Swallowing Dysfunction in Adult Patients with Chiari I Malformation

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J Neurol Surg B 2018;79:606–613.

Abstract

Keywords

- ▶ Arnold–Chiari Malformation
- ▶ deglutition disorders
- ▶ deglutition
- ▶ surveys and questionnaires
- ▶ decompressive craniectomy
- ▶ treatment outcome
- ▶ adult

Background Swallowing difficulties have been reported in patients with Chiari I malformation (CMI) with a prevalence of 4 to 47%, but existing evidence is based only on case reports. We aimed to prospectively study swallowing function in adult patients with CMI before and 3 months after surgical decompression.

Methods We included all adult patients diagnosed with CMI from September 2015 to October 2017 who underwent a planned surgery at Sahlgrenska University Hospital, Sweden. The patients were offered the opportunity to participate in and undergo an assessment consisting of the “Watson Dysphagia Scale (WDS)” and “EORTC QLQ-OG25” written questionnaires in addition to videofluoroscopic examination of swallowing (VFS) before and 3 months after surgery. Demographic data and comorbidities were recorded.

Results Eleven patients were included, nine of which underwent both pre- and postoperative evaluations. Four patients (36%) reported varying degrees of swallowing complaints (mean WDS score, 16). In two of these, there was substantial penetration of contrast material into the laryngeal vestibule on VFS, and in the other two patients, minor swallowing disturbances were observed. Borderline deviations from normal VFS findings were also found in three asymptomatic patients. Although not all VFS deviations completely disappeared after surgery, the patients reported no remaining symptoms.

Conclusion Symptoms of dysphagia and objective abnormalities on VFS are not uncommon in CMI patients. Surgery has the potential to remedy underlying causes of dysphagia, thereby relieving its symptoms.

Introduction

Chiari I malformation (CMI) is defined as the caudal descent of the cerebellar tonsils > 5 mm below the level of the foramen magnum.¹ This definition is controversial as shown

in previous reports where cerebellar tonsillar herniation 3 to 5 mm below the foramen magnum considered borderline CMI and clearly pathologic when it exceeds 5 mm.²

The prevalence of CMI has been estimated to range from 0.8 to 1%,^{3,4} and most patients who become symptomatic present symptoms between the third and fourth decades of life.⁵ The typical clinical manifestation of CMI is an occipital headache of short duration that is aggravated by exertion,

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received
January 12, 2018
accepted after revision
April 15, 2018
published online
May 25, 2018

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Stuttgart · New York

DOI <https://doi.org/10.1055/s-0038-1655758>.
ISSN 2193-6331.

coughing, or laughing.^{5,6} Otoneurological disturbances such as dizziness, pressure in the ears, tinnitus or decreased hearing are less common presentations of CMI.^{5–7} These symptoms could be explained by cerebellar tonsillar descent causing compression and traction of the brain stem and cranial nerves.⁸

The prevalence of dysphagia in CMI patients has been generally examined retrospectively and reported to range from 4 to 47%.^{6,9,10} A few reports have described cases in which dysphagia was the sole presenting symptom of CMI and was improved following occipitocervical decompression.¹¹ Thus, the current evidence regarding whether swallowing dysfunction is a treatable manifestation of CMI is mainly based on retrospective observations.

The subjective symptoms of dysphagia can be evaluated using validated swallowing assessment questionnaires, which assess whether dysphagia is present and, if present, can determine its severity as well as its impact on health-related quality of life.

The gold-standard diagnostic tool for examining oropharyngeal swallowing dysfunction is videofluoroscopic examination of swallowing (VFS). However, this subjective assessment tool has insufficient accuracy due to poor intra- and inter-rater reliability.¹² Objective measures of displacement and timing parameters can be used to adjust for interobserver variability. Comparing objective measurements with known normative data and stratifying by age and sex can help identify abnormal elements on VFS.¹³ Objective measures also provide a reproducible baseline for examining the efficacy of a therapy for treating swallowing function. Certain measures, such as the pharyngeal constriction ratio (PCR), have been validated with high-resolution manometry and are thus reliable surrogate measures of pharyngeal function.¹⁴

The purpose of this study was to prospectively evaluate the occurrence of pre- and postoperative swallowing difficulties in consecutive adult patients with CMI and to determine whether surgical treatment can help such patients. To the best of our knowledge, this is the first study to investigate swallowing function in CMI patients using both subjective and objective assessment tools.

Patients and Methods

Patients

All adult (>18 years old) patients from the region of Western Sweden (population, 1.87 million inhabitants) who were diagnosed with de novo CMI and referred to Sahlgrenska University Hospital for surgery were offered the opportunity to participate in this study. The inclusion criteria were symptoms compatible with CMI and a tonsillar descent of 5 mm or more. We refer patients to a neurologist for clinical assessment before inclusion whenever a nonspecific type of headache is encountered to exclude other causes other than CMI. We excluded patients who were previously diagnosed with swallowing difficulties caused by primary gastrointestinal disorders, patients who underwent an operation in the upper gastrointestinal system, and women who were

pregnant or breastfeeding (a beta hCG test was offered to women of childbearing age before inclusion in the study). We also excluded patients with severe mental illnesses who were unable to perform the tests or otherwise unable to participate.

Fourteen patients with CMI were identified during the inclusion period (September 2015–October 2017). Three patients chose not to participate. All 11 remaining patients provided written informed consent to participate in the study.

Ethics

This study was approved by the regional ethics board of the University of Gothenburg (Diary no. 865–14) and performed according to the principles of the Declaration of Helsinki.

Neurosurgical Approach

The surgical technique consisted of a surgical decompression in line with a standard protocol that involves a suboccipital craniectomy (3 × 3 cm) plus cervical vertebra (C1) laminectomy. The cervical vertebra (C2) laminectomy was performed only in patients with severe forms of cerebellar tonsillar herniation behind the C2 lamina. The dura was opened, and the arachnoid adhesions between the cerebellar tonsils and upper cervical cord and medulla oblongata were released. The dura was then closed using a periosteal and fascia graft obtained from a nearby occipital area. Sutures were placed in a watertight fashion to close the subcutaneous tissue and skin.

Study Design

Clinical data were collected for each patient and registered in a case report form. Swallowing was assessed using swallowing assessment questionnaires (the Watson Dysphagia Scale [WDS] and EORTC QLQ-OG25) and VFS. All assessments were performed both preoperatively and 3 months after surgery. The demographic information and past medical and surgical history obtained from the patients are shown in ►Table 1.

The Watson Dysphagia Scale

WDS is a validated tool for evaluating dysphagia. The test uses nine types of food with different consistencies (ranging from liquids to solid food), and the frequency of problems encountered swallowing the food is recorded as 1 = always, 0.5 = sometimes, or 0 = never. The test results in a score between 0 (no dysphagia) and 45 (severe dysphagia).^{15,16}

EORTC QLQ-OG25

The QLQ-OG25 questionnaire assesses the health-related quality of life of patients with esophagogastric cancer. It was previously applied in randomly selected samples drawn from the Swedish population, and these scores are used as reference values in comparisons with those obtained in the present study.¹⁷ It contains 25 items with six scales for evaluating dysphagia, eating restrictions, reflux, odynophagia, pain, and anxiety in addition to 10 single items. The time frame of the QLQ-OG25 module is “during the last week.”¹⁸ We excluded the following three items that were deemed irrelevant to CMI patients: body image, weight loss, and hair loss.

Table 1 Demographics and medical histories of the included CMI patients (N = 11)

Variable		Number of cases
Age (years), median (IQR)	33 (28)	
Gender	M/F	3/8
BMI (kg/m ²)	≤ 25/ > 25	6/5
Medical history	None	9
	Diabetes mellitus	1
	Asthma	1
Surgical history	None	8
	Non-neurosurgical procedures	3
	Neurosurgical procedures	0

Abbreviations: BMI, body mass index; CMI, Chiari I malformation; IQR, interquartile range.

Videofluoroscopic Examination of Swallowing

The patients were examined using a multipurpose fluoroscopy system (Siemens Artis Zee, Erlangen, Germany) while seated in a lateral position. Fluoroscopy data were digitally stored at a frame rate of 15 frames per second (video matrix, 1024 × 1024). The field of view included the tip of the tongue anteriorly, the pharyngeal wall posteriorly, the soft palate superiorly, and the seventh cervical vertebra inferiorly. A coin with a known diameter (18 mm) was placed under the chin to calibrate spatial measurements. All examinations were performed by a gastro-intestinal radiologist (MA). The patients swallowed six different boluses, four of which consisted of thin contrast medium (3, 5, 10, and 20 mL of Mixobar colon: 1 g Ba/mL mixed with an equal amount of water). For the remaining two boluses, the thin contrast was mixed with “Instant Food Thickener” (Thick & Easy, Hormel Health Labs, Savannah, Georgia, United States) to produce mildly thick and extremely thick consistencies, of which 5 and 3 mL, respectively, was offered to the patients. The subjects were presented with each measured bolus and instructed to hold the bolus in their mouth until they received a command to swallow. The patients swallowed each bolus twice. For the 20 mL of thin liquid, the patients were instructed to drink freely through a straw from a cup.

Analysis of Videofluoroscopic Examination of Swallowing

The same gastrointestinal radiologist who performed the examinations conducted the analyses, and this individual was only blinded to the results of the self-report questionnaires and their temporal relationship with the operations. The digital VFS technique allowed us to analyze the data in a detailed, slow-motion, frame-by-frame manner, and thus, we could obtain measurements from static images. Objective measurements were obtained as described by Leonard et al.¹³ Maximal hyoid displacement (H_{max}) was defined as the distance in centimeter from the anterior aspect of the hyoid bone in the “hold” position to its point of maximal anterosuperior displacement during swallowing.

The pharyngeal area (PA) was determined in the “hold” position by calculating the area (cm²) bordered by the soft palate, the posterior pharyngeal wall, the arytenoid cartilages,

the epiglottis, the valleculae, and the base of the tongue. The PCR was calculated as the ratio between the PA at maximal pharyngeal constriction while swallowing a 20-mL bolus and the PA in the “hold” position for a 3-mL bolus.

The opening of the upper esophageal sphincter (UES_o) was measured in the anteroposterior plane at the narrowest point between C4 and C6 when the bolus induced maximal distention. The duration of UES_o was measured in seconds from the start of the opening of the pharyngoesophageal segment to the time of complete closure of the segment.

To maximize the information gained from VFS, we included some categorical variables that are commonly used to assess swallowing dysfunction. To assess potential penetration into the laryngeal vestibule, we applied Rosenbek’s Penetration-Aspiration Scale (PAS).¹⁹ The PAS is used to describe penetration and aspiration events on a scale from 1 (no material enters the airway) to 8 (material enters the airway and passes below the vocal folds, and no effort is made to eject). The vallecular retention of contrast in the lateral view was scored using four grades.²⁰ Grade 0 indicated no residue, grade 1 indicated that residue occupied <10% of the width of the valleculae, grade 2 indicated that residue occupied between 10 and 50% of the width of the valleculae, and grade 3 indicated that residue occupied >50% of the width of the valleculae. Problems with initiating swallowing, the premature loss of the bolus from the oral cavity and/or delayed onset of the pharyngeal swallowing phase were also subjectively estimated.

Definition of Abnormal Videofluoroscopic Examination of Swallowing Findings

The H_{max} , PCR, and UES_o values were considered abnormal if they were more than one standard deviation from the normal mean for the corresponding sex and age category.^{13,14} A PAS score of 3 or higher was considered pathological because such scores have not been observed in normal materials in the relevant age group.²¹ There is currently no consensus in the literature regarding the limits of normality of post-swallow pharyngeal fluid residue.²² We calculated and compared the pre- and postoperative percentage of swallows with grade 2 or higher residue in the valleculae. Because there was also substantial variability in

the timing and temporal measures of healthy swallowing, we only report a subjective impression of deviations from normal related to problems with initiating swallowing or a delayed onset of the pharyngeal swallowing phase.²³

Statistical Analysis

This is a descriptive study, and data are presented as the mean (standard deviation). No statistical analysis was performed due to the small sample size.

Results

Clinical Presentation of CMI and Outcomes

The clinical presentations of the patients are summarized in ►Table 2.

All four patients with swallowing difficulty reported a problem with fluids. Two of the patients also had problems with swallowing solid food. The reported frequency of experiencing problems with swallowing was 1 to 2 per week in three of the patients and once monthly in one patient. The duration of swallowing complaints ranged between 6 and 36 months. No temporal worsening of symptoms or associated pain was reported. Two patients reported that the bolus temporarily stopped in the throat, while the other two reported it stopped in the chest. Neither regurgitation nor vomiting was reported, and there were no problems with the subsequent passage of the bolus through the gastrointestinal system. The clinical outcomes after surgical decompression were assessed 3 months postoperatively. Preoperative swallowing problems were resolved at

Table 2 Clinical findings in CMI patients (N = 11)

		Number of cases (%)	
		Preop	Postop
Clinical presentation	Headache	9 (81%)	4 (36%)
	Weakness	5 (45%)	1 (9%)
	Sensory loss	5 (45%)	2 (16%)
	Blurred vision	5 (45%)	2 (16%)
	Swallowing difficulty	4 (36%)	0
	Stress and anxiety	2 (16%)	1 (9%)
	Tremor	2 (16%)	1 (9%)
	Gait difficulty	2 (16%)	0
	Dysarthria	2 (16%)	0
	Nausea and vomiting	1 (9%)	0
	Hyperhidrosis	0	0
	Others	0	0
Duration of symptoms	< 6 months	1 (9%)	
	6–< 18 months	5 (45%)	
	≥ 18 months	5 (45%)	
Localization of headache	Occipital	6 (66%)	
	Others	3 (33%)	
Duration of headache	Minutes–hours	7 (77%)	
	Days	2 (22%)	
Headache aggravating factors	Positional (P)	1 (11%)	
	Exertion/Strain (E/S)	3 (33%)	
	P and E/S	3 (33%)	
	Movement	0	
	None	2 (22%)	
Degree of tonsillar herniation (mm)	5–12	6 (54%)	
	12.1–20	4 (36%)	
	> 20	1 (9%)	
Syrinx	No	6 (54%)	
	Yes	5 (45%)	

Table 3 Results of the Watson Dysphagia Scale and QLQ-OG25 in CMI patients with swallowing problems (N = 4)

	Reference value	Preop	Postop	Patients reporting the problem	I/U/W
Symptoms	Mean (SD)	Mean (SD)	Mean (SD)	N	N
Watson scale	–	16 (8)	6 (5)	4	4/0/0
Dysphagia	0.8 (5.5)	22 (15)	11 (9)	3	3/0/0
Eating restrictions	2.9 (9.9)	6 (7)	2 (4)	2	2/0/0
Reflux	6.7 (15.4)	25	25	1	0/1/0
Odynophagia	1.5 (8)	25	0	1	1/0/0
Pain and discomfort	7.6 (17)	25	0	1	1/0/0
Anxiety	–	33 (38)	12 (15)	3	3/0/0
Eating in front of others	1.3 (8.9)	33	0	1	1/0/0
Dry mouth	11.5 (23)	8 (16)	0	2	2/0/0
Trouble with taste	2.6 (12.5)	16 (33)	0	2	2/0/0
Trouble swallowing saliva	1.3 (9.2)	8 (16)	8 (16)	2	1/0/1
Choking when swallowing	3.7 (13.1)	66	0	1	1/0/0
Trouble with coughing	13.7 (23.6)	33	33	1	0/1/0
Trouble talking	2.2 (10.5)	0	0	0	0

Abbreviations: CMI, Chiari I malformation; I/U/W, improved/unchanged/worsened; SD, standard deviation. Reference values were obtained from a randomly selected samples drawn from the Swedish population.¹⁷

3 months after surgery in all patients. One patient presented with cerebrospinal fluid (CSF) collection at the surgical site 6 days after surgery, and this was successfully managed with a temporary CSF diversion using a lumbar drain for 7 days. This was the only complication.

Watson Dysphagia Scale and EORTC QLQ-OG25

Patients with no swallowing problems had scores of zero on both questionnaires. ▶Table 3 shows the pre- and postoperative scores on both the WDS and QLQ-OG25 for the patients who reported swallowing difficulties. The mean WDS in these patients was 16, and the scores on the QLQ-OG25 were worse (“deviation > 1 SD”) than the reference values (obtained from a population-based

study) in all but six domains (i.e., eating restrictions, pain and discomfort, dry mouth, trouble with saliva, coughing and trouble talking). Most scores improved after surgery, and only one patient reported worsening on swallowing saliva.

Videofluoroscopic Examination of Swallowing

The pre- and postoperative mean values of H_{max} and UES_o for each bolus size as well as the normative data for the 3-mL thin bolus obtained from reference¹² are shown in ▶Table 4. The mean values did not obviously deviate from the reported values in the normal adult subjects. The mean PCR during swallowing of the 20-mL liquid bolus was $0.02 \pm .03$ both pre- and postoperatively, similar to the normative data (for

Table 4 Pre- and postoperative mean values of H_{max} and UES_o ($\pm$ SD) for each bolus size as well as normative data regarding the 3-mL thin liquid bolus obtained from reference¹²

	3 mL thin liquid	5 mL thin liquid	10 mL thin liquid	5 mL mildly thick	3 mL extremely thick
H_{max} baseline (mm)	13.5 $\pm$ 3.6	15.5 $\pm$ 3.7	15.8 $\pm$ 3.6	13.1 $\pm$ 4.2	12.0 $\pm$ 6.5
H_{max} post-op (mm)	14.9 $\pm$ 3.9	14.8 $\pm$ 4.0	16.6 $\pm$ 4.3	15.0 $\pm$ 3.8	16.4 $\pm$ 3.8
H_{max} normative data from ref. ⁹ (mm)	18.6 $\pm$ 6.0	–	–	–	–
UES_o baseline (mm)	4.5 $\pm$ 1.8	6.3 $\pm$ 1.9	8.0 $\pm$ 1.7	6.8 $\pm$ 1.3	6.1 $\pm$ 2.0
UES_o post-op (mm)	5.3 $\pm$ 1.6	6.4 $\pm$ 2.3	7.8 $\pm$ 2.1	6.9 $\pm$ 2.0	6.5 $\pm$ 1.6
UES_o normative data from ref. ⁹ (mm)	5.1 $\pm$ 1.5	–	–	–	–

Abbreviations: H_{max} , maximal hyoid displacement; SD, standard deviation; UES_o , esophageal sphincter opening.

individuals younger than 65 years old: $0.03 \pm .03$ for females and $0.04 \pm .03$ for males²⁴).

Of the four patients who complained of subjective dysphagia, one showed a PAS 3 to 5 during 6 of 12 bolus swallows. Penetration with PAS 4 to 5 was observed in five swallows in another patient. No penetration (= PAS 1) was observed in the other two symptomatic patients. One of these patients, however, had a lowered hyoid displacement ($H_{\max}$ lower than one standard deviation from the sex-adjusted mean in normal subjects while swallowing a 3-mL bolus), and this patient also showed a smaller than normal UES opening.¹³ All four symptomatic patients showed signs that they were experiencing difficulty in initiating swallowing of the extremely thick bolus and/or a delayed onset of the pharyngeal swallowing phase. None of the patients had an elevated PCR.

None of the asymptomatic patients had a PAS of 3 or higher. One patient showed a lower than normal range of hyoid displacement as well as a decreased opening of UES during swallowing of a 3-mL liquid bolus. In this patient, a delayed onset of the pharyngeal swallowing phase was also noted. Another asymptomatic patient had a subnormal UES opening, and yet another showed signs of difficulty initiating the oral swallowing phase for the extremely thick bolus.

Nine patients underwent VFS both pre- and postoperatively. The percentage of bolus swallows with vallecular residue of grade 2 or larger decreased from 18% preoperatively to 6% postoperatively. The mean duration of UES opening increased from 0.65 second preoperatively to 0.74 second postoperatively. Signs of difficulty initiating swallowing or a delayed onset of the pharyngeal phase remained unchanged in one patient and were diminished in the remaining patients.

Radiation Dose

The mean entrance skin dose was 1.65 ± 0.85 mGy ($n = 8$).

Discussion

We present the first prospective study exploring changes in swallowing function before and after surgical decompression for CMI. We used both patient-reported complaints and radiological investigations. The frequency of swallowing difficulty reported in the present study (36%) is similar to the ranges reported in previous studies.^{9,10} In the EORTC-QLQ25, the overall scores for dysphagia and eating were worse in 7 out of 12 domains in these patients than in the normal population. Most scores improved after surgery and only one patient had worsening on swallowing saliva. An explanation of difficulty swallowing saliva might be globus pharyngis that is characterized by a nonpainful tightness in the throat or a sense of a lump. Globus is not associated with dysphagia and patients usually have the occurrence of sensation and difficulty swallowing their saliva between meals.²⁵ The patient in this study still had minimal swallowing difficulty despite reporting improvement. In addition, neither tightness in the throat nor sense of a lump has been reported by the patient. The preoperative mean WDS = 16

had improved in all patients at 3 months after surgical decompression (mean = 6).

VFS showed that there was substantial bolus penetration into the laryngeal vestibule in two of the patients with dysphagia. Two of the other symptomatic patients and three of the asymptomatic patients showed minor abnormalities on VFS. Thus, 7 out of the 11 examined patients showed some degree of aberration on VFS.

Yu et al reported that 47% of their adult CMI patients had dysphagia.¹⁰ They found that in CMI patients, neurogenic dysphagia was significantly associated with bulbar and high cervical syringomyelia. In the present study, syringomyelia was found in five patients, two of whom reported swallowing difficulty before surgical decompression. In another study, Yu et al used water swallowing test to evaluate patients both before and 7 to 10 days after surgery and found that dysphagia was significantly improved after surgical decompression in 67.6% of the cases.²⁶

None of our patients presented with dysphagia as the main or sole symptom of CMI. However, such patients have been reported in the literature.^{11,27} All patients initially had difficulty swallowing liquids, which is typical of neurogenic dysphagia, and a similar pattern has been observed in amyotrophic lateral sclerosis and myasthenia gravis.²⁸ In infants and small children, aspirations and oropharyngeal dysphagia are an expected finding in 78% of children under 3 years old with CMI. The differential diagnoses in this age group include gastroesophageal reflux and laryngomalacia.²⁹ Similar to our study, the postoperative outcomes of CMI-associated dysphagia have been previously reported to be excellent.^{6,27,30}

The reasons for the swallowing difficulty observed in CMI are unclear. CMI is a hindbrain anomaly that can negatively affect swallowing. Vascular ischemia, the traction of lower cranial nerves, and compression of the nearby brain stem and cerebellum might explain why dysphagia is associated with CMI.^{8,29} Pollack et al reported that all phases of swallowing were globally impaired on VFS in patients with CMI. They also found that the clinical presentation of these patients varied, especially between adult and pediatric age groups. Dysphagia caused by brain stem dysfunction can precede much more severe presentations of brain stem injury. In addition, a late diagnosis can make symptoms worse, and late-diagnosed patients could lose the cough response to regurgitation, resulting in "silent aspiration," a much more critical condition.⁸ In our study, all patients underwent planned surgical decompression, and no acute deterioration was reported.

The role of the cerebellum in controlling swallowing functions and its involvement in dysphagia has been previously investigated in animal studies. It was found that stimulating areas in the cerebellum such as the ventral parts of the vermis and the fastigial nucleus resulted in the activation and contraction of some muscles involved in the swallowing process.^{31,32} Furthermore, human studies have revealed that the cerebellum is an important contributor to swallowing function.³³ Suzuki et al used functional magnetic resonance imaging of the brain to show that the posterior

regions of the left cerebellar hemisphere are activated upon swallowing in healthy individuals. They also found that regional cerebral blood flow shows a positive response during swallowing.³⁴ Additionally, several reports have described patients with isolated cerebellar pathologies, such as infarctions and hemorrhage, that led to swallowing difficulties.^{35,36} Accordingly, the cerebellum is linked to swallowing functions either directly or via its diverse connectivity with higher cortical areas involved in swallowing.³³

The limitations of our study include the small study population, which was from a single center. The strengths of our study include its prospective design and the fact that we used both subjective and objective tools to evaluate the subjects. In normal individuals, there is a large amount of variability during the oropharyngeal phase of swallowing. It is, therefore, often difficult to define the specific limits that distinguish normal variations from findings that are symptoms of underlying dysphagia in VFS.³⁷ The lack of standardization of the methodology and interpretation of VFS in the literature is an additional source of variability. Hence, the results presented here need to be verified in larger clinical trials.

Conclusion

Dysphagia was found in 4 of 11 (36%) consecutive adult patients with CMI. Preoperative patient-reported dysphagia questionnaires and VFS showed that these patients had variable degrees of swallowing dysfunction. All patients were free from dysphagia after surgery, and the preoperative subjective and objective deviations from normal swallowing had decreased or disappeared postoperatively.

Conflict of Interest

All authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest (such as honoraria; educational; participation in speakers' bureaus; membership, employment, consultancies, stock ownership, or other equity interest; and expert testimony or patent-licensing arrangements), or nonfinancial interest (such as personal or professional relationships, affiliations, knowledge or beliefs) in the subject matter or materials discussed in this manuscript.

Acknowledgment

We would like to thank all the patients who participated in this study. Special thanks are also owed to Gudrun Barrows, Clinical Coordinator at the Department of Neurosurgery, Sahlgrenska University Hospital, for her invaluable help with the administration of this study. This research received funding from Rune och Ulla Amlövs Stiftelse för Neurologisk och Reumatologisk Forskning and Göteborg Läkaresällskap (Gothenburg Medical Society).

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