

Impairment of syntax and lexical semantics in a patient with bilateral paramedian thalamic infarction

Lieve De Witte^a, Ineke Wilssens^b, Sebastiaan Engelborghs^{c,d},
Peter P. De Deyn^{c,d}, Peter Mariën^{a,c,d,*}

^a *Department of Linguistics, Free University of Brussels, Belgium*

^b *Department of Speech Pathology, Middelheim General Hospital, Antwerp, Belgium*

^c *Department of Neurology, Middelheim General Hospital, Antwerp, Belgium*

^d *Laboratory of Neurochemistry and Behavior, Institute of Born-Bunge, University of Antwerp, Belgium*

Accepted 30 August 2005

Available online 13 October 2005

Abstract

Bilateral vascular thalamic lesions are rare. Although a variety of neurobehavioral manifestations have been described, the literature is less documented with regard to accompanying linguistic disturbances. This article presents an in-depth neurolinguistic analysis of the language symptoms of a patient who incurred bilateral paramedian ischemic damage of the thalamus. In the post-acute phase of the stroke, a unique combination of transcortical sensory aphasia with syntactic impairment was found. Because of this atypical semiological association, additional analyses of spontaneous speech were performed. In spite of the typological affinity with the grammatic characteristic of marked simplification of syntax observed in Broca's aphasia, only a wordclass specific, lexical-semantic deficit for verbs was objectified. The hypothesis that lexical-semantic disturbances in our patient might result from a functional deafferentiation of both thalami with the frontal lobe is supported by: (1) associated neuropsychological deficits of frontal origin and (2) frontal-like behavioral disturbances.

© 2005 Elsevier Inc. All rights reserved.

Keywords: Bithalamic stroke; Simplification of syntax; Transcortical sensory aphasia

1. Introduction

A variety of neurobehavioral symptoms have been reported in association with bilateral lesions of the thalamus: (1) transient vigilance disorders, (2) hypersomnia, (3) vertical gaze palsy, (4) persistent anterograde amnesia, (5) speech and language disturbances, and (6) apathy (Bewermeyer, Dreesbach, Rackl, Neveling, & Heiss, 1985; Bogousslavsky, Regli, Delaloye-bischof, Assal, & Uske, 1991; Dehaene, 1982; van Domburg, ten Donkelaar, & Notermans, 1996; Fukatsu, Fujii, Yamadori, Nagasawa, & Sakurai, 1997; Guberman & Stuss, 1983; Krolak-Salmon, Croisile, Houzard, Setiey, & Girard-Madoux, 2000; Reilly, Connolly, Stack, Martin, & Hutchinson, 1992). Although speech and language functions have not been systematically

investigated, dysarthric and aphasic disturbances have been reported after bithalamic lesions (Lhermitte, 1984). On the articulatory level, the deviant speech characteristics have been denoted as hypokinetic dysarthria (Ackermann, Ziegler, & Petersen, 1993; Benabdeljlil, El alaoui faris, Kissani, Aidi, & Laaouina, 2001; Bewermeyer et al., 1985; Dehaene, 1982; Gentilini, De Renzi, & Crisi, 1987; Gerber & Gudesblatt, 1986; Lazzarino & Nicolai, 1988; Malamut, Graff-Radford, Chawluk, Grossman, & Gur, 1992; Reilly et al., 1992). Reduced spontaneous speech and diminished verbal fluency have been described at the linguistic level (Bogousslavsky et al., 1991; van Domburg et al., 1996; Engelborghs, Marien, Pickut, Verstraeten, & De Deyn, 2000; Guberman & Stuss, 1983; Krolak-salmon et al., 2000; Krolak-Salmon, Montavont, Hermier, Millierey, & Vighetto, 2002; Malamut et al., 1992; Rousseaux et al., 1986; Schott, Maugiere, Laurent, Serclerat, & Fischer, 1980). In addition, some authors have reported naming

* Corresponding author.

E-mail address: petermarien@skynet.be (P. Mariën).

difficulties and mild auditory-verbal comprehension disturbances (Bogousslavsky et al., 1991; van Domburg et al., 1996; Engelborghs et al., 2000; Guberman & Stuss, 1983).

We report the findings of in-depth neurolinguistic investigations in a patient who incurred transcortical sensory aphasia (TSA) associated with marked simplification of syntax due to a bilateral paramedian thalamic infarction. The combination of fluent aphasia with syntax simplification is rather unexpected since this grammatical characteristic classically relates to non-fluent aphasia types. To the best of our knowledge simplification of syntax, characterized in our patient by simple sentences and sentence fragments with a complete absence of embedded clauses, has never been reported in association with transcortical sensory aphasia.

2. Case report

A 78-year-old right-handed woman was admitted to our hospital after acute loss of strength in the right arm, diplopia, and speech and language disturbances. Medical antecedents consisted of arterial hypertension and hypercholesterolemia. In addition to fluent aphasia, the clinical neurological examination on admission revealed vertical gaze palsy and restricted adduction of the left eye resulting in divergent strabism due to left oculomotor nerve paresis. Except for a mild paresis of the right arm, muscle strength and tone were completely normal. The finger-to-nose test showed dysmetria of the right arm, the knee-to-heel test was normal. Tendon reflexes at the right arm and leg were increased. Plantar responses were indifferent. Sensory and general physical examination revealed no abnormalities. Computerized tomography (CT) scan of the brain disclosed a bilateral paramedian thalamic infarction. Brain magnetic resonance imaging (MRI) demonstrated bilateral involvement of the medial thalamic nucleus (Engelborghs, Marien, Martin, & De Deyn, 1998; Kretschmann & Weinrich, 1992) as well as a small hyperintense area in the left internal capsula. Extension of the left thalamic lesion to the left red nucleus was shown as well (Fig. 1). A diagnosis of acute bithalamic stroke with extension to the mesencephalon was made. Vertebral artery dissection and significant stenoses were excluded by magnetic resonance angiography (MRA). Trans-oesophageal echocardiography was normal. Antiaggregant secondary preventive treatment as well as intensive physical and speech therapy were started.

After seven weeks the right hemiparesis had recovered completely. The vertical gaze palsy and oculomotor nerve paresis had only partially improved. The aphasia did not resolve.

2.1. Neuropsychological investigations

2.1.1. Methods

Language was assessed according to a time-frame model for the study of aphasia (Alexander, 1989; Mazzochi & Vignolo, 1979). In the post-acute phase of the stroke (lesion phase), extensive formal language investigations were per-

formed by means of standardized language tests, among which: the Aachen Aphasia Test (AAT) (Graetz, De Bleser, & Willmes, 1990), the Token Test (De Renzi & Vignolo, 1962), the Boston Naming Test (BNT) (Kaplan, Goodglass, & Weintraub, 1983; Marien, Mampaey, Vervaeke, Saerens, & De Deyn, 1998), and a semantic verbal fluency task (unpublished norms). Four weeks after the stroke (lesion phase) analysis of spontaneous speech and syntactic competence was performed by means of a method developed by Vermeulen, Bastiaanse, and Van wageningen (1989) and the Werkwoorden- en Zinnen Test (WEZT; Bastiaanse, Maas, & Rispen, 2000).

Cognitive functions were examined during the lesion phase one and a half month after stroke by means of the Mini Mental State Examination (MMSE) (Folstein, Folstein, & Mc Hugh, 1975), parts of the Alzheimer's Disease Assessment Scale (ADAS) (Rosen, Mohs, & Davis, 1984), the Coloured Progressive Matrices (Court and Raven, 1983), the Hierarchic Dementia Scale (HDS) (Cole & Dastoor, 1987), the Rey-Osterrieth Figure (Osterrieth, 1944), and the Judgment of Line Orientation test (JLO) (Benton, Hamsher, Varney, & Spreen, 1983).

2.1.2. Neurolinguistic results

As shown in Table 1, the AAT results for *language comprehension* were within the low normal range at both the auditory and written comprehension level. However, as shown by the subtest results, auditory sentence comprehension and written word comprehension were defective, whereas auditory word comprehension and written sentence comprehension were normal. *Oral language* was fluent and characterized by word-finding difficulties and semantic paraphasias. The spontaneous speech, characterized by simple sentences, sentence fragments with a complete absence of embedded, subordinate clause indicated a marked simplification of syntax (Fig. 2).

On the AAT confrontation naming tests a defective global score of 91/120 was found. The patient obtained a normal result for simple nouns. Naming of colors and compound nouns resulted in a deficient score. Most errors were of the semantic type: semantic paraphasias (e.g., screwdriver = hammer) and semantic neologisms (e.g., cleaner = suckmachine). Although the BNT-score was within the normal range, errors were of aphasic origin: perseverations, circumlocutions (e.g., camel = a sea... with two hunches), semantic paraphasias (e.g., octopus = seal; beaver = rat), and semantic neologisms (e.g., protractor = electric measure organ; hammock = lie folder). In a semantic word fluency task the patient produced a normal number of items. On the AAT sentence construction subtest, in which pictures have to be described depicting complex scenarios, she obtained a defective score. Repetition was normal.

Written language, as shown by the AAT results, was normal.

Summing-up, the aphasia profile of the patient corresponds to a taxonomic diagnosis of TSA: language comprehension was disturbed, speech was fluent and contained a

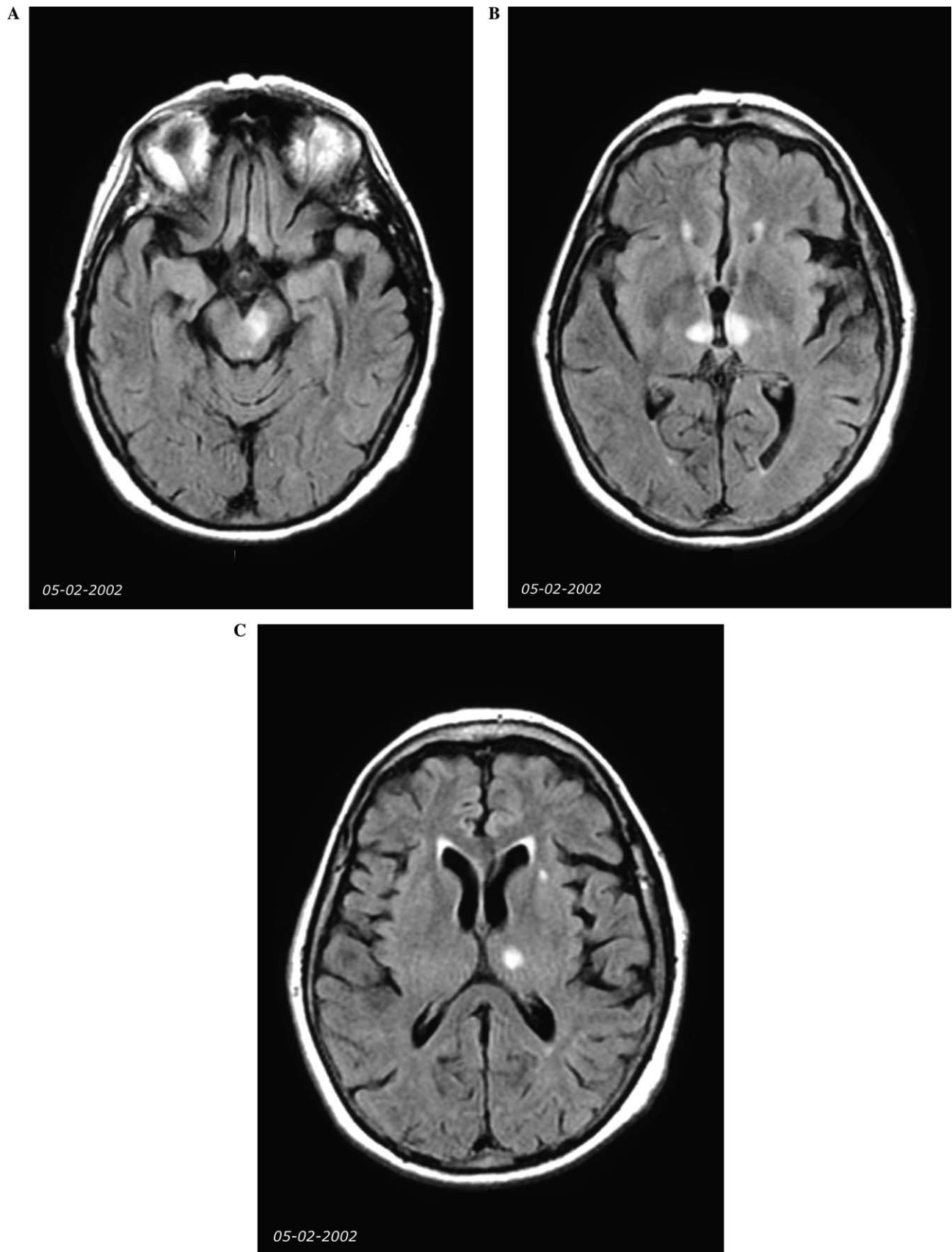


Fig. 1. Brain MRI axial FLAIR slices (A, B, C—from rostral to caudal) performed one month after stroke disclosing bilateral involvement of the medial thalamic nucleus, a small hyperintense area in the left internal capsule and extension of the left thalamic lesion to the left red nucleus.

Table 1
AAT, BNT and word fluency test results

	Score	Max	Mean	SD
<i>Akense Aphasia Test (AAT)</i>				
Language comprehension total	96	120	108.5	10.24
Auditory word comprehension	27	30	26.49	3.30
Auditory sentence comprehension	20	30	26.79	3.41
Total auditory comprehension	47	60	53.28	6.08
Reading word comprehension	21	30	28.30	2.29
Reading sentence comprehension	28	30	26.91	3.39
Total reading comprehension	49	60	55.21	4.90
Token Test	13 errors	0		
<i>Spontaneous Speech</i>				
Communicative behaviour	4	5	4.63	0.54
Articulation and Prosody	5	5	4.63	0.67
Automatisms	4	5	4.59	0.65
Semantic structure	4	5	4.59	0.53
Phonematic structure	5	5	4.54	0.56
Syntactic structure	2	5	4.41	0.55
<i>Imposed Speech</i>				
Repetition total	147	150	144.1	8.07
Phonemes	30	30	28.91	2.09
One-syllable words	30	30	29.22	1.32
Words of foreign origin	29	30	28.94	2.31
Compound words	29	30	28.45	2.22
Sentences	29	30	28.55	1.90
<i>Naming total</i>				
Simple nouns	28	30	27.92	2.90
Colors	23	30	27.69	1.99
Compound nouns	23	30	28.04	2.61
Sentence construction	17	30	25.69	3.72
<i>Written language total</i>				
Reading aloud	28	30	28.95	1.93
Composing on dictation	27	30	28.57	2.75
Writing on dictation	25	30	28	3.67
Boston Naming Test	44	60	49.1	6.76
<i>Verbal Fluency-Semantic</i>				
Animals	14	1 min		
Vegetables	8	1 min		
Clothing	9	1 min		
Means of transport	4	1 min		

high incidence of semantic errors but repetition was intact. In addition, spontaneous speech was characterized by marked simplification of syntax.

Since the association of TSA with simplification of syntax represents an atypical finding, grammatical competence was further investigated.

2.1.3. Additional language analyses

2.1.3.1. Spontaneous language analysis. A sample size of 318 words (Fig. 3) was randomly selected from an extensive corpus of recorded spontaneous speech and compared with the normative data obtained in healthy language users (Vermeulen & Bastiaanse, 1984). Four weeks after the stroke, the corpus was analyzed on both the morphosyntactic and lexical-semantic level.

On the morphosyntactic level, mean utterance length (MLU), number of modal verbs (may, must, can, will, ...) and copulae (be, become, seem, stay, ...), number of subordinate clauses, and the proportion of conjugated verbs were calculated. The number of utterances with conjugated verbs was divided by the number of expressions with one or more verbs. In the lexical-semantic analysis, the number of nouns and lexical verbs as well as the diversity of nouns and lexical verbs represent important variables. To assess lexical diversity, the type token ratio for nouns and lexical verbs was computed. In the type token ratio, which is always calculated on words belonging to the same specific word class (=homogeneous words), the total amount of words belonging to a specific word class (noun, lexical verb) is counted first (token). Then the different, diverse words belonging to that specific word class are counted (=type). Each word is defined as 'different,' 'diverse' when it occurs for the first time. Morphological forms (e.g., 'eating' or 'dogs') are not counted as diverse words when already used in another morphological form ('eat' or 'dog,' respectively). The number of types is then divided by the number of tokens to obtain the type token ratio. Semantic as well as phonological paraphasias were counted as well.

At the morphosyntactic level, analysis of the spontaneous speech sample revealed a mean utterance length (MLU) of 4.53 (245/54) words (normal mean: 8.22; range 5.7–10.7). Thirteen copulae and modal verbs were used (normal range: 4–18). Only one subordinate clause was produced (normal range: 3–15). The ratio of conjugated verbs was 0.63 (23/41) (normal range 0.83–1). At the lexical-semantic level the corpus only contained 34 nouns. The type token ratio for nouns was 0.65 (22/34) (normal mean: 0.75; range 0.54–0.88). Thirty-five lexical verbs were produced. The type token ratio for lexical verbs was 0.48 (17/34) (normal mean 0.74; range 0.63–0.86).

Quantitative analysis of a randomly selected spontaneous speech sample thus showed no marked differences between the number of nouns and the number of verbs. The

Examiner: How did you become ill?

JG: Firstly...free fallen. Then I have called my daughter, my son-in-law, here, my daughter-in-law works here on the ground floor, at the...general...yes I don't know anymore, José, and she called you...mother what's the matter? I come directly as I..., José, I don't know what happened, always turn, always fall... she drove me to hospital.

JG : That knocker... on the door. I think what's that now? Knocker not find... knowing word but not find...and here pain also...always not say. Continuation hospital... there was that large machine, how is it called... also not find... never went in... that was out of order... come to here to Middelheim and stayed here... because José here work.

Fig. 2. Structured conversation (literal English translation).

I am JG, live in the Bisschoppenhoflane..avenue. I was 19 January...90 January euh although ... I telephoned to José, who works here, on euh, the lowest, lowest level, second level, one level ...and who who says :/ mother what **is** the matter /...I come, when I finished working / I telephone to that man /...who **is** in leave euh çois....who **is** in euh in euh in retirement /...euh who telephones to our José. / José your mother telephones. / my mother is very sick /...euh I telephone back, / José comes...José comes than at four o'clock.../ who asks /: mum what **is** the matter? / I don't know what I have euh do I say /...I turn/and I always fall. / that **was** fridays. / she along to Middelheim euh middel... Middelares doing. / who **was** broken that that thing for measuring everything. / I doing to, I to middelheim, here middelheim done. / I know there on my left eye, / everything see double, / on my left eye do they euh ...euh suc a such a plaster, that protects. / I better see /...euh I up, to Middelheim nineteen hundred... euh twenty two./ I up,/ three days euh in Middelheim. / good much noise at night /...euh I back to Middelheim seven, five hundred fifty five./ I there very good./ in the morning self first are washed than self washing./ now with Fleur, a Dutchwoman, good washed,/ Fleur doing nothing,/ I everything do...euh..I everything do./ I go eat, /I eat,/ everything do, /everything eat, /everything spread,/ some bread with cheese some bread with honey,/ with honey is the last ...slice of bread,/ the coffee, 1 cup of coffee, otherwise two./ I must to the doctor, mister Peter. / quietly for hands (laughs). / that woman ask /: are you for a long time widow. / yes I am four, fives no 27 years widow./ my husband 27 years death. man /euh sad, he was 53 is died. / I euh 27 years alone. /I to the...country, /alone on journey,/ with another learn know,/ on journey go euh euh til seven euh two two years with Mariëtte (cries)./ Mariëtte died.

Legend: copula typographically marked in bold; nouns; marked in bold and underlined;/.../=utterance length

Fig. 3. Spontaneous speech sample (*literal English translation*).

diversity in the use of nouns was normal but clearly defective for lexical verbs. Mean utterance length and number of subordinate clauses were deficient.

2.1.3.2. Grammatical analysis. Comprehension and production of verbs and sentences were investigated by means of the WEZT (Table 2). This test consists of several tasks which allow identification and description of grammatical deficits. On the receptive level the test is composed of three subtests including verb comprehension, sentence comprehension, and grammaticality judgment; on the expressive level the test consists of seven subtests: action naming, sentence completion with infinitives, sentence completion with conjugated verbs, oral sentence construction, sentence anagrams, and interrogative sentence anagrams.

On the receptive level the '*verb comprehension task*' examines the ability to comprehend single verbs. In this task the subject is requested to depict one picture from an array of four that matches the verb orally presented by the examiner. The three distractors represent: (1) an object that shares a close semantic relationship with the target verb, (2) an action that is related to the target action and, (3) an object that is related to the action distractor. The 60 items are controlled for frequency, transitivity, and name relatedness with a noun (e.g., 'to hammer,' related to 'hammer' instead of 'to knock'). All these features are indicated on Table 2. A deficient result was found for this subtest (54/60). The majority of errors resulted from lexical-semantic problems. The errors consisted of selection of the semantic distractor of the target verb ($n = 3$) (e.g., target: 'to squeeze,' subject's answer: 'to sift' = a related action distractor with

no name relationship), selection of the distractor noun that closely relates to the target verb ($n = 2$) (e.g., target: 'to grate,' subject's answer: 'a grater' = a distractor with name relationship), and selection of the distractor noun semantically related to the action distractor ($n = 1$) (e.g., target: 'to conjure,' subject's answer: 'a magician' = a distractor with no name relationship to the target word). The '*sentence comprehension task*' contains stimulus sentences with moved constituents as in passives (theme, verb, agent) and sentences in which the order of the thematic roles is canonical as in actives and subject-cleft constructions (agent, verb, theme). These sentence types allow the examiner to investigate whether the order of the thematic roles has an impact on sentence comprehension. The procedure used is analogous to the '*verb comprehension task*': four pictures are presented to the subject while the examiner reads a sentence aloud. The subject is asked to single out the picture that matches the sentence. The distractors consist of: a picture in which the action and the participants are the same as the target picture but in which the thematic roles are reversed, a picture in which a different action is depicted and a picture in which both the thematic roles are reversed and in which the action is different. The result on this subtest was also deficient (34/40). Three errors consisted of role exchanges (e.g., target: 'the grandmother photographs the child,' subject's answer: 'the child photographs his grandmother') and another three errors due to selection of a semantic related action (e.g., target: 'mother pulls the child,' subject's answer: 'mother hits the child'). The '*grammaticality judgment task*' investigates whether the patient is able to parse a sentence. Only irreversible sentences are tested as they can

Table 2
WEZT lesion phase data

WEZT	Score	Max	<i>M</i>	<i>SD</i>	ASD	Cut-off
Verb comprehension	54	60	59.27	1.13	4.66	56
Transitive	28	31				
Name relationship	18	20				
High frequent	26	28				
Intransitive	26	29				
No name relationship	36	40				
Low frequent	28	32				
Grammaticality judgment	49	50	49.70	0.56	1.25	48
Grammatical sentences	24	25				
Non-grammatical sentences	25	25				
Sentence comprehension	34	40	39.77	0.42	13.7	39
Verb-agent-theme	9	10				
Agent-theme-verb	8	10				
Agent-verb-theme	8	10				
Theme-agent-verb	9	10				
Action naming	33	40	38.77	1.73	3.33	32
Transitive	17	20				
Intransitive	16	20				
Name relationship	17	20				
No name relationship	16	20				
High frequent	18	20				
Low frequent	14	20				
Filling in infinitive	8	10	9.57	0.50	3.14	9
Filling in conjugated verbs	9	10	9.65	0.48	1.35	9
Sentence construction	8	20	16.35	2.95	2.83	9
Transitive	4	10				
Intransitive	4	10				
Reversible	2	5				
Not reversible	2	5				
Sentence anagrams – picture	19	20	20	0		20
Active	9	10				
Passive	10	10				
Sentence anagrams + picture	20	20	20	0	0	20
wh-question anagrams + picture	10	20	19.75	0.78	12.5	18
Arguments Who/What	5	10				
Adjuncts	5	10				

be judged correctly without the need to map thematic roles onto the arguments if one has access to the grammatical structure. This subtest yielded a normal result (49/50).

In the ‘*action naming task*’ on the productive level, the patient is asked to name in one word what is happening on the picture shown to him. Forty items are presented that represent agentive verbs. The influence of several linguistic parameters (frequency, transitivity, and name relatedness with a noun) can also be determined. The patient obtained a defective score on this subtest (33/40). Most errors consisted of semantic paraphasias (e.g., target: ‘to play at draughts,’ subject’s answer: ‘to play chess’=an error against an intransitive, low-frequent verb with no name relationship; target: ‘to crawl,’ subject’s answer: ‘to search’=an error against a transitive, high-frequent verb with no name relationship) and the splitting of the target verb in a semantic related noun and verb (e.g., target: ‘to fence,’ subject’s answer: ‘to play with a sword’=an error

against an intransitive, low-frequent verb with no name relationship). In this task the parameter ‘frequency’ rather than ‘transitivity’ and ‘name relatedness with a noun’ seemed to determine the patient’s performance. She obtained 14/20 for low-frequent verbs and 18/20 for high-frequent verbs. The ‘*sentence completion task with infinitives or conjugated verbs*’ comprises two sets of sentences: 10 sentences that require completion with an infinitive verb and 10 sentences that require a finite verb. On the ‘sentence completion task with infinitives,’ our patient obtained a defective score (8/10). Only semantic errors occurred. In the ‘*sentence construction task*,’ the patient is instructed to describe a picture in one sentence. The 20 items that build up this test are controlled for three factors: frequency (all verbs are of high frequency), transitivity (10 transitive and 10 intransitive verbs), and reversibility of thematic roles (5 reversible and 5 irreversible sentences). On this subtest a score of 8/20 was found. Expressive agrammatic features (use of infinite verbs, omission of function words, stereotype sentence construction) and lexical-semantic deficits disrupted performance at the ‘sentence construction task’ (e.g., woman swim instead of the child swims, father falls instead of the baby crawls) (Fig. 4).

‘*The sentence anagram task*’ consists of three subparts. In the ‘*sentence anagram task with pictures*’ the subject is presented a picture and three cards, each of which has a sentence constituent printed on it. The subject is asked to sort these cards into a sentence that matches the picture. This subtest contains 20 items representing 10 reversible and 10 non-reversible sentences. The ‘*sentence anagram task without pictures*’ contains 20 irreversible sentences of which 10 are active and 10 are passive. In this task, three cards with constituents have to be arranged into a sentence. The ‘*interrogative sentence anagram task*’ includes 20 items: five items for each wh-question word (who, what, where and when). The procedure is similar to the sentence anagram task (Bastiaanse, Edwards, Maas, & Rispens, 2003). Only on the interrogative sentence anagram task the patient obtained a pathological score (10/20) as a result of role exchanges and incorrect word order. No difference was found between arguments (5/10) and adjuncts (5/10) (Fig. 5).

Summing-up, formal linguistic analysis of expressive and receptive syntax disclosed: (1) a lexical-semantic impairment at the level of the category of verbs, (2) a discrete mapping problem, and (3) a selective disturbance for interrogative sentence constructions.

2.1.4. Neuropsychological test results

One and a half month post-onset, cognitive functions were formally investigated. Manual preference was assessed by means of the Edinburgh Handedness Inventory (Oldfield, 1971) which revealed a laterality quotient of +100. Orientation, gnosis, praxis, problem solving, arithmetics, memory, and spatial intelligence were normal. On the Mini Mental State Examination the patient obtained a normal score of 26/30 (Crum, Anthony, Bassett, & Folstein, 1993). She obtained percentile 75 on the Raven Coloured Progressive

Target: 'The girl swings' → JG: 'Child in the pull';
 Target: 'The man drinks a glass of wine' → JG: 'Man drinks lemonade';
 Target: 'The boy hits the girl' → JG: 'Man gives wife a push';
 Target: 'The man runs' → JG: 'That man what just';
 Target: 'The boy catches the ball' → JG: 'That man stands in the goal catches the ball';
 Target: 'The boy pushes the girl' → JG: 'The right that pushes the footballer far site';
 Target: 'The farmer cycles' → JG: 'Cycles';
 Target: 'The man smokes a pipe' → JG: 'That man snores';
 Target: 'The child cries' → JG: 'That woman with aversion that cries';
 Target: 'The monkey eats a banana' → JG: 'The monkey sits to see';
 Target: 'The dog bites the cat' → JG: 'That dog noses at that cat';
 Target: 'The girl sleeps' → JG: 'Bear along in bed';
 Target: 'The man runs' → JG: 'That father runs';
 Target: 'The baby creeps' → JG: 'That father falls';
 Target: 'The man paints the woman' → JG: 'That father who paints that woman';
 Target: 'The girl applauds' → JG: 'That woman puts that hand away';
 Target: 'The girl throws the stick' → JG: 'That woman who let the butterfly up';
 Target: 'The clown laughs' → JG: 'That man in that man is clown';
 Target: 'The girl swims' → JG: 'That woman swim';
 Target: 'The child scratches the man' → JG: 'That man pull that hair'.

Fig. 4. Sentence construction (WEZT) (*literal English translation*).

Target: 'Where kisses the boy the girl?' → JG: 'the girl kisses where the boy';
 Target: 'When pushes the witch the magician?' → JG: 'the magician pushes the witch when';
 Target: 'Where draws the man the woman?' → JG: 'the woman when draws the man'.

Fig. 5. Wh-interrogative sentence constructions.

Matrices and normal results were found for 'word recall,' 'commands,' 'naming fingers and objects,' 'constructional praxis,' 'ideational praxis,' 'orientation,' and 'word recognition' of the Alzheimer's Disease Assessment Scale (Zec, Landreth, Vicari, Feldman, & Belman, 1992a, 1992b). No indications for a bucco-labio-lingual, dressing, visuo-constructive, ideational or ideomotor apraxia were found. Visuo-spatial processing as well as visual, tactile, and auditory recognition were normal. Normal results were found on the memory items of the Hierarchic Dementia Scale: 'registration,' 'recent memory' en 'remote memory.' Frontal dysfunctions were objectified on both the behavioral and psychometric level. As evidenced by a severely deficient score on the Stroop Color–Word test (<percentile 5) the ability to inhibit a competing and more automatic response set was severely disrupted. Deficient frontal planning and organisation were indicated by a severely defective result on the Wisconsin Card Sorting Test (zero categories in 128 trials). On the level of mood and behavior a variety of frontal-like disturbances were observed among which, disinhibitory verbal reactions, euphoric mood, loss of initiative, and apathy.

3. Discussion

In our patient, bilateral paramedian ischemic lesions of the thalamus induced a unique combination of cognitive dysfunctions. Indeed, standardized cognitive tests revealed a symptom complex that consisted of: (1) TSA, (2) marked simplification of syntax, and (3) dysexecutive

neuropsychological disturbances. To our knowledge, the combination of TSA with grammatical simplification following bilateral thalamic damage has never been reported before.

Comparison of the MRI images with an atlas of neuroimaging and neuroanatomy (Kretschmann & Weinrich, 1992) confirmed bilateral involvement of the thalamic medial nucleus. Although the presence of a right ataxic hemiparesis might suggest involvement of the ventral lateral nucleus, analysis of the neuroimaging data did not reveal structural damage of this nucleus. Alternatively, right ataxic hemiparesis as well as vertical gaze paralysis and left oculomotor nerve paresis can be attributed to the extension of the left thalamic lesion into the mesencephalon with involvement of the left red nucleus. As thalamoperforating arteries (terminal branches of the paramedian thalamic artery) usually supply the medial and paramedian segments of the thalamus (nuclei intralamellares and nucleus medialis) (Engelborghs et al., 1998), and given the small size of the intralaminar nuclei that certainly limits their identification on MRI images, we cannot rule out that the intralaminar nuclei are involved in the bilateral paramedian thalamic infarction.

Several neuropsychological studies have reported category-specific deficits of grammatical word classes but in none of them the causative lesion was situated in the thalamus (Bak, O'donovan, Xuereb, Boniface, & Hodges, 2001; Berndt, Mitchum, Haendiges, & Sandson, 1997; Breedin, Saffran, & Schwartz, 1998; Luzzatti, Raggi, Zonca, Pistarini, & Contardi, 2001, 2002). In agreement

with these observations, a disruption of verb comprehension, verb production, and a diversity in the use of verbs in spontaneous speech indicated in our patient a lexical-semantic impairment at the verb level. Although spontaneous language analysis was limited to only one speech sample, the omission of obligatory sentence parts, the omission of grammatical morphemes and articles, a decline of the number of conjugated verbs and a reduction of utterance length strikingly resembled the grammatical characteristics typically associated with Broca's aphasia (Schwartz, Saffran, Fink, Myers, & Martin, 1994). In spite of this syntactic deficit, normal results were found on sentence anagram tasks and sentence completion tasks (with conjugated verbs).

The dissociation between normal grammaticality judgment and defective sentence comprehension might in our patient be explained as a discrete mapping problem at the sentence level. Although the patient was able to adjust thematic roles on the basis of the syntactic structure of the sentence, she did not succeed to map these roles into grammatical sentences. In addition, simplification of syntax occurred in the entire subject's spoken language output where integration of lexical-semantic, syntactic, morphological, phonological, and articulatory information is required. As a consequence it might be hypothesized that this grammatic feature in our patient reflects a higher-level cognitive frontal disorder. As simplification of syntax in the absence of agrammatism has been noted with frontal lesions (Nadeau, 1988) but not with thalamic damage, we hypothesize that the syntactic deficit might result from a functional deafferentiation of both thalami with the frontal lobe leading to disruption of the thalamo-frontal circuit that subserves the integration of linguistic information, language planning, and organisation. As most paramedian thalamic artery infarctions involve the medial nucleus and the intralaminar nuclei, disconnection of both the medial nucleus or the centromedian nucleus with the prefrontal cortex might explain the symptoms our patient displayed (Nadeau & Crosson, 1997).

Reviewing the literature on the neurobehavioral consequences of bithalamic lesions since 1980 (Ackermann et al., 1993; Barbizet, Degos, Louarn, Nguyen, & Mas, 1981; Benabdeljlil et al., 2001; Bewermeyer et al., 1985; Bogousslavsky et al., 1991; Calabrese, Haupts, Markowitsch, & Gehlen, 1993; Charles & Fenichel, 1994; Dehaene, 1982; van Domburg et al., 1996; Engelborghs et al., 2000; Gentilini et al., 1987; Gerber & Gudesblatt, 1986; Guberman & Stuss, 1983; Hodges & McCarthy, 1993; Krolak-Salmon et al., 2002; Lazzarino & Nicolai, 1988; Malamut et al., 1992; Reilly et al., 1992; Rousseaux et al., 1986; Schott et al., 1980; Winocur, Oxbury, Roberts, Agnetti, & Davis, 1984) we only encountered 12 out of 41 patients with aphasia (Benabdeljlil et al., 2001; Bogousslavsky et al., 1991, cases 1–2; van Domburg et al., 1996; Engelborghs et al., 2000; Guberman & Stuss, 1983, cases 1–2; Hodges & McCarthy, 1993; Malamut et al., 1992; Rousseaux et al., 1986, cases 2–3; Schott et al., 1980). Seven of these patients

displayed reduced spontaneous speech output (Bogousslavsky et al., 1991, cases 1–2; van Domburg et al., 1996; Engelborghs et al., 2000; Hodges & McCarthy, 1993; Malamut et al., 1992; Schott et al., 1980).

In addition, in 17 out of 24 patients (71%) in whom it was looked for 'psychic akinesia' or a 'loss of psychic self-activation' (Bogousslavsky et al., 1991) was found. This syndrome is characterized by a complex of behavioral alterations such as apathy, asponaneity, indifference, motor inertia, and mental inertia (Ackermann et al., 1993; Bewermeyer et al., 1985, case 4; Bogousslavsky et al., 1991, cases 1–2; Dehaene, 1982, case 2; van Domburg et al., 1996; Engelborghs et al., 2000; Gentilini et al., 1987, cases 2,8; Gerber & Gudesblatt, 1986, case 1; Guberman & Stuss, 1983, case 1; Hodges & McCarthy, 1993; Malamut et al., 1992; Rousseaux et al., 1986, cases 1–3; Schott et al., 1980). In contrast to 'the akinetic state' these behavioral alterations are reversible at external stimulation. In our patient, similar behavioral changes were observed. The frontal-like behavioral disturbances among which abusive verbal reactions, euphoria, loss of initiative, and apathy may consequently be attributed to a functional deafferentiation of both thalami with the frontal lobe.

4. Conclusions

We presented a neuropsychological and neurolinguistic analysis of a unique patient with transcortical sensory aphasia and simplification of syntax due to a bilateral paramedian thalamic ischemic stroke. The hypothesis that the agrammatic symptoms reflecting an underlying lexical-semantic deficit might result from a functional deafferentiation of both thalami with the frontal region is supported by: (1) associated neuropsychological deficits of frontal origin and (2) frontal-like behavioral disturbances.

Acknowledgments

The authors thank the reviewers for their valuable comments that contributed to a significant improvement of the original manuscript. S.E. is a postdoctoral fellow of the Research Foundation—Flanders (F.W.O.-Vlaanderen).

References

- Ackermann, H., Ziegler, W., & Petersen, D. (1993). Dysarthria in bilateral thalamic infarction: A case study. *Journal of Neurology*, 240, 357–362.
- Alexander, M. P. (1989). Clinical-anatomical correlations of aphasia following predominant subcortical lesions. In F. Boller & J. Grafman (Eds.), *Handbook of neuropsychology*. Elsevier Science Publishers.
- Bak, T. H., O'donovan, D. G., Xuereb, J. H., Boniface, S., & Hodges, J. R. (2001). Selective impairment of verb processing associated with pathological changes in Brodmann areas 44 and 45 in the motor neurone disease-dementia-aphasia syndrome. *Brain*, 124(1), 103–120.
- Bastiaanse, R., Edwards, S., Maas, E., & Rispen, J. (2003). Assessing comprehension and production of verbs and sentences: The verb and sentence test (VAST). *Aphasiology*, 17(1), 49–73.

- Bastiaanse, R., Maas, E., & Rispens, J. (2000). *Werkwoorden-en zinnentest*. Lisse: Swets and Zeitlinger.
- Barbizet, J., Degos, J. D., Louarn, F., Nguyen, J. P., & Mas, J. L. (1981). Amnésie par lésion ischémique bi-thalamique. *Revista de Neurologia*, 137, 415–424.
- Benabdelljil, M., El alaoui faris, M., Kissani, N., Aidi, S., Laaouina, Z., et al. (2001). Troubles neuro-psychologiques après infarctus bi-thalamique par thrombose veineuse profonde. *Revue Neurologique*, 157, 62–67.
- Benton, A. L., Hamsher, K. deS., Varney, N. R., & Spreen, O. (1983). *Contributions to neuropsychological assessment. A clinical manual*. New York: Oxford University Press.
- Berndt, R. S., Mitchum, C. C., Haendiges, A. N., & Sandson, J. (1997). Verb retrieval in aphasia. Characterizing single word impairments. *Brain and Language*, 56, 68–106.
- Bewermeyer, H., Dreesbach, H. A., Rackl, A., Neveling, M., & Heiss, W. D. (1985). Presentation of bilateral thalamic infarction on CT, MRI and PET. *Neuroradiology*, 27, 414–419.
- Bogousslavsky, J., Regli, F., Delaloye-bischof, A., Assal, G., & Uske, A. (1991). Loss of psychic self-activation with bithalamic infarction: Neurobehavioral, CT, MRI and SPECT correlates. *Acta Neurologica Scandinavica*, 83, 309–316.
- Breedin, S., Saffran, E., & Schwartz, M. (1998). Semantic factors in verb retrieval: An effect of complexity. *Brain and language*, 63, 1–31.
- Calabrese, P., Haupts, M., Markowitsch, H. J., & Gehlen, W. (1993). The cognitive-mnestic performance profile of a patient with bilateral asymmetrical thalamic infarction. *The International Journal of Neuroscience*, 71, 101–106.
- Charles, P. D., & Fenichel, G. M. (1994). Sneddon and antiphospholipid antibody syndromes causing bilateral thalamic infarction. *Pediatric Neurology*, 10, 262–263.
- Cole, M. G., & Dastoor, D. (1987). A new hierarchic approach to the measurement of dementia. *Psychosomatics*, 28, 298–305.
- Crum, R. M., Anthony, I. C., Bassett, S. S., & Folstein, M. F. (1993). Population-based norms for the mini-mental state examination by age and educational level. *The Journal of the American Medical Association*, 18, 2386–2391.
- Dehaene, I. (1982). Bilateral thalamo-subthalamic infarction. *Acta Neurologica Belgica*, 82, 253–261.
- De Renzi, E., & Vignolo, L. A. (1962). The token test: A sensitive test to detect receptive disturbances in aphasics. *Brain*, 85, 665–678.
- van Domburg, P. H. M. F., ten Donkelaar, H. J., & Notermans, S. L. H. (1996). Akinetic mutism with bithalamic infarction. Neurophysiological correlates. *Journal of Neurological Sciences*, 38, 58–65.
- Engelborghs, S., Marien, P., Martin, J. J., & De Deyn, P. P. (1998). Functional anatomy, vascularisation and pathology of the human thalamus. *Acta Neurologica Belgica*, 98, 252–265.
- Engelborghs, S., Marien, P., Pickut, B. A., Verstraeten, S., & De Deyn, P. P. (2000). Loss of psychic self-activation after paramedian bithalamic infarction. *Stroke*, 31, 1762–1766.
- Folstein, M. S., Folstein, S. E., & Mc Hugh, P. R. (1975). Mini-mental state: A practical method for grading the cognitive status of patients for the clinician. *Journal of Psychiatric Research*, 12, 189–198.
- Fukatsu, R., Fujii, T., Yamadori, A., Nagasawa, H., & Sakurai, Y. (1997). Persisting childish behavior after bilateral thalamic infarcts. *European Neurology*, 37, 230–235.
- Gentilini, M., De Renzi, E., & Crisi, G. (1987). Bilateral paramedian thalamic artery infarcts: Report of eight cases. *Journal of Neurology, Neurosurgery, and Psychiatry*, 50, 900–909.
- Gerber, O., & Gudesblatt, M. (1986). Bilateral paramedian thalamic infarctions: A CT study. *Neuroradiology*, 28, 128–131.
- Graetz, P., De Bleser, R., & Willmes, K. (1990). *Akense Afasietest. Handleiding*. Lisse: Swets & Zeitlinger.
- Guberman, A., & Stuss, D. (1983). The syndrome of bilateral paramedian thalamic infarction. *Neurology*, 33, 540–546.
- Hodges, J. R., & McCarthy, R. A. (1993). Autobiographical amnesia resulting from bilateral paramedian thalamic infarction. *Brain*, 116, 921–940.
- Kaplan, E., Goodglass, H., & Weintraub, S. (1983). *Boston naming test*. USA: Lea & Febiger.
- Kretschmann, H.-J., & Weinrich, W. (1992). *Cranial neuroimaging and clinical neuroanatomy*. New York: Thieme Medical Publishers.
- Krolak-Salmon, P., Croisile, B., Houzard, C., Setiey, A., Girard-Madoux, P., et al. (2000). Total recovery after bilateral paramedian thalamic infarct. *European Neurology*, 44, 216–218.
- Krolak-Salmon, P., Montavont, A., Hermier, M., Milliery, M., & Vighetto, A. (2002). Thalamic venous infarction as a cause of subacute dementia. *Neurology*, 58, 1689–1691.
- Lazzarino, L. G., & Nicolai, A. (1988). Aponia as the only speech disturbance from bilateral paramedian thalamic infarction. *Clinical Neurology and Neurosurgery*, 90, 265–267.
- Lhermitte, F. (1984). Language disorders and their relationship to thalamic lesions. *Advances in Neurology*, 42, 99–113.
- Luzzatti, C., Raggi, R., Zonca, G., Pistarini, C., Contardi, A., et al. (2001). On the nature of the selective impairment of verb and noun retrieval. *Cortex*, 37(5), 724–726.
- Luzzatti, C., Raggi, R., Zonca, G., Pistarini, C., Contardi, A., et al. (2002). Verb–noun double dissociation in aphasic lexical impairments: The role of word frequency and imageability. *Brain and language*, 81, 432–444.
- Malamut, B. L., Graff-Radford, N., Chawluk, J., Grossman, R. I., & Gur, R. C. (1992). Memory in a case of bilateral thalamic infarction. *Neurology*, 42, 163–169.
- Marien, P., Mampaey, E., Vervae, A., Saerens, J., & De Deyn, P. P. (1998). Normative data for the Boston naming test in native Dutch-speaking Belgian elderly. *Brain and Language*, 65, 447–467.
- Mazzocchi, F., & Vignolo, L. A. (1979). Localisation of lesions in aphasia: Clinical CT-scan correlations in stroke patients. *Cortex*, 15, 627–654.
- Nadeau, S. E. (1988). Impaired grammar with normal fluency and phonology. Implications for Broca's aphasia. *Brain*, 111, 1111–1137.
- Nadeau, S. E., & Crosson, B. (1997). Subcortical aphasia. *Brain and Language*, 58, 355–402.
- Oldfield, R. C. (1971). The assessment and analysis of handedness: The Edinburgh inventory. *Neuropsychologia*, 9, 97–113.
- Osterrieth, P. A. (1944). Le test de copie d'une figure complexe: Contribution à l'étude de l'aperception en de la mémoire. *Archives of Neurology*, 30, 205–353.
- Raven, J. C., Court, J. H., & Raven, J., Manual for Raven's progressive matrices and vocabulary scales. 1983, London.
- Reilly, M., Connolly, S., Stack, J., Martin, E. A., & Hutchinson, M. (1992). Bilateral paramedian thalamic infarction: A distinct but poorly recognized stroke syndrome. *Quarterly Journal of Medicine*, 82, 63–70.
- Rosen, W. G., Mohs, R. C., & Davis, K. L. (1984). A new rating scale for Alzheimer's disease. *American Journal of Psychiatry*, 141, 1356–1364.
- Rousseaux, M., Cabaret, M., Lesoin, F., Devos, P., Dubois, F., & Petit, H. (1986). Bilan de l'amnésie des infarctus thalamiques restreints–6 cas. *Cortex*, 22, 213–228.
- Schott, B., Maudiere, F., Laurent, B., Serclerat, O., & Fischer, C. (1980). L'amnésie thalamique. *Revue Neurologique*, 136, 117–130.
- Schwartz, M. F., Saffran, E. M., Fink, R. B., Myers, J. L., & Martin, N. (1994). Mapping therapy: A treatment program for agrammatism. *Aphasiology*, 8, 19–54.
- Vermeulen, J., & Bastiaanse, R., 1984. *Stoornissen in de spontane taal bij afasiepatiënten: een factoranalytisch onderzoek*. Rapport voor Stichting Afasie Nederland.
- Vermeulen, J., Bastiaanse, R., & Van wageningen, B. (1989). Spontaneous speech in aphasia: A correlational study. *Brain and Language*, 36, 252–274.
- Winocur, G., Oxbury, S., Roberts, R., Agnetti, V., & Davis, C. (1984). Amnesia in a patient with bilateral lesions to the thalamus. *Neuropsychologia*, 2, 123–143.
- Zec, R., Landreth, E., Vicari, S., Feldman, E., Belman, J., et al. (1992a). Alzheimer disease assessment scale: Useful for both early detection and staging of dementia of the Alzheimer type. *Alzheimer Disease and Related Disorders an International Journal*, 6(2), 89–102.
- Zec, R., Landreth, E., Vicari, S., Feldman, E., Belman, J., et al. (1992b). Alzheimer disease assessment scale: A substent analysis. *Alzheimer Disease and Related Disorders, an International Journal*, 6(3), 164–181.