

Case Study

Angular gyrus syndrome revisited: Acalculia, finger agnosia, right-left disorientation and semantic aphasia

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Abstract

Angular gyrus (Gerstmann) syndrome is classically described as finger agnosia, right-left disorientation, agraphia and acalculia in association to lesions in the left angular gyrus. Aphasia is not typically described as part of this syndrome. Here we report a 58 year old right-handed male, with an ischemic lesion to the left angular gyrus, who developed sudden loss of speech expression and comprehension, and slowly recovered over the following few weeks. After several months he showed significant improvement on his language skills with only mild word-finding difficulties, but with substantial difficulties in understanding logic-grammatical relationships, comparison adverbs (e.g. bigger-smaller, younger-older etc.), place adverbs (e.g. over-below, on-beneath etc. and time adverbs (e.g. before-after). These language deficits are compatible with a semantic aphasia. Writing difficulties are minimal. In addition, he has important impairments in finger agnosia, right-left discrimination, and in understanding numbers, using numerical concepts, and performing arithmetical operations. We propose that left angular gyrus syndrome should be restated to include acalculia, finger agnosia, right left disorientation and semantic aphasia. A single underlying deficit can account for the simultaneous presentation of these four clinical signs.

Introduction

In 1940 Gerstmann described a syndrome associated to lesions in the left angular gyrus which characteristically included deficits of finger agnosia, right-left disorientation, agraphia and acalculia (Gerstmann 1940). Ever since, the existence of a Gerstmann syndrome has not been free of debate and questioning in literature (Benton 1977, 1992, Botez 1985, Poeck and Orgass 1966, Strub and Geschwind 1983). In part this debate emerges because this syndrome usually unfolds as either, an 'incomplete' tetrad or in association to other cognitive deficits, particularly, aphasia, alexia and perceptual disorders (Frederiks 1985). Even earlier the name 'angular syndrome' was proposed in lieu of the more widely recognized

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'Gerstmann syndrome' (Benson 1979, Strub and Geschwind 1983). The appearance of a Gerstmann syndrome with electrical stimulation of the cerebral cortex in the posterior parietal area supports its angular localization (Morris *et al.* 1984). Mazzoni *et al.* (1990) described a case of a 'pure' Gerstmann syndrome associated with an angular gyrus traumatic damage.

Commonly, investigators have reported the presence of Gerstmann syndrome without aphasia as one of its components (Roeltgen *et al.* 1983, Strub and Geschwind 1974, Varney 1984). However, the existence of a possible semantic aphasia has not been specifically explored nor ruled out (Ardila *et al.* 1989). In 1926 Head described a language alteration whereby he defined an inability to recognize simultaneously the elements within a sentence, and he called it semantic aphasia. In the following years only a small group of researchers referred to this type of aphasia (e.g. Conrad 1932, Goldstein 1948, Zucker 1934). Nearly 40 years later, Luria (1966, 1973, 1976) retook this concept and analysed it extensively. Since then, only a few more authors have shown special interest in studying semantic aphasia and a handful of references have appeared in the literature over the last two decades (Ardila *et al.* 1989, Benson and Ardila 1996, Brown 1972, Hier *et al.* 1980, Kertesz 1979). The *International Neuropsychological Society Dictionary of Neuropsychology* (Loring 1999: 18) defines semantic aphasia as an 'Aphasia subtype described by Henry Head and Aleksandr Luria in which there is an impaired capacity to draw inferences beyond the literal meaning of the word'. Luria (1973, 1976) considered that language deficiencies of semantic aphasia were observed in the following situations: (1) sentences with a complex system of successive subordinate clauses; (2) reversible constructions, particularly of the temporal and spatial type (3) constructions with double negative; (4) comparative sentences; (5) passive constructions; (6) constructions with transitive verbs; and (7) constructions with attributive relations. He also stated that these spatial disorders not only incidentally accompany semantic aphasia, but that semantic aphasia itself, was a defect in the perception of simultaneous structures transferred to a higher symbolic level (Luria 1976). In other words, patients with semantic aphasia have difficulty understanding the meaning of words tinged with spatial or semi-spatial meaning.

The term 'acalculia' was introduced to refer to the impairments in mathematical abilities in case of brain damage (Henschen 1925). Two types of acalculia were distinguished: primary and secondary (Berger 1926). The latter, refers to a calculation defect resulting from an associated cognitive deficit in memory, attention or language, for example. Up to date, several different acalculia subtypes have been described (e.g. Hittmair-Delazer *et al.* 1994, Roselli and Ardila 1996). Among them, anarithmetia represents the real primary acalculia and it is observed in cases of left angular gyrus damage (Ardila and Rosselli 1990, Benson and Weir 1972, Harvey *et al.* 1993, Hecaen *et al.* 1961, Grafman 1988, Rosselli and Ardila 1989, 1998). Luria underlined that the semantic aphasia, that is the inability to use verbally mediated spatial and quasi-spatial concepts, is always associated with acalculia (Luria 1966, 1973).

Arithmetical abilities represent a relatively recent acquired function in mankind. Historically, calculation abilities seem to develop from counting, and in child development this begins with the sequencing of fingers (correspondence construction) (Hitch *et al.* 1987). Finger nomination is usually sequenced in a particular order and this represents a basic procedure found in different cultures around the world, both ancient and contemporary (Ardila 1993, Cauty 1984, Levy-Bruhl

1910/1947). In fact in many contemporary languages a ten- or twenty-base system is evident. From the Latin *digitus*, 'digit' can mean both number and finger. Accordingly, a strong relationship between numerical knowledge and finger gnosis begins to become evident and some commonality in brain activity or anatomy can be expected. Along this reasoning, among the prehistorical man finger agnosia and acalculia could have represented just the same deficit.

Finger agnosia, as initially described by Gerstmann in 1924 (Gerstmann 1940) and later included as classical element of the Gerstmann syndrome, encompasses the inability to distinguish, name, or recognize the fingers not only in their own hands, but also in the examiner's hand or in a drawing of a hand. Patients present difficulties in selectively moving the fingers, both by verbal command or by imitation. Most evident errors are observed with the index, middle and ring fingers. Interestingly, finger agnosia may be associated with toe agnosia as well (Tucha *et al.* 1997). Usually the patient has difficulties in recognizing his/her errors and shows no effort in correcting them. Right-left disorientation involves the inability to identify right and left in one's own body and in that of the examiner. It includes not only linguistic, but also spatial components. As with finger agnosia, right-left disorientation is observed in cases of left posterior parietal damage, and can be included within the left angular gyrus syndrome.

Here we report a patient with a cerebrovascular event involving almost exclusively the left angular gyrus. His exam demonstrated acalculia, finger-agnosia, right-left confusion and interestingly, semantic aphasia. His writing difficulties were minimal. Moderate constructive difficulties and anomia were also observed.

Case presentation

Background

Fifty eight (58) year old, right-handed male, monolingual native Spanish speaker with high-school level education. Until his cerebrovascular accident, he worked as a successful businessman and prestigious politician. Twenty eight months before the current evaluation he suddenly lost language production and understanding. For several weeks following the event he was unable to speak and his level of language understanding was significantly reduced. Except for a mild right facial droop no other significant motor impairment was noted. Speech therapy was initiated and overall his language improved, although remaining with significant word-finding difficulties and paraphasias. Substantial difficulties were present in discriminating antonyms, such as up-down, open-close, to go in-to out, before-after and over-below. In addition, he reported important impairments in understanding numbers and using numerical concepts (e.g. 'I do not understand numbers, particularly long numbers', 'For me the phone numbers are now a mess'; 'When somebody tells me about money, I do not understand well if it is too much or too little'). Despite these limitations he was able to return to his previous working activity, although requiring frequent assistance. He was aware and critical of his deficits. Brain MRI showed a small ischemic lesion involving the left angular gyrus (figure 1 and figure 2).

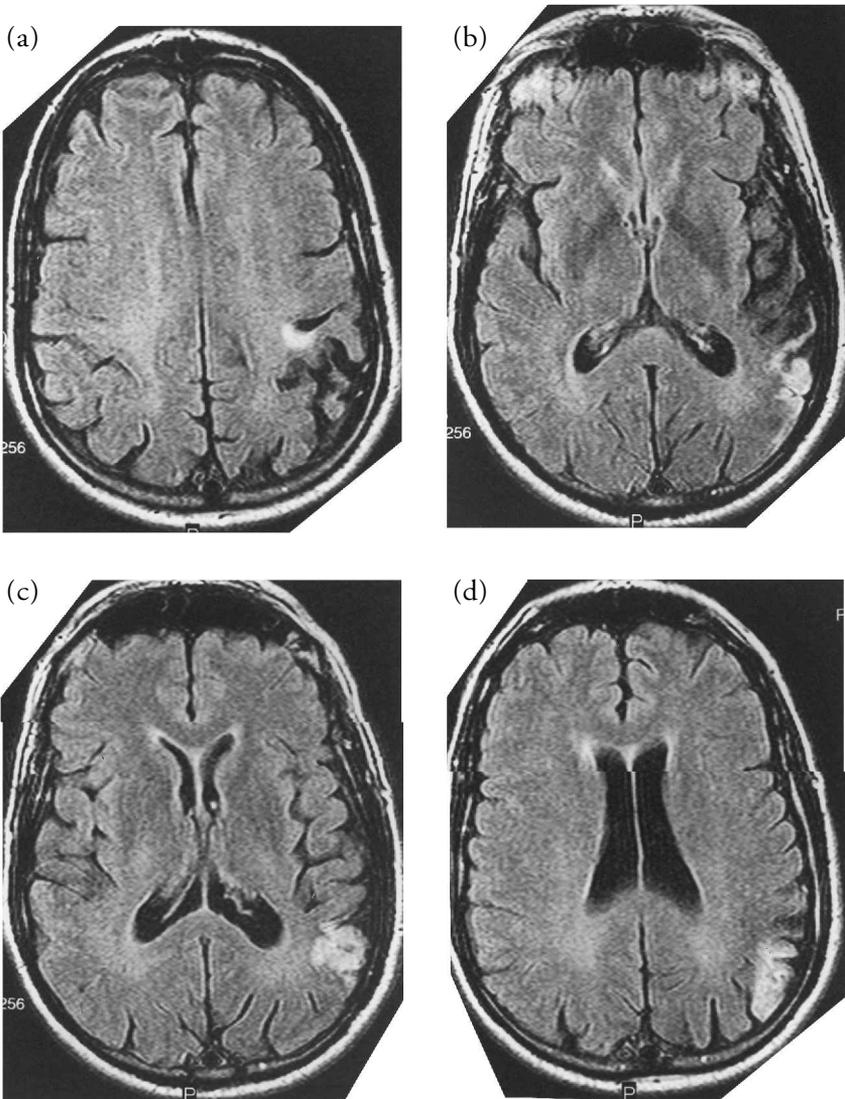


Figure 1. Axial view. Axial FLAIR (fluid attenuated inversion recovery images) sequences show gyriform increased signal along the angular gyrus.

Neuropsychological testing

Table 1 presents a summary of the tests administered to the patient. Most of them are common neuropsychological tests. In the stereognosis test five common objects placed on the hand (match, clip, coin, key, and pencil) were used. In the finger naming and recognition tests four measures were used: (a) naming fingers; (b) pointing to fingers named by the examiner; (c) pointing to own fingers shown in a model; and (d) moving the corresponding finger to the one touched in the opposite hand (right or left) by the examiner. For right-left orientation, three different measures were used: (a) one step command identification of right and left side body parts, e.g. 'show me your left ear'; (b) two step command identification

Table 1. Neuropsychological testing

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1. Multilingual Aphasia Examination (MAE) – Spanish version (Rey and Benton 1991).
 2. Boston Naming Test – Spanish version (Garcia-Albea *et al.* 1986).
 3. Spanish reading, writing and repetition tests (Ardila *et al.* 1994).
 4. Serial Verbal Learning (Ardila *et al.* 1994).
 5. WAIS-R – Arithmetic and Digits subtests (Wechsler 1974).
 6. Calculation Ability Test (Ardila *et al.* 1994).
 7. Stereognosis test.
 8. Finger naming and recognition.
 9. Right-left orientation.
 10. Rey-Osterrieth Complex Figure (Lezak 1995, Rey 1941) – copy and memory conditions.
 11. Praxis ability test. Performance of 10 movements by verbal command.
 12. Understanding of logic-grammatical relationships.
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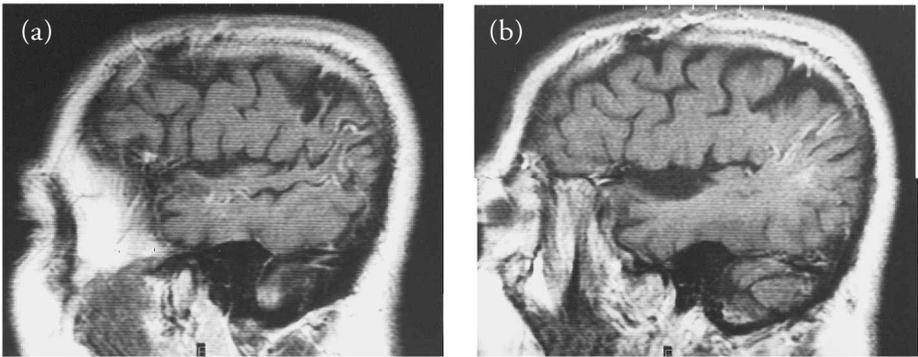


Figure 2. Sagittal view. T1 weighted sagittal images show increase signal in the area of the angular gyrus with mild linear extension of the abnormal signal posteriorly into parieto-occipital area. Abnormal increase T1 signal compatible with blood products and/or calcification.

of right and left side body parts, e.g. ‘with your right hand point to your left ear’; and (c) performance of ‘mirror movements’, i.e. the same movements, from those of the examiner’s sitting in front of the patient (Head’s test). For example, while sitting in front of the patient, if the examiner raised the right hand, the patient must also raise the right hand.

For testing the understanding of logic-grammatical relationships five different tasks were used: (a) Comparative sentences. Sixteen statements orally presented regarding the size of elephants and dogs were used. The patient had to answer ‘yes’ (correct) or ‘no’ (incorrect). The easiest statement was ‘Elephants are bigger than dogs’, the hardest one was ‘Dogs are not smaller than elephants’; (b) Reversible constructions of spatial type. In front of the patient a paper with the drawings of a triangle (above), a circle (middle) and a square (below) was presented. Eleven statements orally presented regarding the spatial location of three geometrical figures were used. For example, ‘The circle is above the triangle’ ‘The square is below the circle’. The patient was required to answer ‘right’ or ‘wrong’ to each statement; (c) Constructions with attributive relations. This task used 10 statements

orally presented regarding kinship relations, for example, 'My brother's father and father's brother is the same person', 'My son's father is my grandfather'; the patient had to answer 'yes' (correct) or 'no' (incorrect); (d) Passive sentences. Four passive sentences orally presented were utilized, for example, 'the sun is illuminated by the earth'; the patient had to answer 'yes' (correct) or 'no' (incorrect); and lastly (e) A series of four orally presented sentences with successive subordinate clauses were used; for example, 'the person who came with the man who had the book is the doctor. Who is the doctor'? The patient had to explain according to the sentence who is the doctor (e.g. 'a person coming with a man who was holding a book').

Results

Speech articulation, speed and voice volume were normal. During conversational speech the patient displayed frequent halting in search of a word that he eventually replaced with a description of the object or its function, circumlocutions. He also displayed semantic and literal paraphasias. No significant agrammatism or paragrammatism was appreciated. In the Controlled Word Association Test from the multilingual aphasia examination (COWAT) he produced a total 29 words using the letters P, T and M (10th percentile) and 17 animal names (25th percentile). His score in the Boston Naming Test (BNT) was below normal with 6 semantic and 12 phonological paraphasias. General results of the testing are presented in the appendix.

Comprehension for simple conversational language was essentially normal. However, he showed difficulties in understanding complex sentences, particularly if they included subordinate clauses. Auditory comprehension of words from the multilingual aphasia examination was normal, but he showed mild deficiencies in following two-step commands in the token test. Identification of body-parts was flawless. He displayed severe difficulties in understanding logic-grammatical relationships, comparison adverbs (e.g. bigger-smaller, younger-older, etc), place adverbs (e.g. over-below, on-beneath, etc) and time adverbs (e.g. before-after). He was unable to understand simple sentences such as 'Peter is older than John; who is younger?', or 'I read the newspaper after having breakfast; what did I do first?'. His overall performance in this tasks was at the chance level (50%).

Repetition of syllables, high frequency words and short sentences (≤ 7 words) was normal, but mild defects were observed in the repetition of long and infrequent words (e.g. *mecanografía*), and sentences with > 7 words. Semantic substitutions were noted occasionally. No errors in identifying written letters, words and sentences were observed. Reading comprehension from the multilingual aphasia examination was normal. Spontaneous writing (written description of plate #1 from the Boston diagnostic aphasia examination) showed a reduced number of nouns and one literal paraphasia in 18 words (the word *inclinada* was written as *inclinade*). Calligraphy was normal. Writing under dictation was normal for frequent words, but one literal paraphasia was observed in low frequency words (the word *dromedario* was written *dromenario*).

Digit span forward and backward score was 3 for each (1st percentile). In the serial verbal learning test using 10 high frequency Spanish words he recalled 3 words in the first trial (1st percentile) and 9 words in the tenth trial (below 20th percentile). Delayed recall was 9/10 (80th percentile). He performed poorly in the

Rey-Osterrieth complex figure-copy condition (5th percentile) and immediate recall (5th percentile). No neglect was observed and his copying strategy was random. Significant omissions in the right half of the figure were noted in recall task.

He correctly identified right and left in his own body with one step commands, but failed in following two-steps commands (e.g. right ear with the left hand). He was unable to mirror the movements performed by the examiner in the Head's test. Naming fingers was normal (e.g. 'What is the name of this finger'? while the examiner touched different patient's fingers), but overall errors were observed in showing fingers (e.g. 'Show me your ring finger'), pointing fingers from a model (e.g. 'point to my index finger', while the examiner presented his hand extended in front of the patient) and showing fingers in the apposite hand (e.g. the examiner touched one patient's finger and the patient was required to show the same finger in the opposite hand) in about 30% of the trials. Stereognosis was preserved. No difficulties in performing movements under verbal command were observed, although, occasionally he used his hand as the object, pointing to a mild ideomotor apraxia.

Significant difficulties were observed in the WAIS Arithmetic subtest. Forward counting was flawless, but he made one omission (1/10) when counting backwards. Reading numbers with three or less digits was normal. However he demonstrated inversions (4908 → 4098) and omissions (10003 → 1003) reading numbers with more than three digits. Writing numbers under dictation was normal. Transcoding from verbal to a numerical code and *vice versa*, was normal except for few literal paraphasias (homophone-orthographic and non-homophonic errors) (382 → *trescientos ochnta y dos*; *ochenta* was wrongly written as *ochnta*; 10 → *dies*, instead of *diez*; 8643 → *ocho mil seisciento curlida y tres*; but crosses out and correctly writes *ocho mil seiscientos cuarenta y tres*) and one decomposition error (10003 → *un mil cero tres*; one thousand zero three. He understood 'greater' and 'smaller' relations when comparing two quantities. Mental arithmetical operations were correct in adding or subtracting small quantities, but he confused adding and subtracting signs. Mental multiplications with two digit figures were impossible. Reading arithmetical signs was correct, except for adding instead of subtracting when performing written operations. Addition and subtraction of 3-digit quantities was normal, but multiplication and division were abnormal. Successive operations were correct, except a corrected error when subtracting 100 - 13. Aligning of numbers in columns during the mathematical operations was normal.

Discussion

Nearly two years after his cerebrovascular accident significant defects in language and calculation were observed. In summary, his neuropsychological testing disclosed naming defects, several paraphasias in conversational language, moderate constructional difficulties and mild ideomotor apraxia. Comprehension difficulties were observed in complex commands, particularly if including elements of a spatial nature and logic-grammatical relationships. Agraphia was minimal, but he had a conspicuous finger agnosia, right-left disorientation and acalculia. Interestingly, the patient could perform oral and written simple arithmetical operations, and solve elementary numerical problems. But he fails in more complex tests, and spontaneously insists in his significant calculation difficulties. According to

Dehaene (1997) and Dehaene and Cohen (1991) his calculation system for simple tasks (elementary calculation) is preserved, but complex calculation system is significantly impaired.

Gold *et al.* (1995) observed a patient with a left angular gyrus syndrome whose ability to name or point to lateralized body parts using verbal labels 'right' and 'left' was not defective, but whose performance was always poor when mental rotation to a command was required. The authors suggested that a defect in horizontal translation, i.e. mental rotation, accounted for the right-left disorientation in their patient. Furthermore, that acalculia and other signs associated with angular gyrus syndrome could also evolve from a deficit in the performance of these mental rotations. Similarly, this deficit in mental rotations could potentially be reflected in the impaired understanding of comparisons, e.g. time and place adverbs, found in semantic aphasia. One could infer that a single underlying deficit, defective mental rotations, may account for right-left disorientation, finger agnosia, acalculia and semantic aphasia, and that their simultaneous appearance in a single clinical syndrome is not coincidental. Notwithstanding, agraphia would still remain unexplained by this unifying underlying mechanism. It is of interest and perhaps not surprising that it is precisely agraphia that usually is missing in the angular gyrus syndrome. When agraphia is present it corresponds more to an apractic agraphia than to an aphasic agraphia, which correlates not exactly to the angular gyrus, but to inferior parietal area (Benson and Cummings 1985).

Right-left discrimination and finger gnosis are strongly interdependent and can even be interpreted as components of the autotopagnosia syndrome. Calculation ability might be interpreted as a type of cognition involving in their origins at least: some type of body knowledge (autotopognosia), spatial concepts and language. The association between spatial knowledge mediated through language and calculation abilities has been strongly emphasized by different authors (e.g. Ardila *et al.* 1989, Luria 1966, 1976). When calculation defects in patients with Broca and Wernicke aphasia were studied using factor analysis, Dahmen *et al.* (1982) were able to identify two different factors: numeric-symbolic and visual-spatial. Milder calculation defects found in Broca aphasia patients were derived from linguistic alterations, while visual-spatial processing defects significantly contribute to the calculation difficulties in Wernicke aphasia. Luria (1966, 1973) also emphasized the underlying spatial conceptualization deficits in the acalculia observed in left-parietal damaged patients and the strong association between acalculia and so-called semantic aphasia.

Some authors have proposed that finger agnosia represents a mild form of autotopagnosia (e.g. Hecaen and Albert 1978). However, dissociation between autotopagnosia and finger agnosia has been reported, and consequently it most likely represents different defects (De Renzi and Scotti 1970). Finger agnosia is a relatively frequent defect, whereas autotopagnosia represents a rather rare syndrome. Further, it has been proposed that finger agnosia might be a polymorphic phenomenon that includes apraxic, agnostic and aphasic aspects. In consequence, different subtypes of finger agnosia can be distinguished: visual finger agnosia, finger constructional apraxia, apractic defects in finger selection, and finger aphasia (anomia) (Schilder and Stengel 1931). The role of parietal lobe in body-knowledge and the disorders of the body scheme in cases of parietal pathology have been particularly emphasized in the literature (e.g. Botez 1985, Critchley 1953). Parietal damage has been associated with asomatognosia in

general, hemiasomatognosia, alloesthesia, finger agnosia, autotopagnosia, asymbolia for pain, apraxia, and the so-called Verger-Dejerine syndrome (Hecaen and Albert 1978).

Pathogenesis of right-left disorientation is not completely understood. Patients with left posterior damaged present more evident difficulties than right posterior damaged-patients (Ratcliff 1979). Right-left disorientation implies difficulties in the application of spatial concepts in the body's lateral orientation. Gold *et al.* (1995) proposed that a defective horizontal mapping could account for right-left confusion and the other components of Gerstmann syndrome as well, because they all share a common dependency on relative horizontal position.

Asymmetry in cerebral organization of cognition represents the most outstanding characteristic of the human brain. LeDoux (1982, 1984) proposed that the primary functional distinction between human hemispheres involves the differential representation of linguistic and spatial mechanisms: while the right posterior parietal lobe is involved in spatial processing, the left is involved in linguistic processing. Spatial mechanisms are represented in both the right and left parietal lobe in human ancestors including nonhuman primates, but in man language is represented in a region of the left hemisphere, the posterior parietal lobe, while spatial functions are represented in the corresponding right hemisphere (De Renzi 1982, Lynch 1980). In consequence, the evolution of language involved adaptations in the neural substrate of spatial behavior (LeDoux 1984). Boles (1991) presenting different tasks (recognition of words, products, locations, dichotic digits, etc.) and using a factor analysis was able to identify different lateralized parietal functions: lexical functions (e.g. word numbers) were associated with left hemisphere, whereas spatial functions (e.g. locations of dots) were correlated with right hemisphere activity.

In conclusion, there seems to exist a rationale for finding a common brain area and similar underlying mechanisms for acalculia, finger agnosia, right-left disorientation and semantic aphasia. A single impaired factor can account for the simultaneous presentation of these four clinical signs.

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Appendix

	Score	
<i>General neuropsychological testing</i>		
MAE: COWAT	34	20th percentile
MAE: Auditory Comprehension	18/18	normal
MAE: Token Test	36/44	3rd percentile
MAE: Repetition of sentences	5/14	2nd percentile
MAE: Reading comprehension	18/18	normal
Repetition high frequency words	7/7	normal
low frequency words	3/5	abnormal
Writing: Spontaneous: Picture description	17/18	mildly abnormal
By dictation: high frequency	10/10	normal
low frequency	6/7	mildly abnormal
Identification body parts	10/10	normal
Boston Naming Test	43/60	below 10th percentile
Serial Verbal Learning: 1st trial	3/10 words	10th percentile
10th trial	9/10 words	20th percentile
Delayed recall	9/10 words	80th percentile
Tactile recognition of objects	5/5	normal
Movements by verbal command	10/10	normal
Rey-Osterrieth Complex Figure: Copy	20/36	5th percentile
Recall	8.5/36	5th percentile

Right-left orientation		
Right-left in own body	5/5	normal
Two-steps commands	1/5	abnormal
Mirror movements (Head's test)	0/10	abnormal
Finger gnosis		
Naming fingers	5/5	normal
Pointing fingers	4/5	abnormal
Pointing fingers from a model	3/5	abnormal
Other hand fingers	3/5	abnormal
<i>Understanding of logic-grammatical relationships</i>		
Comparative sentences (e.g. 'Elephants are bigger than dogs')	8/16	abnormal
Reversible constructions of temporal and spatial type (e.g. 'The circle is over the triangle')	5/11	abnormal
Constructions with attributive relations (e.g. 'My brother's father and father's brother is the same person')	5/10	abnormal
Passive sentences (e.g. 'The sun is illuminated by the earth')	2/4	abnormal
Sentences with successive subordinate clauses (e.g. 'the person who came with the man who had the book is the doctor')	0/4	abnormal
<i>Calculation ability test</i>		
Counting: forwards	10/10	normal
backwards	9/10	abnormal
Reading number up to 3 digits	5/5	normal
more 3 digits	3/5	abnormal
Writing numbers	10/10	normal
'greater' and 'smaller' relations	5/5	normal
Transcoding: verbal to numerical	5/5	normal
numerical to verbal	3/5	abnormal
WAIS-R Digits	Scaled score = 4	abnormal
Arithmetic	Scaled score = 5	abnormal
Mental arithmetical operations		
Adding small quantities	5/5	normal
Subtracting small quantities	3/3	normal
Adding and subtracting large quantities	0/4	abnormal
multiplications (2 digits)	0/3	abnormal
Written arithmetical operations		
Adding and subtracting (3 digits)	4/4	normal
Multiplying and dividing (3 digits)	0/4	abnormal
Arithmetical signs: Reading	4/4	normal
Interpreting	3/4	abnormal
Successive operations: adding (1, 4, 7...)	10/10	normal
subtracting (100, 87, 74...)	4/5	abnormal
Aligning numbers in columns	10/10	normal