which Tony engages with other people and reveals his identity and competence.

Both of the projects described demonstrate the inter-relatedness of communication and identity. A significant purpose of communication for Tony is to be able to represent himself; he wants to engage people in interacting with him in relation to who he is, what he thinks, and what he experiences. This expression of self clearly involves the use of subtle, imaginative communication skills, adapted to meet the constraints and demands of Tony's particular aphasia. To study or to provide therapy for his language without also focusing on his identity is to miss the point of Tony's aphasia. We might be able to describe the impact of aphasia on Tony's language separately from the impact on his identity, but to provide therapy for his communication it is impossible not also to take account of the impact of communication on identity, and of identity on communication.

Tony's intervention needs a range of sciences to underpin it. It requires an understanding of language processing and sociolinguistics to interpret and explore his code switching. The theories and methods of linguistic and cognitive neuropsychological research provide a vocabulary and a method for unpicking his language impairment. The science of research methods contributes to the design of the cognitive neuropsychological investigation and the qualitative interviews used with Tony and his wife in other parts of his intervention. Theories from social science contribute to an understanding of the group dynamics and the interpretation of the health beliefs amongst the members of the group in which Tony participated. Theories underpinning the implementation of augmentative and alternative communication contributed to the understanding of how to develop Tony's communication book. Tony was keen to understand the nature of his stroke and recovery, which relies on neurological and neurophysiological explanations. In summary, Tony demonstrates to us why providing intervention for someone living with aphasia demands that we are informed by a rich and diverse range of scientific disciplines: an unidisciplinary perspective simply does not provide aphasiologists, and especially aphasia therapists, with sufficient tools to approach the task.

SO WHERE DOES THIS GET US?

What is the relevance of this debate about methodology for clinicians and researchers in aphasiology? In this chapter we have been making two principal points: the first is that if our

understanding of aphasia is to progress scientifically then research related to aphasia needs to be conducted using a broad range of methodologies. Without the contributions gathered from the implementation of different methods, our understanding will simply be incomplete. We have set out some of the new challenges being posed to all researchers emanating from the consumer-involvement movement, which will change the social relations of conducting research. In so doing, the weighting of priorities in research will also change and with it the range of research methodologies that we will need to learn to employ.

The second point that we have been making is to illustrate the contribution of qualitative methods in enabling an examination of the assumptions underlying the discipline. For example, within aphasia, as in other chronic conditions and diseases, 'psychosocial issues' have been conceptualised separately from language impairment (Sarno 1993, Byng *et al.*, 2000). This mind/ body dualism is also encapsulated in the WHO framework, which has strongly influenced how interventions are organised (Worrall and Frattali, 2000). However, language (and therefore aphasia) is inextricably bound up with communication, social context and identity suggesting that they cannot be considered separately, in intervention at least.

Tony's therapy, described above, provided evidence of the inter-relation of identity and communication, of aphasia and its impacts. In practice, these were inseparable for Tony and therefore had to be considered as a whole by the therapists working with him. This relationship underlines the importance of ensuring that interventions are not remote from the context of people's lives. This is not the same as just ensuring that all intervention is 'functional' in terms of ensuring the acquisition of basic skills. Rather it relates to the importance for people with aphasia that intervention they receive should relate to the lives they want to lead, the identity they want to reveal, and the people with whom they live (Elman and Bernstein Ellis 1995). Therapists have known this for a long time, but the relationship between communication and identity is not strongly represented in the research literature.

Qualitative methods can open up new areas for intervention and legitimate current areas that are practised by clinicians but not explored or evaluated by researchers. Clinical practice in these areas is reliant on 'custom and practice' for its development, rather than systematic study and development through research. Most clinicians would acknowledge that the output of researchers has enhanced the interventions that they can apply (Byng *et* al., 2000). If researchers are not addressing large areas of practice then those areas can become neglected and acquire 'less important' status (Sarno 1993). If the relationship between communication and identity, and the connection to living with aphasia, are to be explored (and they are being explored), then researched interventions must also take these issues into account.

The imperative to link intervention with impact has been articulated by people with aphasia (Parr *et al.*, 1997) but perhaps most strongly expressed by Frank (1991, p 13) reflecting on the experience of cancer and heart disease:

> 'Medical treatment is designed to make everyone believe that only the disease - that which is measurable and mechanical - can be discussed. I know I am supposed to ask about the disease, but all I feel is the illness. The questions I want to ask about my life are not allowed, not speakable, not even thinkable.'

CONCLUSION

One of the motivations for writing this paper was to address the debated question of whether qualitative methodologies can make a contribution to science. This may seem to be an esoteric question of interest only to a few in the navel-gazing research community. However, we believe that it does have wider implications. The perception of the scientific value of research often determines whether it gets funded in the first place, accepted for publication, read, cited, made use of (as much as research is ever made use of!). Therefore if qualitative research does not appear to have scientific validity to some readerships, then its potential may never reach those who could use it. The credibility of qualitative research does not wholly rest on its relationship to science, but in the biomedical culture within which aphasiology is largely situated, it probably does.

The answer to the question, then, depends on one's conceptualisation of science. If science is simply the rational exploration of the physical, biological and technological world, then rational exploration of the interpretative world is clearly not scientific. Values, interpretations, perspectives belong on the 'mind' side of the body/mind divide. However, accounts of the philosophy of science suggest that broader conceptualisations are constantly evolving and are themselves being challenged (Warburton, 1992). These for example encompass the Kuhnian idea that the history of science is characterised by revolutions in scientific outlook. According to Kuhn *et al.* (1988), scientists work within a dominant paradigm until anomalies are thrown up, then reconfigure the paradigm, just as the theory of relativity replaced the Newtonian paradigm that had prevailed since the Enlightenment. These changes of paradigm, as well as being profoundly creative, made philosophers aware that the fundamentals of a scientific understanding are not a static unchanging set of natural laws, but rather are as much dependent on the community in which they surface as on the nature of reality itself. In other words, science itself is not a naturally-occurring, value-free phenomenon, but is dependent on its own cultural context for meaning.

We would argue that qualitative methods are scientific in that they accord with the Kuhnian concept of science. They are discovery-driven, they are a response to some of the anomalies arising in the field and they provide methods and means appropriate to the nature of the enquiry. It is perfectly possible and valid to measure changes in behaviours using traditional experimental or statistical methods. However, if the purpose of a particular enquiry is to understand the complexities of communication in context, or to explore the experience, meaning and interpretation of aphasia and intervention then additional methods are called for.

Researchers working in aphasiology, as in other scientific disciplines, need to have access to a range of research tools in order to select the methods appropriate to their enquiry. 'The new generation of health research' (Barbour, 1999) will see an increasingly eclectic combination of different methods, as the complexities of illness and intervention are addressed. For this to happen, different ontologies and epistemologies must be articulated, understood and respected. In addition, users will swell the research community and start to generate and shape scientific enquiry for themselves. Such potential synergy and cross fertilisation of disciplines, methods and stakeholders, opens up exciting opportunities for those working in the sciences of aphasia.

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SECTION 4

METHODOLOGY AND EFFICACY IN APHASIA THERAPY RESEARCH

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Chapter 15

SOME PSYCHOMETRIC ISSUES IN APHASIA THERAPY RESEARCH

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INTRODUCTION

Issues of outcome measurement are crucial in clinical care in general and in speech-language pathology in particular (Frattali, 1998). One well-known conceptual framework for the definition of outcomes is the International Classification of Impairments, Disabilities, and Handicaps (ICIDH) from the World Health Organization (WHO) (1980) which conceptualises outcomes with respect to the consequences of some disorder as indexed by impaired psychological functions which then may lead to disabilities as the functional consequence of an impairment as manifested in activities represented by tasks and skills. Handicaps are considered the social consequence of an impairment or disability leading to impoverished or reduced participation of an individual in social life thus limiting or preventing the fulfilment of a role that is considered normal with respect to age, sex, and social as well as cultural standards. The revised ICIDH-2 criteria stress the positive aspects of activities (restricted in case of disabilities) and participation (reduced in case of handicap).

In line with the WHO framework, three broad classes of outcome measures should be distinguished: impairment is assessed by conventional diagnostic tests and instrumental measures (for language functions), disability measures concentrate on functional status (e. g. communication), and handicap is assessed via questionnaires giving the view of the patient and or significant others on quality of life aspects. Irrespective of the methodology with which outcome is tackled - be it experimental, quasi-experimental or non-experimental - measurement

instruments used should be objective, reliable, and valid, i.e. fulfil all the criteria that are deemed necessary for any psychological measurement instrument (see e. g. Gulliksen, 1987; Suen, 1990).

Finally, since the book is primarily concerned with research in aphasia therapy, aspects that should be considered in the development of assessment methods also suited for the thorough examination of individual patients in single-subject therapy research will be dealt with.

SCALES

Three basic attitudes with respect to the construction of psychological measurement instruments may be discerned (Rost, 1988):

(1) The test score of a measurement instrument is chosen to be the simple sum of the item scores. The resulting total raw score is then granted interval scale properties per fiat and it is studied whether the total score provides a good predictor (or not) for some criterion variable. The claim of interval scale properties is taken to be justified the more this simple scaling procedure (compute sum total of item scores) empirically provides a powerful predictor for some relevant criterion measure ('validity pragmatism').

(2) The use of test - or subtest - total scores is justified via deriving standardised or normative scores for some reference population ('norm pragmatism').

(3) Each and every assignment of numbers to persons requires a formal model stating testable assumptions and properties for that assignment ('model priority').

Many scales in neuropsychological assessment and rehabilitation are lacking such a firm measurement basis (cf. Wade, 1992, chapter 2) resp. no one has looked at it. One example for questionable scales can be seen in ten scaled individual functions comprising a neuro-rehabilitation scale (Koppi *et al.*, 1995) which tries to score different aspects of cognitive functioning on 5-point scales. One of these scales is designed to rate 'verbal communication' in the following way (translation by K.W.):

"Summary rating of: aphasia, dysarthria, language comprehension, apraxia, etc. In rare cases, in which comprehension and expression dissociate, the average of both is computed (Example: speechless ALS-patient with completely preserved comprehension is rated 2): 0 = no impairment; 1 = complex sentences with some problems or errors; 2 = simple sentences with severe errors, complex orders are executed erroneously, simple orders can be executed; 3 = some few propositional words and/or execution of most simple orders (related to midline movements) possible."

With such a mixing of receptive and expressive performances, which very often will not be impaired in a similar way and to a similar degree in a given patient, this presumed scale will most probably not measure one unitary (latent) dimension of impairment. This is most obvious from the example of the patient suffering from ALS. The 'compromise' score of 2 is designed to capture some qualitatively different type of impairment as can be easily seen from the description of scale point 2.

With many well-known scales like the Barthel-Index (cf. Wade, 1992) the differential weighting of items, also using different numbers of scale points, has not been justified psychometrically. Furthermore, demonstration of a lack of uni-dimensionality has gone unnoticed even for the famous Glasgow Coma Scale (Koziol and Hacke, 1990).

Since the ground-breaking publication of the probabilistic test theory model (latent trait model) for dichotomous item scores by the Danish mathematician Georg Rasch (1960, 1980) and its several extensions to polytomous items (cf. Wright and Masters, 1982; Rost and Langeheine, 1997) there is no excuse for skipping the stage of analysing the measurement properties, in particular the uni-dimensionality, of a new scale (For an application of the Rasch model to the Token Test cf. Willmes, 1981). Otherwise, assigning a total score across items to some patient may provide invalid information about her/his level of impairment, restriction of activities or participation. Basically, the only restriction is the rather substantial number of participants/patients (usually more than 100) that need to be examined for sufficiently precise parameter estimation and testing of model adequacy.

A well suited model for polytomous items is the *partial credit model* (Masters, 1982), in which threshold parameters with arbitrary distances on a latent dimension are considered. Assuming a left to right ordering of increasing competence, the probability of scoring in category k+1 compared to category k is modelled, and the threshold parameters on the latent continuum designate those points for which the probability for the next higher category just starts to excel the one for the actual category.

In the partial credit model it can be *tested statistically*, whether the total score for a set of items indeed covers all the information (i.e. provides a sufficient statistic) in the full pattern of item scores. Only in case of model fit is the total score fully informative and a scale score with interval scale properties (cf. Wright and Masters, 1982) can be provided for each pattern of responses/item scores.

Differences in threshold parameter differences may be present for different items. In case of ADL- of QoL-ratings by the patients themselves or significant others or staff members using the same scores for all questions, a more restrictive model, the so called *rating scale model*, may be more appropriate (cf. Fig. 1): threshold parameters may have arbitrary distances on the latent dimension but these distances are assumed to be identical across different items. Willmes (1997) has shown - for a sample of n=1769 aphasic patients - the partial credit model to hold within each of the different item groups making up the subtest Written Language of the Aachen Aphasia Test (AAT) with a (m=) 4-point item scale ranging from 0 (no similarity of response with target) via 1 (little similarity with target) and 2 (high similarity with target) to 3 (correct response).

A simple demonstration of the basic property of items making up a one-dimensional scale is provided in Table 1. In the left half of the table a fictitious data set is given with a simple dichotomous scoring (1 = activity can be pursued, o = activity can not be pursued) of nine activities of daily living in a (small) sample of 20 patients. From that raw data set one cannot easily derive information about the scale's properties. But a simple double ordering of activities and patients according to a decreasing number of 1's per row and column reveals a striking patterning of the data: a perfect partition of the data matrix into the upper left part containing only 1's and the rest consisting of only 0's, constituting a so called perfect Guttman scale (cf. Wright and Masters, 1982, p. 4): For each patient there is never a 0 intermingled with a straight sequence of 1's and likewise, no 1 can be found within a sequence of 0's. The total score is fully informative in the sense that it expresses the order up to which *all* activities can be pursued. The activities themselves are also ordered from left to right according to increasing difficulty.

Real data sets will most probably not show such a perfect ordering, but a certain degree of violations will be tolerated in model tests for the dichotomous Rasch model. For items with polytomous scoring the data matrix – after reordering – should reveal a similar partition with the highest score m centred in the upper left corner of the matrix followed by a diagonal band of scores m-1, below it another diagonal band of scores m-2, etc, until there are only 0's in the lower right hand corner.

Finally, it should be noted that the rather common practice of studying the construct validity of scales by subjecting them to a principle components analysis (PCA) may be grossly misleading, in particular when the items are scored dichotomously or with some few categories and the score distribution for each item does not roughly follow a single peaked function with the item mean being close to the middle of the item scale. For the artificial data in Table 1, a PCA with

a subsequent Varimax rotation yields a three-components solution with a high percentage of variance explained (83.6%). All three 'easy' activities load on one component, all three 'moderate' activities on another and the 'difficult' activities on the third component. Therefore, the three components should not be interpreted in a substantial fashion, but rather considered to be an artefact of the analysis method since a PCA may result in a misleading solution that does not capture the obvious linear hierarchy of activities for formal reasons: it is well known that in case of items with low or high difficulties there is a restriction of range for the correlation coefficients that enter the PCA. In case of rather low mean correlations there is a tendency to extract more principle components.

	Activities										**Activities (ordered)**								
Pt	**1**	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**	**Pt**	**4**	**2**	**7**	**1**	**9**	**6**	**3**	**8**	**5**
01	0	1	0	1	0	0	1	0	0	18	1	1	1	1	1	1	1	1	1
02	0	1	0	1	0	0	1	0	0	19	1	1	1	1	1	1	1	1	0
03	0	1	0	1	0	0	1	0	0	20	1	1	1	1	1	1	1	0	0
04	0	1	0	1	0	0	0	0	0	15	1	1	1	1	1	1	0	0	0
05	0	0	0	1	0	0	0	0	0	16	1	1	1	1	1	1	0	0	0
06	0	0	0	0	0	0	0	0	0	17	1	1	1	1	1	1	0	0	0
07	1	1	0	1	0	1	1	0	1	07	1	1	1	1	1	1	0	0	0
08	1	1	0	1	0	1	1	0	1	08	1	1	1	1	1	1	0	0	0
09	1	1	0	1	0	1	1	0	1	09	1	1	1	1	1	1	0	0	0
10	1	1	0	1	0	0	1	0	1	10	1	1	1	1	1	0	0	0	0
11	1	1	0	1	0	0	1	0	0	11	1	1	1	1	0	0	0	0	0
12	0	1	0	1	0	0	1	0	0	12	1	1	1	0	0	0	0	0	0
13	0	1	0	1	0	0	1	0	0	13	1	1	1	0	0	0	0	0	0
14	0	1	0	1	0	0	1	0	0	14	1	1	1	0	0	0	0	0	0
15	1	1	0	1	0	1	1	0	1	01	1	1	1	0	0	0	0	0	0
16	1	1	0	1	0	1	1	0	1	02	1	1	1	0	0	0	0	0	0
17	1	1	0	1	0	1	1	0	1	03	1	1	1	0	0	0	0	0	0
18	1	1	1	1	1	1	1	1	1	04	1	1	0	0	0	0	0	0	0
19	1	1	1	1	0	1	1	1	1	05	1	0	0	0	0	0	0	0	0
20	1	1	1	1	0	1	1	0	1	06	0	0	0	0	0	0	0	0	0

Table 1. Fictitious data for some activities of daily living scale (1/0 activity can/cannot be pursued); left: data as observed; right: data ordered according to decreasing row and column totals

PSYCHOMETRIC SINGLE CASE ANALYSIS

Basic concepts

When data from a profile of standardised test scores are available for a patient, inferential statistical procedures are required to examine whether the true performance level in different subtests is significantly different and whether the performance profile has changed from a first to a second examination. Psychometric single-case analysis (Huber, 1973) offers such a coherent approach for psychological tests with good reliability estimates from a sufficiently large sample and normative data derived from a standardisation sample of no less than about 400 – 500 persons (for an application to the Aachen Aphasia Test see Willmes, 1985).

The basic idea is to treat diagnostic hypotheses about a person's identity of *true* scores in two or more (sub-)tests just like other hypotheses in statistics. Following the so called classical test theory model the observed score X_{ij} of subject i in test j is assumed to be additively composed of the person's true score T_{ij} and and error E_{ij}. Each person is thus characterised by an individual performance level and some degree of (potential) variability around that level. An individual diagnostic examination cannot provide such information concerning variability and repetitive testing is not feasible as well. When, however, assuming homogeneity of the test specific error variance σ^2_{ij} across all subjects i of the reference population for which normative data and reliability information have been obtained, then the test specific error variance is identical to the square of the standard error of measurement, i.e. $\sigma^2_{ij} = \sigma^2(X_{.j})\ (1- \rho_{jj})$, for the particular test j with $\sigma^2(X_{.j})$ denoting the raw score variance in the reference population of interest. The standard error of measurement can be estimated if some reliability estimate is available (internal consistency, split-half or parallel test reliability) and the raw score variance has been reported for the normative sample, information that is regularly available in the test manual of well established tests.

For a comparison of performances across subtests, only standard scores can be employed, since different subtests will usually show differences in difficulty. Norm scores are computed as $Y_{ij} = ((X_{ij} - A_j)/B_j) \times K + L$, with A_j and B_j the mean and standard deviation in the normative sample for test j and L and K the mean and standard deviation of the desired standard norm (e.g. standard scores: L=100, K=10; IQ: L=100, k=15; T-score: L=50, K=10; C-score: L=5, K=2; scaled scores of the WAIS: L=10, K=3).

Diagnostic (null-)hypotheses are concerned with the identity of true scores, in case of only two tests: $T_{ij} = T_{ih}$. But identity of true raw scores only implies identity of true standardised scores if the reliability of both tests is identical (cf. Huber, 1973). In most cases there are differences between subtest reliabilities and therefore an additional transformation has to be carried out on the standardised score Y^{x}_{ij},(x denoting observed score standardisation), the so called *τ-standardisation*, which provides the best estimate Y^{τ}_{ij} of true standardised performance:

$$Y^{\tau}_{ij} = Y^{x}_{ij} /\sqrt{\rho_{jj}} + L(1-1/\sqrt{\rho_{jj}}).$$

The higher test reliability the smaller is the difference between both types of standardised scores, which need to be computed only once for a given test.

When formulating null hypotheses about true standardised scores to be tested with some inferential statistical procedure type-I and type-II errors occur just as with any statistical test. In case of diagnostic hypotheses, commitment of type-II errors, i.e. overlooking some difference in true performance levels, may have more detrimental consequences for a patient than committing type-I errors, i.e. falsely declaring an observed difference in standardised scores to be indicative of a true difference in performance. Therefore, Huber (1973) suggested a more liberal default type-I error level of α=10%.

When comparing subtest performances inferentially one wants to know whether a difference in τ-standardised scores is reliable, i.e. unlikely to be attributable to errors of measurement alone (so called *reliability aspect*, cf. Huber, 1973). In case of a reliable difference between true performance levels, it may additionally be of interest, whether the probability of observing a still larger difference in the reference population is small, e.g. less than 20%. In that case, Huber (1973) defines this difference to be *diagnostically valid*, i.e. potentially indicative of some highly informative piece of diagnostic information. The concept of diagnostic validity also allows for an operational definition of a performance *dissociation* (see below).

Individual profile analysis

When analysing a performance profile composed of *m* subtests, profile level, profile scatter and profile shape can be discerned. In the approach of psychometric single-case analysis the *profile level* is defined to be a weighted mean of the τ-standardised subtest scores; the weights $g_j = \rho_{jj} /(1- \rho_{jj})$ are determined according to the subtest reliabilities, with subtests showing good reliability getting more weight in determining the profile level:

$$h^{\tau}_{i} = 1/G\ \Sigma g_j Y^{\tau}_{ij} \text{ and } G = \Sigma g_j .$$

In order to analyse *profile scatter*, one does an overall comparison of all m subtest performances much like an overall comparison among different conditions in a one-way ANOVA. The test statistic is composed of reliability-weighted squared differences between subtest scores and profile level:

$$\chi^2_{m-1} = 1/K^2 \, \Sigma g_j (Y^\tau_{ij} - h^\tau_i)^2.$$

When the value of the test statistic gets bigger than the 90%-quantile of a χ^2-distribution with m-1 degrees of freedom, significant profile scatter is assumed and the performance profile is termed a *real* profile, i.e. there a differences between true subtest performances.

In case of a real profile, an additional analysis of *profile shape* is required. When no specific (neuro-)psychological hypothesis concerning relations of subtest performances composing the profile are at hand, all $M = m(m\text{-}1)/2$ pairwise comparisons of pairs of subtests (j, h) can be carried out under the reliability aspect using the standard-normal test-statistic:

$$z_{jh} = (Y^\tau_{ij} - Y^\tau_{ih}) / K\sqrt{(1/g_j + 1/g_h)}.$$

Since M post-hoc comparisons have to carried out, an adjustment of the type-I error level for the individual pairwise comparison is required such that the overall type-I error of committing one or more type-I errors among the M comparisons is no bigger than 10%. The well known Bonferroni adjustment could be employed, i.e. testing each pairwise comparison at a reduced (two-tailed) type-I error level of 10%/M. This leads to rather poor power at the level of the individual pairwise comparison. The sequentially rejective procedure of Holm (1979) offers a computationally simple more powerful alternative. The M p-values p_{jh} corresponding to the test statistic values z_{jh} have to be ordered according to increasing values: $p_{(1)} \le p_{(2)} \le \ \ldots \ \le p_{(M)}$. If $M \times p_{(1)} \le 10\%$, the related performance difference between the two respective subtests is declared significant (real) and one proceeds to the next larger p-value; if $M \times p_{(1)} > 10\%$, this pairwise comparison and all other comparisons are declared not significant and the test procedure is stopped. In the next step one examines whether $(M\text{-}1) \times p_{(2)} \le 10\%$; if so, the related subtest comparison is considered to be real and one proceeds to the next step. If $(M\text{-}1) \times p_{(2)} > 10\%$, the procedure stops, etc. With every new step the factor with which the resp. p-value is multiplied is reduced by one thus leading to less conservative comparisons from step to step.

In case of several subtests constituting a performance profile the comparison of all subtest pairs may not be very informative. Contrasting two non-overlapping groups of subtests with m_1 ($\ge$1) resp. m_2 ($\ge$1) in a so-called linear contrast (with $m_1 + m_2 \le m$) may be diagnostically more interesting, e.g. comparing verbal and non-verbal subtests in an intelligence test battery or expressive and receptive subtests in some aphasia test. Technically, the reliability weighted

profile level of each of the two subgroups I and II of subtests h^{τ}_{iI} resp. h^{τ}_{iII} is computed - just as before for the whole profile level – and its difference $\psi^{\tau}_i = h^{\tau}_{iI} - h^{\tau}_{iII}$ determined (cf. Huber, 1973, ch. 9). The corresponding (one-sided) null hypothesis of identical true τ-standardised sub-profile levels is assessed with the standard-normal test statistic:

$$z(\psi^{\tau}_i) = \psi^{\tau}_i / K\sqrt{(1/\Sigma_I g_j + 1/\Sigma_{II} g_h)} .$$

The - usually directional - null hypothesis is rejected if the p-value related to $z(\psi^{\tau}_i)$ is $\leq 10\%$. In case of several a-priori linear contrasts, again the type-I error level for the individual contrast has to be adjusted.

In addition to revealing a linear contrast to be reliable, its diagnostic validity may be of still more relevance, e.g. it might be of interest to compare (a) subtest(s) assessing repetition performance with (some) other language modality subtests for an operational definition of transcortical resp. conduction aphasia in terms of purely quantitative performance levels. The denominator of the general formula is rather involved (cf. Huber, 1973, formula 9.4.7):

$$z(\psi^{\tau}_i) = \psi^{\tau}_i / K\sqrt{\{\Sigma_{I\&II} c^{\tau 2}_j/\rho_{jj} + 2\Sigma\Sigma_{I\&II} c^{\tau}_j c^{\tau}_{j'} \rho_{jj'}/\sqrt{(\rho_{jj}\rho_{j'j'})}\}}$$

with $c^{\tau}_j = g_j/\Sigma_I g_j$ for all subtests in subprofile I, $c^{\tau}_j = -g_j/\Sigma_{II} g_j$ for all subtests in subprofile II, and the double sum $\Sigma\Sigma_{I\&II}$ taken over all subtest pairs with indices $j < j'$.

The diagnostic validity probability is computed as:

$$p(\psi^{\tau}_i) = 1 - \Phi(z(\psi^{\tau}_i)),$$

with $\Phi(z)$ the standard normal cumulative distribution function. If $p(\psi^{\tau}_i)$ is less than 20%, the subtest difference is declared to be diagnostically valid.

For the special case of just two subtests to be compared the test statistic is:

$$z(Y^{\tau}_{ij} - Y^{\tau}_{ih}) = (Y^{\tau}_{ij} - Y^{\tau}_{ih})/K\sqrt{\{1/\rho_{jj} + 1/\rho_{hh} - 2\rho_{jh}/\sqrt{(\rho_{jj}\rho_{hh})}\}} .$$

In this formula ρ_{jh} denotes the correlation between both subtests in the standardisation sample. The diagnostic validity probability is computed as:

$$p_{jh} = 1 - \Phi(z(Y^{\tau}_{ij} - Y^{\tau}_{ih})),$$

with $\Phi(z)$ the standard normal cumulative distribution function. If p_{jh} is less than 20%, the subtest difference is declared to be diagnostically valid.

A free PC-program CASE123 is available from the author to carry out all necessary computations for all test batteries the relevant psychometric information of which has been entered by the program user in an accompanying 'library' of tests.

Intra-individual profile comparisons

Besides analysing one performance profile psychometric single-case analysis procedures also offer a way of comparing two profiles of the same subject or patient who has been examined twice in the course of some neurological condition, possibly before and after some phase of therapy or training, or perhaps in a follow-up examination some time after an intervention to see whether performance has stabilised or deteriorated again. When wanting to compare two performance profiles three steps of analysis may be discerned:

1. Global profile comparison. The test of *profile identity* serves to decide whether the two performance profiles for the same test-battery do not show any substantial differences, neither in profile level nor in profile shape. The test statistic with m degrees of freedom simply comprises the squared differences per subtest between some two examinations denoted 1 and 2, again weighted for reliability:

$$\chi^2_m = 1/2K^2 \Sigma g_j(Y^\tau_{ij1} - Y^\tau_{ij2})^2.$$

Only if the test statistic yields a value larger than the critical 90%-quantile of a χ^2-distribution with m degrees of freedom, there is reason to proceed to the next two steps; otherwise the two profiles are considered to reflect identical true performance levels for each of the subtests comprising it.

2. Comparison of profile levels. For each of the two profiles the profile level computes as explained before and a standard-normal test-statistic serves for a comparison of both levels:

$$z(h^\tau_{i1} - h^\tau_{i2}) = (h^\tau_{i1} - h^\tau_{i2})/K\sqrt{(2/\Sigma g_j)}\ .$$

Depending on whether the diagnostic hypothesis is one-sided (improvement after therapy or due to expected spontaneous recovery resp. deterioration after some progressive disorder) or two-sided (e.g. stability at follow-up) the lower/upper 10%- quantile (±1.28) or the upper/lower 5%-quantile (±1.645) of the standard normal distribution have to be used for comparison. This profile level comparison (just as the other comparisons between time points below) rests on the additional assumption that both examinations with the same test battery can be treated as two parallel measurements.

3. Comparison of profile shapes. For a pure comparison of profile shapes potential differences in profile level have to be adjusted for, again leading to a global test of (level-adjusted) profile identity:

$$\chi^2_{m-1} = 1/2K^2 \Sigma g_j\{(Y^\tau_{ij1} - Y^\tau_{ij2}) - (h^\tau_{i1} - h^\tau_{i2})\}^2.$$

Only if the test statistic yields a value larger than the critical 90%-quantile of a χ^2-distribution with m-1 degrees of freedom, i.e. one decides for differences in profile shape, additional post-hoc comparisons serve to study in detail these differences in profile scatter. Just as with one real profile one can opt for a routine examination of a change of the relation in performance between a pair (j,h) of subtests from examination1 to examination 2 and do this for all M pairs of subtests or select one or a few planned linear contrasts that will be compared each between both examinations to inquire whether the relation between sub-profile levels has changed over the two examinations (see below under 5.)

4. Comparison for individual profile components. A more conventional comparison between two profiles that does not care for the relation between profile components is based on the testing whether the true performance level per subtest has changed between two examinations. Just as with the profile level before, these comparisons may be one- or two-sided. For each of the m subtests, performance is compared via the standard-normal test statistic:

$$z(Y^{\tau}_{ij1} - Y^{\tau}_{ij2}) = (Y^{\tau}_{ij1} - Y^{\tau}_{ij2})/K\sqrt{(2/g_j)} .$$

Since this is a multiple testing situation, control of the overall error level of committing one or more type-I errors can be accomplished again using the Bonferroni approach with 10%/m per subtest or the more powerful sequentially rejective test procedure of Holm (see above).

5. Planned profile comparisons. If a specific change in the performance pattern can be expected as a consequence of some intervention or disease progress, this expected change in relation of subprofile levels from subtest groupings I and II can be examined via comparison of the same linear contrast ψ^{τ}_i for both diagnostic examinations, i.e. by examining whether $\delta^{\tau}_i = \psi^{\tau}_{i1} - \psi^{\tau}_{i2}$ is significantly different from zero using the standard normal test statistic:

$$z(\delta^{\tau}_i) = \delta^{\tau}_i /K\sqrt{2}\ \sqrt{(1/\Sigma_I g_j\ +1/\Sigma_{II} g_h)} .$$

If $z(\delta^{\tau}_i)$ is larger/smaller than the upper/lower 5%-quantile ± 1.645, one decides a specific change in profile shape to hold for the patient of interest.

If both groups of subtests contain just one subtest each, resp. (j, h) , the above formula reduces to:

$$z = \{(Y^{\tau}_{ij1} - Y^{\tau}_{ih1}) - (Y^{\tau}_{ij2} - Y^{\tau}_{ih2})/K\sqrt{(2\sqrt{(1/g_j\ +1/\Sigma g_h)} .$$

When there is no specific diagnostic hypothesis, all M pairs of subtests can be compared that way for changes in subtest performance relations, again employing the Bonferroni- or Holm-approach for a proper control of the overall type-I error level at 10%.

Specific applications in aphasiology

In order to back-up the hypothesis of a selective sparing/impairment of some language function one can examine whether true performance in that specific subtest *s* is reliably *and* diagnostically validly above/below all or a subset of all other subtests. One computes all m-1 (or a smaller subset) pairwise differences $Y^{\tau}_{ij} - Y^{\tau}_{is}$ which all must be reliable (one-tailed) using Holm's procedure. In addition, these differences must be diagnostically valid as well. One computes:

$$z(Y^{\tau}_{ij} - Y^{\tau}_{is}) = (Y^{\tau}_{ij} - Y^{\tau}_{is})/K\sqrt{\{1/\rho_{jj} + 1/\rho_{ss} - 2\rho_{js}/\sqrt{(\rho_{jj}\rho_{ss})}\}}$$

and the corresponding diagnostic validity probabilities p_{js} as shown before. In order not to commit a wrong assignment of selective sparing/impairment all p_{js} should be less than 20%/(m-1).

Effects of a specific therapy regimen may be hypothesised to be best captured by a predicted change of some specific (set of) linear contrast(s) after the intervention period. This can be assessed by carrying out one (or several) specific profile comparisons as under 5. above. This approach has also been chosen in substantiating the assumed presence of primary progressive aphasia (cf. Poeck and Luzzatti, 1988). Besides language impairments revealed in an aphasia test it was shown that a linear contrast comparing language bound subtests of some intelligence test battery with non-language bound ones was reliable and diagnostically valid. In the course of the progressing disease it was expected that the overall profile level would be reduced and more specifically, that the strong discrepancy between both subtest groups would become significantly smaller, although possibly still indicating some superiority of non-language bound subtests.

The profile comparison methods may be used to generate operational criteria for identifying specific responders to some intervention; this may be a more promising approach than only looking for significant group differences.

RANDOMISATION TESTS FOR SINGLE-SUBJECT THERAPY RESEARCH

Conventional statistical test procedures like Pearson's χ^2-test for 2×2-tables or the independent samples t-test have frequently been applied in neuropsychological and other single-case studies to compare a patient's performance for two sets of items with either dichotomous scoring or some metric response variable like reaction time (RT). Unfortunately, these tests may easily

lead to anti-conservative decisions for data in single-case studies; if the item responses are (even only mildly) positively correlated this may easily inflate (even double) the actual type-I error level (cf. Tavaré and Altham, 1983). Edgington (1995) has convincingly argued that interchangeability of observations under the respective null-hypothesis - which is a less stringent requirement than statistical independence - offers a more suitable general starting point for inferential test procedures in single-subject experimental research. He has given a comprehensive account of the class of randomisation tests that may allow for valid statistical tests in case of single-subject research.

The concept of random assignment of examination time points to study conditions is sufficient to allow for valid statistical inferences about effects of different study conditions on a single subject's performance. Items from two or more conditions are randomly ordered; additional blocking or other restrictions on a fully random order may be required. The typical null hypothesis of a randomisation test now is the following: for each time point in the sequence of examination events the response of the respective subject is independent of an influence from the specific condition at that particular examination time point; or stated differently: the relation between examination time point and response is the same for all task conditions. It has almost been forgotten that a randomisation (permutation) test for a single-subject experiment constitutes the first example of a significance test in the pioneering work of the founder of modern statistical inference, R.A. Fisher (1935).

The randomisation test principle will be illustrated for two basic raw data schemes encountered in single-subject research (cf. Table 2). In the independent (item-)samples schema items from two different tasks are put in a joint random sequence and the responses allocated afterwards as in the left part of Table 2. In case of items - or pairs (blocks) of parallel (matched) items - studied under two task conditions it is decided in a random fashion which of the two conditions will be presented first for a particular item resp. which of the parallel items will be subjected to either task condition.

Independent Item Samples				Dependent/Related Item Samples		
Item	Task 1	Item	Task 2	Item	Task 1	Task 2
1	y_{11}	1	y_{12}	1	y_{11}	y_{12}
2	y_{21}	2	y_{22}	2	y_{21}	y_{22}
3	y_{31}	3	y_{32}	3	y_{31}	y_{32}
-	-	-	-	-	-	-
i	y_{i1}	i	y_{i2}	i	y_{i1}	y_{i2}
-	-	-	-	-	-	-
-	-	n_2	y_{n22}	-	-	-
-	-			n	y_{n1}	y_{n2}
n_1	y_{n11}					

Table 2. Two basic data schemes in single-subject research; y_{ij} denotes the response to item i in task j

For two different item sets with a total number of $n = n_1+n_2$ items having been randomly put into an examination sequence, under the null hypothesis 'the response to an arbitrarily chosen item is independent of the task (item set) from which it comes' all potential permutations of responses between both tasks could have been observed with identical probability. For a one-sided test a feasible simple test statistic that is sensitive to mean response differences is the sum of observations for task 1: $T = \Sigma Y_{i1}$.

When the null hypothesis holds, all $N = n!/(n_1!n_2!)$ inter-task permutations are admissible with the same probability of $1/N$. The set (group) of admissible permutations π will be denoted Q_n. The decision about the null hypothesis is based on the exact p-value $p^{\geq}$ which is computed in the following way: For each permutation π from Q_n, the value of the test statistic T(y) is computed for the permuted data $T(\pi y)$, with y denoting the whole set of observations and πy a permutation of them. Then one has to check whether this test statistic value for the permuted data is bigger or equal to the value of the test statistic for the actually observed data. The exact p-value is:

$$p^{\geq} = \#\{\pi \in Q_n \mid T(\pi y) \geq T(y)\}/N,$$

with # denoting the cardinality of the set of permutations fulfilling the inequality. The null hypothesis is rejected in favour of a larger mean response for task 1, if $p^{\geq} \leq \alpha$.

In case of identical items employed in both tasks the set of admissible permutations under the null hypothesis comprises all 2^n intra-item permutations of responses between both tasks which have equal probability of $1/2^n$ when the null hypothesis holds. In case of a one-sided alternative

the same test statistic as before can be used and the exact p-value can be computed accordingly with the same decision rule as before.

Generalisations to more than two tasks are straightforward. In case of dichotomous or ordinal response variables the above tests are equivalent to Fisher's exact test for 2×2 tables or the exact version of the Mann-Whitney U-test in case of independent tasks and equivalent to the exact version of the McNemar test or the exact version of the Wilcoxon signed ranks test. The execution of a randomisation test usually requires a computer because of the large number of computations to be carried out. Edgington (1995, pp. 398-399) provides a list of software and several of the big statistics packages such as SPSS and SAS have options for exact permutation test versions; the most versatile stand-alone package is StatXact (Mehta and Patel, 1999). Willmes (1995) has given a more detailed description of how to use randomisation tests in single-patient research and in determining (double) dissociations.

In single-subject research so called differential carry-over effects cannot be separated from differences between task conditions themselves. A differential carry-over effect is present if the response to some item from a particular task condition is dependent on the specific properties of the items presented before. General carry-over effects which are only dependent on the number of items presented before, such as training, habituation or fatigue effects are not problematic they may only effect the power of a randomisation test.

Randomisation tests have also been suggested by Edgington (1995) for within- and between series single-subject designs such as the A-B design, the A-B-A withdrawal design and extensions thereof as well as the alternating treatments design. The crucial idea for being able to apply a randomisation approach lies in introducing an element of randomness in the time-series design. Without it there is no randomisation basis since the transition from one (baseline) phase to another (treatment) phase is specified for a certain point in time in advance. For a simple A-B design with 20 observational measurement occasions, say, one could require the following: a baseline phase of at least five time points and at least five treatment/intervention time points. This leaves a time window of $Q_n = 11$ time-points (from after obervation 5 till after observation 15) within which the start of the intervention phase could be determined by chance. A randomisation test can be defined by computing e.g. the test statistic $T = 1/n_B \Sigma y_{iB} - 1/n_A \Sigma y_{iA}$ for each of the Q_n admissible equally likely (under the null-hypothesis of no intervention effect) partitions of observations into an A- and a B-phase and defining an exact p-value as before. For conventional p-values the time window for a start of the intervention may however be unrealistically large. The situation becomes better for a withdrawal design in which two time windows can be introduced, one in which the intervention is started and one in

which it is stopped, with the only additional requirement that the actual intervention phase comprises at least some minimum number of examination points in the time-series. Since the two points of change are determined independently of each other, the number of admissible Q_n is larger with the same number of study time points than for the A-B design. A PC-program, SCRT, for the generation of more complicated designs, also with more than one type of intervention, and the subsequent evaluation with a randomisation test is provided by Onghena and Van Damme (1994).

Also for other tests proposed e.g. in Kratochwill and Levin (1992) for single-subject designs randomisation test alternatives can be formulated which can do away with untenable assumptions that are based on notions of random samples from infinite populations of observational units (see also Franklin *et al.*, 1997). One such statistic is the C-statistic used to test for a (trend) departure from the null hypothesis of a time series (with n time-points) made up of just random fluctuations (Tryon, 1982). The C-statistic, which relates the sum of squared differences in responses at adjacent time-points to the conventional variance (i.e. sum of squared differences from the mean response of the time series) among responses. In case of some trend in the data, responses from adjacent time points will tend to be closer together. Under the null hypothesis of 'white noise' all $Q_n = n!$ permutations of responses will be equally likely with probability $1/n!$ and an exact p-value can be defined and computed as before.

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Chapter 16

SINGLE CASES, GROUP STUDIES AND CASE SERIES IN APHASIA THERAPY

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INTRODUCTION

In neuropsychology the case for single case studies has been extensively discussed (see Caramazza, 1986; Shallice, 1988). In essence the argument runs as follows. There are a large number of possible patterns of acquired cognitive impairment. The results of a set of participants can only be meaningfully combined if they suffer from the same underlying impairment(s). If they do not, the findings for the group may not accurately reflect the performance of all the participants – or indeed any of the participants. Therefore, to conduct a meaningful group study it is necessary first to establish first that the participants have the same underlying disorder. This can only be done on the basis of studying each as a single subject.

In addition, participants cannot be grouped on the basis of surface symptoms because these may reflect different underlying disorders. For example, the classical 'syndromes' of Broca's aphasia, Wernicke's aphasia, conduction aphasia and so on are defined in terms of surface features, primarily those evident in spontaneous speech. It is now well established that these surface features can have different underlying causes.

The disadvantage of single case studies is that they seek to establish a set of facts that are true just for a single participant. They do not generalise to other participants. Generalisation, as Coltheart (1984) pointed out, is not to other participants but to an underlying cognitive model. Group studies are typically based on a random sampling model. That is it is assumed that the subjects are a random sample drawn from a particular population defined in some specified way: aphasic people, or Broca's aphasics, or people with aphasia following sub-arachnoid

haemorrhage or whatever. If this is a random sample then the results from the group generalise to the population from which the random sample was drawn.

For studies of aphasia therapy this property seems particularly important. A single case study of treatment can, as will be discussed, establish that a particular kind of treatment has been effective for an individual subject. While this is obviously good for that participant, the result is, in essence *a posteriori*. We know after the event, that treatment has been effective. In order to design therapy, however, we need, ultimately, to be able to predict the therapy that will be effective for an individual patient.

This chapter will consider some of the characteristics of single case and group studies as applied to aphasia therapy. I will argue that studying a series of single cases can have the advantages of both approaches, while avoiding their drawbacks.

SOME BACKGROUND ASSUMPTIONS

First, I assume that, in studying the effectiveness of aphasia therapy what we want to know is which kind(s) of treatment(s) are effective for which kind(s) of patients. This is so that a therapist, with a particular patient (with a particular pattern of impairments) can decide what is/are the most effective treatment approach(es) to use.

Secondly, I will assume, probably uncontroversially, that people with aphasia have a variety of heterogeneous language disorders; that is, there are a (large) number of qualitatively different patterns of language impairment.

The third assumption is that aphasia treatments are qualitatively heterogeneous; there are many different treatment approaches aimed at many different problems.
A consequence of these latter two assumptions is that it is extremely likely that any one treatment will benefit some people with aphasia and not others, and that any one aphasic person will benefit from some treatments and not others.

As a result, it will only be possible to establish what treatments are effective and for whom if, in treatment studies of any kind:

1. The treatment is specified
2. The nature of the patient(s) disorders are specified

3. It is established which of the participants benefited

What therapy studies seek to do is to establish whether patients improve and whether the improvement is due to:

1. Spontaneous recovery
2. "Charm": that is non-specific effects of treatment such as placebo effects or Hawthorne effects.
3. The specific effects of the treatment involved.

All studies of treatment effects seek to differentiate between these as sources of improvement.

GROUP STUDIES

Between-group studies

These studies compare the outcomes for two groups of participants. One can, for example, compare the effects of two treatment regimens; for example Wertz *et al.*. (1981) compared individual and group treatment for people with aphasia. Or, a comparison can be made between treatment from therapists and from volunteers (eg. David *et al.*, 1982), or therapy and no therapy (eg. Lincoln *et al.*, 1984), between treatment soon after onset compared to treatment at some later point (eg. Wertz *et al.*, 1986).

With studies of all of these kinds, when the people with aphasia are allocated at random to groups - and they should be - this is a randomised controlled trial. Where these studies investigate two different treatment regimens that could not be applied to the same people (eg. early versus late treatment) a group study of this kind is the only way that these issues can be empirically addressed.

A second kind of group study can address issues of the kinds of people that benefit from a particular treatment approach: for example, a comparison of the outcomes for men and women, for older and younger people with aphasia, for people with aphasia with different aetiologies, or for people with different types of aphasia. Here random assignment to groups is clearly impossible.

The advantage of between-group studies is that the results of the study can be generalised to the population from which the subjects were randomly drawn if they receive the *same* treatment. But, in practice it is very hard – or impossible – to know what the treatment was.

Group studies almost always involve diverse treatments applied to a diverse group of patients, so that it is usually unclear what the treatment really was. So, for example, in Wertz *et al.*'s (1981) comparison of group and individual treatment, the outcome must depend critically on the content of the group and individual treatment. We would not, presumably, expect the same outcome (marginally better results with individual treatment than group treatment) unless the treatment was the same.

As a result it is impossible to conclude with any confidence that similar results would be found with other subjects (or in other countries, in other centres ...).

Within-subject designs

These involve two different types of treatment applied to one group of subjects. One possibility is to compare two different types of treatment used in different periods. For example Howard *et al.* (1985) compared 'phonological' and 'semantic' therapies used with one group of 12 people with aphasia. This is sometimes called a time series design.

Another form of within-subject design compares treatments aimed at different sets of items. One can, for example, compare the change on treated items with that on untreated items (eg Pring, Hamilton, Harwood and McBride, 1993). Other studies have compared the change on a task on which treatment is focused with another untreated task.

Group studies : the problem

Group studies are usually analysed statistically with analysis of variance (or similar techniques). These statistical techniques assume that the effects of treatment are *homogeneous* across the population (although subject to measurement error). The group results will be meaningless if the treatment effects are heterogeneous (Howard, 1986). If some of the people with aphasia improve and others do not, then the results clearly cannot be generalised to all the individuals in the population. The alternative is to use statistical methods that allow us to identify whether there are significant differences in the size of the treatment effects. If there are, one can then seek to identify what characterises the people that show improvement and those that do not (or, equivalently, those that show most improvement compared to those that show least.

Although it is possible to use statistical tests for homogeneity, they have almost never been employed in studies of aphasia therapy.

TWO EXAMPLES OF MISLEADING RESULTS FROM THE USE OF GROUP STATISTICS.

Here, I will consider two different examples where, because analysis of variance was used as the statistical analysis method, conclusions that were at most partially correct were drawn.

A group study of "semantic therapy" by Pring *et al.* (1993)

In this study, a group of five patients were treated using a task in which they had to match one picture to one from a choice of five written words, where all the distractors were semantically related to the target. Treatment was daily for a period of two weeks. The participants' naming of four sets of items was probed before treatment, at the end of treatment, and four weeks later. The four sets were: 'treated' items, 'related seen' whose names were among the written distractors, 'related unseen' – semantically related items whose names were not distractors in the treatment task, and 'control' items. The outcome for the group is illustrated in Figure 1. Analysis of variance on the results showed that there was a set x test interaction. Subsequent analyses showed that this was because the treated items improved after therapy relative to the other items. There was also significant, but much smaller improvement in the 'related seen' items.

But they are simple, cheap and, when It turns out, though, that this picture is misleading. There are large differences between the participants in the effectiveness of the therapy. What follows concentrates simply on the contrast between treated items and controls at the end of therapy, when the effects of treatment were at their greatest. Figure 2 shows the difference between naming accuracy for the treated items and the controls at the end of therapy for the participants individually and as a group, expressed as an odds ratio, together with a 95% confidence interval for the size of the odds ratio (see Everitt, 1996, pp.165-171 for an introduction to odds ratios). An odds ratio of 1 indicates no difference between treated items and controls. An odds ratio greater than 1 shows that naming of treated items was better than control items.

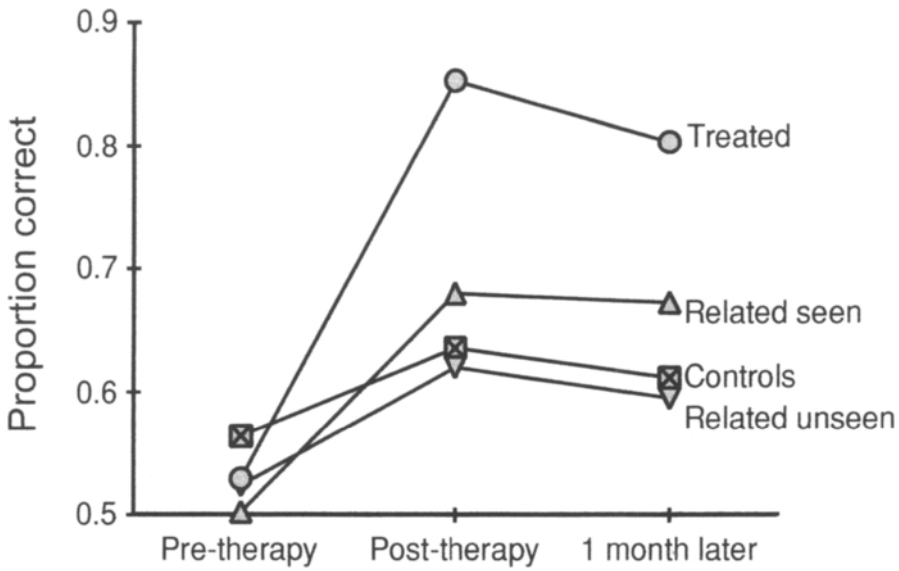

Figure 1. Changes on different sets in the Pring *et al.* (1993) study

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Three of the subjects, MM, PR and BB show differences between treated and control items that are clearly highly significant. The odds ratios for the other two subjects – DL and RH – clearly include 1 within their 95% confidence intervals. That is they show no significant difference in naming treated and control items after therapy. A homogeneity test on the size of the treatment effects shows a significant difference among the participants ($\chi^2(4)=11.18$ p= .025). This means that we can reject the hypothesis that the advantage for treated items relative to controls is the same for all five participants.

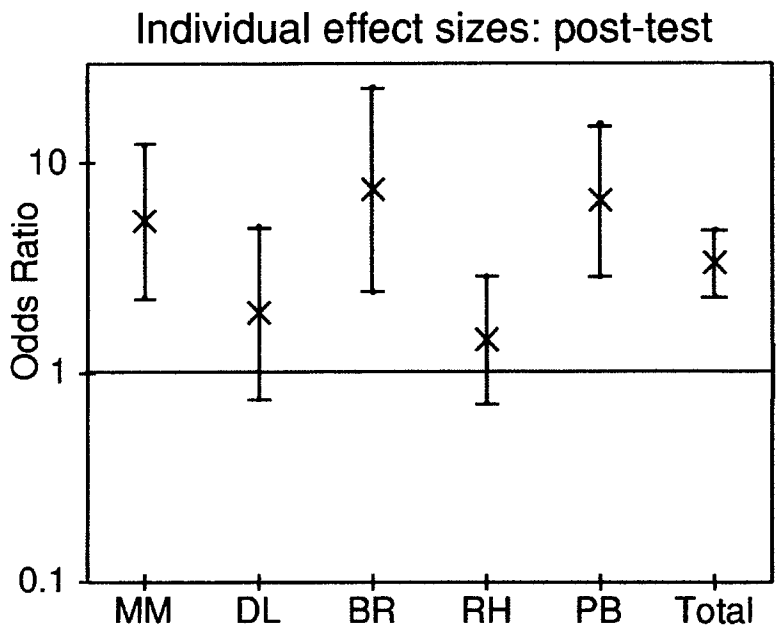

Figure 2. Improvement for individual subjects in the Pring *et al.* (1993) study. Odds ratios are on a logarithmic scale.

The analysis of variance gave a significant result for the patients as a group. This is because the treatment effect was positive in every case, even though it was not significantly better than chance for two of the participants. This result is misleading, because, as I have shown, there is significant heterogeneity in the effects of treatments between the subjects.

The next step would be to seek to understand what differentiated the participants who showed most improvement from those that improved least. It is only once the homogeneity of the treatment effects has been statistically tested, that it is possible to see that this is necessary.

A comparison of 'Semantic' therapy and 'phonological' therapy by Howard, Patterson, Franklin, Orchard-Lisle & Morton (1985)

This study compared semantic and phonological therapy. In 'semantic therapy' items were treated by spoken & written word-to-picture matching and yes/no semantic judgments. In 'phonological therapy' items were treated by word repetition, phonemic cueing and yes/no rhyme judgments. Twelve participants were treated over 1 or 2 weeks. Naming was probed

each day during treatment for the treated items and naming controls that were presented for naming as often as the treated items. The group results are illustrated in Figure 3.

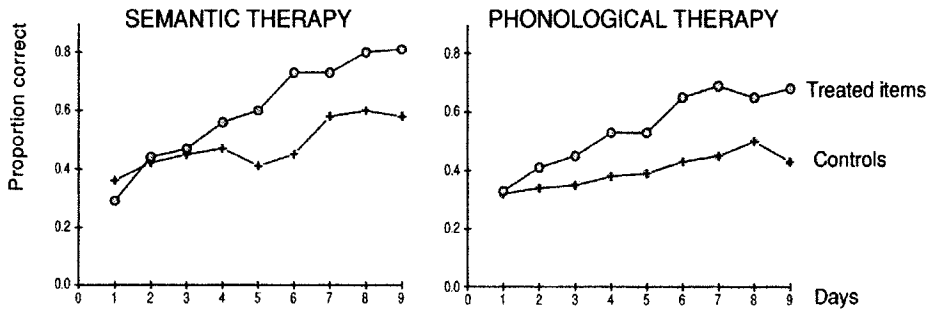

Figure 3. Day-by-day improvement on the treated items and controls in the Howard *et al.* (1985) therapy study

Analysis of variance on these data showed that improvement for the treated items was significantly greater than for the naming controls and that the improvement was equal for the two therapy methods.

This might, however, be for either of two reasons: (i) Some patients benefit from 'phonological' therapy and other patients benefit from 'semantic' therapy (implying different 'mechanisms' for the two treatment effects), or (ii) The patients who benefit from one therapy also benefit from the other (which is consistent with the same 'mechanism' for the effects of the two therapy techniques). The analysis of variance simply does not address such issues, and yet the data gathered in this experiment can.

The rate of improvement for the treated items from the two sets is shown in Figure 4 for the eight participants showing significant improvement. If different patients had benefited from each technique the correlation would have been negative. However, there is a very substantial positive correlation between the effects of the two therapies on naming in the daily probes, showing that the participants who benefited from phonological therapy also benefited from semantic therapy (for further details see Howard, 2000).

This finding is one that was not evident from the analysis of variance. The result was only obtainable because there were real differences among the patients in the effectiveness of therapy. These two examples demonstrate how analysis of the results of patients as a group,

using methods that assume that the effects of treatment are homogeneous, can yield results that are truly misleading.

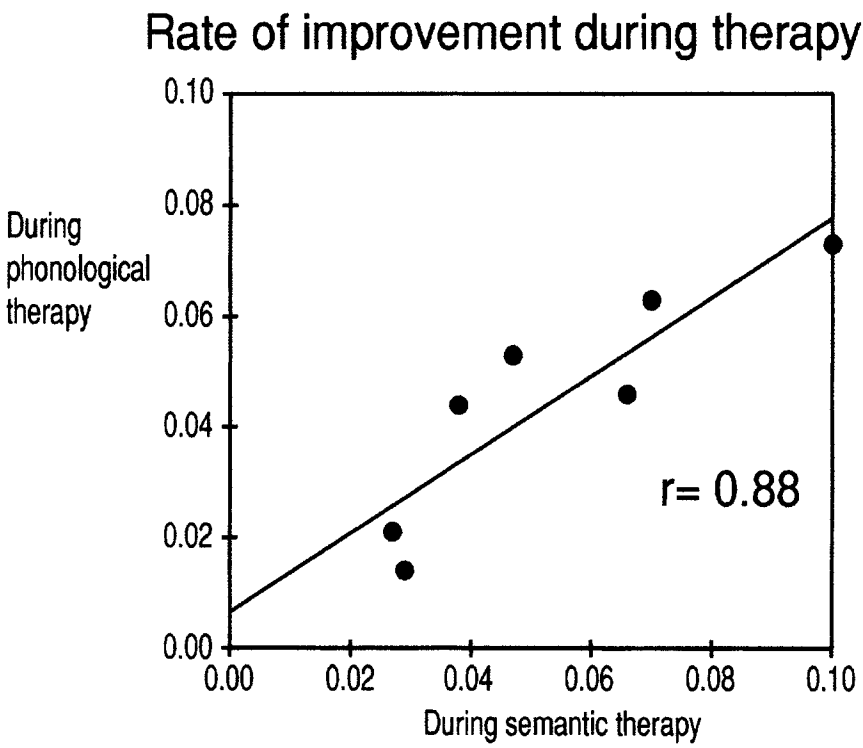

Figure 4. The rate of improvement during semantic and phonological therapy in the Howard *et al.* (1985) study

SINGLE CASE STUDIES

Studies of therapy with a single person with aphasia have some substantial advantages relative to group studies. Given that there is only one participant, it is possible to specify, in detail, the nature of the treatment. It is also possible to specify, in detail, the nature of the participant's disorder. And, when well designed, such studies can show whether an individual patient has improved, and whether improvements are item-specific or whether there is improvement to untreated items, or tasks.

However, the results apply to just this one participant. It is possible to argue that similar results might be found with a participant with the 'same' underlying disorder (eg Coltheart, 1983). But this raises the question of what counts as the 'same' underlying disorder. There is no way that we can, independently of empirical evidence, know the features that identify the people who benefit from a therapy approach and those who do not.

Single case studies are clearly open to the accusation that these are selected results from selected participants. It is conceivable that therapy studies are carried out with a whole set of patients, but only results of patients who appear to improve are actually reported. If that were the case, the improvement may not be causally related to the therapy. It might be a chance improvement (a type I statistical error), or it might be a patient who would have improved under any circumstances.

Certainly there is a selection bias in the single case studies that are published. Failures to find improvement are so inherently ambiguous (was there too little therapy, therapy of the wrong sort, or with the wrong kind of subject?) that they are not individually of great interest.

Table 1. A comparison of some of the characteristics of single case or group studies

Group studies	**Single case studies**
Complicated and expensive	Quick and cheap
In theory easily generalised to a population	Easy to specify accurately both the patient and the therapy
In practice generalisation is limited by heterogeneous therapy	Results only apply to the subject studied
Can be used to compare outcome for different types of patients	But open to replication
Analysis assumes (incorrectly) homogeneous effects	

But they are simple, cheap and, when properly designed, very informative. While, as I have pointed out, the results apply only to the patient studied, they are open to replication, particularly because it is possible to specify the treatment in some detail. Table 1 compares the features of group and single case studies.

CASE SERIES DESIGNS

I have argued that there are problems with both group and single case studies, but both have their advantages. Adopting a case series approach can combine the advantages of both without their disadvantages.

A case series involves a series of participants given the same treatment in the same way. The results of each participant, though, are analysed as single cases. This can then establish whether there is an effect of treatment for each individual subject. The next step is to statistically test whether the effects of treatment are homogeneous. This uses a homogeneity test to investigate the null hypothesis that the treatment effects are equal for all the participants[1]. If they are not, it is then possible to investigate the sources of the differences among the participants in the degree to which they show improvement. At best, this can test *a priori* predictions based on a theoretical account of how the treatment works. Less desirably, one can seek *post hoc* differences between patients who benefit more or less.

For example, Best *et al.* (2002) tested the facilitation effects of four different 'phonological' techniques: word repetition, a spoken CV cue, a written CV cue and a rime cue compared to an extra time control condition. In the experiments, conducted with 11 participants, the subjects named a set of pictures in random order. The pictures that were not named in five seconds were either given a cue or given a further five seconds to name the picture – immediate naming. The pictures were then re-presented for naming without help after a break of at least ten minutes – delayed naming.

Analysis showed that cued items were named better significantly better than controls both at immediate and delayed naming, but that the participants differed in the size of the cue effects

[1] Note that, as always, there may be no significant differences among the sizes of treatment effects either because treatment effects were homogeneous, or because the experiment was not powerful enough to detect differences in the sizes of treatment effects with different subjects.

on a homogeneity test at both immediate naming ($\chi^2(10) = 28.76$, p=.001) and delayed naming ($\chi^2(10) = 23.56$, p=.009; see Figure 5).

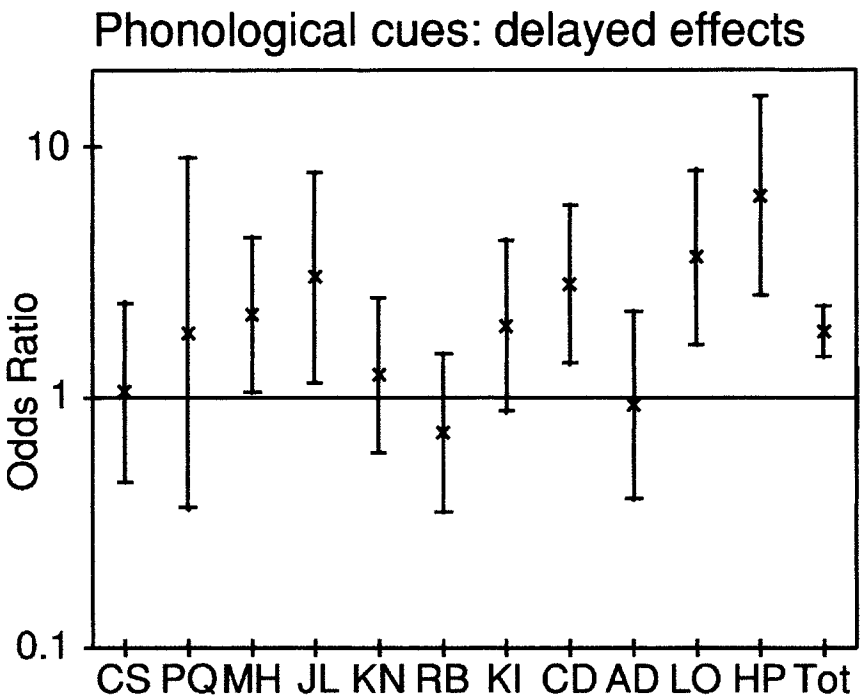

Figure 5. Benefits from phonological facilitation at delayed naming in the Best *et al.* (2002) study for eleven patients individually and for the patients as a group (Tot). The odds ratios are on a logarithmic scale.

So, which patients benefit? Howard (2000) had argued that the effects of these cues should be found with patients whose problem was in the mapping from semantics to phonology. So the patients were divided on the basis of a median split into those with more and less semantic impairment, and those with more or less impairment in phonological output. The prediction was that the largest cueing effect should be found with the people with less impairment at both levels – whose difficulty in word retrieval lay in the retrieval of lexical word forms. And the results confirmed this: at delayed naming there was a three way interaction between improvement, phonological impairment and semantic impairment (z=2.27, p=0.023) with the greatest improvement for the people with less phonological impairment and less semantic impairment (see Figure 6).

For another example of a study that takes a case series approach to the study of treatment effects, see Hickin *et al.* (in press).

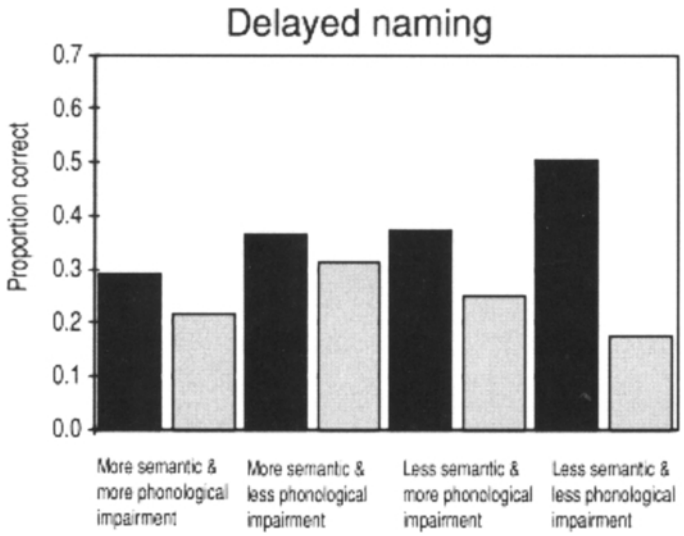

Figure 6. Performance on treated items (black bars) and controls (grey bars) for four groups of participants in the Best *et al.* (2002) facilitation study.

To summarise: case series designs of therapy can be used to identify which patients benefit from a single well-specified therapy. Supported by proper use of homogeneity tests, it is possible to address the null hypothesis that treatment gains are equal for all the participants. They can be used to test specific accounts of how therapies operate. Where the participants are a random sample from a particular population the results can be generalised to that population. There seems to be no reason why this approach should not be more widely adopted.

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Chapter 17

EFFICACY OF APHASIA THERAPY, ESCHER, AND SISYPHUS

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M.C. Escher, the Dutch graphic artist, is recognized for spatial illusions, impossible buildings, repeating geometric patterns, and his incredible techniques in woodcutting and lithography. His intricate repeating patterns, mathematically complex structures, and spatial perspectives all require a "second look." In Escher's work, what you see the first time is most certainly not all there is to see. This may be applicable to the data on the efficacy of treatment for aphasia.

In Greek mythology, Sisyphus was sent to Hades and condemned to roll a huge stone up a hill, only to have it roll down when he reached the top. His name has been used as an eponym in the "Sisyphus Reaction" that applies to an individual who strives against all odds but achieves little sense of accomplishment or satisfaction. If you have conducted aphasia treatment studies, you may identify with Sisyphus.

The relationship among the efficacy of treatment for aphasia, Escher, and Sisyphus, is demonstrated in a recent exchange of opinions between the Danish stroke group, Pedersen *et al.*, and me. In their report on "Aphasia in Acute Stroke: Incidence, Determinants, and Recovery," Pedersen *et al.*, (1995) said, "We found no difference in recovery of aphasia between patients who did and those who did not receive aphasia therapy. The study was not designed to investigate the effect of aphasia therapy...but it could be noted that the result is in line with the majority of previous studies..." (p. 665). What do the studies they cited tell us? Three, David *et al.*, (1982), Meikle *et al.*, (1979), and Hartman and Landau (1987) compared two treatments. None included a no-treatment control group. A comparison of treatments with unknown efficacy will only indicate whether one treatment is the same as, better than, or worse than the other. It indicates nothing about the efficacy of either treatment. A fourth study, Poeck *et al.*, (1989), employed single-treatment groups at three times after onset and

observed significant improvement in treated aphasic patients beyond what could be expected from spontaneous recovery. Two additional studies, Basso *et al.*, (1979) and Shewan and Kertesz (1984), observed significantly more improvement in treated aphasic patients than in self-selected, no-treatment patients. Another study, Wertz *et al.*, (1986), utilizing random assignment of aphasic patients to treatment and no-treatment groups, found significantly more improvement in aphasic patients treated by speech-language pathologists than patients receiving no treatment. Only one of the investigations cited, Lincoln *et al.*, (1984), found no significant difference in improvement between randomly assigned treated and untreated patients. I (Wertz, 1986) responded with a gentle suggestion that Pedersen et al. were misinterpreting the literature they cited. I said, "Three of the studies Pedersen and colleagues cite are inappropriate, because they did not examine the efficacy of treatment. Four studies indicate treatment for aphasia is efficacious. And, one study indicates treatment for aphasia is not efficacious. How Pedersen and colleagues could conclude that their '...result is in line with the majority of previous studies...' is amazing" (p. 130). Pedersen *et al.* response (1996) indicates how effective my gentle suggestion was. They replied, "We will maintain that this statement is an accurate description of the treatment studies that we have listed...No treatment is not a good control, because these patients may have their performances negatively affected by the decision not to treat them...It is only when no difference is found, that it is possible to draw a valid conclusion from a comparison with no treatment" (p. 130). Again, I am prompted to reply, "amazing." Escher is at work. We look at the same literature and interpret it completely differently. And, so too is Sisyphus present. Those eight investigators rolled the stone up the hill called the efficacy of treatment for aphasia only to have it roll back down.

This paper is an exploration of the confusion that exists in aphasia treatment outcome research, and it ponders whether we will ever get that stone to stay at the top of the hill. Its purposes are: to propose some "rules" for conducting and evaluating aphasia treatment research, to examine the "general" question of efficacy of treatment for aphasia, to examine the efficacy of specific treatments, and to draw some conclusions and make some suggestions.

SOME "RULES"

Perhaps some of the confusion in aphasia outcomes research results from application of different yardsticks to measure the results. Robey and Schultz (1998) have provided some "rules" we might employ in our evaluations. They are precise definitions of the

terminology—outcome, efficacy, effectiveness—and an elaboration of the five-phase outcomes research model employed by most scientific disciplines. In addition, I have added the use of "Levels of Evidence" scales employed to rate the quality of the evidence provided by an outcome investigation. We may not agree about what the "rules" should be, but unless we consider what the "rules" might be, confusion will continue. So, let's begin with the terminology.

Definitions

"Outcome," generally, refers to determining a difference between observations made prior to implementing a treatment and observations made after the conclusion of that treatment. Determining the efficacy of a treatment is only one form of clinical outcome research. Another form might be simply selecting a group of aphasic people, evaluating them, treating them, reevaluating them, and comparing the pre- and post-evaluations. This approach would not index efficacy, but it would provide a simple outcome—change between two points in time, pretreatment and post-treatment.

"Efficacy" has a more specific meaning in the realm of outcomes research. As defined by the Office of Technology Assessment (1978), efficacy is the probability of benefit to individuals in a defined population from a medical technology applied for a given medical problem under *ideal* conditions of use. The requirement of *ideal* conditions indicates that an efficacy experiment indicates the *possible* benefits of a treatment, not the *actual* benefits.

"Effectiveness," according to the Office of Technology Assessment (1978), is the probability of benefit to individuals in a defined population from a medical technology applied for a given medical problem under *average* conditions of use. Thus, efficacy indicates whether a treatment *can* work, and effectiveness indicates whether a treatment *does* work, in everyday practice. A rule of outcomes research is that effectiveness experiments are conducted *only after* efficacy has been established.

Another concept that surfaces in outcomes research is "efficiency." It is defined as acting or producing effectively with a minimum of waste, expense, or unnecessary effort, essentially, exhibiting a high ratio of output to input (Wertz, 1998). As with effectiveness studies, efficiency research is done *only after* efficacy has been established.

Are the distinctions among the terminology important? They are if we ever want to reach consensus. Currently, many speech-language pathologists call every outcome study an

"efficacy" study, and many investigators leap to effectiveness and efficiency studies without having demonstrated a treatment's efficacy. Confusion reigns, and it should, because we have not agreed on what we are talking about.

Five-phase Outcomes Research Model

Robey and Schultz (1998) remind us that outcomes research should be programmatic—evolving systematically through each phase in the traditional five-phase outcomes research model. Moreover, they specify how the terminology—outcome, efficacy, effectiveness—is tested in specific phases of the model.

In Phase I, the purposes are to develop the hypotheses that will be tested in later stages of the model; to establish the safety of the treatment; to detect whether the treatment is active, do patients improve; and to define the population for whom the treatment is appropriate. Phase I experiments are brief, they employ small sample sizes, and no control patients are required. Appropriate designs are small group studies, case studies, and single-subject experiments.

If the results of Phase I studies are positive, research continues in Phase II. Here, the purposes are to refine the hypotheses, standardize the treatment protocol, validate the outcome measures, determine the optimal dosage, establish study-patient selection criteria, and develop an explanation for why the treatment may work. As in Phase I studies, Phase II utilizes small-group experiments, case studies, and single-subject experiments, and no controls are required.

Again, if Phase II research is promising, Phase III research is initiated. The purpose is to test the efficacy of the treatment that was developed in Phases I and II. The typical design is a randomized controlled trial where study-patients are assigned, randomly, to treatment and no-treatment conditions. Large sample sizes are necessary to obtain sufficient statistical power with a specified effect size. To obtain the large samples, typically, a multi-center trial is employed.

If the treatment's efficacy is demonstrated in Phase III, Phase IV research is initiated. The purpose is to test the treatment's effectiveness under typical conditions of practice with typical patients. Large samples are required. The designs include a single treatment group with no control, comparison of patients receiving the treatment with patients receiving another treatment, comparison with self-selected no-treatment patients or historical controls, or single-subject experiments with multiple replications.

Phase V research is a continuation of the effectiveness research initiated in Phase IV. Efficiency studies, for example, comparing the intensity and the duration of the treatment, comparing different conditions of quality control, collecting consumer satisfaction data, and collecting quality of life measures, are appropriate. Again, large samples are required, and these can be obtained through single-subject experiments with multiple replications and randomized group comparisons to compare different treatments, different intensities, different durations, and different levels of clinician training.

The five-phase model takes a treatment during a period of discovery, Phases I and II, through a test of its efficacy, Phase III, to tests of its effectiveness and efficiency, Phases IV and V. The path is paved systematically through each successive phase.

Quality of Evidence Scales

Means for evaluating the outcomes obtained in outcome research are called "Quality of Evidence" or "Level of Evidence" scales. Several exist. One is that employed by the American Academy of Neurology (1994). Three classes of evidence are used. Class I evidence comes from one or more well-designed randomized controlled clinical trials. Class II evidence comes from one or more well-designed randomized clinical studies such as case-control or cohort studies. Class III evidence is based on expert opinion, nonrandomized historical control studies, or one or more case reports.

A similar "Level of Evidence" scale was developed by Birch and Davis (1997). It employs three levels of evidence and three consensus levels. Level A evidence is represented by a meta-analysis that includes two or more randomized controlled trials and other studies with good internal and external validity. Level B evidence constitutes a randomized controlled trial with good internal and external validity and that has good generalizability. Level C evidence relies on nonrandomized clinical trials, such as case control or cohort studies, historical controls, or pre- and post-treatment comparisons. The three consensus levels range from "strong consensus," where 90% of the members on a practice guideline panel agree; "consensus," where 75% of the members on a practice guideline panel agree; and "clinical opinion or standard of practice," where the evidence is provided by a study that does not meet level "C" evidence.

THE DATA

The above are some possible "rules" for designing and evaluating outcomes research—precise definitions of the terminology, the five-phase outcomes research model, and levels or quality of evidence scales. We may not agree on them as "*the rules*," but unless some consensus in conducting and evaluating aphasia outcomes research is reached, the current confusion will persist. The following applies these "rules" in evaluating selected literature on the efficacy of treatment for aphasia. Again, we may not agree that these should be the "rules," and they may be wrong. Nevertheless, the following, if wrong, is wrong with a rationale.

Meta Analysis

The highest level of evidence in the Birch and Davis (1997) scale is a meta-analysis. It is a mathematical means for synthesizing independent research findings scattered throughout a body of literature. Its products are the average effect size and its confidence interval which permit an estimate of the degree to which a null hypothesis is false, for example, treatment is *not* efficacious for aphasia, on the basis of all available evidence. Three meta-analyses have been conducted on outcomes in aphasia.

Whurr, Lorch, and Nye (1997) examined 166 studies in 45 papers in their meta-analysis. Their results indicate that "...73% of patients who received treatment improved more than half a standard deviation compared to those who did not receive treatment" (p. 9). And, they concluded that, "...overall, speech and language therapy for adult aphasics yielded positive results" (p. 9).

Robey (1994), in his first meta-analysis, examined 21 aphasia treatment studies. He concluded "...treatment of aphasic individuals (generally considered) by speech-language pathologists is efficacious" (p. 602). In his second meta-analysis, Robey (1998) examined 55 reports on clinical outcomes in aphasia. His results indicated that, "Outcomes for treated individuals are superior to those for untreated individuals in all stages of recovery" (p.184). And, "Outcomes are greatest when treatment is begun in the acute stage of recovery" (p.184). Addressing the influence of treatment intensity, Robey reported that, "Treatment length in excess of two hours per week brings about gains exceeding those that result from shorter durations" (p. 184). Examining the influence of type of treatment, he said, "The average effect size for SWDM..."—the type of treatment advocated by Schuell *et al.*, (1964), Wepman (1951), and Darley (1982) that is typically described as multimodality stimulation—

"...was larger than the overall average" (p. 184) achieved with other types of treatment. And, finally, he observed that, "Large gains are achieved by severely aphasic persons when treated by a speech-language pathologist" (p. 184).

The results of the three meta-analyses are positive. All permit inference that treatment has a positive influence on outcome for aphasic patients.

Treatment versus No-Treatment Comparisons

The randomized controlled trial provides Class B evidence in the Birch and Davis (1997) scale and Class I evidence in the American Academy of Neurology (1994) scale. Four randomized controlled trials, comparing treatment with no-treatment, have been conducted with aphasic patients. Lincoln *et al.*, (1984) conducted the first randomized controlled trial. They concluded that "...there were no significant differences in language recovery in the 104 patients allocated to the treatment group and the 87 allocated to the no-treatment group" (p.1197). Conversely, in the second Veterans Administration Cooperative Study on aphasia, Wertz *et al.*, (1986) concluded that, "Patients who receive language treatment by a speech pathologist improve significantly more than do untreated patients" (p. 658). Katz and Wertz (1997), using random assignment to treatment groups, compared computer-provided reading treatment with no-treatment. Their results indicated that, "The computer reading treatment group made significantly more improvement than the no-treatment group..." (p. 505). And, Elman and Bernstein-Ells (1999), comparing randomly assigned patients to group communication treatment and no-treatment, observed that, "...participants receiving group communication treatment had significantly higher scores on communicative and linguistic measures than participants not receiving treatment" (p. 411).

As indicated earlier, the randomized controlled trial constitutes a Phase III, efficacy study. Three of the four trials permit the inference that treatment is efficacious for aphasia. The exception, Lincoln *et al.*, (1984), observed a negative result--no significant difference in improvement between treated and untreated patients. Even though the designs were similar, other elements differed among trials. For example, study-patient selection criteria, time postonset when treatment was initiated, intensity and duration of treatment, and the nature and content of the treatment differed among trials. The three positive results appear to represent Phase III, efficacy, research. Conditions appeared optimal, a requirement of an efficacy study. The Lincoln *et al.*, (1984) design, however, is enigmatic. It constituted a randomized controlled trial, however, the conditions under which it was conducted resemble effectiveness research, Phase IV, more than efficacy research, Phase III. Few study-patient selection

criteria were employed, thus the samples were less than optimal. The intensity and duration of the treatment prescribed were two hours of treatment each week for 24 weeks. However, less than a third of the patients received the amount of treatment prescribed, a condition more akin to effectiveness, average conditions, research than efficacy, optimal conditions, research. In fact, the authors titled their report "The Effectiveness of Speech Therapy for Aphasic Stroke Patients: A randomized controlled trial."

Other Evidence

The results of other aphasia outcome studies provide additional evidence. Hagen (1973), using a waiting list, and Basso *et al.*, (1979) and Shewan and Kertesz (1984), using self-selected no-treatment groups, all reported that treated patients made significantly more improvement than patients who received no-treatment. These studies represent Level C evidence in the Birch and Davis (1997) model and Class III evidence in the American Academy of Neurology (1994) model. Poeck *et al.*, (1989) compared improvement in treated patients with the amount of improvement attained from spontaneous recovery in previous, untreated patients. They concluded that, "...with intensive treatment, improvement went beyond that expected with spontaneous recovery" (p. 476). Hartman and Landau (1987) compared traditional aphasia treatment with patient and family counseling. Patients in both groups improved, and there was no significant difference in improvement between groups. Unfortunately, Hartman and Landau interpreted their results to imply treatment for aphasia is not efficacious. As indicated earlier, a comparison of treatments of unknown efficacy will not permit inference about the efficacy of either treatment. Perhaps, Hartman and Landau believed the counseling group represented a spurious-treatment, or placebo treatment control. However, these patients received two hours of counseling treatment each week for six months. And, as Robey and Schultz (1998) observe, a placebo form of aphasia treatment cannot exist. Finally, the first Veterans Administration Cooperative Study on aphasia, Wertz *et al.*, (1981), compared randomly assigned patients who received either individual treatment or group treatment. The results indicated that both groups made significant improvement, and individually treated patients made slightly, but significantly, more improvement than group treated patients.

Treatment by Trained Volunteers

Considerable controversy has resulted from investigations that compared speech pathologist delivered treatment with trained volunteer treatment. The results of these studies (David *et*

al., 1982; Meikle *et al.*, 1979; Shewan and Kertesz, 1984; Wertz *et al.*, 1986) are quite consistent. Patients in both groups in all studies improve, and there is no significant difference in improvement between groups. In the two studies that included a no-treatment control, Shewan and Kertesz (1984) and Wertz *et al.*, (1986), the results continue to be consistent. Patients treated by speech pathologists improve significantly more than patients who receive no treatment, however patients treated by trained volunteers do not differ significantly in the amount of improvement attained from either speech pathologist treated patients or patients who receive no treatment. The straight-forward conclusion is that speech pathologist treatment for aphasic patients is efficacious, and volunteer treatment for aphasic patients is not.

Type Of Treatment

Specifying the treatment is a problem that plagues every aphasia outcome study. Howard and Hatfield (1987) said, "...it is axiomatic to every school of treatment that the tasks a patient is asked to do should be determined by his/her particular aphasic symptom complex. So, for different patients, therapy varies both in the sorts of tasks and in their level of difficulty." This heuristic observation appears obvious, however it has not been tested empirically.

Certainly, a variety of treatments exist. These have been grouped by Horner *et al.*, (1994) into: stimulation-facilitation, modality, linguistic, processing, minor hemisphere mediation, and functional. Obviously, different taxonomies would probably result if others did the grouping. Moreover, in the Horner et al. classification, and, perhaps, any classification, it is difficult to confine a treatment to a single category.

Robey (1998) experienced this difficulty when he attempted to code treatment in his second meta-analysis. The acronymic treatments—VAT, MIT, PACE, etc.—were easy. Deciding what constituted multimodality stimulation was more difficult. Several linguistic and cognitive treatments were difficult to differentiate from multimodality stimulation. And, of course, most treatment descriptions were vague or absent, so "not specified" coding was common.

Robey (1998) observed that the most frequently specified treatment was multimodality stimulation, and its effect size in the meta-analysis was 1.39. The most frequently coded treatment was "not specified," and its effect size was 0.81. The effect size for both,

multimodality stimulation and not specified, exceed the benchmark for a large-sized effect. Thus, what Schuell *et al.*, (1964), Wepman (1951), and Darley (1982) recommended, multimodality stimulation, appears efficacious, and so too is something we do not know what to call it.

The comparison of treatments study results are fairly uniform. With one exception (Wertz *et al.*, 1981), group versus individual treatment, all group comparison of treatments studies show no significant differences in improvement between the treatments compared. However, the treatments compared have been sparse. The speech pathologist versus volunteer treatment comparison really does not constitute a comparison of treatments, because both were doing the same thing—multimodality stimulation.

SINGLE-SUBJECT DESIGNS

This review on the efficacy of aphasia therapy has said little about single-subject designs. I agree with Robey and Schultz (1998) that, "Efficacy is a property of a treatment delivered to a population and inference to a population requires a group experiment. Thus, because single-subject experiments do not provide inference to a population, they do not and cannot index efficacy" (p. 805). However, this is not to demean the value of single-subject research. Single-subject designs are particularly valuable for examining whether a treatment is active and for developing hypotheses during Phases I and II in the five-phase outcome research model. In addition, they are useful, with multiple replications, in Phases IV and V, effectiveness, research. And, there is probably no more efficient means for comparing treatments than the single-subject alternating treatments design. So, the question is not, "Group *or* single-subject design?" The question in selecting one or the other, is "What is the question?"

CONCLUSIONS

Given the data to date, we can ask, "What do we know?" The meta-analyses, randomized controlled trials, and other reports suggest that: outcomes for treated patients are superior to those for untreated patients in all stages of recovery; treatment intensity of two hours a week, or more, seems to result in gains that exceed those resulting from less intensity; the optimal duration of treatment has not been specified; the efficacy of a specific type of treatment, except for "multimodality stimulation," has not been demonstrated; severely aphasic people

achieve large gains when treated by a speech-language pathologist; and insufficient data exist to determine the differential effects of different treatments for different types of aphasia.

More importantly, we should ask, "What might we want to know?" Some possibilities are: How much of what kind of treatment for which aphasic patients? What is the validity and believability of the measures we use to index outcome in aphasia? What is the criterion for beneficial change in treated aphasic patients and what are the cost-benefit ratios for the treatment provided?

Darley (1972) asked, "What are the relative degrees of effectiveness of various modes of treatment of aphasia?" (p. 5). The current answer is "not much." But, we have not done our best to find out. Finding the answer may lie in the systematic use of single-case alternating treatments designs in Phase I and II outcome studies. Armed with the evidence about which treatment appears most appropriate for which aphasic people, we could justify the expense of Phase III efficacy research to provide the answer.

Also, Darley (1972) asked, "Are the language gains attributable to therapy worth the necessary investment of time, effort, and money?" (p. 5). Again, the empirical evidence to answer the question has not been forth coming. Most efficacy studies have indexed outcome in terms of reduction in language impairment. There is a need to index outcomes in reduction of communicative disability. Probably, the answer will come from determining the social validity of our outcomes—determining whether improvement can be detected by naïve observers and, specifically, how much improvement is necessary to reach the observer's detection threshold. In addition, patient satisfaction surveys and measures of change in quality of life will be essential in pursuing the cost-benefit answer.

Robey (1998), addressing the last and next eras of clinical outcome research in aphasia, described the past and provided direction for the future. He observed, "Many studies have tested omnibus hypotheses, for example heterogeneous patients improve with the administration of heterogeneous treatments provided on heterogeneous schedules in heterogeneous contexts" (p. 183). He called for, development of focused hypotheses, tested programmatically, for example, replications on tests of dosage, specific populations, certain severities, and treatment protocols.

Like the species, aphasia outcome research has evolved. And, like the species, aphasia outcome research may have reached a stage in evolution where it is a bit portly. Getting in shape may require programmatic research where we apply the terminology precisely and consistently, employ the five-phase outcomes research model systematically, and evaluate our

results with levels of evidence scales. And, when a sufficient body of data has been amassed, it can be submitted to meta-analyses to synthesize the information. Ultimately, unlike Escher, when we look at our results, we will be able to agree that they *are* what they look like. This, of course, will require, like Sisyphus, continuing to roll that stone up the hill called "The Efficacy of Treatment for Aphasia."

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Chapter 18

META- ANALYSIS IN APHASIA THERAPY

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The focus of this chapter will be on the summarization and integration of research data in the treatment of aphasic patients. While our unabashed bias is in favor of a more systematic approach to research integration, we think an overview of the more common and less systematic summary methods will provide a better understanding of the systematic method known as meta analysis, the topic of our presentation. It is also important to recognize that not everyone means the same thing when they use the term 'systematic summary' or 'meta-analysis'. In the USA, the two terms tend to be synonymous and refer to a complete identification, evaluation, and statistical analysis of data a topic or domain of interest. In most of the rest of the world, these terms may be mutually exclusive (see Figure 1).

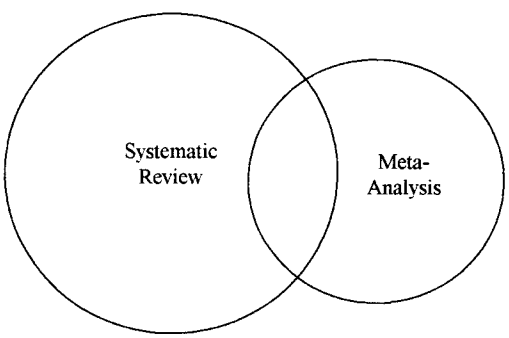


Figure 1. Venn diagram of relationship between systematic review and meta-analysis functions

A systematic summary is the identification and summarization of the body of research in a given domain without the necessarily engaging in any statistical analysis of the data presented in that research. The term meta analysis is applied to the statistical analysis aspect of a review, but would not necessarily be a requirement of a systematic review. For our purposes,

we have chosen to use the terms interchangeably. Our position assumes that there are data available in the body of literature that can be subjected to a statistical analysis at some level. If in the process of identifying, coding, and organizing the data for a review, no studies are found which are amenable to statistical analysis, then the appropriate term would be 'systematic review' however, we have found this to seldom be the case.

The purpose of any kind of research summary is to present an account of the 'state-of-the-art' knowledge base for a given domain. This 'state-of-the-art' knowledge base should provide at least two important general results (1) a basis for estimating the quantitative effects of treatment, and (2) a guide for future research agenda.

METHODS OF SUMMARY

There are four basic methods of integrating results across multiple studies (Hunter and Schmidt,1990):
--Narrative Review,
--Significance Vote Counting Method,
--Cumulation of p-values Method,
--Meta Analysis
Let us take a brief survey of each of these approaches individually moving from the least systematic to most systematic methods.

Narrative Review

The Narrative Review is the least 'quantitative' method of systematic review in that the reviewer defines the parameters of the studies to be included. Typically the reader is not told whether these parameters are determined prior to the collection of available studies or as a result of the reviewer's collection of studies and determining which studies to include and which to exclude. The review tries to find a common thread among the included studies and eliminate the studies that do not meet some a priori criteria for inclusion. These are often the types of reviews produced by students as term paper assignments for a course. There is no attempt to apply any statistical analyses of the data, rather it is a narrative text attempting to make sense of the conclusions drawn from a set of studies related by topic, outcomes, measurement etc.

The advantage of this type of review is the compilation of a set of studies that focus on a specific issue or topic of interest. Thus the conclusions drawn may be generalizable to similar

studies and subjects. The disadvantage of this type of review that the reviewer determines which studies to include and which to exclude in the review thus producing a potentially biased summary based on an individuals preference or point of view. Additionally, the data may be restricted in application to a similarly homogeneous group of subjects which may not reflect the breadth of the individuals identified with topic of interest.

Significance Vote Counting Method

At a slightly higher level of quantitative summary is the significance vote counting method. Here, the reviewer, identifies a set of studies addressing a particular topic, determines whether the result of the individual study shows a significant or non-significant result, tallies up the total number in each category and bases the summary conclusions on the category with the greatest number of representative studies.

The advantage of this approach is that the reviewer is using a quantitative, objective standard to judge the overall effect of the treatment and retains more information for summary. The disadvantage of the significance vote counting method is that no account is made for the quality of the data for factors such as quality of research design or sample size.

Cumulation of *p*-Values Across Studies

This next level of summary is the cumulation of p-values which attempts to collapse the obtained levels of significance across studies. The advantage of this method is that statistical comparisons are made using an objective statistical metric, the significance level. The disadvantage, as Schmidt and Hunter (1990) point out, is that this method fails to account for the magnitude of effect in comparing treated and untreated subjects.

Regardless of the review approach, each of these more traditional summary reviews have common shortcomings that are identified as major concerns (Light and Pillemer, 1984):

- *Subjectivity*. Since one reviewer's opinion of what studies should be included in the summary of a domain may not be the same as another reviewer, the conclusions drawn by each could be diametrically opposed because of the diversity of the database. An even less desirable or explainable situation might occur where two reviewers include essentially the same studies and draw conflicting conclusions.

- *Scientifically Flawed.* The criteria for inclusion of a set of studies is somewhat dependent on the views and interests of the reviewer. Certainly the inclusion of studies reporting both positive and negative outcomes without accounting for sample size, design factors, instrumentation, or effect size makes for a less than precise data set upon which to base one's conclusions.

- *Inefficient.* If the reviewer is attempting to deal with a large number of studies in a narrative format, it becomes increasingly difficult to organize the information in a systematic manner. The sheer volume of information becomes a burden to articulating a coherent organization of data without some way to break the information into even smaller chunks of subjective data resulting in a further dilution of the power of any conclusions.

Meta-Analysis

There is a fourth method of summary, meta-analysis (MA) that provides a mechanism to deal with these types of problems in combining findings by using statistically systematic procedures. It follows the same guidelines of scientific experimentation as the original primary research. At its most global level, MA is a method of combining results from several different studies into a common metric interpreted as a z-distribution. The basic statistic allows for the comparison of the pre- and post-treatment conditions while taking into account such factors as the degree of variability in subject performances, the size of the sample, or the quality of the measures used. The resulting metric is known as the Effect Size (ES) and is a statement of the standardized mean difference expressed in standard deviation units. Many methods are used to quantify the size of an effect. For dichotomous outcomes, relative risk and odds ratio are examples. Typically the term effect size is used for continuous variables where the mean differences are weighted to account for variations in sample size, variance, etc. We will come back to this in more detail later.

The process for conducting this accumulation of data is parallel to the general procedure for conducting a primary study. The meta-analyst must:

a. Define the Problem
b. Describe the Method
c. Analyze the Data
d. Interpret the Data

Defining the Problem

Defining the problem for the meta-analyst is to ask the salient research question or articulate an hypothesis. As with the primary research, the main research questions may be broad in scope. Fratelli (1998) poses several appropriate questions suitable for a meta-analysis including:

"What are the general outcomes of aphasia treatment?
What are the effects of certain variables like severity and age on the outcome of treatment?
What types of treatment are most effective" (p245).

Describe the Method

The second dimension in the MA process involves five basic steps:

Collecting Studies. The first step is the data collection stage. Since the basic unit of analysis is the study, the study can be viewed as analogous to the human subject in a primary experiment. Certainly one of the major concerns of any study is to ensure the inclusion of a representative sample of subjects. That is, we want to include studies investigating the effects of the treatment of individuals with aphasia. In order to insure as representative sample as possible, the reviewer attempts to identify as many studies as possible that provide data on the effects of treatment. These studies may come from a variety of sources both primary and secondary. The sources from which treatment studies are identified might be published articles, books, monographs, dissertations, theses, unpublished reports, working papers, grant reports, or papers presented at conferences. Further this collection process can extend to studies available in any language, not just English. While the quality of the research presented in each of these sources may be quite different, the purpose at the initial stages of the process is to cast a net across as wide a range as possible in order to identify as much information as possible that might be relevant to the study of treatment effects. Studies can be eliminated at a later stage of inclusion based on more objective criteria. The goal is to identify the potentially relevant data.

With the advent of online searches, it might seem that this task has been substantially improved and the process of retrieving studies made much less difficult. In fact, several studies have found that online computer searches identify about 30-50% of the available, useable, and appropriate literature. The result is that the reviewer must invariably resort to some form of hand searching through journals, bibliographies, indexes, or files. Even so, it is certainly probable that the reviewer will not be able to obtain all studies on a give subject. This is known as the 'file drawer problem' (Rosenthal, 1979). The issue is whether or not those studies that are obtained and included are in fact 'representative' of the population of

studies. So, while the reviewer can never know for absolute certain that all studies have been located, there are methods to estimate the number of studies needed in order to insure a representative sample of data and to estimate the number of studies not found in order to alter the size of the effect obtained from the studies included in the analysis. We will not take time to go into detail about these other than to say that this estimate can be based on either the total number of studies included in the meta analysis or the size of the effect obtained in the meta analysis.

Screening Studies. The next step in the process is to conduct an initial screening of all the identified studies in order to determine which studies are, at a minimal level, going to be included in the summary and/or analysis and which are to be excluded. The assumption here is that in spite of all the studies identified, not all will be appropriate for the application of a systematic summary.

Typically, studies are initially excluded if they:

1. Provide no data regarding the pre- and post treatment conditions of a treated group (e.g., anecdotal reports, reviews, position statements).
2. The data are presented for single subject designs and case study reports.
3. The data presented is not amenable to comparison of pre- and post treatment outcomes.

Coding Studies. The next step in the MA process is to quantify the characteristics of each study. This process involves the coding of those variables the reviewer believes might in some way account for some quantity of the treatment effect measured. Minimally, the most one would code or quantify are the subject and treatment characteristics. Here is a list of subject and treatment characteristics that a reviewer might want to identify and assess in determining the degree of potential contribution of each to the treatment effect of speech-language intervention in aphasia (see Whurr *et al.*, 1992).

Subject Characteristics	Treatment Characteristics
Number of Subjects	Clinician Status (degree, training)
Subject Classification	Treatment Method (behavioural)
Gender	Intrumentation
Age	Treatment constellation (group,individual)
Education	Length of treatment Sessions
Marital Status	Number of treatment Sessions
Type of Stroke	Length of treatment program (months)
Site of Lesion	Treatment outcome (Dependent variable)
Time Post Onset	

Additionally, the reviewer could also code each study for Study Characteristics and Design Characteristics. Study characteristics may help identify potential sources of study effects related to sample size, attrition, historical nature of the discipline, qualifications of researchers, or the funding source for an area of study. The Study Characteristics might include: Data Source, Date of Publication, Source of Funding, Number of Subjects, or Attrition Rate (dropout).

The Design Characteristics address concerns regarding the quality of the research included in the meta-analysis. It is important to determine if the quality of the science has a significant bearing on the quantitative outcome and a coding of several design factors can aid the reviewer in making a statistical judgment regarding the scientific character of the included research. The primary characteristics to be coded might be sampling method (random, convenience, etc.), group assignment method (random, matched, nonrandom), and setting (hospital, clinic, etc).

Evaluating Studies. While the basic design characteristics are certainly important, it is also possible to make a judgment of the overall quality of the studies to be included in the review. Several schemes have been developed for ranking the quality of a given study. Most of these ranking systems use some sort of high to low rating of indices of internal validity, external validity, and/or reliability. The importance of this step is to provide a way to determine whether the quality of the study is related to the size of the treatment effect.

Calculating Effects. The next step in the review process is the calculation of the effect size. Over the past 25 years, a number of different formulae for calculating the effect size have been developed and used (Cohen, 1988). However, the vast majority of meta analyses will use one of three basic metrics:

The r-index is a measure of the relationship between continuous variables (e.g., number of sessions and treatment effect),

The d-index is a measure of the difference between a single dichotomous (treated/untreated aphasics) and a single continuous variable (treatment effect).

The odds ratio is a measure of treatment effectiveness which is applied when the variables under study are both dichotomous (treated/untreated aphasics and male/female). It is the odds of an event happening in the experimental group expressed as a proportion of the odds of an event happening in the control group.

The method of calculation is driven by the question being asked by the reviewer and the data being presented in a reviewed study. One metric is not better than another, just more appropriate to the primary data. The most commonly used method in Speech- Language Pathology for calculating the size of the treatment effect has been the d-index expressed as:

$$ES = \frac{Xe - Xc}{Sp}$$

ES = effect size
Xe = mean score for the experimental condition
Xpre = mean score for the control condition
Sp = pooled standard deviation of the experimental and control conditions

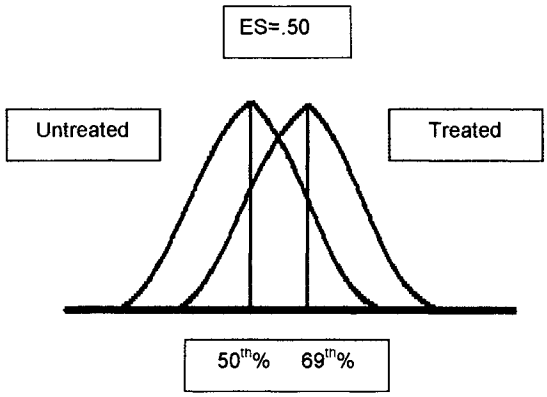
This fundamental equation is directly applied for those studies that report the means and standard deviations for the individual studies. For those studies reporting quantitative results other than means and standard deviations, ES calculations are still possible via significance tests, proportions, or p values. (Glass, McGaw, and Smith, 1981; Cooper and Hedges, 1994; Hedges, 1981; Hunter and Schmidt, 1990). Additionally, there are several statistical programs available to calculate the desired ES values while correcting for different characteristics such as sample size.

Analyze and Interpret the Data

The last step in the MA process is the analysis and interpretation of the effect size itself. The ES is interpreted as a statement of distribution under the normal curve with a 0 value representing the 50th percentile mark. For example, suppose an individual study or group of studies shows and ES=.50. An effect of .50 would be interpreted in the same manner as a z-distribution with the .5 representing a one half (½) positive standard deviation shift and suggest that on average, treated subjects in a study could be expected to improve .5 standard deviations more than the non-treated subjects. This .5 standard deviation improvement represents a 19% advantage suggesting that the average aphasic patient could be expected to move from the 50th to the 69th percentile as a result of treatment.

If this were put in the context of standardized test such as the PICA (Porch, 19XX), it would suggest that the average subject would improve approximately 8 points as a result of

intervention. With the effect size measurement, we now have a quantitative and statistical measure of the degree of effect of treatment.



The qualitative interpretation of the ES value has typically been understood in the context of a three level qualification of 'large', 'medium', and 'small' effects (Cohen, 1988). Cohen defined a large effect size as a value of .8, a medium effect as .5, and a small effect as .2. While other measures of the size of the effect have been proposed, this is still seen as a benchmark for interpreting the obtained effect size.

Published Reviews

There have been at least five MA reviews published in the literature (Greener, Enderby and Whurr, 1998; Robey, 1994; Robey, 1998; Robey, 1999; Whurr, Lorch and Nye, 1992). Four of these reviews utilized different analytical techniques and different data sets (i.e., each review used a different subset of studies). The fifth study (Robey, *et al.*, 1999) conducted an analysis of single subject design studies of intervention. The studies by Roby *et al.*, (1994, 1998) and Whurr *et al.*, (1992) utilized the same effect size index as the basis for their data analyses.

The first study by Whurr *et al.*, (1992) reviewed 45 papers that included 166 comparisons across 1336 treated subjects. The average subject was male (88%) and the achieved effect size was .592, suggesting that the treated group improved more than half a standard deviation as a result of intervention.

Robey *et al.*, (1994) included 41 studies in their review and concluded that the acute stage produced an effect that was almost twice as large as the spontaneous recovery alone. Additionally, they found a small effect size in the chronic stage. Robey *et al.*, (1998) conducted a second review and found that the largest effect sizes were obtained for those subjects in the acute stage receiving more than two hours per week of therapy and treated by a trained speech-language pathologist (SLP).

Greener *et al.*, (1998) conducted a review for the Cochrane Collaboration and using 12 randomized controlled trials representing 976 subjects treated between 1979 and 1983. In this review, the authors utilized both a d-index and an odds-ratio index to analyze the data. In none of the studies did the treated group show a significant difference from the untreated group for those variables that measured speech and language performance. Greener *et al.*, concluded "...that SLT treatment has not yet been shown to be either clearly effective or clearly ineffective within a[n] RCT. Decisions about the management of patients must, therefore be based on other forms of evidence."

However, it can be argued that the degree of effect is in part determined by the metric chosen for analysis. Table 1 presents 10 outcomes analyzed by Greener *et al.*, for which no significant differences emerged. When comparing the performance of the treated and untreated aphasic individuals using a Weighted Mean Difference (WMD). Using the same means and standard deviations for each comparison and applying the formula as presented earlier in this paper, fully half of the comparisons revealed positive values.

While the interpretation of the ES is not a measure of 'significance' it is a statement of the magnitude of effect as a result of treatment. Thus, while the lack of finding by Greener *et al.*, for a significant difference of treatment effect is certainly accurate, it does not suggest that there was no effect as a result of treatment.

Why Different Conclusions from Similar Sources?

The presence of non-complementary conclusions from different studies is, if not surprising, at least an important concern. Such differences are found in the original research arena and the same potential biases that are present when studies are replicated or compared are also present in the meta-analysis summaries. There are at least two major reasons to consider this situation:

(1) Use of different study inclusion criteria: Different results can occur because each study does not use a comparable inclusion criteria in the selection and analysis of the existing research. For example, in Whurr (1992), all studies were included that where there was a comparison design including both experimental and quasi-experimental

Table 1. Means, standard deviations and effect size for 10 outcomes

		Exp	Ctl	*Effect* Size			Exp	Ctl	*Effect* Size
Wertz 86	PICA End of Trial--Overall Score	67.19 (24.64)	61.66 (21.21)	0.237	Prins 89	Lexical Score on STAC end of trial	69.40 (21.8)	74.40 (19.6)	-0.231
Hartman 87	PICA End of Trial-Communicative Ability	10.52 (3.24)	10.65 (3.76)	-0.036	Prins 89	Monosyntactic Score on STAC end of trial	78.70 (45.7)	92.70 (45.1)	-0.295
Hartman 87	PICA End of Trial-Communicative Ability--Non-fluent	9.64 (3.43)	9.03 (3.11)	0.18	Prins 89	Wd Discrm Score on STAC end of trial	27.60 (8.5)	28.40 (6.6)	-0.101
Hartman 87	PICA 3 month Follow-up on Communicative Ability--Non-fluent	10.41 (3.22)	8.76 (3.11)	0.513	Lincoln 84	MAACL end of trial Anxiety Scale	3.00 (3.2)	2.60 (2.6)	0.135
Hartman 87	PICA 3 month Follow-up on Communicative Ability	11.22 (2.88)	10,86 (4.02)	0.101	Lincoln 84	MAACL end of trial Depression Scale	6.90 (6.6)	6.20 (5.8)	0.111

designs, while Greener (1999) utilized only randomised controlled trials for the analysis.

(2) Use of different effect size formula: Different results can also occur when different formulae for calculating effect sizes are used. For example in Whurr (1992) and Robey (1994,1998) used a *d* –index to calculate the magnitude of treatment effects. Greener (1998) used a weighted mean difference (WMD) which uses a different denominator in the formula to express the magnitude of effect.

(3) Outcome criteria selected for analysis: Different results can occur if the reviewers choose to analyse different outcomes. A review of the way in which outcomes were organized for Whurr, Robey, and Greener shows that each study utilized a different categorization/organization of the outcomes that were ultimately meta-analysed.

THOUGHTS ON THE PROVERBIAL 'GOLD STANDARD' —RANDOMISED CONTROLLED TRIALS

Randomised Controlled Trials require that participants be randomly assigned either (a) the experimental group(s) receiving the intervention(s) being tested or (b) the control group receiving no treatment, an alternative treatment or placebo. This allows for the assessment of the relative effects of intervention. The RCT (randomised controlled trial) has taken on the power of the 'Gold Standard' of research design of choice and the 'best evidence' methodological procedure. Any non-RCT is viewed as "riding the crest of a slump," that is, the non-RCT may be acceptable in the larger scheme of research, but no matter how one views research methodology, a non-RCT is still a second best methodology in many research programs.

This methodological debate ultimately focuses on the question of the degree to which one is willing support the findings of the non-RCT over the RCT research. For example, would the purchaser of speech and language services be willing to deny speech and language services on the basis of an 'insufficient evidence' finding as reported by Greener who used only RCT studies? Are the other meta-analytic studies mitigating a similar result because they included other non-RCT designs.

No good evidence that a treatment works, is not the same as saying a treatment does not work. A clear distinction between lack of benefit and the lack of evidence of benefit needs to be understood and made plain in any review. Summary phrases of clinical evidence have changed from "the review found no difference" to "the review found no evidence of

difference" We need better information on what constitutes clinically important differences in the major outcome(s) for each intervention.

A key aim of clinical evidence is to emphasise the important trade offs between advantages and disadvantages of different treatment options. A complete systematic review needs to address the issues of the effects of intervention both positive and negative results with both benefits and harms of intervention presented to the reader.

While clinical professionals, methodologists, and statisticians may argue over whether or not meta-analysis provides the 'best evidence', the fact remains that a well designed experimental study will provide a greater degree of confidence in findings and conclusions, differences across studies not withstanding, than quasi-experimental, pre-post single group, single subject design, or case study design. This is not to be construed as a denigration of all non-experimental research. Other research designs may provide a more useful and appropriate understanding of a particular problem. Single subject design research, for example, is perhaps most powerful as a statement of theory veracity and a prospectus for more focused research on an issue. Similarly, some would urge that only the RCT should be used as Greener (1998) did. The exclusive use of RCT's for trying to understand the state-of-knowledge is an attempt to place the 'best evidence' at the forefront of the scientific and clinical community. Unfortunately, in a behavioural science such as Aphasia therapy, the use of RCTs is often limited due to ethical, practical, or economic considerations. However, these limitations are not sufficient reason to abandon a 'best evidence' view of the research arena.

The data as presented in the available meta-analyses and systematic reviews argue strongly for the development and implementation of a rigorous, well defined, study of the speech and language effects in aphasia therapy. Further, a systematic research agenda that would include the development of single subject, comparative, and quasi-experimental studies as preliminary methodologies to inform the development of a more convincing knowledge base of intervention effects provides the basis for a greater impact of intervention without rejecting important contributions to the issue.

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