Epileptic Disord 2016; 18 (1): 77-82

Deterioration of dyslexia after non-dominant temporal lobectomy for drug-resistant epilepsy

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Received September 30, 2015; Accepted January 27, 2016

ABSTRACT – We present a patient with drug-resistant right-sided temporal lobe epilepsy, caused by a ganglioglioma of the parahippocampal gyrus. Preoperatively, the patient was also known to have dyslexia. A right-sided anterior temporal lobectomy, including complete lesionectomy, was performed. Several months after the otherwise uncomplicated procedure, the patient complained about visual memory disturbances, accompanied by increased reading and spelling problems. Postoperative neuropsychological examination revealed deterioration of the visual memory functions, compared to the preoperative assessment, and consequently provided a possible explanation for worsening of the pre-existing dyslexia. In this case report, we hypothesize on the cause of this unusual deterioration and present recommendations to be included in the preoperative epilepsy surgery evaluation for patients with verbal or reading disorders such as dyslexia.

Key words: epilepsy surgery, dyslexia, complication, temporal lobe epilepsy, visual memory

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doi:10.1684/epd.2016.0806

Drug-resistant temporal lobe epilepsy (TLE) is characterized by persistent seizures despite the administration of several first-choice antiepileptic drugs. For these patients, anterior temporal lobectomy (ATL) and amygdalo-hippocampectomy is a successful treatment option with 70-80% of patients demonstrating a favourable seizure outcome (Wiebe *et al.*, 2001; York *et al.*, 2003; Joo *et al.*, 2005).

Associated risks of ATL include, among others, partial but permanent contralateral visual field deficits and potential cognitive decline, such as language or memory impairment (Wiebe *et al.*, 2001; York *et al.*, 2003; Helmstaedter *et al.*, 2004). Partial resection of the language-dominant (mostly left) temporal lobe may give rise to verbal (memory) decline, whereas resections of the non-dominant temporal lobe are sometimes associated with a deterioration of visual memory function (Lee *et al.*, 2002; Vaz, 2004; Sherman *et al.*, 2011).

We report on a patient with drug-resistant temporal lobe epilepsy and a right-sided ATL, including lesionectomy, complicated by postoperative deterioration of pre-existing dyslexia and above average intellectual abilities. Tailby *et al*. described reading deficits as a common co-morbidity in epilepsy patients in general, with a prevalence of 10% (Tailby *et al.*, 2014). Postoperative decline in naming and reading accuracy after ATL is a recently documented complication in the paediatric population and seemingly unrelated to laterality (Lah and Smith, 2015). This independence of laterality was not described in an earlier review on naming outcome in adult epilepsy patients undergoing ATL (Ives-Deliperi and Butler, 2012).

A postoperative deterioration of dyslexia after right-sided ATL has not been described in the literature so far. This unusual complication is of importance for future preoperative language evaluation in TLE patients with known dyslexia.

Case study

A 22-year-old right-handed female was referred to our outpatient clinic for evaluation of epilepsy surgery. Medical history reported an uncomplicated perinatal period, no febrile seizures, no developmental problems, and no history of cerebral trauma. She was diagnosed with dyslexia at primary school. At the time of referral, she was a student at university. The preoperative neuropsychological assessment, of which the results are summarised in *table 1*, confirmed her dyslexia, classified as phonological dyslexia.

The patient experienced her first seizure at the age of 20, followed by complex partial seizures occurring with a frequency of two a week. Seizures started with staring, an upward eye deviation, and lowered consciousness. Seizures were not preceded by an aura. Sometimes seizures were accompanied by automatic behaviour or motor automatisms. The postictal period was characterized by tiredness, drowsiness, and amnesia for the seizure. No phatic disturbances were reported by the patient or her relatives during the postictal period.

Despite treatment with valproic acid, levetiracetam, and lacosamide, a long-lasting seizure-free period was not achieved. Neurological examination was without abnormalities.

MRI of the brain showed a right-sided lesion in the parahippocampal gyrus (*figure 1*) Three months later, a follow-up MRI showed no increase in the size of this lesion. Based on the radiological findings, a low-grade glioma was suspected. Since urgent resection of the lesion was not required, a multidisciplinary work-up for epilepsy surgery was performed.

The preoperative interictal video-EEG showed bilateral frontotemporal epileptiform activity, predominantly on the left side. Ictal video-EEG was not conclusive due to a variable bitemporal ictal onset (left in one seizure, right in another, and no clear lateralization in two seizures) and bilateral temporal involvement during all seizures. Temporal localization was suspected based on seizure semiology, without clear lateralization. The patient did not speak during the precise preictal phase of any of the registered seizures, therefore a speech arrest or dysphasia could not be observed. In the late ictal and postictal phase, a dysphasia was noted but

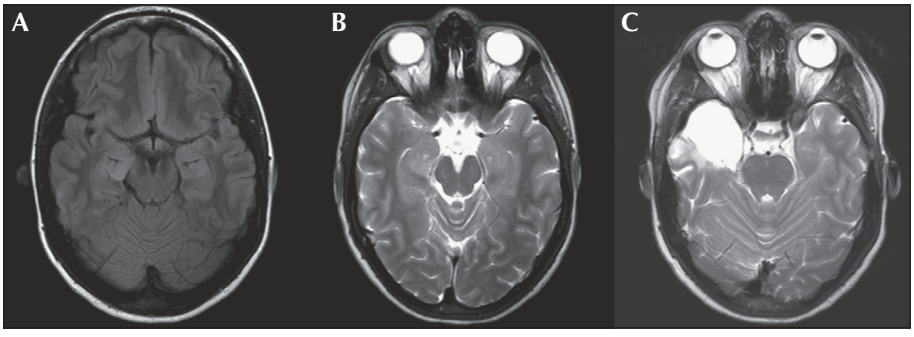

Figure 1. MRI performed in 2012. The FLAIR images (A) show a hyperintense lesion in the right parahippocampal cortex. On the T2-weighted images (B), the lesion appears slightly hypointense. There was no contrast enhancement. Note the impression of a slightly swollen mesiotemporal cortex with little compression on the mesencephalon. (C) Postoperative defect with no abnormalities in the left temporal lobe.

Table 1. Preoperative and postoperative neuropsychological language and memory test results are summarized. The relative score for the WMS IV scores are also included. A clear decline in language but only slightly in general memory function is noted. However, the deterioration in visual memory is evident. UNKA: word production test; WMS IV: Wechsler Memory Scale IV; WAIS-IV-NI: Wechsler Adult Intelligence Scale; N.A.: non-applicable; N.P.: not performed.

Test used	Preoperative score	Preoperative value	Postoperative score	Postoperative value
Language				
Word fluency (UNKA)	61	Average	47	Low average
Dictation (number of faults)	6	N.A.	23	N.A.
Reading task (number of faults)	0	N.A.	3	N.A.
Naming (D-Kefs Color Wordtest)	10	Average	5	Low average
Reading (D-Kefs Colow Wordtest)	8	Average	5	Low average
Memory				
Auditory Memory Index (WMS IV)	97*	Superior	87*	High average
Visual Working Index (WMS IV)	81*	High average	39*	Average
Visual Working Memory (WMS IV)	96*	Superior	N.P.	N.P.
Immediate Memory Index (WMS IV)	92*	Superior	68*	Average
Delayed Memory Index (WMS IV)	92*	Superior	92*	Superior
15-word memory test short term	9**	High average	9*	High average
15-word memory test long-term	8**	Average	8**	Average
Complex figure recall	60-70*	Average	23-30*	Low average
Intelligence (WAIS-IV-NI)				
Total IQ	116	High average		
Verbal Comprehension Index	100	Average		
Perceptual Reasoning Index	121	Superior		
Working Memory Index	105	Average		
Information Processing Index	128	Superior		

*score presented as percentile; **score presented as decile.

language lateralization could not be determined due to bitemporal spread of epileptiform activity on scalp EEG.

In order to define seizure lateralization, a bitemporal implantation of three subdural temporal strips and one hippocampal depth electrode was required. Subsequently, the patient underwent an extraoperative ECoG recording during a two-week registration period. A total of 15 habitual seizures and 28 subclinical seizures were registered. All seizures started in the right hippocampus, with or without rapid spread of the epileptic discharge to the contralateral side. Interictally, epileptiform abnormalities were seen in both hippocampi. No language disturbances were observed during many registered seizures. The other seizures were accompanied by speech arrest with, at that moment, already left hemisphere cortical spread of the epileptiform activity. Because of these ictal language deficits related to left-sided EEG abnormalities, no further assessment of language lateralization was performed.

Preoperative neuropsychological assessment showed an above average total IQ (116) with a significant difference between an average index score on verbal comprehension (100) and an above average index score on the perceptual reasoning index. Word fluency scores and reading performance were reported as low average. The cognitive profile was characterized by average memory functions, except a low average score on the index for visual working memory. The results of the neuropsychological assessment are presented in *table 1*.

Subsequently, a tailored ATL, en bloc amygdalohippocampectomy, and complete lesionectomy in the parahippocampal gyrus was performed. The direct postoperative course was without complications. No seizures were reported during the postoperative hospital stay. Histopathological evaluation showed a ganglioglioma (WHO grade I).

In the first postoperative year, one complex partial seizure and incidental epigastric rising sensations were reported, suggesting a postoperative ILAE class II outcome.

In the first few months after surgery, the patient started complaining about visual memory disturbances. Retrospectively, the patient mentioned the need for visual cues in remembering textual information. Also, she mentioned experiencing more problems in creating these visual cues postoperatively. The difficulties in remembering textual information impeded the patient's activities in daily life, and increased spelling and reading difficulties were noted.

The preoperatively known dyslexia worsened over months after surgery. A postoperative neuropsychological assessment was performed and compared to the preoperative assessment. The assessment showed a decreased memory capacity in general, although significantly more pronounced in visual memory. Also, the patient's performance on standardized reading and spelling tasks showed a three-fold increase in spelling mistakes.

Postoperative MRI did not provide a potential explanation for the worsened reading and spelling tasks (*figure 1C*). The postoperative EEG showed subtle left-sided fronto-centro-temporal focal, sporadic epileptiform activity. Compared with the preoperative EEG, these abnormalities were clearly less pronounced, making this unlikely as an explanation for the deterioration of the dyslexia.

Discussion

Worsening of dyslexia has not previously been described as a complication of right-sided (non-dominant) ATL. A decline in verbal memory after dominant ATL is not unusual (Engman *et al.,* 2004; Joo *et al.,* 2005; Sherman *et al.,* 2011; Baxendale *et al.,* 2013). Deterioration of accuracy of naming and reading after ATL was recently reported in a paediatric population and found to be unrelated to laterality (Lah and Smith, 2015). In a review on ATL in an adult epilepsy patient population, a decline in naming was not found following non-dominant-sided ATL (Ives-Deliperi and Butler, 2012).

Dyslexia is characterized by difficulties with accurate and/or fluent word recognition, resulting in poor spelling and decoding abilities (Shaywitz and Shaywitz, 2005). A deficit in the phonological component of language causes these difficulties, leading to unexpected poor reading and spelling relative to other cognitive abilities. Phonological processing requires adequate frontal lobe executive functions (Vanasse *et al.,* 2005). Not surprisingly, phonological reading difficulties are significantly more common in frontal lobe epilepsy (Vanasse *et al.,* 2005). Also, phonological deficits in reading, as seen in dyslexia, have been described in TLE (Chaix *et al.,* 2006; Tailby *et al.,* 2014). An aetiological role for the left (most often dominant) temporal lobe is suggested based on fMRI studies showing a decreased perfusion in the left temporal neocortex and increased activity in the left inferior frontal region in adults and children with dyslexia (Eckert, 2004; Vanasse *et al.,* 2005). In addition, Tailby *et al.* found that reading difficulty is more common, with a prevalence of ~10%, in epilepsy patients with a focal onset primarily in the temporal lobe (Tailby *et al.,* 2014).

One could hypothesize a right temporal compensatory role for left temporal dysfunction in patients with dyslexia. Shaywitz *et al.* studied young adults with reading disabilities, such as dyslexia (Shaywitz *et al.,* 2003). Based on fMRI results, they suggested that a reading improvement relies on a compensated neural system in the right hemisphere. In our patient, the worsened reading difficulties may have been related to a disrupted compensating neural system after right-sided ATL.

In this specific case, the patient mentioned the need for visual cues to remember textual information. We believe that the visual memory pathway, the input of which starts in the right mesial temporal lobe, is an important compensating mechanism in patients with lower verbal and textual performance. In a review by Vaz (2004) on memory decline after right-sided ATL for TLE, visuospatial memory decline was found in two thirds of the included studies. The worsened dyslexia might therefore be due to disruption of her visual compensation mechanism by the right-sided ATL. This hypothesis is supported by the evident postoperative decline in visual memory function (*table 1*).

In this case, no preoperative Wada testing or functional MRI (fMRI) for language lateralization was performed. Evidence of the benefit of routine use of the Wada test or fMRI in the preoperative evaluation for epilepsy surgery is currently limited (Baxendale, 2009; Bauer *et al.,* 2014). After careful consideration of the risk-benefit ratio, a Wada test or fMRI was not performed in this right-handed patient lacking ictal signs of right-sided speech or language involvement, in whom a standard right-sided ATL was to be performed.

Non-conclusive language dominance based on fMRI would have resulted in a subsequent Wada test. This test could have revealed the side of language predominance and the memory capacity of the hippocampus that should not be resected. However, it would be difficult to identify the impact of visual memory loss on the pre-existing dyslexia during the short testing time frame. Although the latter would prevent postoperative phatic disturbances, it would not have prevented visual memory disturbances which might have been important in this case.

Retrospectively, one could argue that in drug-resistant epilepsy patients with a known language or speech

impairment, such as dyslexia, the preoperative hemispheric dominant (language) lateralization paradigm should include a Wada test or fMRI. An atypical or right-sided language dominance on fMRI would have resulted in a subsequent Wada test or performing an awake craniotomy, respectively. Although the latter would prevent postoperative permanent dysphasia, it would not prevent visual memory disturbances which might have been important in this case. The same holds true for a subsequent Wada test which could reveal the side of language predominance and the memory capacity of the hippocampus that should not be resected. However, it would be difficult to identify the impact of visual memory loss on the pre-existing dyslexia during the short testing time frame.

In conclusion, we have presented a case of evident deterioration of dyslexia after right-sided ATL as a result of drug-resistant temporal lobe epilepsy due to a right mesiotemporal gangioglioma. We suggest that this was caused by a combination of: (1) postoperative disruption of a right-sided cortical compensating neuronal system for language; and (2) postoperative disruption of the hippocampal visual memory mechanism. We hypothesize that due to disruption of these mechanisms, insufficient visual support was generated for sequential information structuring. fMRI or Wada testing should be included as part of the preoperative counselling for patients with a known reading disorder, however, in this case, we believe that this would not have prevented the unusual complication described. □

Supplementary data.
Summary didactic slides are available on the www.epilepticdisorders.com website.

Disclosures.
None of the authors have any conflict of interest to disclose.

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TEST YOURSELF

(1) In a patient submitted to right anterior temporal lobectomy what is the possible cause of the deterioration of dyslexia?

(2) Would it have been more appropriate to perform a preoperative Wada test or fMRI for language lateralization? Would this have prevented the unusual complication?

Note: Reading the manuscript provides an answer to all questions. Correct answers may be accessed on the website, www.epilepticdisorders.com, under the section "The EpiCentre".