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Case Report Unusual atypical language lateralization

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ABSTRACT

Determining the language-dominant hemisphere is essential for planning epilepsy surgery. A 60-year-old righthanded woman with epilepsy since age 16 failed a partial right anterior lobectomy at age 21. Later, a brain MRI found extensive right-sided cortical dysplasia and periventricular heterotopia. Subsequently, prolonged video-EEG monitoring localized her seizures to the right temporoparietal region. Functional MRI was inconclusive in lateralizing her language, prompting a Wada test, which strongly lateralized language to the right. This unique case of atypical language representation in a right-handed individual with an extensive right-hemispheric congenital malformation and seizure focus illustrates the important thorough presurgical language assessment. © 2016 The Authors. Published by Elsevier Inc. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

In the general population, 92–96% of right-handed individuals and 50–70% of left-handed individuals are left hemispheric-dominant for language [1].

Language lateralization is an essential component of the surgical evaluation of patients with medically intractable epilepsy. Determining the language-dominant hemisphere is essential for tailoring the resection of the epileptogenic zone and predicting functional outcome [2]. Over the past half-century, a multitude of tests were developed in an attempt to reliably lateralize and localize eloquent areas of the brain involved in speech production and comprehension. Among those, the gold standard for language and memory lateralization in patients with epilepsy is the Intracarotid Amobarbital Procedure (IAP) or the Wada test [3]. Newer techniques, including fMRI and MEG, have since been developed to replace the Wada, providing a noninvasive method to serve the same purpose. With refined language tasks, fMRI results are greatly concordant with those of the Wada test, which has led many centers to rely on fMRI solely for language lateralization. However, occasional clinical situations will still necessitate the Wada test.

In patients with epilepsy, and specifically those with dominant hemispheric seizure foci and early seizure onset age, language processing tends to reorganize outside of its typical brain regions. Various factors have been suggested to increase the chances of the reorganization of language functions intrahemispherically and interhemispherically, sometimes leading to atypical language representations.

We report a 60-year-old right-handed woman with epilepsy since the age of 16. The patient's seizures were described as frequent staring spells, at times associated with manual and oral automatisms and occasionally followed by secondarily generalized tonic–clonic seizures. In 1972, at the age of 21, the patient underwent a partial right anterior lobectomy to resect a "lesion" suspected to be the cause of her epilepsy. The patient was reportedly seizure-free for approximately 2 years before her seizures recurred. Antiepileptic medications failed to control the patient's seizures over the years, and she was referred for a presurgical evaluation. In December 2008, a brain MRI showed extensive cortical dysplasia and heterotopia involving the right temporal lobe, the right lateral ventricle, and the white matter adjacent to the right lateral ventricle. Additionally, it showed a resection cavity in the right temporal lobe and increased volume of the right hippocampus compared with that of the left. Subsequently, a prolonged video-EEG monitoring session suggested an epileptogenic zone in the right temporoparietal region. As part of the evaluation, a functional MRI was performed in October 2009 which was inconclusive as to language lateralization and localization. Although the likelihood of right language dominance was too low owing to her right-handedness and congenital right-hemispheric malformation of cortical development, the equivocal results of the fMRI prompted further evaluation of language and memory with a Wada test. The Wada test lateralized language to the right hemisphere while memory was bilateral. An invasive video-EEG monitoring session with a subdural grid and multiple depth electrodes was carried out in an attempt to further localize the epileptogenic focus and proceed with surgical resection. The results of the latter evaluation were concordant with previous findings, confirming the right temporoparietal epileptogenic focus. However, careful mapping failed to reveal language areas underlying the subdural grid. Being unable to precisely localize language in the right hemisphere, a unanimous decision was reached not to proceed with surgery and to continue managing the patient with medications since the data gathered would have poorly predicted surgical outcome in terms of language and speech (Fig. 1).

This case illustrates the importance of the Wada test in determining language dominance prior to epilepsy surgery, especially when fMRI is

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Fig. 1. MRI brain T 1 axial cut. Arrow pointing to periventricular heterotopia. It also shows resection cavity in the right temporal lobe.

inconclusive or shows evidence of atypical language representation. Reports of individuals with epilepsy due to right-hemispheric lesions studied for language dominance are rare. One large study compared language dominance in right- and left-handed epileptic subjects with hemispheric lesions, classified as "early" and "late". There was no difference in left-hemispheric dominance for language in right-handed patients with lesions in the left hemisphere compared with that in patients with no lesions. However, left-handed patients with congenital or perinatally acquired left hemispheric lesions were predominantly right hemisphere-dominant for language. Only two cases with righthemispheric lesions and right language dominance were described in this large series. Both patients were left-handed, and only one patient had an early lesion described as a right frontal cortical dysplasia while the other had a late-onset right parietoocciptal stroke [3]. Our unique case does not fall in any of the categories described above. Some of the disadvantages and complications of the Wada test are outweighed by the benefit of providing crucial information that can either result in favorable outcomes or prevent disastrous ones.

Conflict of interest

The authors declare that there are no conflicts of interest.

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