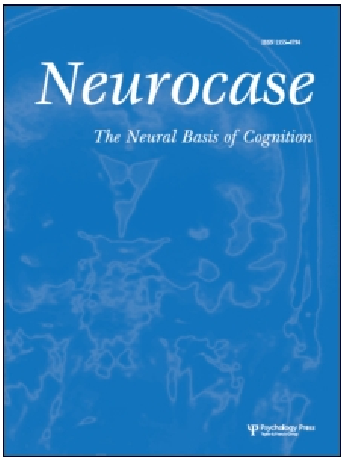

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Speaking without Broca's area after tumor resection

Monique Plaza [a]; Peggy Gatignol [a]; Marianne Leroy [a]; Hugues Duffau [a]
[a] Laboratoire de Psychologie et Neurosciences Cognitives (UMR CNRS 8189), Université Paris Descartes, Service de Neurochirurgie, CHU Gui de Chauliac, Montpellier, France

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Speaking without Broca's area after tumor resection

Monique Plaza, Peggy Gatignol, Marianne Leroy, and Hugues Duffau

Laboratoire de Psychologie et Neurosciences Cognitives (UMR CNRS 8189), Université Paris Descartes, Service de Neurochirurgie, CHU Gui de Chauliac, Montpellier, France

We present the case of a right-handed patient who received surgical treatment for a left frontal WHO grade II glioma invading the left inferior and middle frontal gyri, the head of the caudate nucleus, the anterior limb of the internal capsule and the anterior insula, in direct contact also with the anterior-superior part of the lentiform nucleus. The tumor resection was guided by direct electrical stimulation on brain areas, while the patient was awake. Adding a narrative production task to the neuropsychological assessment, we compared pre-, peri- and post-surgical language skills in order to analyze the effects of the tumor infiltration and the consequences of the left IFG resection, an area known to be involved in various language and cognitive processes. We showed that the tumor infiltration and its resection did not lead to the severe impairments predicted by the localization models assigning a significant role in language processing to the left frontal lobe, notably Broca's area. We showed that slow tumor evolution – the patient had been symptom-free for a long time – enabled compensatory mechanisms to process most language functions endangered by the tumor infiltration. However, a subtle fragility was observed in two language devices, i.e., reported speech and relative clauses, related to minor working memory deficits. This case study of a patient speaking without Broca's area illustrates the efficiency of brain plasticity, and shows the necessity to broaden pre-, peri-, post-surgery language and cognitive assessments.

Keywords: Language; Inferior frontal gyrus; Working memory; Plasticity; Tumor; Neurosurgery.

INTRODUCTION

Historically, language has been considered as localized in two major brain areas, Broca's anterior frontal area – for production – and Wernicke's posterior temporal area – for comprehension. The Lichtheim–Geshwind model (Geshwind, 1967; Lichtheim, 1885) was based on clinical information from aphasia consecutive to sudden trauma or vascular stroke, and anatomical information from post-mortem observations. As severe language impairments were observed in cases of temporal and left frontal lesions, especially when Broca's and Wernicke's areas were involved, the first functional model of language advocated a temporal frontal network. Today, the complex networks underlying language can be explored through neuroimaging techniques based on fMRI and diffusion tensor tractography, bringing support to less static and more connectionist approaches. Thus, areas originally thought to be specifically specialized for language have been shown to be involved in different cognitive and perceptual processing not necessarily related to language, and speech does not exclusively emanate from the Wernicke-Broca's language network (Démonet, Thierry, & Cardebat, 2005; Etard et al., 2000; Ffytche & Catani, 2005; Vigneau et al., 2006).

Neuropsychological studies on the consequences of acquired brain lesions in children and adults have suggested that the classical aphasia

Address correspondence to Monique Plaza, PhD, Laboratoire de Psychologie et Neurosciences Cognitives (UMR CNRS 8189), Université Paris Descartes, 71 Avenue Edouard Vaillant, 92773 Boulogne-Billancourt cedex, France. (E-mail: monique.plaza@univ-paris5.fr).

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model shows great variability in outcomes, according to age, etiology, lesion site, lesion size and initial impairment severity (e.g., Anderson, Morse, Catroppa, Haritou, & Rosenfeld, 2004; Anderson, Catroppa, Morse, Haritou, & Rosenfeld, 2005; Aram, 1999; Bates et al., 2001; Clark, Manes, Anotun, Sahabian, & Robbins, 2003; Coelho, Max, & Tranel, 2005; Damasio & Damasio, 1989). While sudden lesions affecting specialized areas often result in severe aphasia, the clinical pattern is different in the case of slow-growing lesions such as Low Grade Gliomas (LGG). Here slow tumor evolution allows for compensatory mechanisms to develop, with the brain recruiting other areas for the processing of the endangered functions, i.e., perilesional and/or controlateral homologous regions (Belin et al., 1996; Blasi et al., 2002; Crosson et al., 2005; Desmurget, Bonnetblanc, & Duffau, 2006; Léger et al., 2002; Musso et al., 1999). Neurosurgical resection under electrical stimulation of WHO grade II Glioma highlight such processes which can be directly observed within cortical and sub-cortical areas (Bonnetblanc, Desmurget, & Duffau, 2006; Duffau, 2005, 2006). This surgery technique adds useful data to those obtained through functional imaging techniques, enriching neuropsychological case and group studies (e.g., Plaza, Gatignol, Cohen, Berger, & Duffau, 2007).

In the case presented here, FV, a right-handed adult aged 27, received surgical treatment for a WHO grade II Glioma (LGG). The tumor invaded the left inferior and middle frontal gyri, the head of the caudate nucleus, the anterior limb of the internal capsule and the anterior insula. The tumor was also in direct contact with the anterior-superior part of the lentiform nucleus. The surgical resection concerned the pars triangularis, the pars orbitalis and the anterior part of the pars opercularis, i.e., Broca's area (see below for clinical details). The clinical and theoretical objective of this case study was to analyze the effects of the tumor infiltration and the consequences of the left inferior frontal gyrus resection since this area is known to be activated by various linguistic, cognitive and sensory-motor tasks (Bonnetblanc et al., 2006; Bookheimer, 2002; Devlin, Matthews, & Rushworth, 2003; Greewe et al., 2006; Grodzinsky, 2006; Heim et al., 2005; Koechlin & Jubault, 2006; Musso et al., 1999; Smith & Jonides, 1999; Tettamanti et al., 2002; Tettamanti & Wenninger, 2006; Voets et al., 2006). Research on brain-damaged adults and children has shown that narrative competence is particularly sensitive to disruptions in frontal attention processes, causing deficits in complex language assembly procedures and in executive function (e.g., Alexander, 2006; Bates et al., 2001; Chapman et al., 1992; Liles, 1993; Plaza, 1998; Plaza, Guitton, & Le Normand, 1998; Reilly, Bates, & Marchman, 1998; Reilly, Losh, Bellugi, & Wulfeck, 2004). A narrative production task was thus added to the classical neuropsychological assessment to better evaluate the patient's linguistic profile. Narratives provide a quasi-naturalistic measure of spontaneous language as well as a relevant context for comparing several aspects of discourse in typical and atypical populations. On the basis of longitudinal analysis of the patient's performances, we hypothesized that (a) the tumor infiltration may have been compensated for by plasticity mechanisms and (b) the resection of the left IFG resection would lead to subtle impairments, in narrative production more specifically. Finally, this case study raises the interesting question of how Broca's area can be 'replaced' by other structures for its cognitive and language processing skills.

METHODS

The patient

FV was a 27-year-old right-handed man working as a computer engineer. Right-handedness was documented using the Edinburgh inventory questionnaire (Oldfield, 1971). He had had word retrieval difficulties for 4–5 years, in marked increase for the past 2 years when he first consulted for generalized seizures. The neurological examination was normal, without either somosatosensory or motor deficit, but the MRI revealed a tumor invading the left frontal lobe. Though put on antiepileptic medication, his partial seizures and language difficulties intensified. The patient then accepted to undergo surgical resection with electrical stimulation technique while awake. Eleven months elapsed between the first seizure and the surgical tumor removal. After surgery, FV had 70 speech therapy sessions for 6 months, training working and episodic memory, verbal and non verbal reasoning, flexibility and inhibition mechanisms. Speech therapy began 1 month after surgery.

Procedure

Neuroimaging

Pre- and postoperative anatomical MRI were performed to establish the exact lesion location and the extent of the surgical cavity.

Surgical mapping

Intra-operative mapping was done under local anesthesia with direct electrical stimulation technique already described by the authors (Vigneau et al., 2006). Sensory-motor and language functions were assessed. First the patient was asked to count repetitively from 1 to 10 in order to identify the areas essential to speech production, namely those indicating complete anarthria when stimulated. Second, the DO 80 picture naming test (Metz-Lutz et al., 1991) was used to detect anomia and naming impairment. Each site was tested three times, three trials being enough to establish whether a cortical site is essential for a particular cognitive function (Ojemann, Ojemann, Lettich, & Berger, 1989). To avoid seizures, the same cortical site was never stimulated twice in a row. To ensure successful tumor removal while sparing functional areas, the limits of the resection were progressively set so as to preserve functional pathways in the immediate vicinity of the surgical cavity. Such a procedure minimizes residual morbidity while enhancing the quality of the resection — thus improving patient survival by minimizing the anaplastic transformation of WHO grade II glioma (Duffau, 2005).

Pre-operative neuropsychological assessment procedure

Ten days before surgery, FV was administered an extensive neuropsychological assessment including the following tests: *WAIS-R 7-SF* (Ward, 1990; Wechsler, 1981); *Gröber and Buschke* (Gröber, Buschke, Bang, & Dresner, 1988); *Rey–Osterreith complex figure* (Rey, 1959, 1970); *Boston Naming Test* (Kaplan, Goodglass, & Weintraub, 1983); *Verbal fluency* (Cardebat, Doyon, Puel, Goulet, & Joanette, 1990); *Chapman–Cook speed of reading test*; *Weintraub and Mesulam Cancellation Test* (Weintraub & Mesulam, 1985); *Geometric figures and cube of BEC* 96 (Signoret et al., 1988); *The Hooper Visual Orientation Test* (Hooper, 1958); *Stroop Test* [Modified version: Chatelois, Pineau, Belleville, & Peretz, 1993; Stroop, 1935); *Wisconsin Card Sorting Test* (Berg, 1948; Heaton, Chelure, Talley, Kay, & Curtiss, 2002); *Ruff Figural Fluency Test* (Ruff, Light, & Evans, 1987); *Corsi blocks* (Milner, 1971); *Ruff 2 and 7* (Ruff, 1992). The neuropsychological assessment systematically included three language tests (Boston Naming Test, Chapman–Cook Naming Test and Verbal fluency Test) allowing the neuropsychologist to correlate verbal and non verbal skills, i.e., 'left' and 'right' hemispheric processing.

Longitudinal language assessment procedure

Language processes were assessed on the day before and after surgery with the following tasks:

1. the standardized *Boston Diagnosis Aphasia Examination*, *BDAE* (Goodglass & Caplan, 1983), adapted in French by Mazaux and Orgogozo (1982), administered on the day before, as well as 5 days after surgery and 3 months later;
2. the written narrative task of the *BDAE*, 'Cookies', which in one minute requires the subject to write a short story from a picture representing a scene;
3. the Picture naming test *DO 80* (Metz-Lutz et al., 1991) consisting of 80 black and white drawings of objects controlled for frequency, familiarity, age of acquisition and level of education, administered on the day before surgery, during surgery, as well as 5 days after surgery and 3 months later;
4. the experimental narrative ask elicited by the wordless picture book *Frog, where are you?* (Mayer, 1969) used in previous studies by the authors (Plaza, 1998; Plaza et al., 1998), and administered on the day before surgery, 5 and 12 days after surgery as well as 3 months later. This storybook represents a boy and his dog looking for their frog which escaped during the night. While looking for the frog in the forest, the boy and his dog meet several animals that interfere with their search. In the end, the boy and his dog find the frog which is with a friend and several baby frogs. The story ends with the boy and his dog heading home with a baby frog as their new companion. As he looked through the pictures, the patient was asked to tell the story in his own way so that anybody listening to the registered story could understand it. The experimenter kept silent during

the session. All recordings were timed, registered and transcribed orthographically using the convention of the CHAT system to facilitate lexical analysis (different words used and type/token ratio) by the CLAN computer program (MacWhinney & Snow, 1991). For each transcript, as we have done in previous data, overall story length, lexical, syntactic, cohesion, descriptive coherence and evaluative devices were scored.

Overall story length was scored from the total number of propositions used by the narrator. A proposition corresponding to a single event is defined as composed of a verb and its arguments.

Lexical analysis represented the number of tokens, different words, and the ratio of nouns and verbs compared to the total number of words.

Syntactic analysis scored three types of clauses, i.e., **sequential** (e.g., 'the boy sleeps in the room'), **subordinate and coordinate** (e.g., 'the boy and the dog saw that the frog escaped', 'the dog was afraid because he saw the bees flying towards him', 'the boy is looking into the hole while the dog is smelling the flowers', and **relative clauses** (e.g., 'the boy's dog, who is looking at the tree, suddenly falls').

Cohesion referred to the 12 situations composing the story, i.e., (1) it is night time; (2) the boy and the dog are looking at the frog; (3) they are sleeping; (4) the frog is escaping; (5) they wake up and discover that the frog has escaped; (6) the dog is falling through the window; (7) they are looking for the frog in the forest; (8) they meet bees; (9) an owl; (10) a reindeer; (11) they discover the frog's family; and (12) they are heading home with a baby frog.

Descriptive coherence took into account the 65 events suggested by the pictures.

Evaluative devices were composed of (a) **reported speech** (indirect, e.g., 'he asks the dog to be quiet' and direct, e.g., 'he asks the dog "be quiet", please'), (b) negative qualifiers (e.g., 'he did not see the reindeer'), (c) causal connectors (since, because, as) and (d) frames of mind (desire, intention, perception, emotion and knowledge). We calculated the ratio of each device compared to the total number of propositions.

Besides, a qualitative analysis was conducted on errors or speech difficulties (e.g., paraphasia, incoherence or planning disorder) in the patient's and control group's productions.

Control group and statistical data

Since the narrative task is experimental and not standardized yet, the patient's narrative production was compared with that of 10 control subjects matched in age (26–29 years, mean age: 27; 3) and socio-professional status (post-graduate studies). The means and standard deviations were computed for each measure in the control group, serving as a basis to establish the patient's *z* scores.

RESULTS

Intraoperative functional data

After the incision of the dura matter and the identification of the tumor boundaries through ultrasonography, electrical functional mapping was performed on the awake patient. Several significant cortical sites were detected with stimulation (Figure 1) i.e., in 5, primary somatosensory finger area, inducing dysesthesia (retrocentral gyrus); in 12, primary motor face area, inducing facial movements (precentral gyrus); in 10 and 11, premotor ventral area, eliciting complete speech arrest (rolandic operculum); in 13, dorsal premotor cortex, eliciting semantic paraphasia. The inferior frontal gyrus was removed up to the anterior and middle part of the insula, which was also partly removed without impairing language processing (Figure 2). After the resection of the pars orbitalis (BA 47) and the pars triangularis (BA 45), the anterior half of the pars opercularis (BA 44) was also removed without eliciting any language disorders. The cortical limit for the resection was established according to the eloquent sites previously identified by the stimulation. Sub-cortically, after opening the frontal horn of the ventricle, a deep functional boundary was set by the head of the caudate nucleus, in 41, where the stimulation induced persevering responses. Next to this landmark, the pathway coming from the premotor ventral cortex, in 42 was detected with anarthria induced by the stimulation, and the resection went further, in the fibers composing the posterior wall of the surgical cavity. As to the deep postero-superior boundary of the resection, the limit was set at the inferior occipito-frontal fasciculus, connected to the dorso-lateral prefrontal cortex, in 40, where the stimulation elicited semantic paraphasia. The anterior frontopolar cortex was completely removed. Thus, the resection stopped within functional boundaries, both at cortical and sub-cortical levels.

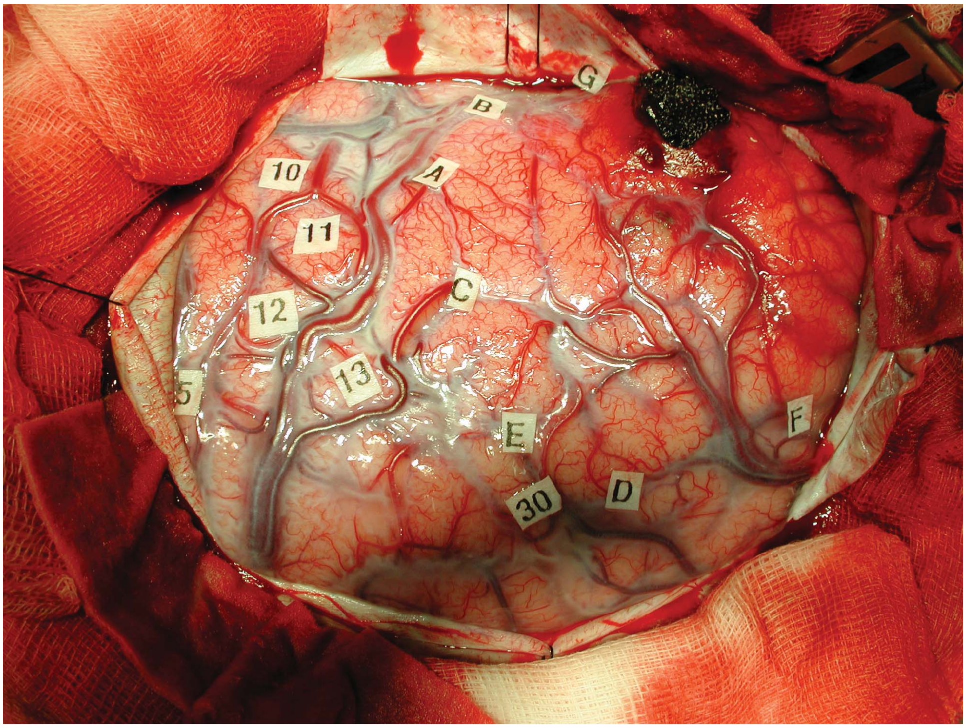

Figure 1. (Preoperative MRI) reveals a left precentral glioma, invading the left inferior and middle frontal gyri, the head of the caudate nucleus, the anterior limb of the internal capsule, the anterior insula and regions in contact with the anterior-superior part of the lentiform nucleus.

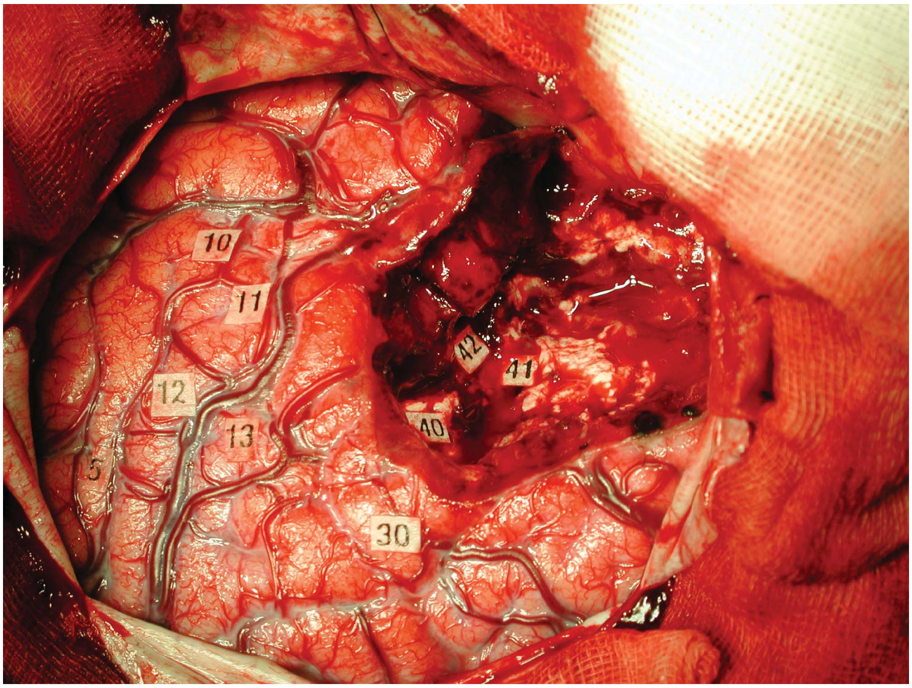

Figure 2. (At the beginning of the brain mapping) shows the sites directly involved in speech production as detected by electrical stimulation.

Pre- and post-neuropsychological data

Neuropsychological assessment before surgery (Table 1)

FV had a normal Verbal IQ (99) and a superior Performance IQ (130), which suggested a significant inter-hemispheric discrepancy, to the detriment of the left hemisphere (WAIS-R 7-SF). This pattern was coherent with the location of the damaged brain areas. However, only slightly low scores were noted in semantic memory (9 on 'information' subtest of WAIS-R), in verbal attentional/short-term memory and in working memory tasks (9 on 'digit span' subtest of WAIS-R). Moreover, verbal and visual abstraction were efficient, as well as processing time (13 on 'similarities' and 12 on 'picture completion' subtests of WAIS-R), planning and visuo-constructional skills (17 on 'block design' subtest of WAIS-R). Semantic controlled encoding, recognition and long-term retrieval of a word list (16 items) were normal, witnessing to good episodic verbal memory (16/16 on recognition of Gröber and Buschke test). However, some learning slowness (short-term retrieval limitation) on the last two immediate recalls was observed. The non verbal episodic memory tasks (Rey–Osterreith complex figure) showed good encoding and learning (35/36, although FV was not previously informed of the complex copied figure recall). These elements suggested a functional preservation of the bilateral frontal and temporal areas (Papez circuit) involved in episodic memory, except for slight frontal learning/retrieval difficulties.

Concerning language, oral understanding (oral orders) and written comprehension (Chapman–Cook speed of reading test) were efficient (12/12) and spontaneous speech was fluent and informative. However, internal lexical access was impaired, more in its left hemisphere component (literal verbal fluencies: 'P' letter: 11 items), than in its right counterpart (semantic verbal fluencies: 'Animals' category:

TABLE 1
Pre-surgical neuropsychological assessment

	NON VERBAL scores ("right" hemisphere)	*Neuropsychological tests*
IQ	130	WAIS-R 7-SF
Memory		
Semantic memory	12/19	Picture completion WAIS-R
Short term memory	6	Corsi block
Working memory	6	Corsi block (backward)
Episodic memory		
Encoding	35/36 (copy)	Rey Osterreith complex figure
Short term retrieval learning	31/36	
Long term retrieval	28/36	
Recognition	24/24	
Praxis		
Constructional praxis	35/36	Copy of Rey Osterreith complex figure with planning help
	17/19	Block design WAIS-R
Visuo-spatial abilities		
Cancellation		
Cancelled items	60/60	Weintraub and Mesulam
Strategy of exploration	non typical	
Visuo-construction	35/36	Copy of Rey Osterreith complex (planning help)
	12/12	Copy of BEC geometrical figures and cube
Lines bisection	right bias: 1 to 5 mm	
Organization	27/30	Hooper test
Gnosia	60/60	Object naming
"Frontal" functions		
Fluency	112 (perseverative behavior = 0)	Ruff figural fluency test
Visuo-motor planning	17/19	Block design WAIS-R
Abstraction	12/19	Picture completion WAIS-R
Working Memory	6	Corsi block (backward)

(*Continued*)

TABLE 1
(*Continued*)

	VERBAL scores ("left" hemisphere)	*Neuropsychological tests*
IQ	99	WAIS-R 7-SF
Memory		
Semantic memory	9/19	Information WAIS-R
Short term memory	9/19	Digit Span WAIS-R
	5	Digit Span
Working memory	9/19	Digit Span WAIS-R
	5	Digit Span (backward)
Episodic memory		
Encoding	16/16	Grober and Buschke
Short term retrieval learning	15 (11 + 4); 16 (12 + 4); 16 (13 + 3)/16	
Long term retrieval	16 (15 + 1)/16	
Recognition	16/16	
Language		
Lexical access		
Spontaneous speech	ok	
Naming	**42/60** (4 semantic paraphasias; 7 anomias; 4 circumlocutions; 3 delayed responses)	Boston Naming Test
Semantic fluency	21	Animal category
Literal fluency	11 (+2 perseverations)	"P" letter
Comprehension		
Oral understanding	ok	Orders
Written comprehension	12	Chapman-speed of reading test
Transcodage		
Word Reading	16/16	Self made lists
Non-word reading	6/6	
Word dictation score	**14/21**	
Word dictation phonological score	21/21	
Non-word dictation	6/6	
Non-word repetition	6/6	
Calculation	**9/19**	Arithmetic WAIS-R
	2/2	Writing calculation on failed
"Frontal" functions		
Inhibition	89″; no error	Stroop test
Flexibility	93″; no error	Stroop test
Fluency	21	Animals category
	11 (+2 perseverations)	"P" letter
Abstraction	13/19	Similarities WAIS-R
Working Memory	5	Digit Span (backward)

21 items). Lexical access assessment showed anomias, semantic paraphasias, circumlocutions and delayed responses (42/60 on the Boston Naming Test). Besides, FV reported lexical access troubles for 4 or 5 years previous to surgery, aggravated the past 2 years. A 'surface' dysgraphia during irregular and low frequency word dictation was also observed (14/21), whereas non-word reading and repetition were preserved. These results suggested a frontal inferior and temporal superior perturbation in the left hemisphere.

Calculation abilities were efficient but slow (9 on 'arithmetic' subtest of WAIS-R), due to verbal working memory limitation (backward digit span). FV operated on a visual mode (he used a visual diagram for solving mental arithmetic problems). Neither limb (significant, non significant, reflexive, non reflexive gestures), nor visuo-constructional (Rey–Osterreith complex figure copy with planning help), melokinetic (keyboarding) praxis troubles, pathological psychomotor slowness ('digit symbol' subtest of WAIS-R), were observed suggesting parietal, premotor and motor functional preservation. Despite an atypical visual exploration strategy (Weintraub and Mesulam Cancellation Test), and a discrete bias to the right (1–5 ml, i.e., non pathological) during line bisection, no neglect, agnosia, visual organization (Hooper Test), visuo-constructional or planning deficits (Rey complex figure) were observed. The neuro-visual assessment suggested functional right hemisphere (post-rolandic) preservation.

In summary, the impairment was not of a heavy quantitative nature but involved light deficits in verbal fluency ('P' letter and 'Animals' category), auditory attentional and verbal working memory ('digit span' *WAIS-R subtest*), especially during the mental arithmetic task (increased resolution times in 'arithmetic' *WAIS-R* subtest). All the other 'executive' skills were efficient (*Stroop test; Wisconsin Card Sorting test; Ruff Figural Fluency Test; 'Block design', 'Similarities', 'Pictures completion' subtests of WAIS-R; Backward Corsi blocks; Ruff 2 and 7*).

Neuropsychological assessment after surgery

Generally, apart from difficult clinical situations, the neuropsychological assessment only intervenes eight months to 1 year after surgery. FV had no complaint after surgery and was not preoccupied by his cognitive health.

TABLE 2
BDAE and DO 80

Tasks	*Day –1*	*Day +5*	*Day +3 months*
Picture pointing	72/72	72/72	72/72
Body parts	20/20	18/20	20/20
Auditory instructions	14/15	14/15	14/15
Reasoning	10/12	8/12	10/12
Articulation	7/7	5/7	7/7
Fluency	7/7	7/7	7/7
Sequence repetition	14/14	13/14	14/14
Series	9/9	9/9	9/9
Automatic recitation	2/2	2/2	2/2
Words	10/10	10/10	10/10
Concrete Sentence repetition	7/8	8/8	8/8
Abstract sentence repetition	8/8	6/8	7/8
Oral words	30/30	30/30	30/30
Sentence reading	10/10	10/10	10/10
Definition	30/30	30/30	30/30
Picture naming	105/105	105/105	105/105
Body parts	30/30	30/30	30/30
Aphasic phonemic	2	0	0
Jargon	0	0	0
Letter discrimination	10/10	10/10	10/10
Verbal recitation	8/8	8/8	8/8
Spelled words	8/8	8/8	8/8
Word/picture matching	10/10	10/10	10/10
Text reading	10/10	10/10	10/10
Writing	3/3	3/3	3/3
Automatic	46/46	46/46	46/46
Dictation	15/15	15/15	15/15
Spelling	10/10	9/10	10/10
Graphic evocation	10/10	8/10	9/10
Sentence spelling	12/12	9/12	12/12
Description	4/4	4/4	4/4
Music song	2/2	2/2	2/2
Rhythm	2/2	2/2	2/2
DO 80: score	77/80	80/80	80/80
DO 80: time	123 s	129 s	88 s

BDAE, DO 80 on Day –1, Day +5, Day +3 months

The comparative results (Table 2) showed that FV, whose language capacities were unequally efficient on the day before resection, had slightly lower results on Day +5, and improved 3 months after resection. Slight difficulties were observed on Day –1 in auditory instructions (93.3%), logical and reasoning (83.3%), concrete sentence repetition (87.7%) and picture naming (96.2%). On Day +5, slightly lower results were observed in 9 tasks, but lexical access was accelerated (tapped by the *DO 80* time). On Day +3 months, only 4 tasks showed below normal results, i.e., on auditory instructions (93.3%), logical and reasoning (83.3%), abstract sentence repetition (87.5%) and written picture naming (90%).

Written narrative 'Cookies' on Day –1, Day +5, Day +3 months

From Day –1 to Day +3 months, the results showed an overall improvement in all tasks (Table 3).

On Day –1, descriptive coherence was slightly defective (60%) with language errors such as persevering frozen forms, incapacity tokens ('I don't know'); incorrect syntactic forms erroneously using relative pronouns without any following proposition ('the woman who is washing the dishes [. . . *proposition*]') or using verb without subject ('[*subject*] looking outside'). FV's Handwriting was clumsy, with irregular letter drawing.

TABLE 3
Narrative written BDAE task elicited by the picture 'Cookies' in 1 min

Day –1	*Day +5*	*Day +3 months*
Je ne sais [1]pas ce que fait la femme (**I don't know** [1]what the woman is doing).	Ce sont deux petits **garçons** [1] (They are two little **boys**[1]).	Au moment où la mère faisait la vaisselle **sans attention** [1] (While the mother washed the dishes **without attention[1]**)
Le petit garçon attend **je sais pas** [1]quoi (The little boy waits **I don't know** [1]what).	Un prend un gâteau (One takes a cake).	En regardant par la fenêtre (Looking through the window).
Le tabouret est **en train de** [2]tomber (The stool is falling).	Il est en train de [2] donner à une petite fille (he is giving [2] a little girl).	L'eau de l'évier se mit à déborder (The water in the sink started to overflow).
Et la femme **qui**[3] est **en train de** [2]faire la vaisselle (And the woman who is washing the dishes).	Pendant ce temps, la mère **qui** [3]fait la vaisselle (In the meantime, the mother **who**[3] washes the dishes).	Pendant ce temps, son fils et sa fille essayaient de voler des gâteaux dans le placard du haut (In the meantime, her son and daughter tried to steal cakes in the top cupboard).
En regardant dehors (Looking outside).	Est en train de regarder dehors (Is looking outside).	Son fils avait dû monter sur une **chaise** [2] (her son had to stand on a **chair**[2])
Et **que**[3] son évier est **en train de** [2]déborder (And that her sink is overflowing).	Pendant que son évier déborde (While her sink overflows).	**Qui** [3] était en train de basculer (**that** [3] was falling over).
Narrative coherence: 6/10 [1] and [2] : perseverative forms. [1] : Incapacity tokens (« I don't know »). [3] syntactic erroneous forms with « qui » (who) and « que » (that) **Irregular handwriting**	**Narrative coherence**: 8/10 [1] semantic error : the characters are a boy and a girl. [2] pronoun omission (« le », it) [3] relative clause correctly written **Regular handwriting**	**Narrative coherence**: 10/10 [1] verb omission : « without **paying** attention » [2] semantic paraphasia : « chair » instead of « stool » [3] relative clause correctly written **Regular handwriting**

On Day +5, narrative coherence improved (80%) but the patient made one semantic error (mentioning two *boys* while the characters were a *boy and a girl*), he incorrectly used relative pronouns (e.g., 'In the meantime the mother who is washing the dishes [*lacking proposition*]') and he omitted one complement pronoun ('he is giving [*it*] to a little girl').

On Day +3 months, narrative coherence was perfect (100%) but the patient omitted one verb ('while the mother was washing the dishes without [*paying*] attention'), he also madeone semantic error ('chair' instead of 'stool'). His handwriting was harmonious.

ORAL NARRATIVE PRODUCTION

Global and lexical patterns of narrative production

The comparative intra-subject results (Table 4) showed (a) a stability of verbal fluency (words/sec), a decrease in discourse (number of tokens) and lexicon diversity (number of different words used by the patient) immediately after surgery and (b) a general improvement 3 months later. The noun and verb ratios remained stable. The inter-subject comparison showed that FV's results were within normal limits in all measures at each testing session.

Syntactic skills

The comparative intra-subject results (Table 5) showed (a) a stability of complex clause use, a decrease in discourse content (number of propositions), and number of sequential clauses and above all a significant decrease in the use of relative clauses, immediately after surgery and (b) a general improvement *except for relatives* 3 months later. The inter-subject comparison showed that FV's results were within normal limits in all measures at each session, *except for relative clause use,* where he scored significantly lower than the control group (z score = –1.37; p = .01).

Narrative cohesion and descriptive coherence

All situations were reliably accounted for by FV at any time, and by the control group as well (Table 6). Similarly, concerning descriptive coherence, FV recounted most events suggested by the pictures. Like most subjects in the control group, he showed descriptive coherence but for five details systemically omitted at each session. But since those five details were also omitted by most subjects in the control group, FV did not differ on this point.

TABLE 4
Global and lexical pattern of narrative production in FV and the control group

	Day −1	*Day +5*	*Day +12*	*Day +3 months*	*Control subjects Mean and SD*
Fluency	2.6 words/s	2.5 words/s	2.3 words/s	2.9 words/s	1.8 (0.9)
Tokens	507	400	447	507	655.1 (308.7)
Different words	205	165	162	193	238.6 (98.1)
Type Token Ratio	0,40	0.41	0.36	0.38	0.37 (0.03)
Different nouns	39	36	35	39	47.9 (19.5)
Ratio nouns	19%	21.8%	21.6%	20.2%	20.4% (2.3%)
Different verbs	58	48	47	56	76.8 (36.6)
Ratio verbs	28.2%	29%	29%	29%	31.5% (2.8%)

TABLE 5
Syntactic skills in FV and the control group

Type of clause	*Day −1*	*Day +5*	*Day +12*	*Day +3 months*	*Control subjects*
Number of propositions	61	44	47	63	85.7 (47.9)
Sequential clauses	31	21	22	34	45.1 (24.8)
Complex clauses	30	31	25	30	40.6 (24.3)
Relative clauses	8	1	1	0	10.2 (6.7)
Ratio sequential clauses	50.8%	50%	46.8%	53.1%	53.7% (7.8%)
Ratio complex clauses	49.2%	50%	53.2%	46.2%	46.3% (7.8%)

TABLE 6
Narrative cohesion and descriptive coherence in FV and the control group

	Day −1	*Day +5*	*Day +12*	*Day +3 months*	*Control subjects*
Spoken elements	11/12	12/12	12/12	12/12	11.8 (0.6)
Spoken events	48/65	45/65	50/65	49/65	51.8 (7.3)

Evaluative devices

The evaluative analysis (Table 7) highlighted FV's *poor use of reported speech (direct and indirect discourse) before and after surgery;* his scores were significantly lower than those of the control group (z scores = −1.22, −1.32, −1.12; p = .01). In contrast, he used more negative qualifiers after surgery and produced a great number of tokens about mental states, scoring relatively better than the control group at each session.

Pattern of narrative errors

FV's semantic and phonological errors were not relevant, since he corrected them, as did subjects in the control group. By contrast, his planning was more erratic, leading to a minor narrative incoherence due to word substitutions.

TABLE 7
Evaluative elements in FV and the control group

	Day −1	*Day +5*	*Day +12*	*Day +3 months*	*Control subjects*
Direct or indirect discourse	1	0	2	1	13 (9.8)
Ratio	1.6%		4.2%	1.5%	15.1%
Negative qualifiers	5	3	5	10	6.3 (4.2)
Ratio	8%	6.8%	10.6%	15.8%	7.3%
Causality	1	0	0	1	2.4 (2.4)
Mental states	20	16	16	21	15.3 (10.8)
Ratio	32.7%	40%	34%	32.8%	17.8%

DISCUSSION

In the present paper, a detailed evaluation of the patient's pre- and post-surgical profile was enriched by narrative production data not regularly provided by classical neuropsychological assessment. On the basis of a longitudinal analysis of the patient's performances, we originally hypothesized that (a) the tumor infiltration may have led to impairments partially or completely compensated for by plasticity mechanisms and (b) the left IFG resection would lead to subtle impairments in narrative production, more specifically.

Before resection: tumor consequences

Before surgery, FV displayed minor neuropsychological deficits, as shown by PIQ/VIQ discrepancy with low scores in verbal fluency, learning/ retrieval difficulties, verbal working memory limitation (*digit span*) and lexical access trouble (*Boston Naming Test*: anomias, semantic paraphasias, circumlocutions, delayed responses). Just below average results in *DO 80* (96.2%) were observed as well as in three *BDAE* tasks requiring auditory working memory, i.e., concrete sentence repetition (87.5%), auditory instructions (93.3%), logical and reasoning (83.3%). *In narrative production*, five lexical disturbances were observed, i.e., one semantic paraphasia, three preposition substitutions and one phonological error. The observed lexical disturbances could have been expected, since the left inferior frontal gyrus is specifically involved in individual word processing (Fiebach & Friederici, 2004). However, the patient displayed efficient verbal speed in spite of slightly weaker lexical access. He also correctly processed all aspects of narrative, notably recruiting language assembly procedures such as syntactic strategies and lexical repertoire, respecting script cohesion (the 12 situations composing the story) and descriptive coherence (picture/speech matching), using evaluative devices – notably negative qualifiers (which introduce the virtual axis) and mental states, as described by Theory of Mind. In contrast, the *BDAE* written narrative task (elicited by the single picture 'Cookies' under time constraint) highlighted difficulties in descriptive coherence, syntactic planning and handwriting. Thus, the complex coordination of visual picture analysis, lexical/syntactic retrieval and handwriting was impaired.

As shown in Figures 3 and 4, the surgical resection was very large, close to removing the left inferior frontal gyrus, which was completely 'silent' for naming during stimulation. The sites directly involved in speech naming production were the premotor ventral area (complete speech arrest), the dorsal premotor cortex (semantic paraphasia), the head of the caudate nucleus (persevering responses), the premotor ventral cortex (anarthria), the inferior occipito-frontal fasciculus, connected to the dorso-lateral prefrontal cortex (semantic paraphasia). Thus, before resection, plasticity mechanisms recruiting adjacent and in some cases controlateral regions (Knecht et al., 2000) allowed the brain to compensate for the left inferior frontal gyrus incapacitation due to tumour growth.

Compensatory mechanisms: their efficiency and limits

After the tumor resection which preserved regions directly involved in speech naming production, the patient quickly retrieved good language abilities. On Day +5, some *BDAE* scores were slightly below normal limits (due to surgical after-effects, as observed in all patients) but most scores were average 3 months later. The naming speed increase observed on Day +3 months in the *DO 80* picture naming test was parallel to the speech acceleration observed in the narrative task from Day –1 to Day +3 months. Speech acceleration could be partially attributed to retest effect, though its amplitude suggests a motor and phonological planning improvement following tumor removal, speech-therapy and brain decompression, linked to the arrest of epilepsy episodes. The patient experienced generalized seizures 11 months before surgery and despite anti-epileptic drugs, namely *Valproat* and *Gabapentin*, partial seizures intensified, accompanied with language difficulties. After surgical resection, epilepsy stopped, which led to a reduce prescription of anti-epileptic drugs.

The narrative task longitudinal analysis highlighted the patient's stable syntactic competence illustrated by the number of correct complex utterances, the preservation of verbs as action markers between pre- and post-assessments. Lexical abilities were shown to improve after a period of impoverishment (–20% at Day +5 and Day + 12), and overall story length increased after a fall at Day +5 and Day + 12. The patient also preserved script cohesion

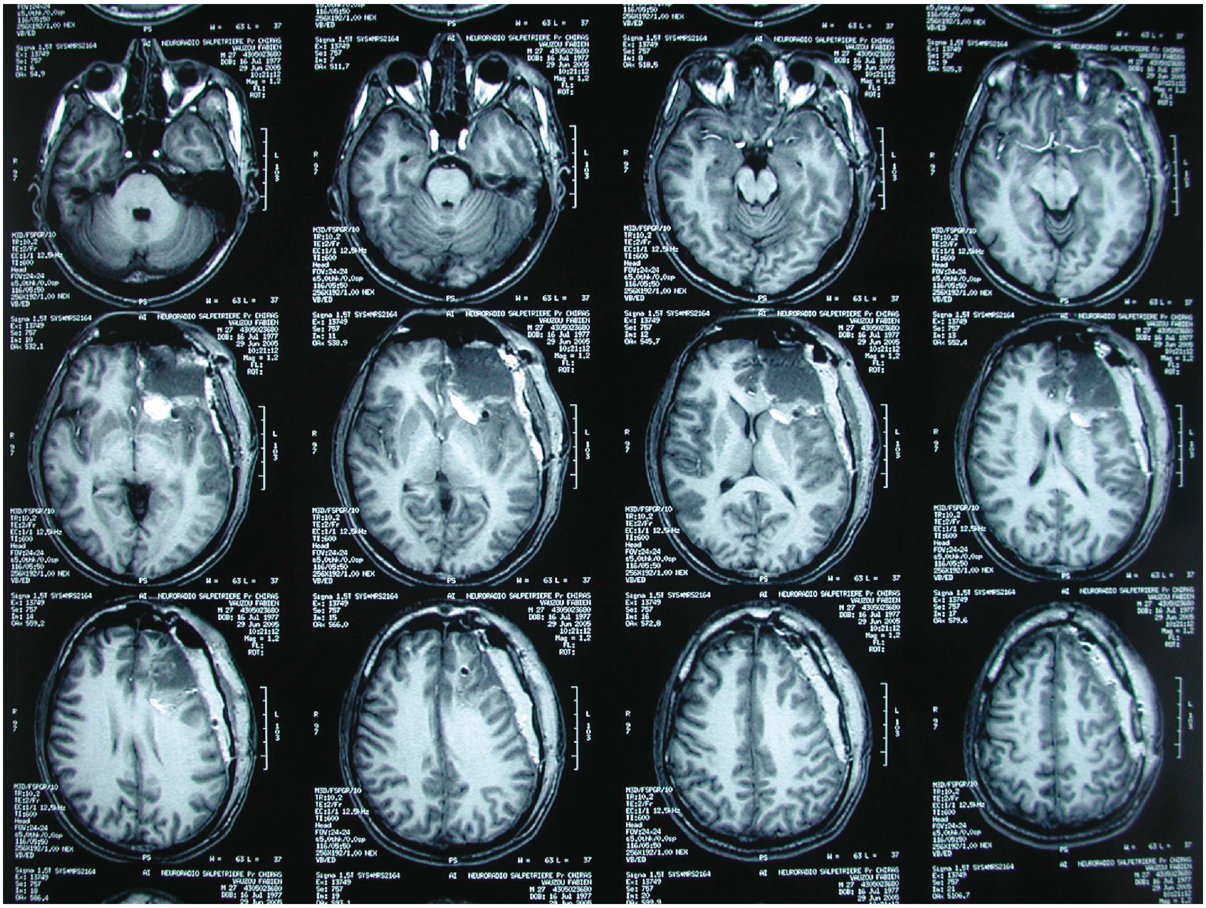

Figure 3. Post resection figure shows that the resection concerned the left frontal lobe, notably the pars triangularis, the pars orbitalis and the anterior part of the pars opercularis.

and descriptive coherence, efficiently using negative qualifiers and evoking mental states. Moreover, the narrative evaluative assessment showed that FV was able to express his own point of view, introduce emotional factors, specify virtual and potential features and then give a personal meaning to the story (Bamberg & Damrad-Frye, 1991). In the written narrative task, descriptive coherence and syntactic strategy, which were defective before resection, dramatically improved afterwards.

However, the patient's narrative production remained impaired in subtle ways. *At the syntactic level, FV partially 'lost' the use of the relative pronominal strategy which allows speakers to produce complex sentences including more than two subject sites* (e.g., 'The boy's dog, which has put its head into the box, goes to the window and falls outside'). This relative clause deficit suggested a formal linguistic vulnerability which could be explained by the resource limitation hypothesis.

At Day +5 and Day +3 months, FV had just below average results in BDAE tasks requiring working memory involvement, i.e., auditory instructions (93.3% at both evaluations), abstract sentence repetition (respectively, 75 and 87.5%), logics and reasoning (respectively, 66.6 and 83.3%). In parallel, during narrative production, he tended to avoid sentences including various subject sites. The left IFG, specifically the upper part of BA 44, is known to be critically involved in verbal-auditory working memory (Paulesu et al., 1993; Wallentin, Ropstorff, Glover, & Burgess, 2006). Length and complexity of relative clauses increase demands on syntactic production. The speaker's performance is determined by the fact that language processing is a sequential process in time. It depends on the distance between an element original position in the syntactic hierarchy and its actual position in the sentence. Once a word has been produced as a moved element, the speaker has to keep this

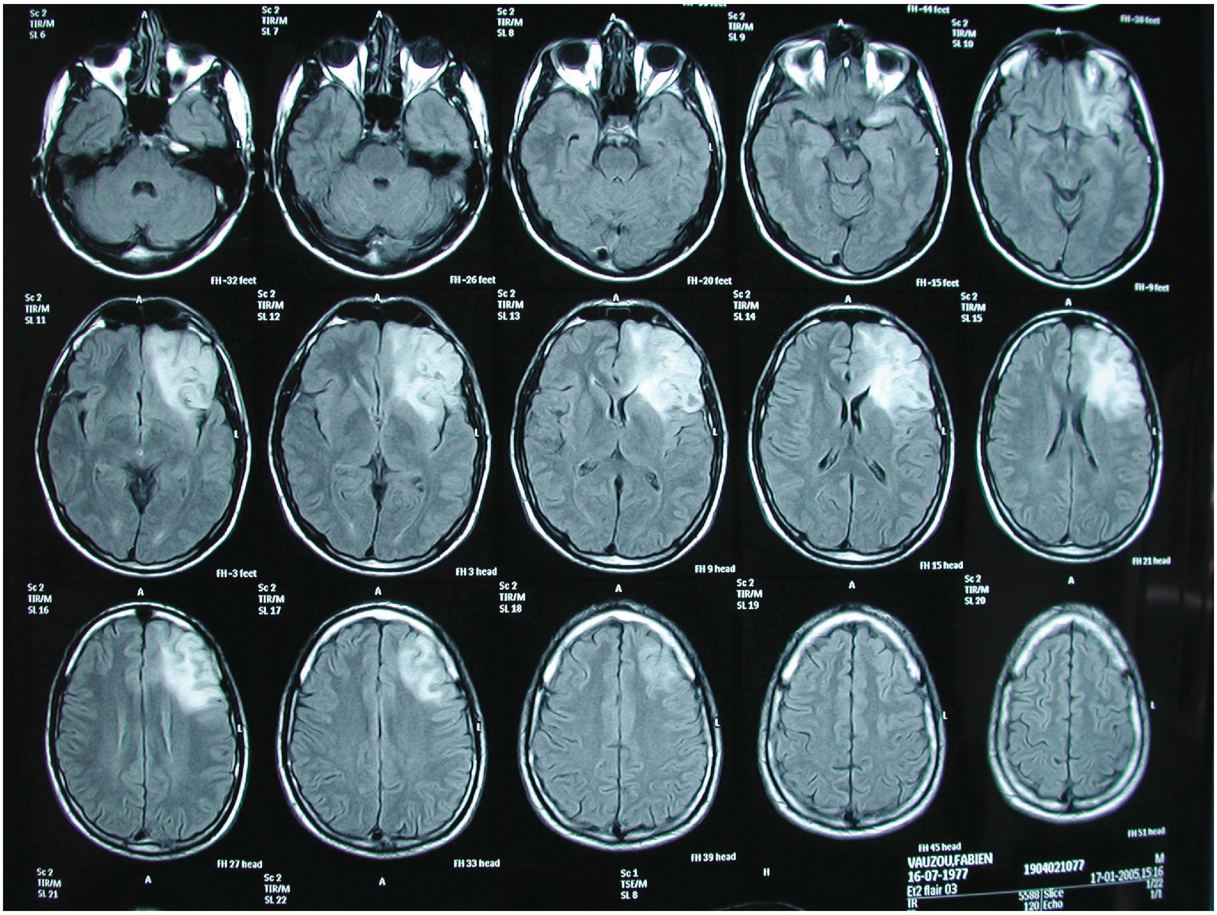

Figure 4. Post resection MRI shows that the resection concerned the left frontal lobe, notably the pars triangularis, the pars orbitalis and the anterior part of the pars opercularis.

element active in the working memory specialized for syntactic features. Processing demands increase as a function of the distance between non-local dependencies. The core region of BA 44/45 apparently supporting these processes in syntactic parsing (Friederici, 2006), seems to be involved in the same way in syntactic production too. In the written narrative task, difficulties with relative and embedded clauses were observed on Day −1, and disappeared on Day +5 and Day +3 months. The task undoubtedly increased the difficulty involved in the visual processing of one single picture, the handwriting and the overall sentence planning under time constraint.

Along with impaired lexical access (Faidiga & Craighero, 2006), Broca's aphasics show disturbed syntactic production, with grammatically simplified speech, missing function words and morphemes (Caplan, 2006). Globally this was not FV's case. *However, in the written narrative task, he omitted some words and in the oral task, he did not use many causal conjunctions,* although he produced many complex clauses. He only used one causal conjunction before surgery, and another 3 months later. Conjunctions are grammatical function words that underlie inter-sentential connections. They are complex tools, for they do not have a referent in an extra-linguistic context and are used for both semantic and pragmatic functions (Bloom, Lahey, Hood., Lifter, & Fiess, 1980). Developmental studies showed that young children initially join sentences by juxtaposing them without conjunction (Miller, 1981) since for children, conjunctions function as index items without autonomous meaning (Orsolini, 1993). Clinical studies also showed that dyslexic children tend to use a similar 'economic' pattern without conjunctions (Plaza, 1998), just as aphasic adults, and this can be related to working memory limitations. FV tried to compensate for his vulnerability about causal connectors by using sequential clauses introduced by 'then', implicitly requiring the auditor to infer causality. But, since the control group did not use more causal connectors, the lack of causal conjunctions from Day –1 to Day +3 months could not be considered as atypical in our patient.

At the evaluative level, FV did not use reported speech (direct and indirect). The rarity or absence of reported speech suggested that FV did not represent speech within speech. The animated subjects of the story (the boy, the dog, the frog, the stag, the owl, the bees . . .) did not have any inner speech and they did not talk to each other, although they were endowed with a large range of perceptions, desires and emotions. We hypothesized that the poorness of reported speech could correspond to the 'social-emotional change' observed in the patient's behavior post surgery. He presented a slight anosognosia and did not acknowledge his difficulties at first and he also appeared more indifferent than preoperatively.

The discrepant pattern of evaluative devices in FV's narrative shows that *Broca's area is involved in auditory verbal working memory, and could be required in the subtle process of representing speech within speech.* As suggested by recent data, the superior part of the IFG (BA 44) is involved in the activation of internal speech representations, notably mobilized for correct identification of speech stimuli (Zekveld, Heslenfeld, Festen, & Schoonhoven, 2006). Though Broca's area is not 'necessarily' involved either in the process of translating social intent into speech (Gentilucci, Bernardis, Crisi, & Volta, 2006) or in the linguistic representation of actions (Hamzei et al., 2003; Rizzolatti, Focassi, & Gallese, 2001), its resection could be presumed to lead to transitory disturbances of 'mirroring' speech devices within narrative discourse.

CONCLUSION

Despite a massive resection of the inferior frontal gyrus, the patient did not exhibit the severe language impairments predicted by the localization theory of the brain. The present case confirms the relevance of connectionist approaches based on studies of slow-growth tumors, which demonstrate that compensatory mechanisms start before surgery, in reaction to tumor infiltration, and consolidate during and after surgical procedures (Bonnetblanc et al., 2006). The patient recovered his functional preoperative status within 3 months following surgery, with no neurological deficit, and could resume a normal socio-professional life. The patient's positive outcome can be attributed to the slow evolution of his tumor and the ensuing compensatory processes the results of which were clearly revealed by cortical and sub-cortical electrical stimulation. Whereas the compensating role of the controlateral right hemisphere has already been observed especially in slow-growing lesion such a LGG (Desmurget et al., 2007), the present identification of several structures essential to language within the remaining left hemisphere *support the crucial role of perilesional areas in functional compensation.* It is worth noting that cortical sites alone (i.e., the postero-inferior part of the pars opercularis, the ventral and dorsal premotor cortex, the dorso-lateral prefrontal cortex, the posterior insula) cannot account for the patient's language recovery following surgical resection. Indeed, sub-cortical mapping also showed that the preservation of both white matter pathways and deep grey nuclei prevented the occurrence of permanent postoperative aphasia. Such cerebral plasticity brings strong support to connectionist brain processing models, claiming to the existence of parallelly distributed cortical/sub-cortical networks (Devlin et al., 2003).

But the case also demonstrates the need to undertake fine-grained language and neuropsychological analyses before, during and after surgery, in order to specify the remaining disturbances and define new rehabilitation techniques. Thus, future studies should be undertaken to develop the assessment of syntactic skills, working memory and emotional processing during peroperative session, in the hope of preventing the subtle deficits noted here. In pre- and postoperative evaluations, the addition of a narrative production task to the neuropsychological assessment is of particular interest, usefully complementing the standardized assessments, by focusing on both narrative microstructure (syntax, lexicon, story length), macrostructure (initiation, maintenance and resolution of story components, i.e., executive processing) and psychosocial/emotional features. Narrative production appears as a 'marker' of subtle impairments that could otherwise go unnoticed.

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