

Pearls & Oysters: Selective postictal aphasia

Cerebral language organization in bilingual patients



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ABSTRACT

Background: Ictal and postictal language dysfunction is common and strongly predictive of language laterality in monolingual patients. For bilingual patients, selective dysfunction has been reported for a single language with focal cerebral lesions, electrical brain stimulation, and intracarotid sodium amytal.

Methods: Two right-handed Ukrainian-English bilingual patients with left perisylvian structural lesions, late onset complex-partial seizures, and postictal aphasia for English are presented and discussed with regard to mechanisms of selective aphasia and factors contributory to language lateralization in bilingual patients.

Results: Ukrainian was the native language of both patients with English acquired after 7 years of age. Regular/video-EEG showed left temporal epileptogenesis. A 56-year-old man, who had a left hemorrhagic stroke at age 50 and had not spoken Ukrainian for 40 years, was unable to speak English for ~20 minutes postictally but had global preservation of Ukrainian. A 71-year-old woman, who had a left temporal epidermoid cyst and had not spoken Ukrainian since childhood, had 10- to 15-minute postictal expressive aphasia in English but not Ukrainian and preservation of comprehension in both languages.

Conclusions: These cases are instructive and consistent with the literature on cerebral organization of language in bilingual individuals. For both patients, postictal aphasia with preservation of Ukrainian is consistent with findings from clinical and experimental studies indicating that later age of second language acquisition (>6 years) rather than language proficiency is a primary factor in language laterality. Second, global aphasia in the man with a late lesion vs expressive aphasia with preservation of comprehension of English in the woman with a prenatal/early lesion is consistent with the atypical language laterality described for individuals with left-sided lesions sustained prior to age 5. Although neither Wada test nor fMRI was done to assure left hemisphere laterality of spoken Ukrainian and English, this is probable, and the selective postictal aphasia observed for both patients is consistent with the differential intrahemispheric organization reported for the two languages of bilingual individuals. Possible mechanisms of selective postictal aphasia, e.g., active inhibition, and differences in language networks in bilingual patients are discussed. *Neurology*® 2008;71:e14-e17

Ictal and postictal language dysfunction is common and strongly predictive of language laterality in monolingual patients. For bilingual patients, selective dysfunction has been reported for a single language with focal cerebral lesions, electrical brain stimulation, and intracarotid sodium amytal. We report two right-handed Ukrainian-English bilingual patients with left perisylvian structural lesions, late onset complex-partial seizures, and postictal aphasia for English, and discuss mechanisms of selective aphasia and factors contributory to language lateralization in bilingual patients.

CASE HISTORIES **Case A.** The first patient was a 56-year-old, right-handed bilingual man born to Ukrainian parents, who first learned to speak English at the age of 7. At 50 years of age, he sustained a hemorrhagic stroke involving the left perisylvian region due to a ruptured anterior communicating aneurysm. Several

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Disclosure: The authors report no disclosures.

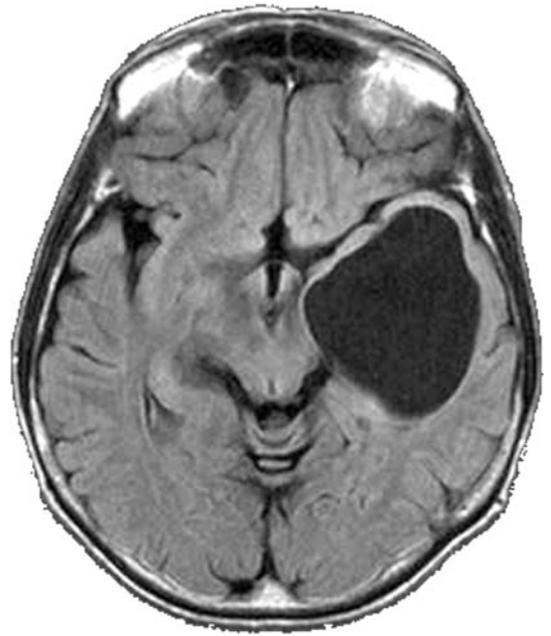
months later, he began having seizures. The semiology of his seizures was characterized by recurrent episodes of confusion lasting 1–2 minutes with secondary progression to generalized tonic-clonic seizures. Seizures were stereotypically followed by inability to speak English for 15 to 20 minutes with preservation of his native Ukrainian. Interestingly, he had not spoken Ukrainian for the past 40 years and his daily communication, even at home, was entirely in English. Unfortunately, the content of his verbal output could not be translated during the postictal period. On admission, cranial CT showed encephalomalacia of the left frontotemporal operculum from the previous stroke. EEG recording displayed paroxysmal epileptiform discharges emanating from the left temporal region. Ultimately, total control of seizures was achieved by carbamazepine monotherapy.

Case B. This was a 71-year-old, right-handed, bilingual woman born to Ukrainian parents. She learned English after beginning school. At 52 years of age she presented with recurrent episodes of disorientation that occasionally evolved into generalized tonic-clonic seizures lasting for 10 minutes. Postictally, her comprehension of English remained intact but her responses to questions were in normal Ukrainian as translated by an interpreter. This unilingual expressive aphasia in English lasted for 10–15 minutes with gradual recovery. Of note is that her day-to-day communications were mainly in English for the last 50 years. The cranial CT revealed a left frontotemporal perisylvian cyst which was subsequently resected and identified as an epidermoid cyst (figure 1). EEG recording showed interictal epileptiform discharges and frequent focal seizures originating in the left temporal region (figure 2). She continues to have infrequent seizures following surgery.

DISCUSSION The approach to an analysis of selective aphasia in multilingual patients can be facilitated using the following conceptual parameters, as discussed below: laterality of language function, intrahemispheric organization of language, patterns of language recovery, and higher cortical control and switching mechanisms.

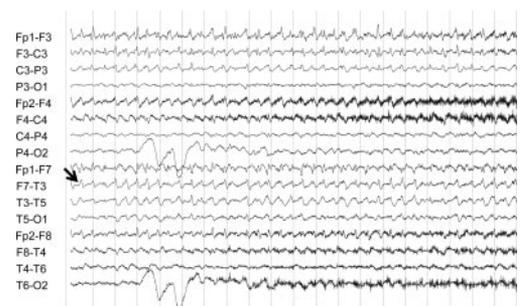
Laterality of language functions in multilingual individuals. The cortical representation of different languages in multilingual individuals is controversial because of inconsistent findings in the literature.^{1–3} Two key hypotheses have been proposed on the basis of experimental studies of normal multilingual subjects: the age and the stage hypotheses.¹ The age hypothesis emphasizes the age of second language (L2) acquisition as most important in L2 laterality; acquisition after 6 years

Figure 1 Cranial CT revealing a left frontotemporal perisylvian cyst which was subsequently resected and identified as an epidermoid cyst



results in identical lateralization of L1 and L2. The stage hypothesis emphasizes that degree of L2 proficiency is the primary determinant of language laterality. In a meta-analysis of 66 behavioral studies of language laterality, monolingual subjects and late bilingual subjects showed a reliable left hemispheric dominance across language tasks regardless of proficiency, whereas early bilingual subjects demonstrated a tendency for bihemispheric contributions.^{1,2} Left hemispheric dominance was more prominent in those less proficient in L2, especially if the L2 was English. Hence, earlier acquisition of L2 predicts a divergent pattern of cerebral lateralization with right hemispheric contributions. Both of our patients were

Figure 2 EEG recording showing interictal epileptiform discharges and frequent focal seizures originating in the left temporal region



late bilingual individuals who acquired English after the age of 6 and accordingly would have left hemisphere dominance for both languages. The dual aphasia with rapid L1 recovery in strict left-sided lesions argues in favor of the contention that age rather than proficiency was the primary determinant of language laterality in our patients.

Intrahemispheric organization of language in multilingual subjects. Various clinical studies support the differential localization model in bilingual patients where two languages occupy distinct but overlapping loci within the dominant hemisphere. Electrocortical stimulation of the dominant perisylvian region in bilingual patients revealed different but overlapping areas for each language.²⁻⁴ Sequential recovery of two languages over different time courses was also observed after the Wada test.⁵ At the lexical level, fMRI studies confirmed the shared neural substrates for L1 and L2 with distinct neuronal populations displaying language specific responses.⁶ The lesions in both of our patients spanned gross anatomic landmarks and disruption of both languages would have been expected. The sequential recovery may be explained by the differential localization model with limited seizure propagation to the neural population of English and relative sparing of Ukrainian. The fact that Case B postictally comprehended both languages but spoke only English could implicate minimal posterior seizure spread and/or right hemisphere representation of comprehension associated with her epidermoid cyst acquired at an early age. Nevertheless, the concept of separate but overlapping topography for each language remains too simple to explain this phenomenon. The other possibility is that the complexity of the network for each language could be different and perhaps related to the time of acquisition. A language acquired earlier may have a more extensive and thus more resistant network, whereas one established later would have a more superficial and easily disrupted network.

Patterns of language recovery in multilingual patients. Six major patterns of language recovery in bilingual patients have been historically observed.^{7,8}

1. Parallel recovery when both languages are impaired and restored at the same pace.
2. Differential recovery when languages recover at different rates relative to their premorbid levels.
3. Selective recovery (30%) with recovery of one language but not the other.
4. Antagonistic recovery where one language recovers to a certain extent before regression occurs as the other language begins to recover.
5. Successive recovery where recovery of L2 begins after L1 has recovered.

6. Mixed recovery with mutual interference between languages during recovery process.

These descriptive categories emphasize the possible phenomenologic patterns of recovery and do not correlate with specific anatomic or pathologic processes. Both of our patients had not been speaking Ukrainian for many years, and the second patient responded appropriately to commands in English and correctly answered, in Ukrainian, questions presented in English. The recovery in both cases was difficult to categorize due to their short-lasting deficits in English and the inherent difficulties in conducting formal neuropsychological assessments postictally to quantify the relative recovery for both languages.

Higher cortical control and switching mechanisms. Volitional alternation of verbal output between two languages is under sustained higher cognitive system control. Involuntary intrusions of linguistic elements from different languages represent pathologic switching or mixing. Pathologic switching is a higher cortical disorder of communication that usually relates to frontal lobe lesions. Pathologic mixing is an aphasic disorder that is generally associated with fluent aphasias due to left postrolandic lesions, and in bilingual aphasics with dementia.⁹ The anterior loop of language that mediates language planning comprises the cortico-subcortical circuit between the prefrontal cortex and basal ganglia in the dominant hemisphere.¹⁰ This loop is the neuroanatomic device for language selection control, and functional neuroimaging demonstrates resolution of pathologic switching and mixing with functional restoration of this cortico-subcortical loop.¹¹ Both patients had structural lesions spanning the frontotemporal operculum with minor involvement of the white matter connecting the frontal lobe with the basal ganglia. Neither mixing nor switching was observed interictally or postictally.

Postictal aphasia. The mechanism underlying postictal aphasia (PA) may be related to active inhibition or postictal exhaustion induced by ictal activities in language areas and their connections.¹² The literature is relatively scarce in substantiating the prevalence of PA in both monolingual and bilingual patients. In one series of 68 patients with temporal lobe epilepsy (TLE), PA was found in 51.6% and correctly lateralized the seizure onset to the dominant hemisphere in 81.3%.¹³ In another review, 92% of patients with TLE with PA had their seizures originating from the dominant temporal lobe.¹⁴ In another series of 64 patients with TLE, the positive predictive value of PA in lateralizing seizure onset to the dominant side was 80%.¹⁵ The seizures in both of our patients orig-

inated electrographically from the left frontotemporal region and correlated with PA for both languages. Nonetheless, objective evaluations of PA are difficult due to the overlap with postictal behaviors like postictal confusion and fatigue that can confound any linguistic analysis of postictal speech. This will be further complicated in cases of bihemispheric speech representation or with seizure propagation to the contralateral hemisphere or, conceivably, in multilingual patients. The immediate recovery of Ukrainian may reflect the relative resistance of the native language to postictal mechanisms, or simply and incidentally, relates to limited seizure propagations into their neural population in both patients.

CONCLUSION Focal seizures originating from the dominant perisylvian regions resulted in postictal aphasia involving mainly the second language in two late bilingual patients. The diffuse lesions in both patients do not conform to isolated anatomic or language-specific territories. Postictal inhibition may selectively involve one circuit subserving one language. This corroborates the notion that neurobiological substrates mediating two languages share a common topographic anatomy with relatively distinct functional circuits. Arguably, disruption of distinct switch mechanisms rather than an isolated functional circuitry of one language could be the defining pathology of this rather unusual phenomenon.

CLINICAL PEARLS

1. Monolingual and late bilingual patients have a reliable left hemispheric dominance across language tasks regardless of proficiency, whereas early bilingual patients demonstrate a tendency for bihemispheric contributions.
2. The differential localization model of bilingual speech indicates that the two languages occupy distinct but overlapping loci or functional circuits within the dominant perisylvian region.

REFERENCES

1. Hull R, Vaid J. Bilingual language lateralization: a meta-analytic tale of two hemispheres. *Neuropsychologia* 2007; 45:1987–2008.
2. Ojemann GA, Whitaker HA. The bilingual brain. *Arch Neurol* 1978;35:409–412.
3. Rapport RL, Tan CT, Whitaker HA. Language function and dysfunction among Chinese-and English speaking polyglots: cortical stimulation, Wada testing and clinical studies. *Brain Lang* 1983;18:342–366.
4. Roux FE, Lubrano V, Lauwers-Cances V, Trémoulet M, Mascott CR, Démonet JF. Intra-operative mapping of cortical areas involved in reading in mono- and bilingual patients. *Brain* 2004;8:1796–1810.
5. Berthier ML, Starkstein SE, Lylyk P, Leiguarda R. Differential recovery of languages in a bilingual patient: a case study using selective amygdala test. *Brain Lang* 1990;38:449–453.
6. Klein D, Zatorre RJ, Chen JK, et al. Bilingual brain organization: a functional magnetic resonance adaptation study. *Neuroimage* 2006;31:366–375.
7. Pearce JM. A note on aphasia in bilingual patients: Pitres' and Ribot's Laws. *Eur Neurol* 2005;54:127–31.
8. Paradis M. Bilingualism and aphasia. In: Whitaker H, Whitaker HA, eds. *Studies in Neurolinguistics*. New York: Academic Press, 1977;3:65–121.
9. Fabbro F. The bilingual brain: cerebral representation of languages. *Brain Lang* 2001;79:211–222.
10. Fabbro F, Skrap M, Aglioti S. Pathological switching between languages after frontal lesions in a bilingual patient. *J Neurol Neurosurg Psychiatry* 2000;68:650–652.
11. Marien P, Abutalebi J, Engelborghs S, De Deyn, PP. Pathophysiology of language switching and mixing in an early bilingual child with subcortical aphasia. *Neurocase* 2005;11:385–398.
12. Adam C, Adam C, Rouleau I, Saint-Hilaire JM. Postictal aphasia and paresis: a clinical and intracerebral EEG study. *Can J Neurol Sci* 2000;27:49–54.
13. Leutmezer F, Baumgartner C. Postictal signs of lateralizing and localizing significance. *Epileptic Disord* 2002;4:43–48.
14. Gabr M, Luders H, Dinner D, Morris H, Wyllie E. Speech manifestations in lateralization of temporal lobe seizures. *Ann Neurol* 1989;25:82–87.
15. Privitera M, Kohler C, Cahill W, Yeh H. Postictal language dysfunction in patients with right or bilateral hemispheric language localization. *Epilepsia* 1996;37: 936–941.

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Neurology 2008;71:e14-e17

DOI 10.1212/01.wnl.0000325017.42998.d1

This information is current as of August 11, 2008

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