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Language monitoring in multilingual patients undergoing awake craniotomy: A case study of a German–English–French trilingual patient with a WHO grade II glioma

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A B S T R A C T

In this paper, we report a case study of a 31-year-old multilingual female (LK) who presented with a left prefrontal brain tumour (WHO grade II glioma). LK is a late trilingual person whose first language is German. She had been learning English and French for 10 years when she moved to France at the age of 20 and now mostly uses French. German (L1) and French (L3) were assessed using a selection of sub-tests taken from the MT 86, the French version of the BDAE, the ECOSSE, the MEC, the German BAT, and, a non-standardized German adaptation of parts of the MEC. Preoperatively, LK had no language deficit. She was operated on under awake craniotomy, and both languages were mapped. Direct intraoperative electrical stimulation mapping showed that i) L1 and L3 were represented by both distinct and overlapping areas within the left (dominant) inferior frontal cortex, but shared the same subcortical tracts, and ii) the left dorsolateral prefrontal cortex was engaged when switching from one language to another. Since surgery, the patient has been followed longitudinally, with six-monthly assessments of her language skills using the same test battery. Her L1 and L3 language skills have been intact for 24

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months postoperatively. After presenting the behavioural and brain mapping data, we discuss their relevance with respect to the organization of language skills within the frontal cortex and deep frontal structures.

1. Introduction

WHO Grade II gliomas are infiltrating, primary tumours of the brain parenchyma. They represent approximately 15% of gliomas in adults and their peak incidence is in young adults aged 30–40 years (25% of all cases). Their treatment consists of the removal of as much of the tumour as possible, without impairing the patient's quality of life. Therefore, when these tumours are located in the dominant hemisphere, within or in the vicinity of the eloquent language cortex, awake surgery with direct intraoperative electrical stimulation mapping (ESM) is used in order to preserve the patient's language skills (Ojemann, Ojemann, Lettich, & Berger, 1989). This technique allows most of those tumours to be aggressively resected without any long-term language deficit (Sanai, Mizadeh, & Berger, 2008).

As several clinical studies have shown that bilingual aphasics do not necessarily manifest the same language disorders with the same degree of severity in both languages, it has been hypothesized that languages in bilinguals are sustained by at least partially distinct microanatomical systems located within the same gross anatomical areas (Paradis, 2004). Stimulation mapping studies support this view, and most (if not all) neurosurgeons who have operated on bilinguals with intraoperative language monitoring under awake craniotomy recommend testing all the languages in which the patients are fluent (see Giussani, Roux, Lubrano, Gaini, & Bello, 2007, for a review).

Nonetheless, the number of ESM studies addressing this topic is still very small. Most of them have investigated the cortical representation of languages (Lucas, McKhann, & Ojemann, 2004; Ojemann & Whitaker, 1978; Rapport, Tan, & Whitaker, 1983; Roux et al., 2004; Roux & Trémoulet, 2002; Serafini, Gururangan, Friedman, & Haglund, 2008; Walker, Quinones-Hinojosa, & Berger, 2004), although some have dealt with the role of subcortical language pathways (Bello et al., 2006; Moritz-Gasser & Duffau, 2009), and others with language switching (Kho et al., 2007; Moritz-Gasser & Duffau, 2009). A common denominator of these studies is the scarcity of behavioural data.

The present study investigated the neural substrates of the processing of a third language (French) and a native language (German) in a late trilingual who underwent left frontal tumour surgery via a craniotomy under awake conditions. The aim of the study was to determine whether distinct L1 and L3 areas might be distinguished within the inferior frontal cortex (IFC). Subtests of the Bilingual Aphasia Test (BAT; Paradis & Libben, 1987) were included in our patient's comprehensive language assessment, in order to demonstrate that functional languages can be preserved for the long-term in bilinguals undergoing WHO grade II tumour resection under awake craniotomy. We discuss what can be learnt from pre-, intra-, and postoperative language monitoring with respect to multilingual brain organization.

2. Illustrative case

2.1. Clinical presentation and neuroimaging

A 31-year-old trilingual female (LK) was referred for a neurosurgical assessment of a mass discovered on imaging after an initial seizure. Structural MR imaging was performed (Fig. 1), showing a mass lesion located in the middle frontal gyrus, in the vicinity of the eloquent inferior frontal and precentral cortices. The mass was of low signal intensity on T1-weighted imaging, but high signal intensity on T2-weighted and FLAIR imaging. A cyst was present, and there was no evidence of contrast enhancement. Given the imaging characteristics of the lesion, the obvious diagnosis was a WHO grade II glioma.

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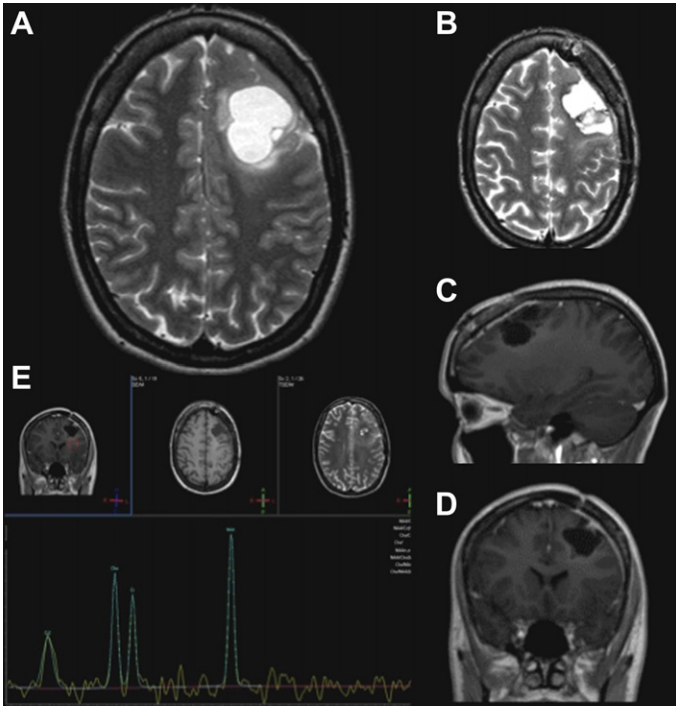

Fig. 1. MR imaging. This figure shows pre- and postoperative MR imaging of the left prefrontal WHO grade II glioma harboured by our patient (LK). A: preoperative T2-weighted MR image shows a left prefrontal tumour in the axial plane. B, C, D, E: 6-month postoperative MR imaging: (B), T2-weighted image in the axial plane; (C), T1-weighted image in the sagittal plane; (D), T1-weighted image in the coronal plane; (E): single-voxel 1H NMR spectroscopy in the deep white matter of the surgical cavity. These images show no residual tumour.

2.2. Multilingual profile and preoperative examination

LK's first language (L1) is German, the only language spoken in her family during her childhood in Germany. She was formally taught English (L2) from the age of 10 years and French (L3) from the age of 12 at secondary school. Between the ages of 18 and 20, she used both languages professionally for her work as a trilingual secretary. At 20, she moved to France, where she mostly speaks French, even with her children. She only uses German with certain friends and had not used English for the past 13 years.

LK's neurological examination was considered to be normal. She had no motor, sensory or visual field impairment. Her constructional skills, as assessed by copying a figure depicting intersecting pentagons and drawing a clock face, were intact. The Edinburgh inventory indicated that LK is right-handed (Oldfield, 1971).

Given the role of the different languages in her present life, we decided to concentrate the language assessment on German (L1) and French (L3). French was assessed using a selection of sub-tests taken from the MT 86 (Nespoulous et al., 1986), the French version of the BDAE (Mazaux & Orgozo, 1981), the ECOSSE (Lecocq, 1996), and the MEC (Joanette, Ska, & Côté, 2004). It is standard practice within our neurosurgical department to combine all these subtests to in order to arrive at a comprehensive language assessment of a reasonable length that covers a variety of aspects of language use and knowledge. German was assessed in a similar way, using subtests selected from the German BAT (Paradis & Lindner, 1987), and a non-standardized German adaptation of parts of the MEC was developed. The complete battery used in this study is described in Table 1. Preoperative testing with these French and German language batteries was performed two weeks before surgery. It showed that the patient was highly proficient in L1 (German) and L3 (French), with normal performances in both languages, but relatively weak performance on the formal verbal fluency task in L1 (Table 1).

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Table 1
Behavioural data in L1 (German) and L3 (French) collected before surgery, and 6, 12, and 24 months postoperatively.

Task	Preoperative test November 2008		Postoperative test June 2009 (M6)		Postoperative test December 2009 (M12)		Postoperative test December 2010 (M24)	
	German	French	German	French	German	French	German	French
Naming[c]								
-nouns	10/10	25/25	10/10	24/25	10/10	25/25	10/10	25/25
-verbs	10/10	6/6	10/10	6/6	10/10	6/6	9,5/10	5/6
Word repetition[b,d]	20/20	20/20	20/20	20/20	20/20	10/10	20/20	20/20
Sentence repetition[b,d]	7/7	15/16	7/7	16/16	7/7	16/16	7/7	16/16
Word comprehension[b,c]	18/18	9/9	18/18	9/9	18/18	9/9	18/18	9/9
Sentence comprehension[b,c,e]	71/71	43/44	71/71	42/44	70,5/71	40/44	71/71	43/44
Narrative speech[b,c]	normal	normal	normal	normal	normal	normal	normal	normal
Definition of metaphors[a]	15/15	15/15	15/15	14,5/15	15/15	14,5/15	15/15	14/15
Semantic categories[a]								
-judgement	5/5	23/24	5/5	22/24	24/24	23/24	23/23	23/24
-justification		11/12		10/12	12/12	11/12	11/11	11/12
Linguistic prosody[a]								
-comprehension	12/12	10/12	12/12	10/12	11/12	8/12	12/12	10/12
-repetition	12/12	11/12	12/12	12/12	12/12	12/12	12/12	12/12
Emotional prosody[a]								
-comprehension	11/12	12/12	12/12	12/12	12/12	12/12	12/12	12/12
-repetition	12/12	12/12	12/12	11/12	12/12	12/12	11/12	12/12
Indirect speech acts[a] (interpretation)	15/15	14/15	15/15	15/15	14/15	14/15	14/15	14/15
Verbal fluency[a]								
-formal	15	20	24	23	31	22	19	27
-semantic	30	27	36	28	29	27	28	29
Conversation[a]	34/34	34/34	34/34	30/34	34/34	34/34	33/34	34/34

[a] Test used MEC (Joanette et al., 2004).
[b] Test used German BAT (Paradis & Lindner, 1987).
[c] Test used MT86 (Nespoulous et al., 1986).
[d] Test used BDAE (Mazaux & Orgozo, 1981).
[e] Test used ECOSSE (Lecocq, 1996).

Additionally, LK performed a picture-naming task the day before surgery. Stimuli were taken from the Centre for Research in Language International Picture-Naming Project corpus (CRL-IPNP, Szekely et al., 2005). We used a subset of 80 items featuring 40 objects and 40 actions chosen from the original corpus. A further selection of stimuli was then made in order to construct personalized blocks of items in both languages for the intraoperative mapping session.

2.3. Intraoperative electrical stimulation mapping

Brain mapping was undertaken during asleep-awake craniotomy. A neuronavigation system (Brainlab®, Munich, Germany) was used to define tumour location. A left frontal craniotomy was performed during the asleep period of the craniotomy, which exposed the cortex surrounding the tumour area, the left IFC and the motor cortex.

Intraoperative cortical and subcortical electrical stimulation mapping was used to localize the critical cortical areas and functional tracts in the patient under the awake condition. This technique has been described in detail elsewhere (Bello et al., 2006; Ojemann et al., 1989; Roux & Trémoulet, 2002). The brain was directly stimulated, using the bipolar electrode of the NIMBUS i-Care® multifunctional stimulator (1-mm electrodes 5 mm apart; Newmedic®). The initial current amplitude of 1 mA was gradually increased in steps of .5 mA. We used biphasic square-wave pulses of 1 ms at 60 Hz, with a maximum train duration of 4 s. Language mapping was performed in French and in German using the lowest current (2.5 mA) that produced speech arrest in the inferior ventral motor cortex (i.e., blocking number counting without simultaneous motor response in the mouth or pharynx in both languages).

While the patient performed a picture-naming task in either L1 or L3, different cortical sites were randomly selected and then electrically stimulated. We recorded whether the stimulation interfered

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with picture-naming or not (i.e., leading to speech arrest, anomia, or paraphasia). Stimulation-positive sites were interpreted as being essential for the task, so that the procedure resulted in a map of sites that were essential for either L3 (French) picture-naming, L1 (German) picture-naming, or both.

Cortical mapping revealed five language sites (Fig. 2). Three of these were common to both languages, eliciting the same type of error, whereas the other two specifically impaired L3 but not L1 production. Stimulation of one of the latter (no. 10, Fig. 2), located within the inferior frontal gyrus (IFG), elicited semantic paraphasia in French but not in German, while stimulation of the other site, located within the posterior and lateral part of the middle frontal gyrus (no. 2, Fig. 2), provoked unintentional language switching from French (L3) to English (L2). The stimulation mapping data for L1 and L3 are summarized in Table 2. Resection was then performed according to the anatomical and functional cortical-subcortical boundaries we had established (the subcortical level was directly stimulated to identify the white matter language pathways). Interestingly, subcortical ESM in the white matter in the posterior and lateral depth of the surgical cavity also generated reproducible language disturbances (speech arrest and transient limited speech in French and German), but no difference between L1 and L3 processing was found in the subcortical structures.

2.4. Postoperative course

Immediately after surgery (Day 1 post op.), LK experienced transient aphasia with severely impaired speech expression in both languages. Language production was reduced to a few words and automatisms, combined with anomia as well as phonemic paraphasia. In addition, there were major disturbances in reading and writing. However, she had no oral language comprehension impairment. No major dissociation was observed between the patient's performances in German and French, which seemed equally impaired, though no normalized testing was applied to either language. Control MR imaging was performed immediately after surgery, showing subtotal tumour removal, as a FLAIR signal

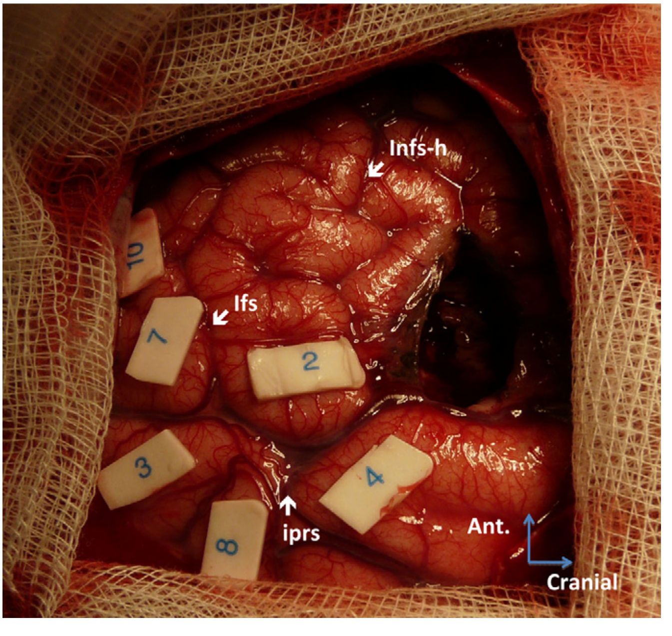

Fig. 2. Stimulation mapping of the left inferior frontal cortex (post-resection). This figure shows the left inferior frontal cortex of our patient (LK), as exposed after the craniotomy, and following tumour resection. Cortical sites producing language impairment during stimulation mapping were labelled with a sterile ticket in order to gain a clearer intraoperative picture. All of them disrupted language production, except for site no. 4. The various types of language interference are described in Table 2. Ifs, inferior frontal sulcus; Infs-h, intermediate frontal sulcus (horizontal part); iprs, inferior precentral sulcus.

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Table 2
Stimulation mapping data in relation to L1 (German) and L3 (French).

	Mapping sites	Object naming task	
		L1 (German)	L3 (French)
Region			
Brodmann area			
Broca's area			
BA 44/6	3	Speech arrest	Speech arrest
BA 44	7	Anomia	Anomia
BA 45	10	No error	Semantic paraphasia
Premotor cortex			
BA 6	8	Speech arrest	Speech arrest
BA 6	4	No error	No error
Dorsolateral prefrontal cortex			
BA 9	2	No error	Single instance of involuntary switching (L3/L2)

Mapping sites are shown in Fig. 2.

abnormality was still present at the deep posterior part of the cavity. The pathological examination confirmed a WHO grade II oligodendroglioma. No other treatment was administered to the patient.

At discharge from hospital (Day 12 post op.), LK exhibited almost fluent speech in French and in German, despite word-finding difficulties and paraphasic errors. Reading was normal, but writing was still impaired. These aphasic deficits were observed during conversational interaction between LK, her family and her surgeon (VL). French language rehabilitation was prescribed. After a few sessions, LK decided to stop rehabilitation because she found that the type of therapy on offer did not match up to her language skills and did not allow her to progress. Three months later, her naming performance was almost back to normal, although a longer production time was observed in both languages in the outpatient clinic.

At the first comprehensive postoperative assessment (6 months post op.), LK was tested with the same test battery that had been used preoperatively. The results showed a slightly lower performance in French when they were compared with those of the presurgery assessment (Table 1). For most of the sub-tests, her performance remained within the normal range, except for the conversational skills measures taken from the MEC (Joanette et al., 2004), which were slightly below standard deviations. Her performance in German was absolutely normal.

At the second comprehensive postoperative assessment (12 months post op.), her French conversational skills were back within the normal range (Table 1). However, slightly impaired performance was found on the comprehension of linguistic prosody and sentence comprehension. While the former might have been due to technical difficulties in the presentation of the prosody task, the results from the sentence comprehension task seemed to indicate some difficulties with complex sentence structures in French, as all the errors involved embedded relative clauses. Again, her performance in German was normal, apart from one hesitation in the sentence comprehension task.

At the third comprehensive postoperative assessment (24 months post op.), her performance was within the normal range for both languages (Table 1).

MR imaging was carried out at 6, 12, 18, and 24 months post op. The radiologist noted no progression of the disease. MR imaging performed at the 6-month follow-up is shown in Fig. 2.

On the whole, behavioural data showed that verbal behaviour was close to normal 6 months after surgery. At that point, slight persisting difficulties affected the later learnt L3 more than L1. From 12 months after surgery to the present time, her performance in both languages has been within the normal range.

3. Discussion

We investigated the language performance of a late and highly proficient German–French–English trilingual female suffering from a WHO grade II glioma. Direct intraoperative ESM data showed that i) L1 (German) and L3 (French) were represented in both distinct and overlapping areas within the left (dominant) IFC, but shared the same subcortical tracts, and ii) the left dorsolateral prefrontal cortex (DLPFC) was involved when switching between languages. Twenty-four months after the subtotal removal of the tumour, taking care to preserve these functional areas, behavioural data showed that L1 and L3 language skills were intact, as assessed by a comprehensive language examination, which included BAT subtests in addition to the standard language protocol (MT 86, BDAE, ECOSSE, MEC) we use for monolinguals. Above and beyond the benefit for the patient of still being able to use L1 and L3 in her everyday life, the combination of ESM and a rigorous assessment provided valuable data that enhanced our understanding of the organization of language skills within the frontal cortex and deep frontal structures more fully. These points are discussed below.

3.1. What can we learn from intraoperative language monitoring?

3.1.1. Involvement of shared and specific structures

Researchers have long wondered whether the processing of different languages involves the same anatomical areas belonging to a single system, or whether each language constitutes an independent system relying on distinct anatomical areas (Paradis, 2004). Studies of bilingual aphasic patients exhibiting i) different aphasic symptoms that vary across languages, and/or ii) different recovery patterns in their different languages first prompted researchers to raise the question of distinct cortical and subcortical structures for languages (Fabbro, 2001; Fabbro & Paradis, 1995; Paradis, 2001). One of the first mapping methods used to assess the cerebral representation of linguistic functions was direct ESM. Investigating bilinguals, Ojemann and Whitaker (1978) found cortical sites where both languages were equally disrupted, sites where one language was more disrupted than the other, and sites where one language was disrupted but not the other. Larger cortical areas were observed for languages in which the patients were less proficient, and language-specific areas were noticed in the frontal and temporoparietal regions. To date, 10 ESM studies have been dedicated to the study of cortical-subcortical language organization in bilingual patients with epilepsy or brain tumours (Bello et al., 2006; Lucas et al., 2004; Kho et al., 2007; Moritz-Gasser & Duffau, 2009; Ojemann & Whitaker, 1978; Rapport et al., 1983; Roux et al., 2004; Roux & Trémoulet, 2002; Serafini, Gururanga, Friedman, & Haglund, 2008; Walker et al., 2004). All the authors identified language-common areas and language-specific areas by stimulating cortical structures, while Bello et al. also pinpointed language-specific white matter fibre tracts by stimulating subcortical structures (Bello et al., 2006). In our case study we, too, located language-common areas and language-specific areas in the frontal cortical structures we tested, but we only found language-common tracts in the deep frontal subcortical structures we tested (see Section 2.3).

Since the 1980s, extensive use of scanning techniques and computer technology exploring the relationship between blood flow and neural activity (PET, fMRI) have allowed researchers to investigate the neural bases of language in the normal brain. The first study to focus on bilinguals was conducted by Klein, Zatorre, Milner, Meyer, and Evans (1995), who carried out PET investigations of 12 French–English bilinguals. They found no difference in brain activation during word processing except for a left putaminal activation, which was present in the second acquired language, and they suggested that this putaminal activation was related to increased demands on articulation processes, when speaking a L2 learnt late in life. In subsequent studies, conflicting results emerged with respect to the bilingual brain, even though factors such as age of onset of bilingualism, proficiency and exposure, which could influence the way in which the two languages are represented (Paradis, 1994), were taken into account. While several authors found that two different languages activate the same regions (Chee, Tan, & Thiel, 1999; Hernandez, Dapretto, Mazziotta, & Bookheimer, 2001; Hernandez, Martinez, & Kohnert, 2000; Illes et al., 1999; Klein et al., 1995; Klein, Milner, Zatorre, Zhao, & Nikelski, 1999; Perani et al., 1998), others found that different languages may also activate separate regions (Dehaene et al., 1997; Kim, Relkin, Lee, & Hirsch, 1997; Marian, Spivey, & Hirsch, 2003; Simos et al., 2001; Yetkin, Zerrin Yetkin, Haughton, & Cox, 1996). As most of the studies that found activation in the same regions featured tasks involving single word processing, and many of the studies that reported activation in separate regions involved the processing of sentences or discourse, it has been suggested that the inconsistency of results might be due to the fact that lexical processing relies on declarative memory and is therefore likely to be similar for both first and later learnt languages, whereas sentence processing depends on

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procedural memory and is thus likely to differ between a first and a later learnt language (e.g. Paradis, 1994, 2004, 2009; Ullman, 2001).

Nevertheless, a recent meta-analysis of haemodynamic studies by Indefrey (2006) found reliable differences between patterns of L1 and L2 activation for subgroups of bilinguals in the shape of stronger activation during L2 processing, despite the fact that all five experiments compared in this meta-analysis were based on word production tasks. Thus two studies involving participants with late L2 onset and variable L2 exposure reported stronger activation of the left posterior IFG (BA 44, 47) in L2 picture-naming compared with L1 picture-naming (De Bleser et al., 2003; Vingerhoets et al., 2003). By contrast, the participants in the other three studies reporting no differences between L2 and L1 picture-naming (Hernandez et al., 2000, 2001; Rodriguez- Fornells et al., 2005) had early L2 onset and lived in L2-dominant environments. Both onset and exposure might, therefore, explain the difference in the results. Our finding – showing that naming in the latest learnt, but currently most frequently used language (L3) relies on a larger cortical area in the left posterior IFG (BA 45) than L1 processing - is in line with these data.

Furthermore, the posterior IFC has been shown to be involved in phonology, semantics, syntax and sentence processing, as well as in comprehension and working memory (Vigneau et al., 2006). There are even strong arguments for a relative functional sublocalization within Broca's area and the left IFC, with the more posterior regions being involved in phonological processing (BA 6/44/45) and the more anterior areas being predominantly involved in semantic processing (BA 45/47), while the dorsal part of the pars opercularis may be dedicated to syntactic processes (Poldrack et al., 1999; Vigneau et al., 2006). Regarding bilinguals, Klein et al. (1995) suggested that L2 semantic processing generates more cortical activation than L1 semantic processing. Similarly, Marian et al. (2003) showed that the L2 activates a larger cortical area in the IFG than the L1 during both lexical and phonological processing. Wartenburger et al. (2003) found greater activation during grammaticality judgements in late learners, whereas Golestani et al. (2006) found greater activation for the processing of syntax but not for single words in late learners. Together, these results suggest that in this population of late bilinguals, L2 processing tends to require more cortical resources than L1 processing, for both single word and sentence processing. Our mapping data partially corroborate these results, as we identified a site within the IFG where L3 but not L1 semantic processing was disrupted.

This report is in line with the general concept that shared and separate neural networks may support different types of language processing, both at the cortical and subcortical levels. However, we acknowledge that this latter point needs further investigation, as subcortical ESM data in bilinguals are very scarce.

3.1.2. Language switching

The second basic question in the neuropsychology of bilingualism concerns the neural correlates of language switching and control, that is, the areas that are engaged when bilinguals switch from one language to the other. Penfield and Roberts (1959) postulated the existence of a separate language switching mechanism controlling access to different languages. In recent years, a growing number of studies have made it possible to identify several brain areas that seem to be involved in language switching. Price, Green, and von Studnitz (1999) conducted a PET investigation of German–English bilinguals performing translation and language switching tasks. They found that the most active areas during translation were located, bilaterally, in the anterior cingulate gyri, the putamen and the head of the caudate nuclei. They also observed activation of Broca's area and the dominant supramarginal gyrus during language switching. In an fMRI study of picture-naming in Spanish–English bilinguals, Hernandez et al. (2001) found increased activation of the DLPFC in switch trials compared with non-switch trials. These and other results (e.g., Holtzheimer, Fawaz, Wilson, & Avery, 2005) led Hernandez (2009) to claim that the DLPFC plays a major role in language switching. This claim is corroborated by our data showing involuntary switching from L3 to L2 during stimulation of the left DLPFC (BA 9), even though this occurred only once. ESM studies are undoubtedly an interesting means of improving our understanding of language switching mechanisms, but data are still scarce. One patient who switched from French to Chinese during stimulation of the posterior part of the left IFG, and another patient who switched from French to English during stimulation of the left dominant posterior temporal lobe have been described so far (Kho et al., 2007; Moritz-Gasser & Duffau, 2009). These authors also collected

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subcortical stimulation data on the role of the superior longitudinal fasciculus and postulated the existence of a large-scale distributed network subserving language switching, which would rely on an executive system (partially mediated by the prefrontal cortex) controlling a more dedicated language system involving temporoparietal areas in addition to Broca's area (Moritz-Gasser & Duffau, 2009). These preliminary results clearly call for larger series of bilingual patients to be studied under similar circumstances with ESM.

3.2. What can we learn from pre- and postoperative language monitoring?

The previous sections have indicated how intraoperative data from our ESM study can enhance our understanding of bilingual brain organization. In this section, we discuss how pre- and postoperative assessments can provide additional information on multilingual processing.

3.2.1. Preoperative language monitoring

A better understanding of how different languages are represented in the human brain can be obtained from studies of multilingual patients who have sustained brain lesions. Patients with WHO grade II gliomas classically present with seizure, a normal neurological examination, and normal personal and professional lives (DeAngelis, 2001). However, although it has been reported that these patients have normal higher functions in 90% of cases (DeAngelis, 2001), the course of the clinical symptoms in this early stage of the disease is imperfectly known, and patients often exhibit a slight deficit in language and cognitive functions when a comprehensive examination is performed (Duffau, Gatignol, Mandonnet, Capelle, & Taillandier, 2008; Teixidor et al., 2007). In the present case study, the presence of a slight deficit in the formal fluency task was noted in German, but not in French, in the preoperative period, which is not unusual for frontal lesions (Benzagmout, Gatignol, & Duffau, 2007; Tucha, Smely, Preier, & Lange, 2000). Moreover, this differential impairment might indicate a differential representation of these languages.

3.2.2. Immediate postoperative language monitoring

Patients with WHO grade II gliomas who have undergone surgery within or in the vicinity of speech areas, using awake craniotomy and direct intraoperative ESM, often exhibit an immediate postoperative deficit, which generally resolves itself within 3–6 months after continual improvement (Sanai et al., 2008). The risk of a deficit is higher for those patients in whom a functional tract has been pinpointed by means of subcortical mapping (Bello et al., 2006). Hence, these patients should allow researchers to track different patterns of recovery during this period.

So far, as most ESM studies of bilinguals have focused on intraoperative mapping, detailed postoperative examinations have rarely been reported and are hard to compare. Nonetheless, in Bello et al. (2006)'s study, subcortical mapping revealed more L1-specific functional tracts in patients who experienced a decrease in fluency immediately after surgery, mainly affecting the L1. Similarly, Gatignol, Duffau, Capelle, and Plaza (2009) described two patients who had cortico-subcortical stimulation mapping during WHO grade II glioma surgery and who showed stronger deficits in L1 in the immediate postoperative period. Taken together, these observations may support Kainz's theory (as cited in Fabbro, 2001, p. 213) and Fabbro's hypothesis (Fabbro, 2001) about the cortical and subcortical representation of languages, whereby subcortical lesions result in greater impairment of the patients' L1 and poorer recovery. This does not, however, seem to have been the case for our patient, who demonstrated similar patterns of language impairment in L1 (German) and L3 (French). Therefore, as none of these studies, including ours, have provided sufficient data for comparison, further research is required.

Similarly, involuntary language switching has occasionally been observed in the immediate postoperative period (Gatignol et al., 2009; Moritz-Gasser & Duffau, 2009) and in the latter study was assumed to corroborate intraoperative findings (Moritz-Gasser & Duffau, 2009). These cases are not surprising, given that pathological unintentional language switching had previously been described in two patients with frontal lobe lesions who had no other linguistic impairments (Fabbro, Skrab & Aglioti, 2000; Meuter, Humphreys, & Rumiati, 2002). Nevertheless, we did not observe any language switching or even mixing in our patient in the immediate postoperative period. Further investigations on pathological switching are clearly needed.

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3.2.3. Long-term follow-up

Overall, our behavioural data indicate that skills in both languages can be preserved in the long-term despite the aggressive resection of a WHO grade II glioma within or in the vicinity of eloquent areas. After a short period of temporary deficits, all aphasic symptoms resolved themselves. Nevertheless, WHO grade II gliomas grow continuously prior to malignant transformation (Mandonnet et al., 2010). This is why assessments of both languages will be conducted every 6–12 months in order to allow early detection of any modification in the patient's linguistic performance, which might correspond to tumour recurrence. In which case, we may observe differential patterns again.

3.3. Limitations of the study

3.3.1. Limitations related to the stimulation technique

Direct intraoperative ESM yields valuable data that complement behavioural and functional brain imaging data. This is of particular interest for the brain mapping of language functions in bilingual or multilingual individuals, where ESM can help us to disentangle the micro-areas engaged in the processing of each language (Paradis, 2004). Nevertheless, it is an invasive technique applicable only to neurosurgical patients with brain pathologies. The "region of interest" for mapping is determined by the extent of the craniotomy, which can be more or less targeted according to the surgeon's habits and preoperative planning (Ojemann et al., 1989; Sanai et al., 2008). In the context of WHO grade II gliomas, which are generally slowly developing infiltrating tumours, functional reorganization may largely modify "normal" brain functioning within the considered region (Lubrano, Draper, & Roux, 2010) and probably far beyond (Guye, Bettus, Bartolomei, & Cozzone, 2010).

In addition, as it had been shown that language interference may vary with the intensity of stimulation (Pouratian, Cannestra, Bookheimer, Martin, & Toga, 2004), we can speculate that differential stimulation thresholds have a variable impact on the processing of different languages. Within the same area, one language could be disrupted at a given stimulation intensity and another language at a higher intensity, just as threshold influences activation in functional imaging.

3.3.2. Limitations related to our behavioural protocol

Integrating the German BAT into our standard local protocol allowed us to perform a comprehensive assessment of preoperative and long-term language performances. However, French BAT data were not available for a valid comparison, thus constituting a major shortcoming of our study. In the future, the BAT will be integrated into our standard language examination for all the patient's languages, providing us with a quantification and classification of the disorders for each language. By so doing, it will allow us to make a direct comparison of performances in the different languages known by the patient – something that researchers have so far been unable to do. Furthermore, we are preparing to integrate not just single words but larger linguistic units, too, into the intraoperative protocol of verbal tasks.

4. Conclusions and perspectives

Our illustrative case study revealed differential segregation of the IFC for the semantic processing of two languages, and its implication in language switching. The anatomical and functional correlates of bilingual processing are a major focus of interdisciplinary research, and direct intraoperative ESM is a reliable method of investigating brain organization in bilingual neurosurgical patients. In our opinion, it is all the more informative as a rigorous behavioural assessment is performed before, during and after the operation. However, to date and to the best of our knowledge, bilingual aphasia in neurosurgical patients has been studied using different test instruments that do not allow for direct comparison between languages (see Paradis, 2004). So far, the BAT has mostly been employed by aphasiologists and researchers, and apparently has not reached the neurosurgical clinical setting, as most clinicians still use the same tests for bilingual assessments as they do for monolingual ones (e.g., BDAE, AAT, and MT 86 for French speakers). We have shown that it is feasible to introduce the BAT, now easily available online, into surgical clinical practice. We believe that, due to the course of language impairment in WHO grade II gliomas, it is extremely worthwhile combining ESM data with longitudinal behavioural studies and other noninvasive mapping techniques such as functional MR imaging.

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