



Case Report

A case of aphasia due to temporobasal edema: Contemporary models of language anatomy are clinically relevant

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ABSTRACT

Background: Understanding the anatomy of language in the human brain is crucial for neurosurgical decision making and complication avoidance. The traditional anatomical models of human language, relying on relatively simple and rigid concepts of brain connectivity, cannot explain all clinical observations. The clinical case reported here illustrates the relevance of more recent concepts of language networks involving white matter tracts and their connections.

Case Description: Postoperative edema of the ventral occipitotemporal cortex, where modern network models locate a crucial language hub, resulted in transient severe aphasia after a subtemporal approach. Both verbal comprehension and expression were lost. The resolution of edema was associated with complete recovery from phonetic and semantic dysfunction.

Conclusion: Complete aphasia due to a functional disturbance remote from the areas of Broca and Wernicke could be explained by contemporary neuroanatomical concepts of white matter connectivity. Knowledge of network-based models is relevant in brain surgery complication avoidance.

Keywords: Aphasia, Connectivity, Language, Networks, White matter tracts

BACKGROUND

The current understanding of language representation in the human brain has been reshaped by recent advances in brain imaging^[5,22] and perioperative functional assessment during awake brain surgery.^[19] Classical associationist models^[23] [Figure 1] are giving way to network concepts. In these networks, specialized cortical regions have different but related functions. They interact via white matter tracts which are parallel and bidirectional.^[6] These network concepts may more satisfactorily represent the anatomical infrastructure of higher cognitive functions, including language.

Aphasia has been reported after subtemporal extra-axial approaches, without explanation of the underlying pathophysiology, besides possible vein of Labbé injury, or ill-defined temporal lobe edema mentioned in some cases.^[4,8,9,16,24,25] In the case reported below, severe speech dysfunction

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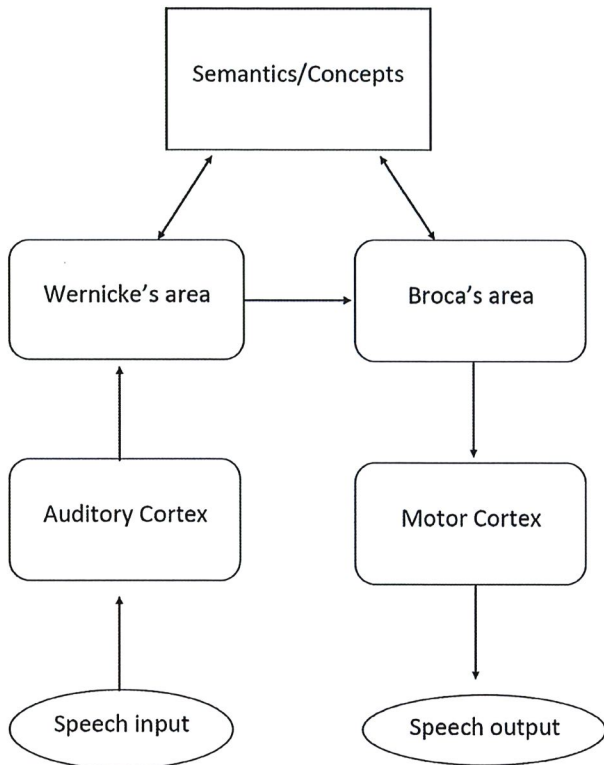


Figure 1: The traditional Wernicke–Lichtheim model of language connectivity. The depicted connectivity permits serial, mainly unidirectional information processing. Semantic concepts are formed by higher associative systems distributed throughout the associative cortex, harboring conceptual representations of distinct modalities associated with a particular object.

occurred with transient basal temporal edema after a surgical subtemporal approach and could be explained using contemporary concepts of language anatomy.

CASE DESCRIPTION

A 44-year-old woman with a history of whole-brain radiotherapy at age 9 for leukemia presented with slight dizziness. Left-sided headache was unchanged since childhood radiotherapy. Magnetic resonance imaging (MRI) showed three meningiomas, including one petroclival one showing growth at 1-year follow-up [Figure 2]. The patient was referred for surgery. Detailed preoperative language assessment did not reveal any deficits in verbal comprehension, fluency, reading, or naming, only slight attention deficits. The meningioma was uneventfully removed through a subtemporal transtentorial approach.

Immediate postoperative language function appeared normal. Over the first postoperative days, the patient developed progressive speech impairment, reaching mutism on the 2nd day. Five days after the operation, word-finding

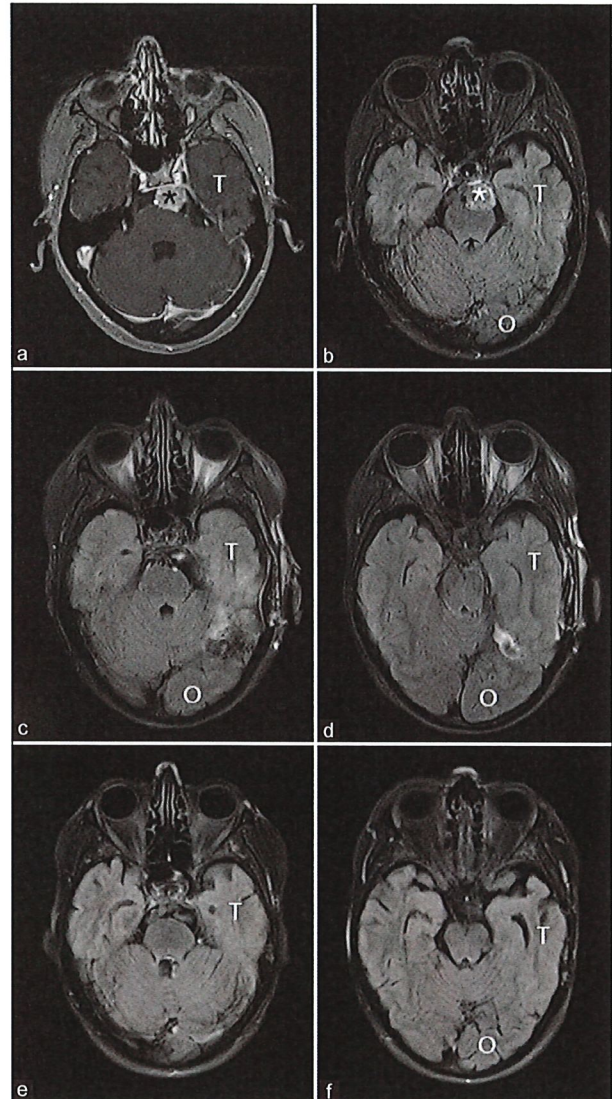


Figure 2: Preoperative magnetic resonance imaging (MRI); a: T1 + gadolinium, b: T2-fluid-attenuated inversion recovery (FLAIR), and asterisk: clival meningioma. Postoperative MRI, c and d, day 3, e, and f 1 year; T2-FLAIR. T: Temporal lobe. O: Occipital lobe.

difficulties and deficient verbal comprehension persisted: the patient was incapable of naming objects or repeating sentences. Reading tests demonstrated verbal perseveration (VP).

Postoperative MRI 1 and 3 days after surgery showed an area of T2 hyperintensity at the inferior occipitotemporal junction [Figure 2], where a retractor had maintained a cottonoid to protect the surgical corridor, in the absence of any other parenchymal anomaly.

Over the following days, language improved rapidly and, after 6 weeks, reading and comprehension had almost

recovered: the semantic function was normal, phonemic fluency remained slightly deficient. At 1-year follow-up, these residual deficits resolved and language function completely returned to the preoperative state. MRI confirmed the expected resolution of the inferior temporal T2-hyperintensity. MRI diffusion tensor tractography was obtained 4 years after surgery to document the anatomy of white matter tracts involved in language. After the fusion of postoperative images and tractography, the lesion was located at the junction of the inferior longitudinal fasciculus (ILF), the inferior fronto-occipital fasciculus (IFOF) and the superior longitudinal fasciculus (SLF) [Figure 3].

DISCUSSION

In the described case, severe mixed aphasia reaching mutism was attributable to a transient functional disturbance at the ventral temporooccipital junction. This area is located remote from the components of the classical Wernicke–Lichtheim model of language connectivity: the “language areas” of Broca and Wernicke and their direct connection through the arcuate fasciculus.^[23] In this traditional model, strongly anchored in medical education and neurological reasoning, human language arises from the activity of a sensorimotor “reflex machine.”^[10] It depends on clearly defined cortical areas connected through a single white matter pathway, allowing serial information processing [Figure 1]. This traditional model only unsatisfactorily explains the spectrum of clinically observed speech dysfunctions, including the present case.

Network concepts, more than unidirectional information processing, can provide satisfactory models for brain function. In short, parallel and bidirectional white matter connections of functionally related cortical areas (so-called “edges”) provide the anatomical basis for integrative processes and dynamic interactions. Critical cortical epicenters (so-called “hubs”) integrate plurimodal information originating in unimodal regions.^[21] Hubs are, therefore, highly eloquent. Cognitive processes, including language, depend on interactions between hubs connected through subcortical white matter tracts.^[13]

White matter tracts have traditionally been investigated through postmortem gross dissection, fiber tract-tracing in animals, human functional imaging, and *in vivo* mapping by diffusion tensor imaging.^[11] In addition, intraoperative direct electrical stimulation of the brain during awake craniotomies provides a direct clinical assessment of the exposed cortex and white matter.^[19] Its correlation with neuropsychological examinations and anatomofunctional neuroimaging has significantly contributed to the evolving neuroanatomical concept of human language.^[6]

According to this concept, language processing can be explained by a dual-stream architecture analogous to that of

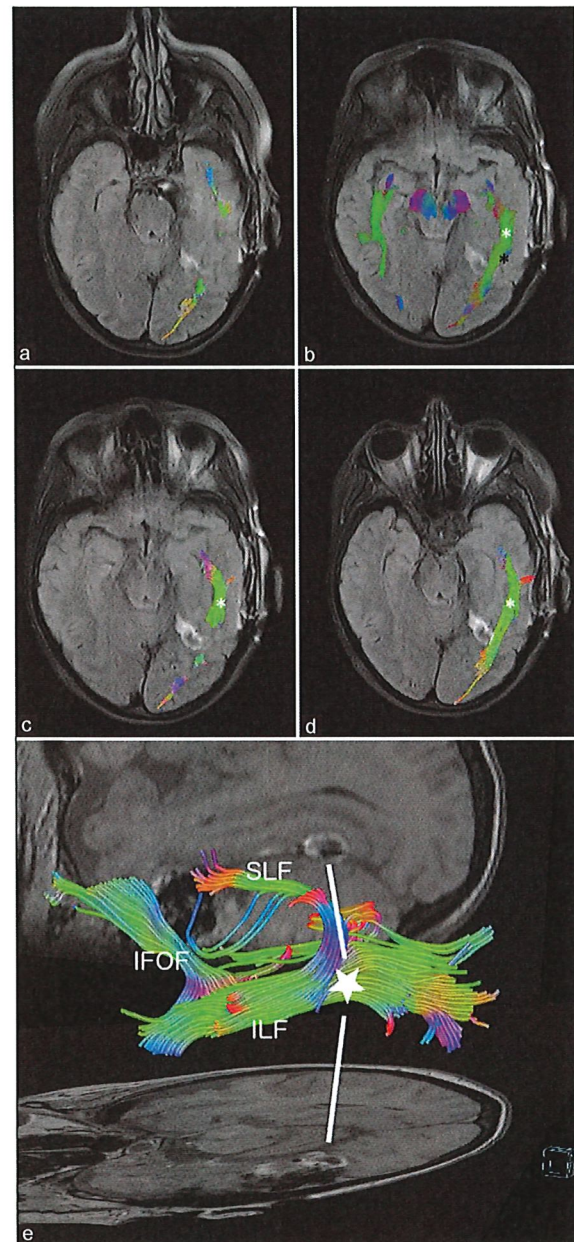


Figure 3: Diffusion tensor imaging tractography 4 years after surgery, fused with initial 2D postoperative fluid-attenuated inversion recovery (FLAIR) magnetic resonance imaging (MRI) (syngo.via, Siemens, Munich, Germany). The ILF and inferior fronto-occipital fasciculus (IFOF) are part of the ventral stream and join posteriorly. The superior longitudinal fasciculus (SLF) represents the dorsal stream. (a-d): the reconstructed inferior longitudinal fasciculus (ILF) (white asterisk and green fibers) and posterior part of the IFOF, including their junction with the SLF (black asterisk and blue fibers), are depicted on craniocaudal axial FLAIR slices in relation to the temporobasal hypersignal. (e): 3D VRT reconstruction of the SLF, IFOF, and ILF in relation to the temporal FLAIR hypersignal (white star and lines, at the junction of the three tracts) on axial and sagittal images.

the visual system.^[12,17,20] The two streams unite the frontal lobe with the temporal, parietal, and occipital lobes [Figure 3]. The dorsal stream, carried by the SLF, can be subdivided into anterior and posterior parts versus its deep part (arcuate fasciculus). It appears to be involved in the processing of auditory and somatosensory inputs and their conversion into working phonological and articulatory representations.^[11] The ventral stream is carried by two bundles: a direct pathway formed by the IFOF and an indirect pathway formed by the ILF and the uncinate fasciculus (UF).^[6] The ventral stream relates to various aspects of semantic processing.^[2,6,17,18]

Finally, parallel pathways of one stream may be, at least partially, functionally redundant. This provides an explanation, for example, for the well-known tolerance of anterior temporal lobectomy in the “dominant” hemisphere. The surgically inevitable loss of the indirect bundle (ILF-UF) of the ventral stream may, at least partly, clinically be compensated for by the direct pathway (IFOF),^[7] but when both streams are compromised, language may be severely impaired.

At the cortex of the ventral occipitotemporal junction, the dorsal stream (SLF) connects with both bundles of the ventral stream (ILF and IFOF) [Figure 3].^[15,26] This hub may, therefore, subserve integration of (plurimodal) linguistic information.^[5] Its disturbance, disrupting the anatomical connection of all language pathways, with no possibility for redundancy, may therefore result in global aphasia, as seen in the present case. In addition, the observed VP corroborates the recent suggestion of an implication of the IFOF in a wide network subserving VP, including deep gray nuclei as well as white matter pathways within the ventral stream.^[14]

In summary, the function of the ventral occipitotemporal cortex as a crucial hub in current neuroanatomical language models explained complete transient aphasia with loss of both verbal comprehension and expression. Phonological, articulatory, and semantic function recovered with the resolution of occipitotemporal edema.

CONCLUSION

The present case of a complete loss of speech function could be satisfactorily explained by contemporary neuroanatomical concepts of white matter connectivity. Knowledge of network-based models is clinically relevant in brain surgery complication avoidance.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

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