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Single Case Report

Mapping the neural substrates of verbal and sign language: A single-case study using direct electrical stimulation

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ABSTRACT

This case report investigates the cortical and subcortical representations of verbal and sign language in a bilingual patient who uses both spoken and signed modalities, assessed intraoperatively during awake surgery. Although spoken language is regularly mapped during awake craniotomies, other language modalities are rarely reported. We performed direct electrical stimulation (DES) during both spoken and sign language tasks in peritumoral regions of the left temporoparietal lobe. The language abilities of the patient were intraoperatively assessed using verbal object naming and sign recognition. Our findings demonstrate that cortical regions, such as the supramarginal gyrus, play a crucial role for both verbal and sign language. However, the specific sites within this region that elicit DES-positive responses differ between the two language modalities. Similarly, subcortical disconnections highlight the overlap between sign and verbal language, particularly in major language pathways, while also emphasizing the specialized role of motor pathways in sign language processing. Clinically, our results emphasize the importance of tailoring DES protocols for intraoperative mapping to individual patient needs, and theoretically, they enhance our understanding of the roles of the supramarginal gyrus and the corticospinal tract in language comprehension.

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1. Introduction

Awake craniotomy with intraoperative brain mapping is a well-established technique used to maximize the extent of tumor resection while preserving essential neurological functions, particularly in eloquent brain areas (De Witt Hamer et al., 2012; Duffau et al., 2003; Sanai & Berger, 2008). This approach is crucial in surgeries involving language areas (i.e., typically left perisylvian regions). Traditionally, language mapping during awake surgery has focused on spoken language, providing significant insights into the cortical and subcortical organization of this cognitive function (Duffau et al., 2005; Ojemann et al., 2008; Rofes et al., 2024; Tate et al., 2014). However, languages can vary significantly in their structure and modality, raising the question of whether different forms of language, such as spoken and sign languages, share the same cortical and subcortical representations.

The application of direct electrical stimulation (DES) to patients who use sign language presents unique challenges and opportunities, both from a clinical and a theoretical perspective. Sign languages are fully-fledged natural languages with complex grammatical structures and features that are similar to spoken languages (Sandler & Lillo-Martin, 2006). The extent to which their neural correlates overlap with those of spoken languages, however, remains an open question. Studies of functional Magnetic Resonance Imaging (fMRI) suggest that, at large, single word production relies on similar cortical areas for spoken and sign language (Emmorey, 2021; Emmorey et al., 2007). This overlapping network includes traditionally language-relevant areas such as the left inferior frontal gyrus (IFG). However, differences in cortical activation have also been identified; for example, the left supramarginal gyrus (SMG) and post–central gyrus (PoG) appear to be more engaged in sign language production compared to spoken language (Emmorey et al., 2007). The involvement of the SMG more prominently in sign language can be attributed to its potential role in integrating the various spatial, temporal, and configurational phonological elements of signs, such as locations on the body, hand movements, and handshapes (Emmorey, 2021; Emmorey et al., 2007; MacSweeney et al., 2002). Bilateral SMG is also engaged more in signers during processing of phonological violations (Cardin et al., 2016; Emmorey, 2021). In sign language, a phonological violation during awake surgery could involve incorrect handshapes, where the shape of the hand does not conform to the expected form for a particular sign.

While the existing literature provides examples of the cortical representation and overlap of sign and spoken languages, the vast majority of this evidence is based on research using fMRI. However, fMRI studies primarily measure brain activity through changes in blood flow, a measure that does not provide information regarding white matter connectivity. As a result, the subcortical overlap between these two language modalities remains even less understood. A few studies have contrasted microstructural properties of the white matter between deaf and hearing adults, reporting decreased white matter volume around auditory areas, whereas others report differences in parts of the corpus callosum (Emmorey et al., 2003; Hribar et al., 2014; Leporé et al., 2010). However, these studies focus on comparing structural properties of white matter between congenitally deaf and non-congenitally deaf individuals, without the opportunity to investigate the connectivity of the two language modalities in real-time within the same individuals.

Contrary to methods such as fMRI, intraoperative brain mapping can assess the neural underpinnings of different language modalities, providing real-time and causal data with high spatial resolution and offering insights that are challenging to obtain via non-invasive methods alone (Mandonnet et al., 2010). However, due to the complexity of awake craniotomies and the limited number of suitable cases, there has been little information derived from awake surgery studies. To the best of our knowledge, there are only three existing case studies, presenting conflicting evidence. Corina et al. (1999) reported a case of a deaf user of American Sign Language (ASL) undergoing DES mapping. Their findings demonstrated that Broca's area is involved in the motor execution of sign language, indicating that the linguistic specificity of this region is not confined to speech behavior alone. Additionally, they observed unusual semantic-phonological errors with stimulation to the SMG, suggesting its role in binding linguistic features for language production. This seminal study, however, was limited to mapping only ASL intraoperatively, as the patient was an exclusive user of sign language. As a result, it can provide evidence regarding the neural substrates of sing language processing, but not in relation to the substrates of spoken language.

Two recent case reports had the opportunity to explore the representation of both language modalities, yielding conflicting results. In one study, Lau et al. (2023) mapped subcortical areas of the posterior temporal lobe and identified overlapping sites for both spoken English and ASL in an adult who was deaf since childhood. All three DES-positive sites elicited phonological paraphasias during naming of pictures of signs or objects with induced errors including neologisms and phoneme substitutions. Based on this mapping outcome, the authors concluded that the neural representation of sign and spoken language is identical. Different findings were reported by Barua et al. (2024), who mapped the anterior temporal lobe and the IFG during spoken object naming and naming using British Sign Language. In contrast to the previous report, Barua et al. (2024) identified differential maps for the two language modalities, proposing distinct neural pathways and regions specialized for each form of language.

These reports vary not only in their findings but also in terms of the craniotomy sites and the specific tasks used during language mapping. They also vary in relation to the induced errors and the definition of linguistic errors during spoken and sign language tasks. Also, previous studies did not leverage the positive sites identified through DES to investigate the potential white matter disconnections caused by the stimulation. As a result, the subcortical connectivity associated with sites for different language modalities remained unexplored. In this report, we therefore present findings from an awake surgery in which both spoken and sign language modalities were mapped. Additionally, we employed virtual DES-lesion tractography to explore the disconnection of white

matter tracts in relation to language modality. Our evidence helps reconcile the discrepancies in previous literature and provide causal data regarding the representation of sign and spoken language in the brain.

2. Methods

2.1. Case presentation

We report the case of a 55-year-old, right-handed male with normal hearing, fluent in spoken German and German Sign Language (GSL), who was diagnosed with a left temporoparietal glioblastoma (WHO grade IV, IDH-wildtype). The patient acquired spoken German as his L1 and GSL later in adulthood. Although no formal proficiency assessments were available, he reported using GSL alongside spoken German daily for communication at home for the past 15 years. The study was conducted in accordance with institutional ethical guidelines (ID: 2026-3606). Written informed consent for participation and publication was obtained from the patient.

The patient presented with communication difficulties in both spoken and written language that intensified over the period of six weeks. Preoperative patient consultation confirmed the presence of word-finding difficulties, phonological paraphasias during spontaneous speech, and minor issues with language comprehension. Preoperative language and cognitive assessment took place 4 days prior to surgery and demonstrated minor difficulties with verbal naming and repetition in the absence of a formal aphasia diagnosis according to the Aachen Aphasia Test (Huber et al., 1980). Our cognitive screening revealed additional issues with short-term and working memory, as revealed by digit span tasks included in the DemTect test (Kalbe et al., 2004). Table 1 lists the raw preoperative performance of the patient in a series of tasks.

2.2. Image acquisition and connectome reconstruction

Prior to the operation the patient received high-quality structural images with a 3T Siemens Magnetom Skyra (Siemens, Erlangen, Germany) to aid surgical planning. Specifically, T1 MPRAGE images (slice thickness = 1 mm, no inter-slice gap, number of slices = 176, TR = 2000 msec, TE = 2.45 msec, FoV = 192 × 256), T2 (slice thickness = 5 mm, inter-slice gap = 5.5 mm, number of slices = 30, TR = 7150 msec, TE = 98 msec, FoV = 201 × 230), and Diffusion Tensor Imaging (DTI; slice thickness = 5 mm, inter-slice gap = 5.5 mm, number of slices = 30, TR = 6000 msec, TE = 64.2 msec, FoV = 230 × 230) along 64 directions with b = 1000 sec/mm^2 were acquired. Thirty B0 volumes were also acquired. The complete imaging protocol additionally included T1 images with contrast enhancement and Fluid-Attenuated Inversion Recovery (FLAIR) images to improve lesion visualization and lasted approximately 45 min.

Preoperative MRI revealed a 14.4 cm^3 big, non-contrast-enhancing lesion in the left temporoparietal lobe, mainly affecting the posterior superior temporal (STG) and the SMG. Post-operative biopsy confirmed a grade 4 glioblastoma. DTI software (Brainlab Elements, Munich, Germany) and previously published guidelines for clinical tractography (Fekonja et al., 2019) were used to reconstruct the language connectome around the tumor. DWI preprocessing was conducted in Brainlab Elements and included brain masking, denoising, and eddy-current/motion correction (Veraart et al., 2016). Diffusion tensors were then estimated, and deterministic tractography was performed using Brainlab's default parameters (step size 1 mm, 20° max. angulation, minimum length 50 mm, FA: .19). All relevant tracts were reconstructed using a two-ROI approach (Fekonja et al., 2019). For the arcuate fasciculus (AF), the seeding ROI was placed on the axial plane in the posterior temporal lobe at the level where the superior temporal sulcus (STS) becomes visible on the sagittal plane. The refining ROI was positioned on the coronal plane just inferior to the most ventral point of the central sulcus. For the inferior fronto-occipital fasciculus (IFOF) and inferior longitudinal fasciculus (ILF), the seeding ROI was placed on the coronal plane over the posterior occipital cortex, aligned with the point where the cerebellum terminates on the sagittal plane. The refining ROI for the IFOF was placed in the left prefrontal cortex, whereas for the ILF it was positioned in the anterior temporal lobe to isolate long-range temporo-occipital fibers.

Our reconstruction of the AF revealed that the tumor was in direct contact with the tract, whereas the IFOF and the ILF were located 7 mm and 9 mm away from the tumor mass, respectively. These distances correspond to the minimum Euclidean distance between the tumor boundary and each tract on the axial plane, computed using the Object Management tool in Brainlab Elements 3.2.1. Fig. 1 displays the tumor mass alongside relevant language pathways.

2.3. Intraoperative language mapping and fiber tracking

The intraoperative language mapping protocol consisted of object naming and sign recognition. Given the constraints of the patient's positioning on the operating table, we opted to use sign recognition as the intraoperative task for evaluating sign language. This decision was made because testing naming using sign language would require bimanual coordination and this was not feasible under the restricted conditions of the surgical environment. Both tasks were tested twice pre-operatively and any objects or signs that were not correctly or effortlessly named were excluded from the intraoperative assessment (see also Table 1).

The surgery followed the asleep-awake-asleep protocol. The craniotomy was performed during the asleep stage and

Table 1 – Results of preoperative assessment. Raw scores are presented (score achieved/total score).

Task	Pre-operative accuracy
Object naming	68/75
Action naming	60/74
Sign recognition	14/14
Sentence comprehension	14/16
Sentence production	21/25
Word repetition	19/30
Sentence repetition	23/30
Word reading comprehension	24/30
Sentence reading comprehension	24/30

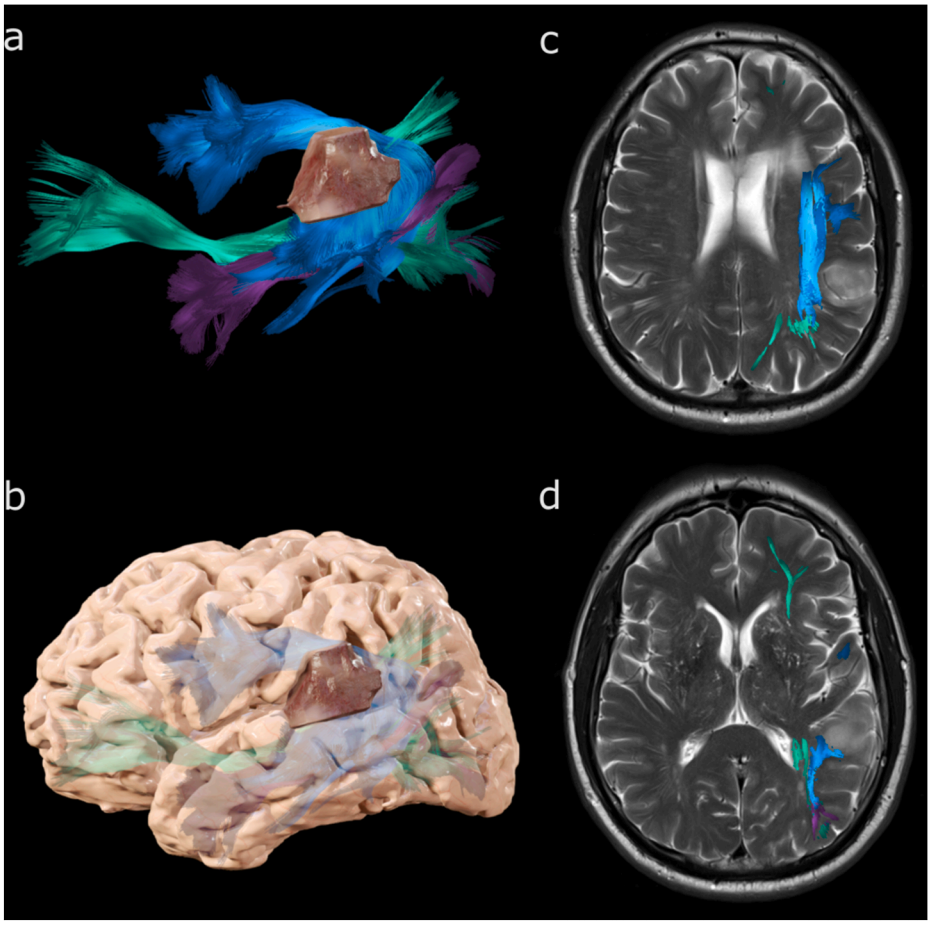

Fig. 1 – Tumor segmentation and fiber-tracking of language-relevant pathways. Blue: Arcuate Fasciculus. Green: Inferior Fronto-Occipital Fasciculus. Purple: Inferior Longitudinal Fasciculus. (a) 3D reconstruction of the tumor and the language-relevant fiber-tracts. (b) 3D reconstruction of the tumor and pathways alongside the cortical surface mesh of the patient. (c & d): Representative T2 axial slices.

the patient was awoken for language mapping. The craniotomy provided access to the tumor and to limited adjacent peritumoral regions, including parts of the left SMG and the ventral PoG. Fig. 2 illustrates the location of the craniotomy in relation to the tumor and the exposed cortex. DES mapping was conducted using a bipolar probe with a 5-mm interelectrode spacing, delivering biphasic square-wave pulses at 60 Hz with a 1 msec phase duration, and an amplitude of 5 mA. The stimulation was applied in 4-s trains, with a 4-s interval between each stimulation. The entire exposed cortex was first mapped with object naming and then with sign recognition. DES stimulation and picture presentation occurred simultaneously without delay. Each cortical site was stimulated 3 non-consecutive times and was only considered positive if 2 out of 3 stimulations yielded the same type of error.

During the surgical procedure, the patient's head was securely fixed using a Mayfield clamp and registered to individual MRI scans via a neuronavigation system (Brainlab Elements, Munich, Germany). This setup allowed for precise intraoperative mapping and post-operative evaluation. Each DES-positive point was identified on the cortical surface using a neuronavigation pointer and automatically registered to the patient's individual T1 MPRAGE dataset via the "acquire point" function within the Brainlab Elements cranial neuronavigation system (Brainlab AG, Munich, Germany). This procedure enabled precise marking of the x-, y-, and z-coordinates for each positive stimulation site directly on the individual MRI (Proelss et al., 2025). The extracted coordinates were then used for subsequent visualization and analysis.

We used the VAN-POP object naming task, a standardized naming task for intra- and pre-operative language mapping (Ohlerth et al., 2020). The task items are controlled for key psycholinguistic variables, including word frequency, age of acquisition, number of syllables, and picture complexity. All stimuli consist of black-and-white line drawings and have been validated and used in previous TMS and DES mapping studies (Ntemou et al., 2024; Ohlerth et al., 2021; Rofes et al., 2024).

For the sign naming task, we used pictures of signs (e.g., Fig. 4) provided by the patient and his family. We aimed to select concepts matching those in the object naming task. However, because this was a medical emergency and surgery took place only 48 h after the preoperative assessment, full item matching was possible for only 5 out of 16 sign items.

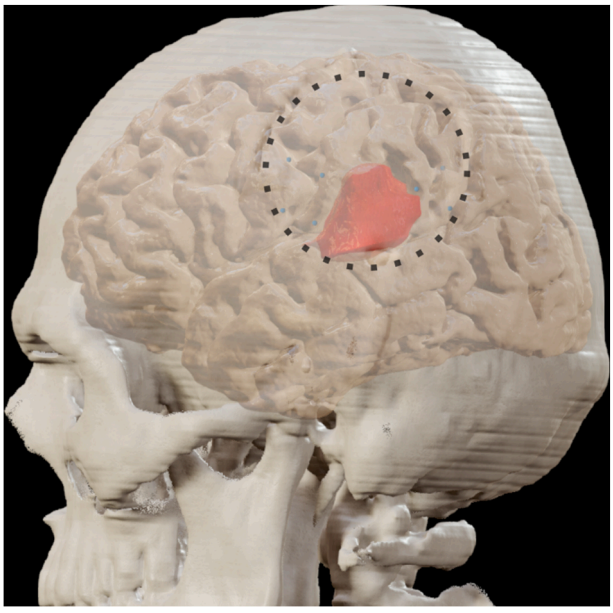

Fig. 2 – 3D rendering showing the craniotomy location and size, the tumor mass, and sites with positive responses during DES. positive stimulation sites: blue points, tumor: red mass, craniotomy: dotted circle.

For both object naming and sign naming, two randomized lists of items were prepared to minimize order effects. Object naming items were repeated on average 3 times, and sign naming items 4 times. Repetitions were necessary because the entire craniotomy area had to be mapped and the higher repetition rate for signs was due to the smaller available item pool. Both tasks assessed visual recognition of object or sign depictions, access to semantic and phonological information of concepts and overt production of words with the same lead–in phrase (i.e., "Das ist …" [This is …]). Similarly to the object naming task, sign recognition did not require overt motor execution beyond speech articulation and did not involve sign production. Both tasks included concepts with a maximum of four syllables (e.g., Zi-ga-ret-te [cigarette]).

Error classification during verbal naming followed previously published guidelines for pre- and intraoperative language mapping (Ntemou et al., 2024; Picht et al., 2013; Rofes, Spena, et al., 2017). Error categories included the following: 1) anomia, e.g., "Das ist ein …" [This is a …], 2) hesitation, e.g., "Das ist ein…uhm…Auto" [This is a…uhm…car], 3) semantic paraphasia (e.g., "Das ist ein Fahrrad" [This is a bicycle] for the target "car"), 4) phonological error, e.g., "Das ist ein *Ato" a non-existing word for the target "Das ist ein Auto" [This is a *par] for the target [This is a car] or with an existing word "This is a **b**ear" for the target "This is a **p**ear", 5) no response, e.g., lack of intelligible response), 6) articulation error, e.g., "Das ist ein A-A-Auto" [This is a c-c-car] and 7) mixed errors (i.e., a combination of different categories).

During naming of signs, the same error categories were largely applicable as in object naming. For example, misidentifying the sign for *car* as *bicycle* was classified as a semantic paraphasia, and failing to produce the target word (e.g., *Das ist ein …*) was classified as anomia. The main difference between the two modalities concerned phonological errors. Spoken verbal errors such as *Ato* for *Auto* were considered *spoken phonological errors*, originating at the word-form level, just as in object naming (Rofes et al., 2019).

However, since sign languages have their own phonological system, defined by parameters such as manner of handshape and location, errors in processing these visual-gestural features could also occur. Such errors were classified separately as *sign phonological errors*, in order to distinguish them from phonological errors in spoken output. These sign-based errors reflect difficulties in identifying or interpreting the formational features of the sign itself, rather than in producing the corresponding spoken word.

To identify potentially disconnected fibers associated with each language task, we enlarged the exact (x, y, z) coordinate of each positive stimulation point by a 5-mm radius to form a seed region of interest (ROI). This expansion was applied to the stimulation coordinate itself, not in addition to the physical volume of the bipolar probe. These ROIs were then used to dissect the connectome and track fibers that were potentially disconnected due to DES. This procedure has been previously used for fiber tracking of positive stimulations yielded from navigated Transcranial Magnetic Stimulation (nTMS) and more recently DES (Frey et al., 2012; Mandonnet et al., 2020). Following previously published fiber tracking parameters for stimulation-based seed ROIs, we thresholded our virtual dissections using the following values: FA = .11, minimum length = 80 mm, and maximal angulation 80° (Fekonja et al., 2019; Frey et al., 2012; Mandonnet et al., 2020). To compute the percentages of disconnection for each language-relevant tract around the glioma, we converted the tracked disconnections into binary NIfTI files. We then computed the overlap with pre-traced fiber bundles, such as the AF and the IFOF, to determine the extent of disconnection in terms of percentages. This method has been previously used to compute disconnections due to lesions based on standardised atlases and was recently also used for virtual DES-lesions (Coletta et al., 2024; Griffis et al., 2021). To the best of our knowledge, this is the first time a similar method has been used to compute disconnections of patient-specific connectomes with virtual lesions.

3. Results

3.1. *Intraoperative mapping and DES-error description*

Stimulation of the ventral post–central gyrus (vPoG) induced anomia and semantic errors during object naming (Fig. 3, sites 1 & 4). Stimulation of the inferior SMG and the posterior STG induced phonological and semantic paraphasias during naming of objects (Fig. 3, points 2 & 3). Out of these 4 sites that were positive for object naming, only one was also positive for naming of signs and when stimulated yielded an anomia (site number 2). Three additional sites were identified as positive for sign language. All three were located in the SMG, in closer proximity to the glioma compared to object naming sites. When stimulated, they yielded sign phonological paraphasias (point 5), hesitations (point 6), and anomias (point 7). The remainder of the craniotomy area was also stimulated, but no

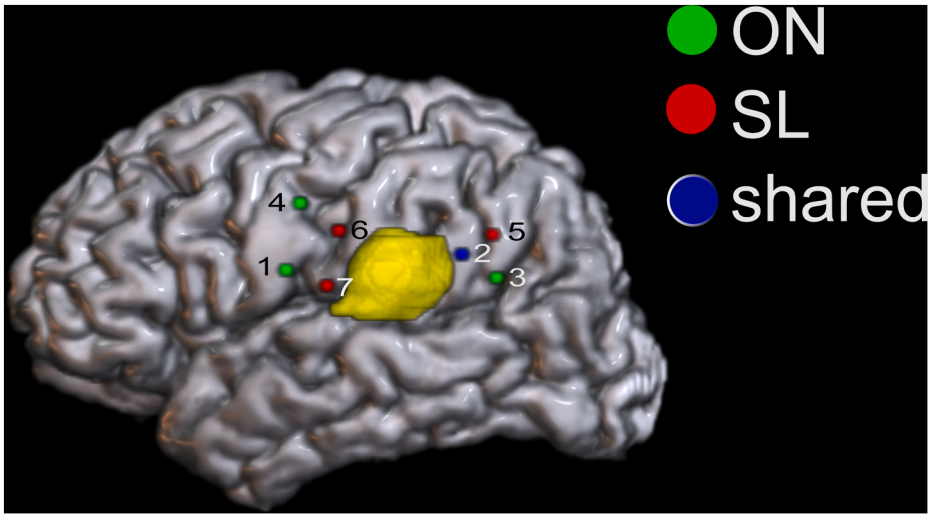

Fig. 3 – 3D cortical surface visualization of the individual patient's left hemisphere with DES-positive stimulation sites. Stimulation sites have been color-coded according to language task. Green: positive sites only for ON, Red: Positive sites only for SL, Blue: Positive sites for both ON and SL, ON: Object naming, SL: Sign language.

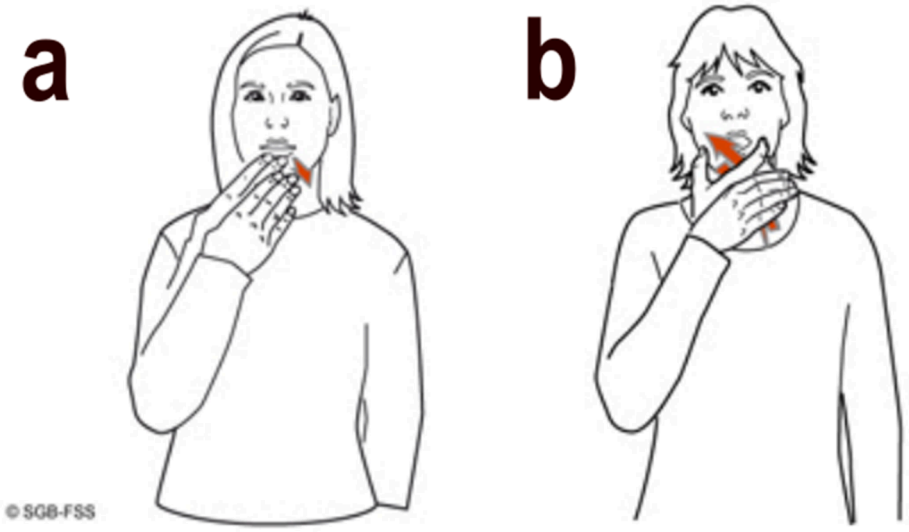

Fig. 4 – Representative signs for "thank you" (a) and "apple" (b) in GSL.

additional positive sites were identified for either task. Fig. 3 illustrates all DES-positive sites according to task on the left hemisphere individual surface mesh of the patient.

At site number 5 (Fig. 3), where stimulation induced a sign phonological error, when presented with the sign for "thank you" (Fig. 4a), the patient recognised it as the sign for "apple" (Fig. 4b). Although these concepts seem unrelated, an examination of the GSL signs reveals that both involve a hand movement starting near the mouth and moving outward (Fig. 4). The distinction lies in the hand configuration: the sign for "thank you" uses a straight palm, whereas the sign for "apple" employs a more closed palm, that resembles a half circle. The same site was negative for verbal object naming.

3.2. *Tractography*

The unique DES-positive sites for each language modality were used as seed ROIs. For object naming sites, 1, 3, and 4 were used and for sign recognition sites, 5, 6, and 7 were employed as ROIs for fiber-tracking (see Fig. 3 for site locations). Both sets of unique sites showed disconnections in parts of the arcuate fasciculus (AF) and the inferior fronto-

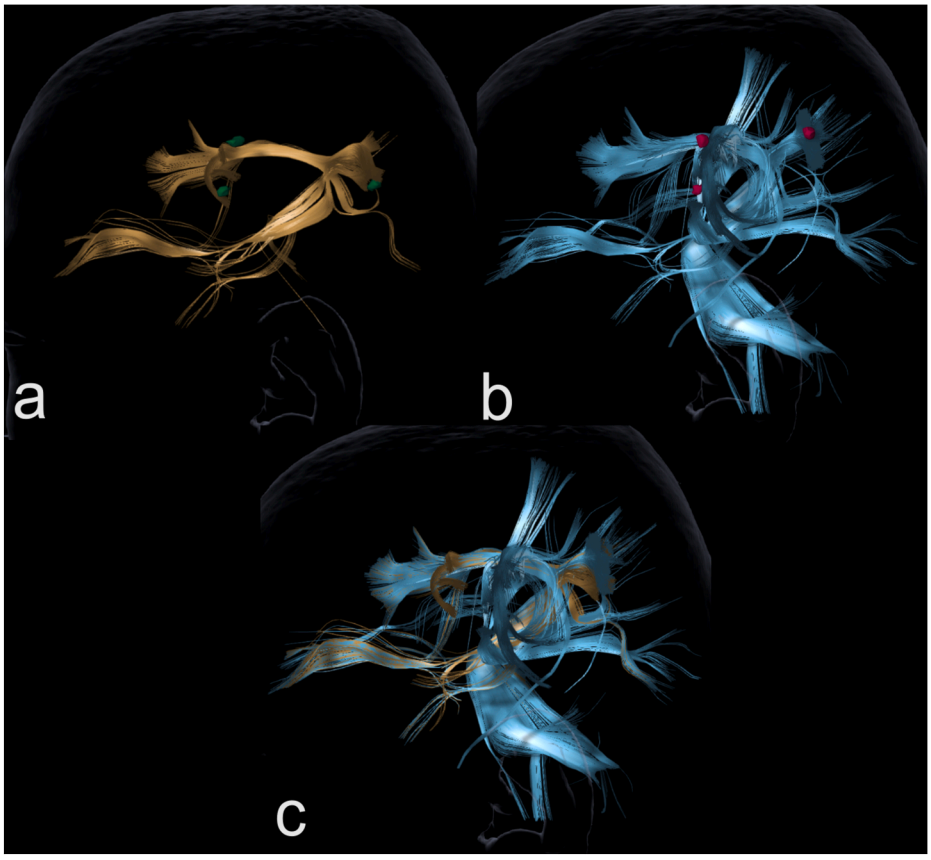

Fig. 5 – Visualization of disconnected fibers and unique DES-positive sites for object naming (a), sign recognition (b), and their overlap (c).

occipital fasciculus, albeit with varying degrees of disconnection (IFOF; Fig. 5a and b). Exclusive object naming sites disconnected on average 7% of the AF and 5.2% of the IFOF, whereas sign recognition sites disconnected on average 11.5% of the total volume of AF and 6.7% of the total IFOF volume. Fig. 6 visualizes overlapping and differential disconnected fibers that belong to the AF or the IFOF according to language modality.

In addition to the AF and IFOF, the positive sites of sign recognition showed disconnection of parts of the cortico-spinal tract (CST) and cerebellar tracts (Fig. 5b). However, this pattern was not observed for the traced disconnections related to object naming positive DES-sites, which did not overlap with either the CST or cerebellar tracts (Fig. 5a). The glioma was removed while preserving DES-positive sites and the postoperative MRI confirmed a gross total resection (Fig. 7).

4. Discussion

The patient of the present study, a bilingual user of both spoken German and GSL, presented a unique opportunity to explore the neural substrates of these distinct forms of communication. Using DES, we identified both shared and distinct cortical sites for the two language modalities. To our knowledge, this is the first study performing tractography from DES-positive regions to examine the connectivity of verbal and sign language, revealing significant subcortical overlap as well as notable differences between language modalities.

Although fMRI data predominantly indicate that the same cortical regions are activated for both spoken and sign languages, our findings using DES only partially align with these observations (Blanco-Elorrieta et al., 2018; Emmorey, 2021). We identified only one overlapping DES-positive point that when stimulated induced errors during both object naming and sign recognition (Fig. 3, site 2). This cortical site was located in the inferior SMG and when stimulated induced phonological paraphasias during object naming and anomia during sign recognition. The rest of our DES-positive sites, although located within the same cortical regions, were not identical for the two language modalities. For example, positive sites were identified for both modalities when stimulating parts of the SMG. However, the positive site for object naming appeared inferior to the DES-positive site for sign recognition.

Our observation that DES induced errors for tasks in the same cortical regions aligns well with previous fMRI findings in both signers and non-signers. The resolution of DES during awake surgery, however, can be higher than that of fMRI, as it does not require activation averaging according to atlas

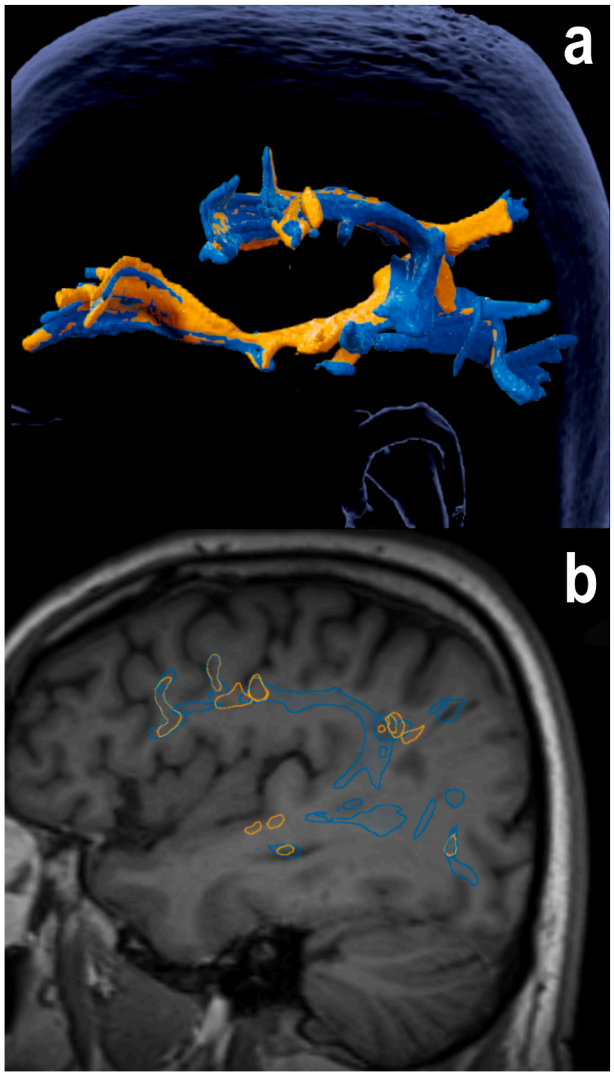

Fig. 6 – Disconnected fibers belonging to the AF or IFOF for object naming (orange) and sign recognition DES-sites (blue). a: 3D reconstruction, b: representative sagittal slice.

regions or voxel clusters. Additionally, DES demonstrates which cortical sites are essential for a given cognitive function, whereas fMRI indicates which active voxels participate in a cognitive process. This means that, for example, although the inferior frontal gyrus appears to be active in multiple fMRI studies of language processing, it might not be essential for fluent language production as revealed by lesion symptom mapping and DES studies (Duffau et al., 2014; Mandonnet & Duffau, 2021; Ntemou et al., 2023).

Previous case reports of intraoperative language mapping have yielded mixed findings, identifying either overlapping or distinct cortical sites for spoken and sign language (Barua et al., 2024; Lau et al., 2023). Although Lau et al. (2023) reported identical representations of sign and spoken language over the AF and IFOF, the elicited errors were restricted to phonological paraphasias at the word level during object naming and sign recognition (e.g., neologisms) and did not include phonological errors specific to sign language structure. In contrast, Barua et al. (2024) did not identify any overlap between spoken and sign language, although their DES-positive sites were derived from a semantic fluency task, which probes a different set of cognitive and linguistic processes than those examined here. We opted for object naming and sign recognition, two tests with similar sensory, cognitive, and articulatory demands. As described in the Methods, both tasks assessed visual recognition of object or sign depictions, access to semantic and phonological information of concepts and overt production of words with the same lead–in phrase (i.e., This is …). In contrast to semantic fluency tasks, naming tasks are more suitable for intraoperative use and are therefore more commonly used, as they are trial-based, can be time-locked to stimulation onset, and place fewer demands on executive control and sustained attention (Rofes, Mandonnet, et al., 2017; Shao et al., 2014). Given that previous DES-based case studies have employed tasks with different cognitive and sensory demands, varied operational definitions of sign-language-specific errors and stimulation sites, a direct comparison across studies remains challenging.

Our results reconcile previous discrepancies by demonstrating both overlap and divergence in cortical and

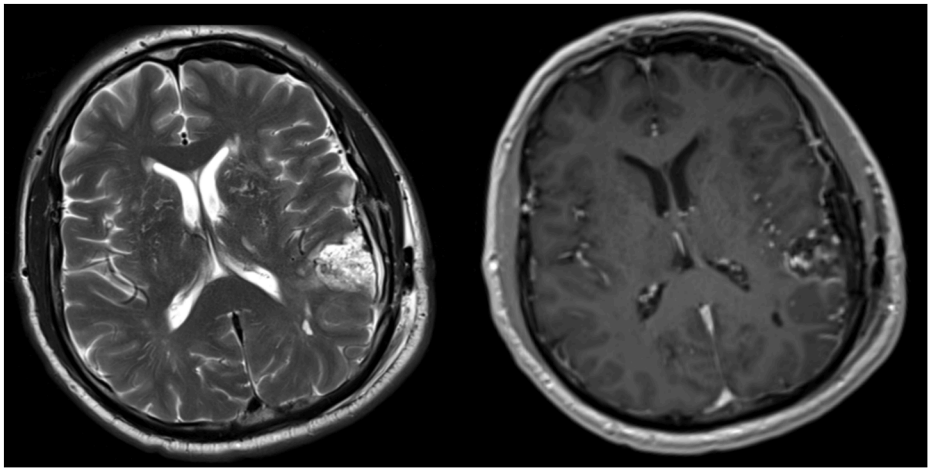

Fig. 7 – Axial view of postoperative T2 (left) and T1-weighted (right) MRI scans.

subcortical representations within the same individual. At the cortical level, stimulation of the same site within the SMG impacted both language modalities and caused anomic errors. However, sites that induced verbal phonological and sign phonological errors were distinct. Subcortically, the disconnected fibers indeed overlapped with both the AF and the IFOF, irrespective of language modality and as previously reported by Lau et al. (2023). The main difference at the subcortical level was that sign language sites additionally exhibited disconnections in the CST and cerebellar tracts, potentially including the cerebellothalamic tract.

The involvement of motor and sensory regions has been documented in fMRI studies, primarily during the production of signs and, to a lesser extent, during the comprehension of sign language (Corina & Knapp, 2006; Emmorey, 2021; Emmorey et al., 2007). This observation aligns with the theory of the mirror neuron system, which posits that sensorimotor regions are engaged not only during the execution of actions but also during the observation of human actions (Buccino et al., 2001, 2004). In our study, although tasks for both language modalities identified DES-positive regions in the post–central gyrus, only the sites relevant to sign recognition showed disconnections in the CST. This suggests that the sensorimotor system's engagement during sign language processing is more pronounced and potentially more critical compared to spoken language. The unique disconnection of the CST during sign recognition tasks underscores the specialized role of motor pathways in sign language, likely due to its inherent reliance on motor and visual-spatial processing.

In a seminal single-case study, Corina et al. (1999) assessed a signer during intraoperative naming of objects using signs. Although their study did not evaluate the representation of spoken language, their findings for sign language bear striking similarities to ours. For instance, the authors reported that stimulation of a site, which appears to be nearly identical site 5 in our study, produced a phonological paraphasia. In their case, the patient generated a sign that was phonologically similar to the target. Our study echoes this observation but extends it to the context of sign recognition. When stimulated at this site, our patient produced a word that was phonologically similar in terms of sign articulation, with the two signs differing only slightly in their manner of articulation (Fig. 3). This finding provides causal evidence for the role of the SMG in the phonological processing, extending beyond spoken language to include sign language as well (Corina et al., 1999; Corina & Knapp, 2006; Oberhuber et al., 2016). Furthermore, it demonstrates that this phonological function is crucial not only for the production of sign language but also for its comprehension, as evidenced by our use of a sign recognition task. This observation, however, should be interpreted with caution, as it is based on a single positive stimulation site. Nevertheless, given the inherent within-subject control of DES and the fact that intraoperative stimulation typically yields a limited number of positive sites in comparison to non-invasive methods (e.g., navigated TMS or fMRI; Bährend et al., 2021; Ille et al., 2015; Picht et al., 2013), the presence of a focal effect remains informative.

An additional factor that may contribute to the patterns observed in this case is the patient's language history. Unlike early bilinguals or congenitally deaf signers typically described in the literature, our patient acquired GSL in adulthood and only later integrated it into daily communication with a family member. Although no formal proficiency measures were available, the late age of acquisition is relevant, as several neurosurgical and neuroimaging studies have shown that later-learned languages may exhibit more spatially distinct cortical representations compared to early-acquired ones (Fernández-Coello et al., 2016; Perani et al., 2003; Połczyńska et al., 2016). Consequently, some of the non-overlapping DES sites identified for spoken and signed modalities may reflect not only modality-specific sensorimotor demands of sign language but also differences related to the patient's acquisition timeline (for a review see Martín-Fernández et al., 2022). Nevertheless, the presence of overlapping cortical and subcortical sites indicates that, even when acquired in adulthood, a sign language used regularly in daily life can engage the same core perisylvian regions as the first language. At the same time, however, it may still rely on partially distinct microanatomical systems within these shared regions.

Despite the novel insights offered by this case, several limitations should be acknowledged. As a single-case study, the findings cannot be generalized without caution, particularly because DES provides information only from surgically exposed areas and therefore cannot capture the broader functional network. In addition, although DES offers unique causal evidence, translating stimulation sites onto the patient's MRI is not entirely straightforward and may introduce spatial imprecision. We attempted to mitigate this by registering all stimulation coordinates directly onto the patient's preoperative MRI using a state-of-the-art neuronavigation system, thereby reducing operator dependence and variability (see Section 3.1). Even so, the resulting maps inevitably reflect the constraints of surgical exposure, task selection, and intraoperative conditions, all of which shape the cortical and subcortical regions that can be evaluated. These considerations should be taken into account when interpreting the present findings and when designing future studies.

5. Conclusion

In this study we presented the unique case of a bilingual individual proficient in both spoken German and GSL to investigate the neural substrates of these distinct forms of communication. Using DES, we identified both shared and distinct cortical and subcortical sites for the two language modalities, performing also tractography dissections from DES-positive sites to examine the connectivity of verbal and sign language. Our findings reveal significant subcortical overlap as well as important differences between the neural correlates of the two modalities, partially aligning with existing fMRI data but also highlighting the higher resolution of DES in pinpointing essential cortical sites for specific cognitive functions.

Notably, sign language sites exhibited disconnections in parts of the CST and cerebellar tracts, underscoring the specialized role of motor pathways in sign language processing. This study extends previous observations of phonological

paraphasias during sign production to sign recognition, providing causal evidence for the role of the SMG in phonological processing for both spoken and sign languages. On a theoretical level, our findings contribute to a deeper understanding of the neural basis of different forms of language and at a clinical level, they highlight the importance of patient-tailored DES protocols for intraoperative mapping.

Disclosures

The authors have no personal, financial, or institutional interest in any of the materials or devices described in this article.

Consent for publication

The patient of this case report gave informed consent for publication.

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Scientific transparency statement

DATA: Some raw and processed data supporting this research are publicly available, while some are subject to restrictions: https://github.com/EffyNtemou/Verbal_and_sign_language_using_DES_ProcessedDataMNI/tree/Coordinate_transformation.

CODE: All analysis code supporting this research is publicly available: https://github.com/EffyNtemou/Verbal_and_sign_language_using_DES_ProcessedDataMNI/tree/Coordinate_transformation.

MATERIALS: No study materials supporting this research are publicly available.

The authors provided the following justification for restricting access to materials: '*As described in the paper, the stimulus materials used in this study were not developed by the authors but are standardized, copyrighted materials originally developed and published by* Ohlerth et al. (2020) *and* Huber et al. (1980). *The authors do not hold the copyright to these materials and therefore do not have the legal right to redistribute them via a public repository. For this reason, we provide a citation to the original publications describing the materials, which is the appropriate access point for these instruments. For access to the psychometric instruments, please, contact the authors of the original studies.*'

DESIGN: The authors indicated that reporting the sample size determination, data exclusions, and inclusion and exclusion criteria was not applicable to this research. They provided the following justification: '*This is a case report of a single patient. Sample size calculations are not applicable. All relevant details regarding the patient have already been described in the manuscript.*'

PRE-REGISTRATION: No part of the study procedures was pre-registered in a time-stamped, institutional registry prior to the research being conducted. No part of the analysis plans was pre-registered in a time-stamped, institutional registry prior to the research being conducted.

For full details, see the *Scientific Transparency Report* in the supplementary data to the online version of this article.

Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.cortex.2025.12.010.

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