Speech arrest with stimulation may not reliably predict language deficit after epilepsy surgery

Abstract—The authors present a patient in whom electrical cortical stimulation of the posterior temporal cortex induced speech arrest, comprehension deficits, and other language-related impairments. This area was ultimately resected because of persistence of a severe seizure disorder. No postoperative aphasia was observed despite the cortical stimulation results, and the patient is since seizure free. These findings question the well-established principle that corticography directly reflects local cortical functions in all patients.

NEUROLOGY 2006;66:1–1

M. Seeck, MD; A.J. Pegna, PhD; S. Ortigue, PhD; L. Spinelli, PhD; C.A. Dessibourg, MD; J. Delavelle, MD; O. Blanke, MD; C.M. Michel, PhD; T. Landis, MD; and J.-G. Villemure, MD

fn1 The decision regarding whether, and to what extent, pathologic cerebral tissue can be surgically removed depends on successful identification of the functionally essential cortex, which must be spared. The location of language functions show considerable interindividual variability and must be individually determined.[1-3] Despite recent progress in functional neuroimaging methods, intraoperative or extraoperative mapping by electrical stimulation is still considered to be the gold standard, and the decision to operate is usually based on this invasive procedure. Here we present a patient in whom the posterior language cortex was localized with electrical cortical stimulations, but in whom resective surgery of the region was performed with no subsequent aphasia.

Case report. The patient is a 20-year-old, right-handed (100% right-handed as determined by the Edinburgh handedness questionnaire[4]) woman who had pharmacoresistent epilepsy since age 7 years. Her neurologic examination results were normal. Loss of consciousness and clonic movements of the right hemibody, followed by postictal aphasia, characterized her first seizures. Later, she also presented acoustic auras. A presurgical workup was performed, revealing an active epileptogenic focus over the left posterior temporal region. A high-resolution MRI revealed a focal transmantle dysplasia in the posterior aspect of the left superior and middle temporal gyrus (figure 1). F1

Before surgery, the neuropsychological examination was characterized by several deficits in language-based tasks, suggesting a moderate aphasic anomia. Indeed, spontaneous speech was fluent but with circumlocutions due to her word-finding difficulties. Her results on the Boston Naming test (BNT) were below the normal limits (14/34 on an abridged version of the test). Oral comprehension and written language were unimpaired, as the remainder of the examination.

Immediately after the seizures, naming, comprehension, and reading were marked by additional difficulties and hesitations. Thus, the neuropsychological evaluation suggested the presence of a language deficit consequent to the left temporal epileptic focus, and phase II evaluation with chronically implanted subdural electrodes was conducted (figure 2). F2

Invasive EEG recordings revealed consistent seizure onset in the posterior superior and middle temporal gyri. Functional mapping with bipolar electrical stimulation was performed at all grid electrodes. The following tasks were used: spontaneous speech, naming (e.g., of common objects), oral comprehension (simple and complex orders), writing, and reading of short stories or texts. Established stimulation parameters were used (bipolar stimulation of 50-Hz pulse rate, pulse duration 300 μs, train duration 2 to 5 seconds, intensity 1 to 11 mA). Stimulation was gradually increased until either a response or afterdischarges occurred. If no responses were elicited until a maximal intensity of 11 mA, stimulation was discontinued at that site. Stimulation of several contacts over the left temporoparietal cortex repeatedly resulted in speech arrest, as well as comprehension and other difficulties in language-related functions (see figure 2). On the basis of the electrical stimulation findings, both language areas were identified in typical cortical sites in this patient.

Resection of the anterior aspect of the superior and partially middle temporal gyrus was performed under local anesthesia as well as perioperative cortical stimulation and speech monitoring. Three subpial transsection incisions over the speech interference area were performed as well to improve postoperative seizure control. The histopathologic analysis revealed severe cortical dysplasia with balloon cells.

Postoperatively, the seizures stopped for 1 week, but recurred after 4 weeks with the same frequency as preoperatively. Clinical assessment of the patient's oral comprehension and spontaneous speech showed no obvious modification. On the BNT, she remained at her presurgical level of performance, with 15 of 34 correct responses. All other cognitive functions tested (written language, praxis, visual gnosis, executive function, and memory) also remained unchanged.

A reintervention was considered on the basis of nearly absent aggravation of the patient's language capacities. Intraarterial Amytal injection (IAT) in the left and right carotid artery suggesting bilateral language representation with predominance in the left hemisphere, except for oral comprehension, which apparently relied more on right than left hemispheric functions. The seizure disorder worsened, and on the basis of the IAT and the relatively good postoperative outcome in terms of language functions after the subtotal resection, a complete resection was performed 8 months later (see figure 2).

A brief bedside examination conducted the day after the second resection showed no change in the patient's language capacity. A formal postsurgical neuropsychological examination was performed 3 and 6 months later. This revealed a worsening of her word-finding difficulties observed both in her spontaneous speech, marked by a greater amount of circumlocutions, and in the BNT (7/34). The remainder of the psychometric neuropsychological assessment was perfectly comparable to the preoperative baseline.

The patient has since been seizure free (follow-up 2 years, 4 months).

From the Presurgical Epilepsy Evaluation Unit, "Functional Neurology and Neurosurgery Program" of the Universities of Geneva and Lausanne (M.S., L.S., C.A.D.), Departments of Neurology (A.J.P., S.O., O.B., C.M.M., T.L.) and Radiology (J.D.), University Hospital of Geneva; and the Department of Neurosurgery (J.-G.V.), University Hospital of Lausanne, Switzerland. C.A.D. is also affiliated with the Institute Pédagogie Curative, University of Fribourg, Switzerland.

Supported by SNF grants (nos. 3200B0-104146, 3200-068105).

Disclosure: The authors report no conflicts of interest.

Received March 17, 2005. Accepted in final form November 1, 2005.

Address correspondence and reprint requests to Dr. Margitta Seeck, Department of Neurology, Hôpital Cantonal Universitaire de Genève, 24 rue Micheli-du-Crest, CH-1211 Genève 14, Switzerland; e-mail: margitta.seeck@hcuge.ch

<article article-type="brief-report"> •
<article-id pub-id-type="doi">10.1212/01.wnl.0000199254.67398.a7</article-id>

AQ: 1

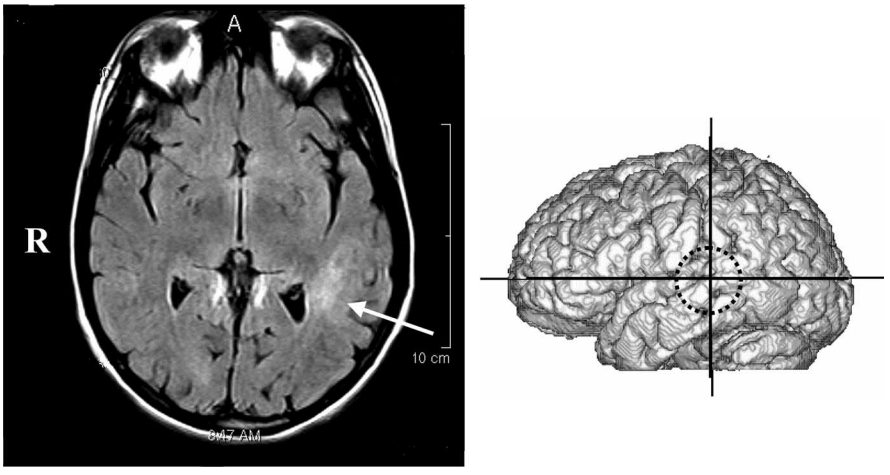

Figure 1. Left: MRI of our patient shows the focal transmantle dysplasia between the ventricle and the superior and middle temporal gyrus in the left hemisphere. Fluid-attenuated inversion recovery image sequence shows a large area of hyperintensity in the left mid to posterior temporal region (white flash). Right: Abnormal area (encircled) projected onto the left hemisphere of the reconstructed patient's MRI.

Discussion. Although language functions were impaired or blocked during electrical stimulation of contacts over the left temporoparietal cortex, postoperative (Wernicke) aphasia has not occurred since the resection of this brain region. A moderate aggravation of the preexisting word-finding difficulties was noted, but spontaneous speech and oral comprehension remained preserved.

The deficits obtained by subdural electrical stimulation are similar to those described in previous reports on electrical stimulation of the posterior language cortical area.[5] Our observations suggest that the stimulated cortex could be part of a larger network and that electrical stimulation may interfere not only with underlying cortex, but also with remote cortical language functions. For example, in this patient, oral comprehension seemed to be subserved mainly by the right hemisphere, as suggested by the IAT, but comprehension deficits were also clearly induced by electrical stimulation of the left temporoparietal cortex.

The observation that electrocorticography may not necessarily reflect local cortical function is also suggested by previous cortical stimulation studies. Equal occurrence of comprehension deficits during stimulation of the Broca and Wernicke areas, i.e., the specific dichotomy "language production–frontal cortex vs comprehension–posterior temporal cortex" as seen in stroke patients has been reported to be less reliably observed with electrical stimulation.[5]

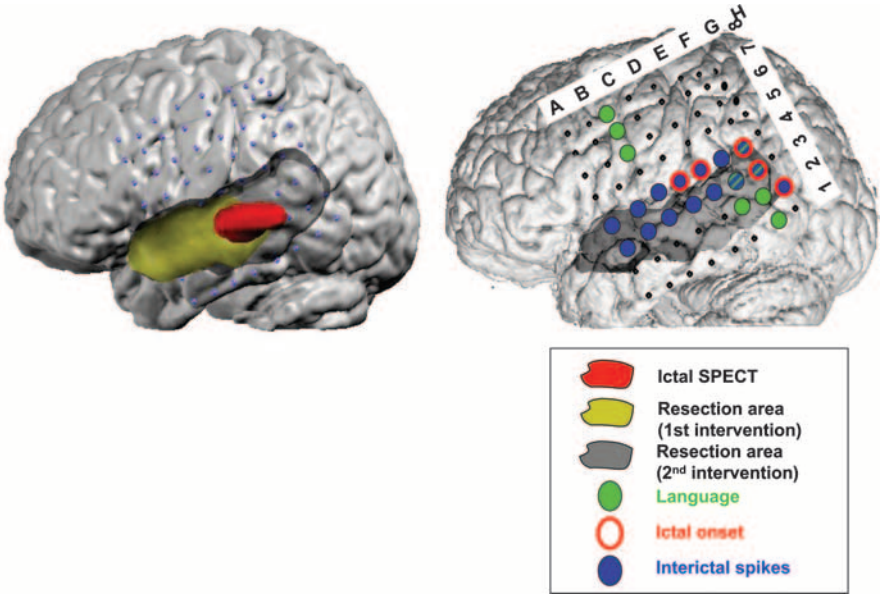

Figure 2. Left: Left hemisphere of patient's brain. Yellow shaded area corresponds to the tissue resected during the first operation; gray shaded area corresponds to the second operation 8 months later. Right: Results of the electrical cortical stimulation. Grid electrode (8 × 8) positions determined by coregistration of the preimplantation and postimplantation MRI. The contacts where electrical stimulation interfered with language functions are in green. The blue contacts indicate the zone of interictal spikes, whereas the contacts encircled in red correspond to the ictal onset zone. Contacts filled with green and blue stripes indicate the presence of both the interictal epileptogenic zone and area of stimulation-related speech interference. In contact G2–G3, and the adjacent contacts G1–G2 and F2–F3, the patient showed speech arrest and could not name the objects on the cards or made errors in naming these objects. These findings were obtained with currents between 5 and 7 mA. Oral comprehension was unimpaired in G1–G2, but interrupted in G2–G3 and impaired in G3–G4. In contrast, stimulation of contact F2–F3 (7 mA, 2 seconds) also produced speech arrest, but oral comprehension was not disturbed. Reading and writing were similarly impaired in most contacts. For example, the patient wrote "vognage" instead of "voyage." This type of error was found during stimulation of contacts G2–G3, G3–G4, and F2–F3. Stimulation of contacts C6–C7 and C7–C8, covering mainly the middle frontal gyrus, resulted also in speech arrest and impaired oral comprehension as well as difficulties in reading and writing. No other movement (tongue, mouth, arm) was elicited or impaired by the stimulation.

The type and the timing of the lesion may explain the absence of posterior temporal implication in speech and comprehension in our patient. Dysplasia with balloon cells was found in the histopathologic analysis, which may point to early cortical reorganization. A recent study found evidence that cortical lesions with balloon cells are less likely to harbor physiologic activity as compared with dysplastic lesions without balloon cells.[6] Transfer of language to the right hemisphere has been found after early injuries in young children, i.e., before age 5 to 9 years, although dysplasia is not necessarily associated with language relocation.[7-9] Because some aggravation was noted after the second operation, the resected region participated to a certain degree in language-related tasks; however, its resection did not lead to major speech or comprehension alterations. Plasticity as a result of the first intervention seems a less likely mechanism, given that the patient had almost no changes in her neuropsychological test results after the first operation and that the delay between both interventions was relatively short.

Our results do not imply that cortical stimulation is of no value. The intraoperatively or preoperatively identified language cortex should remain preserved in patients with an acquired lesion or in whom a Wada test result or other evidence clearly suggests unilateral left hemispheric language lateralization. Significant transient or even persistent postoperative neuropsychological deficits have been reported to occur in patients in whom the resection was too close to the posterior language cortex.[10]

Acknowledgment

The authors thank Dr. Zaim and Dr. Murray for comments on the manuscript.

References

1. Penfield W, Jasper H. Vocalization and speech. In: Epilepsy and the functional anatomy of the human brain. Boston, Little & Brown; 1954: 109–113.
2. Ojemann GA. Individual variability in cortical localization of language. J Neurosurg 1979;50:164–9.
3. Lesser RP, Lüders H, Morris HH, et al. Electrical stimulation of Wernicke's area interferes with comprehension. Neurology 1986;36:658–663.
4. Oldfield RC. The assessment and analysis of handedness: the Edinburgh inventory. Neuropsychologia 1971;9:97–113.
5. Schäffler L, Lüders HO, Beck GJ. Quantitative comparison of language deficits produced by extraoperative electrical stimulation of Broca's, Wernicke's, and basal temporal language areas. Epilepsia 1996;37:463–475.
6. Marusic P, Najm IM, Ying Z, et al. Focal cortical dysplasias in eloquent cortex: functional characteristics and correlation with MRI and histopathological changes. Epilepsia 2002;43:27–32.
7. Duchowny M, Jayakar P, Harvey AS, et al. Language cortex representation effects of developmental versus acquired pathology. Ann Neurol 1996;40:31–38.
8. Boatman D, Freeman J, Vining E, et al. Language recovery after left hemispherectomy in children with late-onset seizures. Ann Neurol 1999;46:579–586.
9. Janszky J, Ebner A, Kruse B, et al. Functional organization of the brain with malformations of cortical development. Ann Neurol 2003;53:759–767.
10. Haglund MM, Berger MS, Shamseldin M, Lettich E, Ojemann GA. Cortical localization of temporal lobe language sites in patients with gliomas. Neurosurgery 1994;34:567–576.