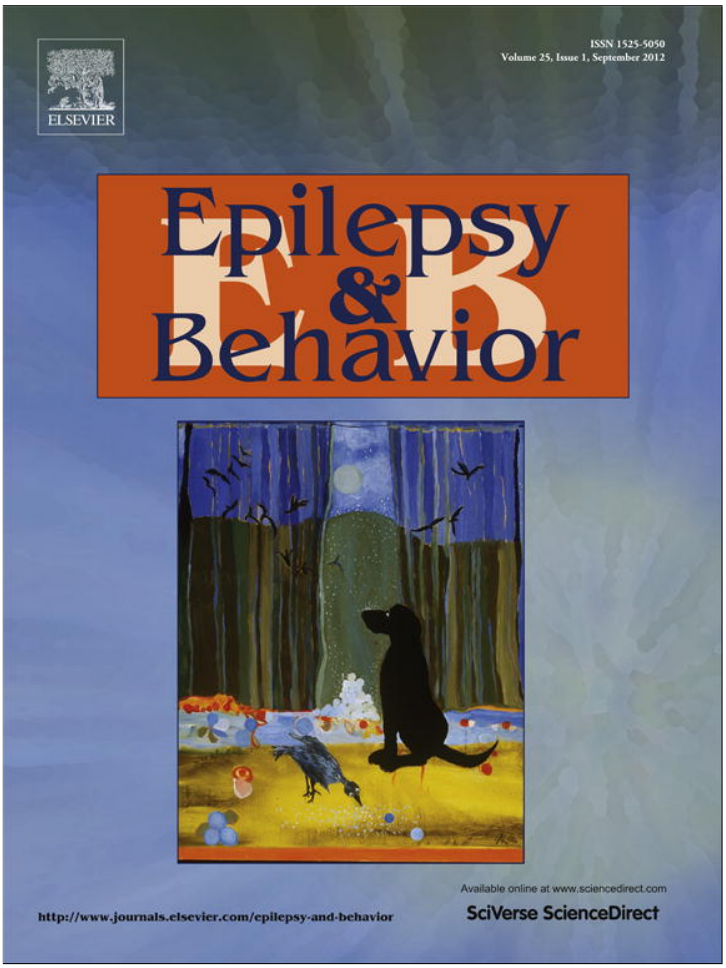

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Case Report

Crossed aphasia after right anterior temporal lobectomy. A case report

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ABSTRACT

The occurrence of crossed aphasia as a complication after temporal lobe epilepsy surgery is extremely rare. We report the case of a 47-year-old right-handed patient with drug-resistant mesial temporal lobe epilepsy (MTLE) who developed a transitory aphasic syndrome after a right temporal anterior lobectomy. This syndrome was characterized by anomia, poor verbal fluency, verbal perseveration, and verbal comprehension difficulties. He also showed writing difficulties, reading substitutions, and calculation task errors. The patient was regularly assessed with language tasks, and showed a spontaneous and progressive recovery of his symptoms, with remaining naming difficulties. We discuss the role that epileptogenic zone could play in cortical reorganization of the language systems.

1. Introduction

Temporal lobe epilepsy (TLE) surgery could affect cognitive status in some patients. The assessment of these patients has allowed the identification of different cognitive mechanisms and the understanding of the relationships between brain and behavior. Brenda Milner described the case of HM who became amnesic after bilateral medial temporal lobe resection [1]. Since that time, it was also found that some patients showed memory deficits in verbal material after language dominant left temporal lobe resection and non-verbal material after right temporal lobe resection [2], but this relation is less clear for non-verbal material [3].

More recently, Jones-Gotman et al. [3] compared memory performance after standard temporal lobectomy vs selective amygdalohippocampectomy showing that learning and retention were disturbed regardless of the extension of the resected area. Helmstaedter [4] described that those patients who have a better cognitive status before surgery show greater losses than patients with poor memory baseline performance.

With respect to language performance, our research group found [5], in the presurgical evaluation, impairment in 46% of patients with TLE and hippocampal sclerosis without relation to hemispheric dominance for language. It has been observed that TLE may "disrupt the semantic retrieval network" affecting different language processes [6]; therefore, it is possible to find changes within this network after surgery.

Many studies have described deficits in naming ability after left anterior temporal lobectomy [7–9], with an average rate of decline of 34% [10] that can occur as a result of interruption of two different components: lexical-semantic or word production [11]. There is also some evidence of atypical language dominance (between 4% and 20%) in patients with right hemisphere epilepsies with two predominant patterns observed: an incomplete left hemispheric dominance or a bilateral distribution of language [12,13]. However, the occurrence of crossed aphasia (CA) is extremely rare, with an incidence of approximately 2% and only a few cases have been reported [14,15]. This entity was defined by Bramwell in 1899, and refers to an acquired language disturbance resulting from a right hemisphere lesion in right-handed patients.

We report the case of a 47-year-old right-handed man with a drug-resistant MTLE who developed an aphasic syndrome after a right temporal anterior lobectomy with progressive and spontaneous recovery of symptoms.

2. Case report

E.S. is a 47-year-old right-handed man with 7 years of formal education and employed as a factory worker. He was diagnosed with drug-resistant mesial temporal lobe epilepsy. Video-EEG monitoring was performed (Stellate Harmonie, BioScience) over 5 days, with 32 recording electrodes using the international ten-twenty system of electrode placement. Longitudinal and transverse bipolar electrodes and referential monopolar electrodes were used. Interictal EEG results showed right anterior temporal spike-and-sharp wave paroxysms (F8–T4–FT10 electrodes), occasionally spreading to the left temporal area. Five seizures were registered and characterized

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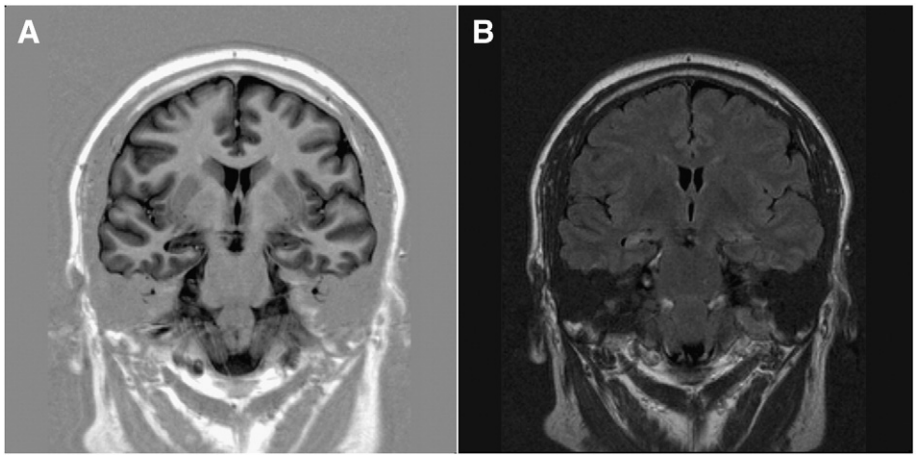

Fig. 1. Preoperative MRI scans showing right hippocampal atrophy, loss of the hippocampal internal structure, and increased signal intensity on (A) coronal inversion-recovery and (B) FLAIR weighted images.

by simple autonomic partial onset (epigastric sensation) followed by impairment of consciousness, movement arrest, and early right-hand automatisms. An unstructured interview and an ictal consciousness inventory [16] were used to assess ictal and postictal behavior. No language deficits occurred during or after the seizures. Ictal EEG changes started 500 ms before the clinical onset, characterized by synchronic rhythmic 5-Hz activity in the region of the right anterior temporal lobe (F8–T4–FT10 electrodes). It was concluded that the epileptogenic zone was localized to the right mesial temporal lobe.

Magnetic resonance imaging studies showed morphological and signal abnormalities indicating right hippocampal sclerosis (HS), consisting of hippocampal atrophy, loss of the hippocampal internal structure, and increased signal intensity on T2 or FLAIR weighted images [17]. Slightly decreased volume of the left hippocampus that did not meet the criteria for HS was also observed [18] (Fig. 1). Except for a deficit in verbal memory, there was no other evidence of clinical correlation with this feature, nor suspicion of bilateral epilepsy.

Patient E.S. had his first seizure at 8 months of age. No history of prenatal or early childhood diseases was present. He is strongly right-handed with no family history of left-handedness. He was treated with first generation and new generation antiepileptic drugs with no seizure control in spite of adequate dosing schemes and doses.

A standard right temporal resection was performed. The surgery included a 3.5-cm cortical resection and a 5-cm “en bloc” amygdalohippocampectomy (Fig. 2).

Presurgical neuropsychological assessment showed normal values in naming tasks, processing speed, attention span, and working memory. He had deficits (z values equal or lower than −2) in memory tasks for both verbal and non-verbal material as well as in verbal semantic fluency tasks (Fig. 3). To determine handedness, we used the Edinburgh Handedness Inventory [19,20] and the Grooved Pegboard Test [21]; results showed right-hand dominance. We did not use fMRI or the Wada test to assess language dominance before surgery because the patient did not show any sign of left language dominance.

Patient E.S. underwent surgery on April 2011 after which he developed an aphasic syndrome characterized by anomia, poor verbal fluency, verbal perseverations, and verbal comprehension difficulties. He also presented writing difficulties, reading substitutions, and calculation task errors, without other neurological deficits. The clinical features observed in E.S. could be classified as an amnesic (anomic) aphasia, because it was fluent, without articulatory disorders, and showed mainly anomia and circumlocutions that improved with phonological cues [22]. Comprehension, reading, and writing were slightly compromised [23]. The histopathological analysis showed mesial temporal damage with loss of hippocampal neurons higher than 50% involving all areas (Wyler score IV) [24]. An MRI study was performed a week after surgery, showing surgical sequelae affecting the right temporal lobe and no displacement of the midline or other complication (Fig. 2).

The patient was regularly assessed after surgery, using different language tasks that included a naming task (Boston Naming Test — BNT [25,26]). He was also evaluated in reading (sentence reading

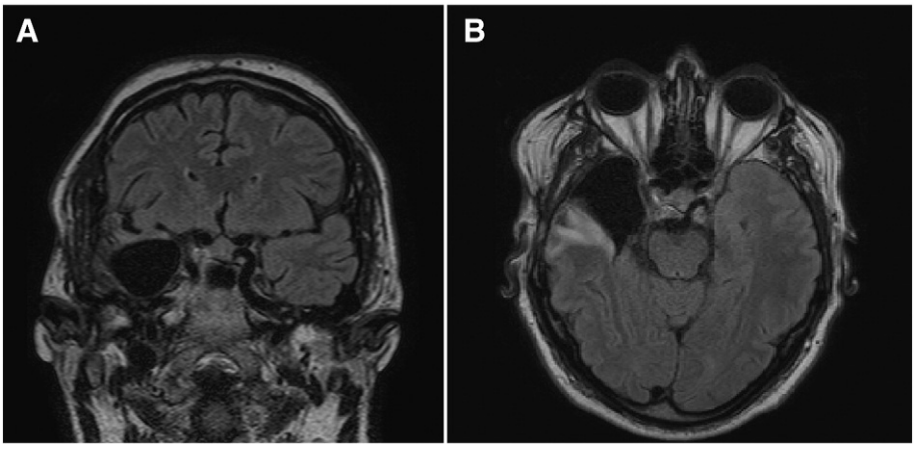

Fig. 2. Postsurgical sequelae associated with right temporal resection in (A) coronal and (B) axial FLAIR images.

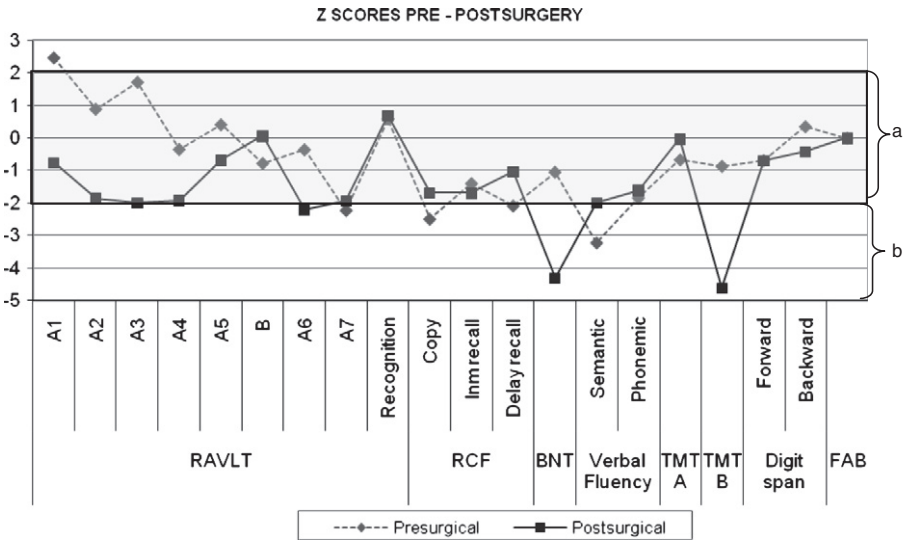

Fig. 3. Neuropsychological assessment: pre- and postsurgical z-scores. Note: a z score ≥2 = normal values, b z score ≤ −2 = Deficit. RAVLT = Rey auditory verbal learning test; RCF: Rey complex figure; BNT: Boston naming test; TMT: Trail making test; FAB: Frontal assessment at bedside.

and comprehension), writing (spontaneous and dictation), comprehension, and calculation (addition, subtraction, division, and multiplication) (subtests Western Aphasia Battery) (Table 1).

At 7 days post-surgery, the patient produced 10 spontaneously correct responses in the BNT using circumlocutions and perseverative responses. The patient showed substitutions (i.e., caja — capa, le — se) and paralexias (pastelisis instead of pastelitos) when asked to read a short story. He was unable to write down his name correctly and also had difficulties writing single words and phrases (i.e., when asked to write down TAZA, he wrote TACA; then he wrote MEMA, instead of MESA; MELTOLO instead of PELOTA). His spontaneous speech was full of perseverations, diminished verbal fluency, and comprehension difficulties. He also had errors in subtraction, division, and multiplication tasks, but addition was preserved (Table 1).

At 9 days post-surgery, the patient was able to write down his name correctly but still had difficulties when asked to write down phrases. In the BNT, he could name 22 images, but had an increased number of circumlocutions and definitions. No perseverations in responses were observed. Difficulties in subtraction and multiplication remained. Two days later, writing difficulties persisted for phrases and adding letters to a given word (Casa — Casada). The use of circumlocutions and definitions remained during naming tasks (Table 1). The patient was also assessed at 16 and 23 days after surgery, showing no writing difficulties and using fewer paraphasias (Table 1).

One month after surgery, writing, reading, and calculation difficulties were no more observed, but naming difficulties remained (Table 1).

Table 1
Postsurgical language assessment results.

Days after surgery	BNT-spontaneously produced correct responses	Correct responses after phonemic cueing	Reading	Writing	Calculation
7	10	18	20/40	3.75/16	6/24
9	22	25		11/16	12/24
11	26	26		11/16	
16	28	22			
23	29	24			
1 month after surgery	24	19	40/40	15/16	24/24
2 1/2 months after surgery	32	17	39/40	15/16	24/24
6 months after surgery	33	17	37/40	15/16	24/24

Two and a half months after surgery, the patient had a better performance during naming tasks, showing self-facilitating responses during complex orders (Table 1).

At 6 months follow-up, neuropsychological assessment was made according to our Epilepsy Centre's Neuropsychological Protocol [5]. We found memory deficits for verbal material, naming deficits with anomias, circumlocutions, and slowed performance in attentional switching tasks (TMT B). The patient performed well in writing, reading, and calculation tasks, with a correct intonation and prosody, and a minimum amount of paralexias. He had no significant changes in naming tasks compared with the last assessment (two and a half months after surgery) (Table 1). Although E.S. was evaluated repeatedly with the same naming test, a high test–retest reliability over short intervals (1 to 2 weeks) ($r = 0.91$) in healthy adults and reliability of 0.94 in patients with epilepsy after 8 months [27] in the BNT have been observed.

Compared with his basal cognitive status, the patient did not show memory deficits for non-verbal material (Fig. 3).

To identify possible lesions in the left hemisphere, an MRI was repeated at 6 months after surgery, showing right anterior and mesial temporal lobe surgical sequelae with no structural changes or vascular lesions in the left temporal lobe. Also, we performed an fMRI using language paradigms (semantic and phonemic fluency, verbal generation, and sentence completion), showing left hemisphere cortical activation. Fluency paradigms showed activation in the left antero-inferior frontal cortex (BA44 and BA45), left temporal superior cortex (phonemic fluency) and left posterior dorsolateral cortical association areas. Verbal generation paradigm showed left infero-lateral frontal cortex activation (BA44) and supramarginal and angular gyrus in the left temporal region (BA22). Sentence completion showed featureless activations affecting left antero temporal regions. One of the limitations for analyzing these results is the lack of a previous fMRI that would have helped us to identify cortical reorganization.

Our patient has been free of auras and epileptic seizures and returned to work 6 months after surgery. Until now, there have been no changes in medication.

3. Discussion

We observed the occurrence of a crossed aphasia with a spontaneous and progressive recovery in a right-handed 47-year-old man who had right TLE and who underwent right temporal lobe surgery. Postsurgical left hemisphere lesions due to ischemia, vasospasm or other causes

related to the surgical procedure that would explain the symptoms by the patient were not found.

We considered E.S. to have left language dominance based on strong right-handedness, no family history of left-handedness, and no presurgical difficulties in more than one language skill (reading, naming, comprehension, repetition, and speech) [28]. Although it is possible that MRI findings justify the presurgical cognitive profile (verbal and nonverbal cognitive impairment), like the slightly decreased volume in the left hippocampus that did not meet the criteria of HS [18], we did not give relevance to this finding. In our experience, approximately 18% of the population with an epileptogenic zone in the non-dominant hemisphere and memory impairment [5] showed a deficit in both memories.

Patient E.S. showed postsurgical activation in the left frontal and temporal cortical areas, however, since we lack presurgical fMRI, we cannot affirm, as it seems, if he had only left language dominance or if he had an original bilateral dominance. Furthermore, fMRI studies are useful to determine hemisphere dominance [29], but are less sensitive to determine bilateral atypical bilateral representation. Also, his progressive, spontaneous, fast, and almost complete recovery of postsurgical aphasia supports the assertion that the main language representation is in the left hemisphere. It would be possible that E.S. had an atypical bilateral representation that could have been altered after surgery but was rapidly compensated due to an independent and more represented system on the left side [30–32].

In conclusion, with the experience obtained from this case, in the future, we will suspect atypical language representation in patients with right TLE when any of the following are present: early epilepsy onset (<3 years old), ambidexterity, left-handedness family history, bilateral HS or right HS with slight MRI changes in the hippocampus in the dominant hemisphere, and presurgical verbal memory deficits [28,33–35,30]. In these cases, an exhaustive study for language dominance will be performed using fMRI and/or Wada tests, and a selective amygdalohippocampectomy will be considered as surgical treatment.

Appendix A. Supplementary data

Supplementary data to this article can be found online at http://dx.doi.org/10.1016/j.yebeh.2012.05.014.

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