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Language improvement after awake craniotomy in a 12-year-old child: illustrative case

Ellen Collée, MA,[1] Djaina Satoer, PhD,[1] Barbara Wegener Sleeswijk, MA,[2] Markus Klimek, MD, PhD,[3] Marion Smits, MD, PhD,[4] Marie-Lise Van Veelen, MD, PhD,[1] Clemens Dirven, MD, PhD,[1] and Arnaud Vincent, MD, PhD[1]

Departments of [1]Neurosurgery, [3]Anesthesiology, and [4]Radiology & Nuclear Medicine, and [2]Ear, Nose, and Throat, Hearing and Speech Center, Erasmus MC – University Medical Center, Rotterdam, Zuid-Holland, The Netherlands

BACKGROUND Although the standard procedure to treat adult patients with lesions in eloquent brain areas is awake craniotomy with direct electrical stimulation, this procedure is not often used in children because of feasibility concerns. Some studies have shown that the procedure is feasible in children. They reported the postoperative language ability, which was not based on standardized language tests for children. To give an objective overview of preoperative assessment of the language ability of a child before and after this procedure, the authors described the perioperative course, including standardized language tests for children and the awake surgery setting, of a 12-year-old child undergoing awake craniotomy with brain mapping for the indication of cavernoma in the left somatosensory cortex close to the motor cortex.

OBSERVATIONS The patient performed better on language tests after surgery, showing that his language ability improved. He also cooperated well during the entire perioperative period. His mother was present during the awake surgery, and the patient tolerated the surgery well.

LESSONS The authors conclude that awake craniotomy is indeed feasible in a child and that it can even result in an improved postoperative language outcome. It is, however, crucial to carefully assess, inform, and monitor the child and their proxies.

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KEYWORDS awake craniotomy; brain mapping; pediatrics; language testing; language improvement

The standard treatment for adults with lesions in eloquent areas of the brain is resection via awake craniotomy with direct electrical stimulation. This procedure is used for different indications, such as epilepsy, tumors, or vascular malformations (e.g., cavernous hemangioma/cavernoma).[1] Because the patient is awake during the procedure, neurological and higher cognitive functions (e.g., language, motor) can be monitored. This results in a larger extent of resection, which in the case of cavernoma resection is also related to better seizure-free results after surgery.[2,3] It also results in maintenance of neurological and cognitive functions and quality of life.[4]

The awake procedure is scarcely used in children because it can be psychologically challenging. Additionally, uncertainties concerning the technical side of the surgery (safety, anesthetic procedure) and feasibility in general exist.

However, some studies in children investigated the feasibility of the procedure and the anesthetic management possibilities and reported on how to adapt the surgery for children. These studies vary from case reports[5–7] to larger studies describing 6 to 10 children[8,9] up to 28 children[10] ≥7 years old. They conclude that awake surgery in children is indeed feasible as long as they are carefully prepared and monitored.

In general, no permanent postoperative cognitive deficits were found in these studies. These outcomes were only briefly described and based on observations, except for Delion et al.,[8] who also administered standardized language tests. They used the Boston Diagnostic Aphasia Examination[11] pre- and postoperatively and an object naming test based on the Oral Denomination 80 (DO80)[12] intraoperatively. However, these tests are designed for adults. Delion

ABBREVIATIONS CELF-5^{NL} = Clinical Evaluation of Language Fundamentals 5; DIMA = Diagnostic Instrument for Mild Aphasia; DuLIP = Dutch Linguistic Intraoperative Protocol; EEG = electroencephalography; MRI = magnetic resonance imaging.

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et al.[8] report the performance level based on these adult norms, which may not be accurate for children. The raw scores, which may enable one's own interpretation, were not provided. Therefore, interpretation of these results is difficult.

Administration of standardized language tests specifically for children would give more insight into the language ability of the child in the perioperative period, and it can exclude the possibility of a developmental language disorder. Additionally, assessment should include the language tests used in the awake surgery setting. In this way, the child can be optimally prepared for, monitored during, and followed after an awake surgery.

We describe the perioperative course of a 12-year-old child undergoing awake craniotomy with brain mapping for resection of cavernoma in the left somatosensory cortex, in close proximity to the motor cortex. A detailed language assessment consisting of standardized language tests for children and for the awake surgery setting was used.

Illustrative Case

A Dutch 12-year-old, right-handed boy attended secondary school (first year, higher general secondary education/preuniversity education) in The Netherlands. He had developed epileptic seizures 1.5 years earlier. During seizures, there were occurrences of mouth and lip convulsions, tingling in the fingers of the right hand and the right foot, contractions and tremors in the right arm, and salivation in the right corner of the mouth. Speech production was impaired but language comprehension remained intact. After a seizure, speech was no longer impaired. Three cavernomas (left occipital, left parietal, and right frontal lobe) were present. When the seizures started to occur more often and anticonvulsants did not stop them (only diminished them slightly), additional assessments were performed to give more insight into a possible treatment plan to stop the seizures.

Contrast-enhanced three-dimensional T1-weighted and susceptibility-weighted magnetic resonance imaging (MRI) showed that the largest cavernoma was localized in the left postcentral gyrus (Fig. 1A and B). Electroencephalography (EEG) showed that epileptiform abnormalities sporadically occurred in the lesion in the left parietal lobe while the patient experienced tingling in the right hand. This sensation was sometimes also reported when no clear abnormalities on the EEG were seen. Given this information, in combination with the effects of the seizures on the right hand, it was assumed that the epilepsy was symptomatic localization-related to the lesion in the left parietal lobe.

This cavernoma was localized in close proximity to the eloquent motor cortex, with the lesion in the postcentral gyrus obliterating the central sulcus and compressing the precentral gyrus, as shown by brain activation adjacent to the lesion during a bilateral finger-tapping task during a functional MRI scan (Fig. 1C). Additionally, diffusion tensor imaging fiber tracking showed a close relation between the lesion and the corticospinal tract (Fig. 1D). Resection via awake craniotomy with direct electrical stimulation was suggested, if the patient were able to cooperate sufficiently.

Preoperative Procedure

The patient was treated by a multidisciplinary team consisting of pediatric specialists and experienced awake surgery specialists. Preoperatively, the patient was extensively evaluated and informed by anesthesiologists and neurosurgeons. The patient was tested by clinical linguists.

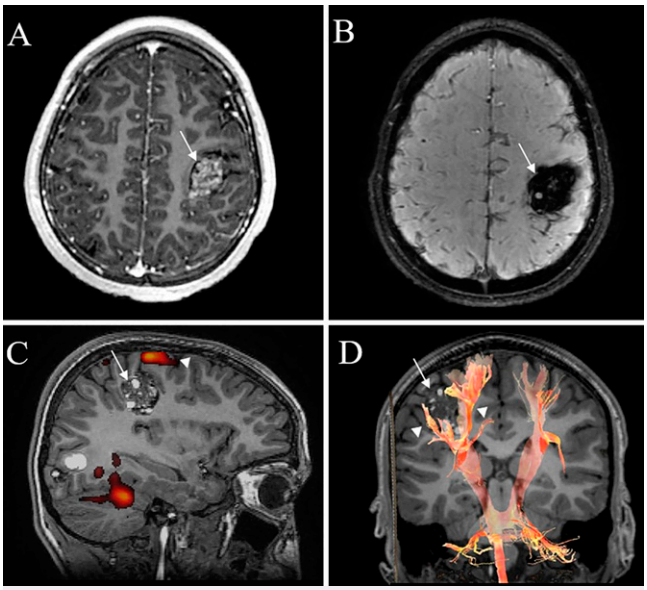

FIG. 1. Preoperative scans of the left parietal cavernoma. **A:** Axial postcontrast T1-weighted image showing a typical "popcorn effect" of the cavernoma (*arrow*). **B:** Axial susceptibility weighted image showing signal loss of the lesion (*arrow*), consistent with hemosiderin deposition, typical of cavernoma. **C:** Sagittal T1-weighted image with functional MRI finger tapping activation indicating the primary motor cortex (precentral gyrus; *arrowhead*) as an overlay. The lesion causes mass effect on the precentral gyrus, which is located anterior to the lesion (*arrow*). **D:** Coronal precontrast T1-weighted image with fiber tracking as an overlay showing the close proximity of the lesion (*arrow*) to the corticospinal tract (*arrowheads*).

The patient and mother reported mild word-finding difficulties. Additionally, the mother emphasized that the patient's articulation was often not clear, but the patient himself did not report any articulation difficulty. There were no complaints concerning reading, spelling, and language comprehension. The patient reported that his handwriting was untidy but that this had always been the case. The patient did not have any difficulty keeping up at school. No disorders in language acquisition or speech therapy had ever been noted or needed.

The Clinical Evaluation of Language Fundamentals 5 (CELF-5^{NL}),[13] a test to diagnose language and communication difficulties in children aged 5 to 18 years, was administered. On all production and comprehension subtests an average or above average score was found, except for a low score on a production task in which sentences had to be formulated (Table 1). Language production in general (89 measured by the Expressive Language Index) was just within the limits of an average score (86–114), whereas language comprehension (127 measured by the Receptive Language Index) was above average ($\geq$115; Table 1), showing a large difference between the two domains. All index scores were intact, indicating that no language or communication deficit was present.

Based on these scores, additional language production tests at different linguistic levels were administered to prepare for intraoperative monitoring (Table 2). Some new, unpublished subtests of the Dutch Linguistic Intraoperative Protocol[14] (DuLIP) were used, for which no norms were available. However, even without comparing

TABLE 1. Pre- and postoperative scores on the CELF-5^{NL} divided by subtest, corresponding levels, and change in levels

Subtest	Preoperative			Postoperative			Preoperative→ Postoperative Change
	Scaled/ Standard Score	Percentile	Level	Scaled/ Standard Score	Percentile	Level	
Word categories	14	91	Above average	14	91	Above average	=
Following instructions	15	95	Above average	13	84	Above average	=
Formulating sentences	5*	5	Low	10	50	Average	↑
Repeating sentences	10	50	Average	15	95	Above average	↑
Word definitions	9	37	Average	9	37	Average	=
Combining sentences	10	50	Average	11	63	Average	=
Semantic relations	13	84	Above average	13	84	Above average	=
Index							
Core Language Score	103	58	Average	120	91	Above average	↑
Receptive Language Index	127	96	Above average	122	93	Above average	=
Expressive Language Index	89†	23	Average	111	77	Average	=‡
Language Memory Index	100	50	Average	117	87	Above average	↑

Standard scores subtests: ≤6 = low; 7 = below average; 8–12 = average; ≥13 = above average.
Standard scores index: ≤70 = very low; 71–77 = low; 78–85 = below average; 86–114 = average; ≥115 = above average.
* This is a low score.
† Borderline average score (close to below average scores of ≤ 85).
‡ While the level is the same pre- and postoperatively (average), the standard score has improved from 89 (borderline average) to 111 (high within the average range).

performance to norm scores, the patient seemed to score high on semantic tests: object naming, odd picture out, and semantic association (visual/written presentation). Additionally, he scored above average on semantic fluency, for which suitable norms were available. According to adult norms (age: <55 years; education: ≤12 years), the patient scored below average on two out of three auditory phonology tests (word repetition of simple and compound words) of the Diagnostic Instrument for Mild Aphasia[15] (DIMA). Errors consisted of stammering with self-correction and a staccato response. Additionally, the patient scored below average on a test for spontaneous speech in context/syntax of the DIMA (sentence completion). Errors consisted of no response and a delayed response. Some unintelligible speech was observed in spontaneous speech and during some of the tasks.

In summary, the preoperative language tests showed no deficits in language production or language comprehension, except for a low score on formulating sentences. Notably, the patient scored high on language comprehension but lower on language production (just within the range of average scores), measured by the CELF-

TABLE 2. Pre- and postoperative scores on language tests for intraoperative monitoring, the corresponding levels or percentages correct, and change in levels

Test	Preoperative		Postoperative		Preoperative→ Postoperative Change
	Raw Score	Level/% Correct	Raw Score	Level/% Correct	
DuLIP object naming	71/71	100%	71/71	100%	=
DuLIP odd picture out	19/20	95%	25/25	Average	=
DuLIP semantic association	15/15	100%	22/25	Average	=
Semantic fluency (animals)	29	Above average	31	Above average	=
DIMA					
Word repetition, e.g., "*con**struc**tie*" (construction)	9/10	Below average	10/10	Average	↑
Word repetition (compounds), e.g., "***feest**verlichting*" (party lights)	9/10	Below average	10/10	Average	↑
Nonword repetition, e.g., "*an**á**to*"	10/10	Average	10/10	Average	=
Sentence completion, e.g., "*Ik luister naar …*" (I am listening to …)	8/10	Below average	9/10	Average	↑
Total	36/40	Average	39/40	Average	=

New unpublished subtests of the DuLIP were administered, for which no norms were available yet. Postoperatively, some subtests of the DuLIP were used and interpreted with the existing norms for adults. Note that all interpreted scores are based on adult norms.

5[NL]. In other production tasks at word (repetition) and sentence level (sentence completion), lower but not impaired scores (below average) were found. The complaints by mother and patient concerning word-finding and articulation were not objectified during these assessments.

The patient was motivated and willing to undergo the awake procedure with his mother being present in the operating room. Extra time was spent with the mother to inform and prepare her as well. The patient's family was very supportive. After extensive evaluations and testing and considering the positive attitude of the patient and the support of his family, the patient was expected to be able to cope during awake surgery and we decided to go ahead with the awake procedure.

At this point, the patient used two anticonvulsants daily: carbamazepine (morning: 200 mg; evening: 300 mg) and levetiracetam (morning: 500 mg; evening: 250 mg). Midazolam nasal spray (7.5 mg) was used when necessary.

Intraoperative Procedure

An asleep-awake-asleep procedure with direct electrical stimulation was performed. Stimulation was applied with a bipolar electrode at the cortical level. The patient's mother was present in the operating room at the induction and awake part of the procedure for reassurance and support. Word repetition (two syllables) was administered during cortical stimulation. Contractions of the right hand and fingers were elicited during stimulation (Fig. 2). Additionally, dysarthria was elicited once, but it was not reproducible. During resection, semantic tasks such as object naming, odd picture out, and short fluency tasks related to his interest/age (e.g., sports, car brands, crisps flavors) were alternated with spontaneous speech and motor tasks (e.g., finger tapping, squeezing, making a fist, giving thumbs up). Performance on these tasks was good and stable. A focal seizure occurred, manifesting as short jolts in the right hand. Ice water was applied directly onto the brain surface and the seizure stopped. At the end of resection, a deterioration in motor function occurred. The ability to extend the fingers of the right hand was reduced, while the patient remained able to squeeze and make a fist. Language performance remained stable throughout resection. The patient tolerated the procedure well; he was very motivated and cooperative.

Postoperative Procedure

Directly after surgery, the patient developed paresis and tingling of the right hand. Tingling was also present in the right foot. Over the next few days, some short focal seizures occurred in which the mouth and right hand twitched. Two days after surgery, the anticonvulsants were changed. Levetiracetam was stopped and carbamazepine was increased to 300 mg twice daily. Midazolam nasal spray (7.5 mg) was still used when necessary.

The patient received physiotherapy to train the hand function. The patient did not have any seizures after surgery until the anticonvulsant carbamazepine was lowered and stopped. After increasing the dose again (200 or 300 mg, depending on patient's needs, twice daily), no more seizures occurred. A postoperative MRI 3 months after surgery showed that the resection was complete (Fig. 3).

At the 3.5-month follow-up, the patient was feeling well, was able to concentrate, and performed well at school. His mother indicated that he was more cheerful and energetic than before surgery and that his handwriting had improved. No complaints concerning language comprehension, language production, or reading were present.

During this follow-up, the patient was assessed with the same tests as performed preoperatively by the same clinical linguists. On all production and comprehension subtests of the CELF-5[NL], average to above average scores were found (Table 1). Language comprehension and language memory (as measured by the Receptive Language Index and Language Memory Index) were above average. Language production (as measured by the Expressive Language Index) was average, with a standard score (111) on the high end of the average range (86–114). Compared to preoperative test performance, the patient improved on formulating and repeating sentences, the Core Language Index, and the Language Memory Index (Table 1). The intact scores indicated that no language or communication deficit was present after surgery.

Published and unpublished subtests of DuLIP, semantic fluency, and DIMA were administered and interpreted with adult norms when possible (Table 2). The patient scored average to above average on semantic tests (object naming, odd picture out, semantic association [visual], semantic fluency). Average scores were also found on the DIMA phonology tests (word repetition, word repetition compounds, nonword repetition) and on a test for spontaneous speech in context/syntax (sentence completion; one delayed response occurred). Compared to preoperative test performance, the patient improved on word repetition, word repetition compounds, and sentence completion (Table 2). Some unintelligible speech was still occasionally observed during spontaneous speech and some tasks.

At the 5-month follow-up, hand function had improved to almost preoperative level, but tingling or cramps in the right hand occurred

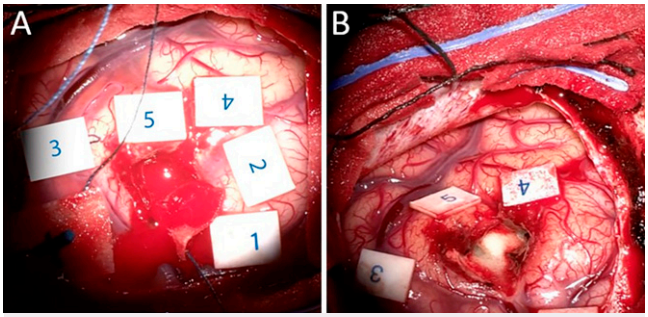

FIG. 2. The intraoperative positive stimulation points marked by numbers before (**A**) and after (**B**) resection.

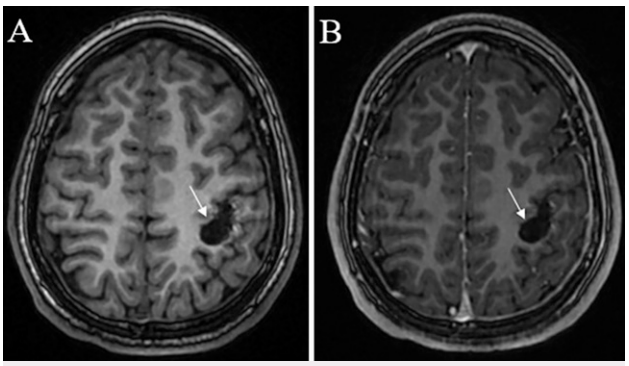

FIG. 3. Postoperative MRI scans of the resection cavity (*arrows*) at 3 months. **A:** Axial precontrast T1-weighted image showing the resection cavity without residual cavernoma. **B:** Axial contrast-enhanced T1-weighted image showing the resection cavity without residual cavernoma.

daily. No seizures had occurred since changing the medication. He took 200 mg of carbamazepine twice daily.

A year after surgery, the patient's mother reported that the patient was doing well. He performed well at school. His hand function had recovered to baseline level. Sporadically, mini seizures of 2 seconds occurred when there was less structure in the patient's daily life (e.g., during Christmas break when he did not attend school).

Discussion

Observations

We described the perioperative course of a 12-year-old patient with cavernoma who underwent awake craniotomy with direct electrical stimulation. Extensive language testing was performed pre-, intra-, and postoperatively. The patient scored similarly on many language tests postoperatively compared to preoperatively. Interestingly, he did not decline on any language tests and even improved on some language tests (Tables 1 and 2). He improved from a preoperative low score on formulating sentences to a postoperative average score, moving from the 5th to the 50th percentile. He also improved from a preoperative average score on repeating sentences to a postoperative above average score, moving from the 50th to the 95th percentile. Language production (as measured by the Expressive Language Index) remained average but improved from a standard score of 89 (borderline average) to 111 (high within the average range). The large preoperative difference between the borderline average language production and the above average language comprehension resolved postoperatively. Language memory (as measured by the Language Memory Index) and the Core Language Index improved from average to above average. The patient also improved on two DIMA phonological tests (word repetition of simple and compound words) and the DIMA spontaneous speech in context/syntax test (sentence completion) from below average to average. These higher scores showed that the language production of the patient improved after surgery. Such a language improvement, based on standardized tests, has not been reported in a child undergoing awake surgery before. This result is important because it shows that awake craniotomy can indeed protect and even improve language function in children, as reported in adults as well.[16] To assess the child's language ability even more accurately in the future, intraoperative language tasks designed for children are needed.

The patient tolerated the awake procedure well. This has previously also been reported in other studies.[10–15] In our opinion, the tolerability is partly due to extensive assessments, providing information, careful monitoring, and attitude of the patient and family. The patient was exceptionally motivated and willing to undergo the procedure. His family was very involved and supportive. The patient's mother accompanied and supported him during most of the surgery, which contributed substantially.

Lessons

Based on this illustrative case, we suggest that awake craniotomy in a child is feasible and can even result in an improved postoperative language outcome. However, every child eligible for this procedure should be carefully assessed, informed, and monitored by a multidisciplinary team (pediatric and awake surgery team) because not all children may tolerate the procedure well. Support of a child's proxies is crucial as well.

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Disclosures

Dr. Klimek reported personal fees from Paion AG, fees as member of a data safety monitoring board and personal fees from De Tijdstroom, and royalties as a textbook editor outside the submitted work. No other disclosures were reported.

Author Contributions

Conception and design: Collée, Satoer, Wegener Sleeswijk, Klimek, Van Veelen, Dirven. Acquisition of data: Satoer, Wegener Sleeswijk, Klimek, Smits, Vincent. Analysis and interpretation of data: Collée, Satoer, Wegener Sleeswijk, Smits. Drafting the article: Collée. Critically revising the article: all authors. Reviewed submitted version of manuscript: Collée, Satoer, Wegener Sleeswijk, Klimek, Smits, Dirven, Vincent. Approved the final version of the manuscript on behalf of all authors: Collée. Anesthesiological Care: Klimek, Vincent.

Correspondence

Ellen Collée: Erasmus MC – University Medical Center, Rotterdam, Zuid-Holland, The Netherlands. k.collee@erasmusmc.nl.