SHORT REPORT

Aphasia after hemispherectomy in an adult with early onset epilepsy and hemiplegia

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A 55 year old left handed man with left hemisphere subcortical encephalomalacia, seizures, language impairment, and right hemiparesis from a motor vehicle accident at age five was evaluated for epilepsy surgery. The patient continued to speak and followed commands during a left intracarotid amobarbital test (IAT). Left functional hemispherectomy resulted in expressive aphasia. Based on postoperative outcome, language was bilateral. The injury after primary development of language function, the predominantly subcortical lesion, and the late timing of surgical intervention well past development and plasticity may have been factors in the emergence of postoperative aphasia.

In patients with epilepsy and hemiplegia caused by severe left hemisphere injury sustained before six years of age, language function usually develops in the right hemisphere.[1] The ''transferred'' language is usually not normal,[2] but typically there is no significant worsening after left hemispherectomy carried out for treatment of intractable epilepsy.[3] We report a patient with early brain injury and hemiplegia who had persistent expressive aphasia following left hemispherectomy at 55 years of age. The patient's bilateral preoperative language function was not suspected preoperatively from a left sided intracarotid amobarbital test (IAT).

CASE REPORT

A 55 year old man with right hemiparesis and intractable seizures caused by head trauma (a motor vehicle accident) at five years of age was evaluated for epilepsy surgery. The patient was in coma for a month after the accident, and a left fronto-temporal subdural haematoma was evacuated. Following the accident, he had spastic hemiparesis and language decline with reduced vocabulary, word finding difficulties, and regression to two word sentences. Handedness shifted from right to left. Seizures started one month after the accident.

Seizures started with tingling in the legs, nausea, and fear; these symptoms were followed by unresponsiveness, automatisms, forceful right head and eye version, and secondary generalisation. Aura frequency ranged from 30 to 140 per month. Seizures with evolution into automatisms occurred 15 times per month and secondary generalised convulsions occurred two times per month.

Neurological examination revealed a right spastic hemiparesis with decreased power (4/5) in the proximal upper and lower limb, and no power (0/5) in his right hand, without preserved fine hand movements. Gait was hemiparetic with circumduction of the right leg. Formal neuro-ophthalmological visual field testing revealed a non-specific inferior arcuate scotoma in the right eye. The visual field of the left eye was normal.

Continuous digital video-EEG monitoring at age 53 showed left fronto-temporal, left fronto-centro-parietal, and left parietal interictal sharp waves. Four habitual seizures were recorded with ictal EEG lateralised to the left hemisphere, maximum in the left posterior head regions. EEG seizures started with rhythmic slow waves in the theta range, lateralised to the left with a left temporo-parietal maximum, and evolved within 15 to 20 seconds into rhythmic widespread left hemispheric polyspiking. Spread to the contralateral hemisphere was seen within 30 seconds.

Magnetic resonance imaging (MRI) showed a cystic cavity within the left hemisphere involving the left caudate nucleus, putamen, external capsule, and corona radiata, as well as parenchymal volume loss with prominence of the sulci in the suprasylvian and inferior frontal regions (fig 1).

Language rating during a left sided IAT was based on performance in different components of language (spontaneous speech, comprehension tested by requesting the patient to execute simple commands, repetition of words, confrontation naming, and reading). Additionally, the patient was monitored for paraphasic and dysarthric errors.[4] Memory testing was done by presentation of 16 words and objects during the procedure (pictures, designs, function words, and object words or sentences).[5]

After injection of 100 mg amobarbital the patient continued to count while his right sided hemiparesis worsened from 4/5 in the proximal right arm and leg at baseline to left sided hemiplegia (0/5) lasting 137 seconds. He then slowly recovered and strength returned to baseline after a total of 272 seconds. The patient was able to follow commands, repeat words, name all presented objects (six), and read all presented words and sentences (six) during the 137 seconds while hemiplegia was complete. Four additional items (one picture, one design, one object word, and one function word) were read and named by the patient during recovery from hemiparesis. Good filling of both the left anterior and the left middle cerebral artery was seen on angiography, with no crossover to the other hemisphere and no abnormal vessel pattern. An EEG during the left hemispheric IAT showed left hemispheric delta slowing with a frontal maximum lasting 300 seconds. On memory testing 30 minutes after recovery from the hemiparesis, the patient remembered 14 of 16 items presented during the time of the hemiparesis. In comparison, he remembered 16 of 16 items at baseline. Right IAT was not done to avoid complications caused by coma following injection of the intact hemisphere and because of the risk of infarction of the remaining functioning cortex.

Neuropsychological assessment for epilepsy surgery revealed word finding difficulties in the patient's conversational speech and on formal testing. Additionally, the patient showed reduced word fluency, diminished sentence repetition, slow complex visual scanning, and inefficient problem solving. Verbal intellectual tasks were completed below the level of their non-verbal counterparts (table 1). Fine manual dexterity was diminished in the patient's dominant left hand.

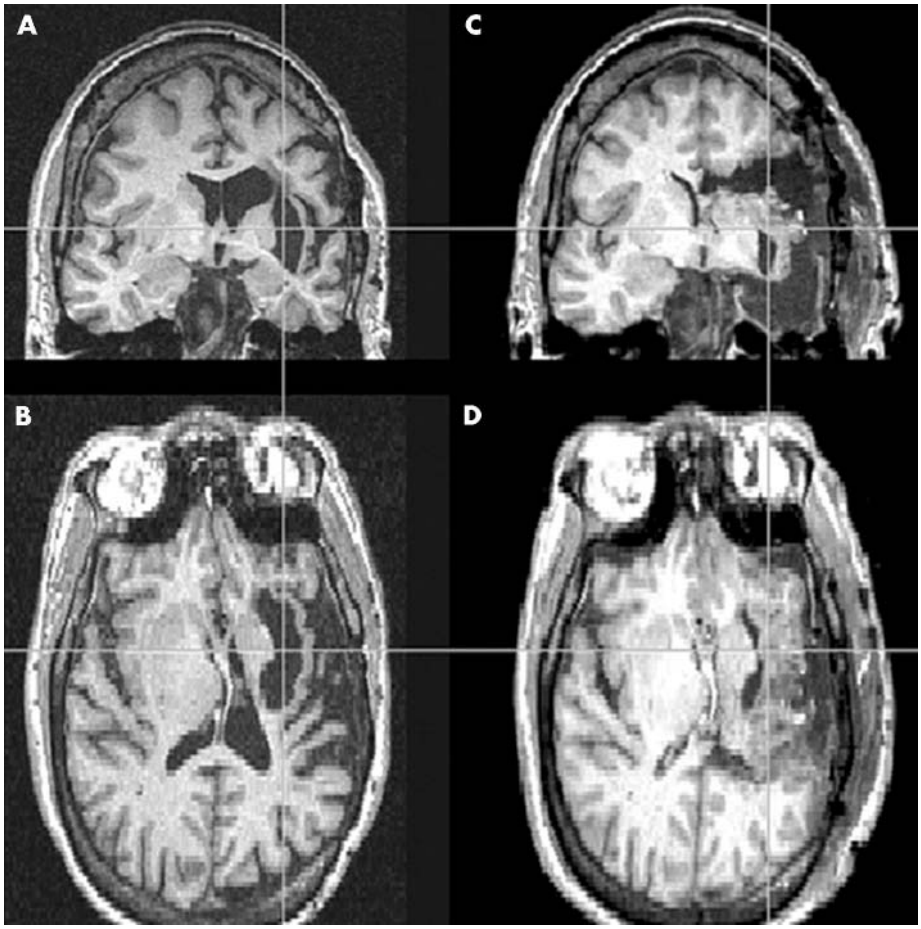

Figure 1 Panels A and B: Coronal and axial T1 weighted images before hemispherectomy, showing a cystic cavity of CSF signal intensity within the left cerebral hemisphere which involves the left caudate nucleus, putamen, external capsule, and corona radiata adjacent to the body of the left lateral ventricle. There is parenchymal volume loss with prominence of the sulci in the suprasylvian-inferior frontal gyrus regions and compensatory dilatation of the left lateral ventricle with mild midline shift towards the left. There is also evidence of previous left frontal temporal craniotomy. The findings are compatible with encephalomalacia and surrounding gliosis caused by the previous motor vehicle accident. (Siemens Magnetom Vision: time of repetition (TR), 11.4; time of echo (TE), 4.4.) Panels C and D: Coronal and axial T1 weighted images after hemispherectomy, showing a left temporal lobectomy including a left amygdalohippocampectomy and a large parenchymal defect in the dorsal inferior aspect of the left frontal and anterior parietal lobes extending into the left lateral ventricle. No evidence of right hemisphere injury after surgery was seen (Siemens Magnetom Sonata: TR, 11.08; TE, 4.3). The preoperative and postoperative images were co-registered.

Table 1 Preoperative and postoperative neuropsychological test scores

Test	Preoperative	Postoperative
WAIS-III, VIQ	92	81
WAIS-III, PIQ	97	87
WAIS-III, FSIQ	94	83
WMS-III, auditory immediate index	108	89
WMS-III, auditory delay index	111	97
WMS-III, visual immediate index	97	71
WMS-III, visual delay index	97	72
WMS-III, immediate memory index	103	76
WMS-III, general (delayed) memory index	111	87
WMS-III, working memory index	91	66
WRAT-III, reading index	98	90
Boston naming test, raw score*	47	53
Token test, raw score	38	25
Sentence repetition test, raw score	9	3

*The patient was overexposed to the Boston naming test as a result of his postoperative speech therapy; thus this result is misleading—the apparent improvement on this measure reflects increased familiarity with the test and not an improvement in confrontation naming as a result of surgery. FSIQ, full scale intelligence quotient; PIQ, perceptual intelligence quotient; VIQ, verbal intelligence quotient; WAIS-III, Wechsler adult intelligence scale, third edition; WMS-III, Wechsler memory scale, third edition.

After informed consent about the risks and benefits of surgery, the patient and his family agreed to a left functional hemispherectomy. Histopathology was consistent with remote infarction.

Immediately after surgery, the patient had expressive aphasia with two to three words per utterance, yes/no confusion, restricted grammar, impaired naming, relatively good comprehension for simple material, and poor repetition. After 30 months of intensive speech therapy, the patient still misuses or omits pronouns and prepositions and has extreme word finding difficulties and problems initiating speech, with multiple paraphasias and syntax errors.

Neurological examination eight months after surgery revealed expressive aphasia, a right sided spastic hemiparesis with decreased power in the proximal upper and lower limbs (4–/5), and no power in the right hand (0/5). Fingerperimetric visual field testing revealed right hemianopia.

Neuropsychological follow up eight months after surgery showed marked declines from baseline on language tests including aural comprehension and sentence repetition, as well as on measures of general intellectual functioning, working memory, problem solving, and verbal and visual memory. Qualitatively, he had marked word finding

difficulties and reduced comprehension, requiring frequent repetition of instructions and pantomiming (table 1).

The patient was completely seizure-free for three months after surgery. He then started to have rare auras characterised by nausea. Seizure frequency 32 months after surgery is one aura per month.

DISCUSSION

Left hemispherectomy led to unexpected language difficulties in our case. Bilateral speech was not suspected from the unilateral IAT. The language difficulties were not transient, as seen with resection of the supplementary motor area, and could not be explained by buccofacial apraxia caused by the parietal resection. In retrospect, factors for the occurrence of unpredicted aphasia included first, the predominantly subcortical location of the early brain injury; second, the timing and onset of the lesion after left sided language development has begun; and third, the late hemispherectomy in middle age with minimal remaining plasticity.

The type and location of the lesion may influence interhemispheric language transfer. For example, in contrast to large destructive vascular lesions,[6] early low grade tumours may not result in language transfer but lead to continued left hemispheric language development in regions separate from the neoplasm.[7] Additionally, the mainly subcortical location of our patient's lesion may have resulted in incomplete transfer of language dominance to the other hemisphere, in contrast to other patients with more complete language shift after widespread cortical lesions—for example, hemidecorticectomy for treatment of Rasmussen's syndrome,[3] perinatal infarctions,[6] Sturge-Weber syndrome,[6] malformation of cortical development,[8] and hemispherectomies.[2] The preoperative impairments in neuropsychological language testing also suggest that language transfer was incomplete. Moreover, residual functional capacity in the left hemisphere is suggested by intact preoperative visual fields.

Incomplete inactivation of subcortical, deeper white matter tracts by amobarbital may also have accounted for the negative IAT result. Comparisons of amobarbital and lignocaine (lidocaine) during superselective cerebral angiography have shown that amobarbital acts more strongly on grey matter, with little effect on nerve fibres and subcortical white matter.[9] Additionally, the IAT most probably did not inactivate posterior cerebral regions that were subsequently resected because of their cerebral perfusion patterns (fig 1, panels C and D).

Furthermore, the time of onset of lesions may have played an important role in our case. Language transfer to the right side after brain injury before the age of one year is much more likely than later in life.[10] Language transfer is commonly seen in hemispheric lesions acquired before the age of six years,[1 6] and rare cases of language transfer have even been reported in adolescent patients.[3]

Most hemispherectomies are done in the first decade of life and few data are available from adolescents and adults. Reduced neuronal plasticity at the age of 53 in our patient may also have contributed to the aphasia. In younger patients with some bilateral language representation, developmental plasticity may partially protect against worsened postoperative language outcome.

Some patients with bilateral independent language representation can continue to talk during right and left sided amobarbital injection.[11] Right IAT may have clarified our patient's bilateral language function and contributed some additional information for preoperative informed consent. However, the risks for contralateral IAT in patients with hemispheric injury include infarction of the remaining intact hemisphere or postinjection coma caused by suppression of the functioning cortex. Careful selection of patients undergoing IAT is crucial owing to a complication rate of 1% during this invasive procedure,[12] with complications mainly seen in patients over 40 years, and because of the possibility of a non-predictive IAT in patients with atypical language representation. Less invasive speech mapping paradigms besides the IAT may complement language lateralisation in the future.[13]

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