Dysphagia after pediatric functional hemispherectomy

Clinical article

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Object. Functional hemispherectomy is a well-recognized surgical option for the treatment of unihemispheric medically intractable epilepsy. While the resultant motor deficits are a well-known and expected consequence of the procedure, the impact on other cortical functions has been less well defined. As the cortical control of swallowing would appear to be threatened after hemispherectomy, the authors retrospectively studied a pediatric population that underwent functional hemispherectomy for medically intractable epilepsy to characterize the incidence and severity of dysphagia after surgery.

Methods. A retrospective cohort (n = 39) of pediatric patients who underwent hemispherectomy at a single institution was identified, and available clinical records were reviewed. Additionally, the authors examined available MR images for integrity of the thalamus and basal ganglia before and after hemispherectomy. Clinical and video fluoroscopic assessments of speech pathology were reviewed, and the presence, type, and duration of pre- and postoperative dysphagia were recorded.

Results. New-onset, transient dysphagia occurred in 26% of patients after hemispherectomy along with worsening of preexisting dysphagia noted in an additional 15%. Clinical symptoms lasted a median of 19 days. Increased duration of symptoms was seen with late (>14 days postoperative) pharyngeal swallow dysfunction when compared with oral dysphagia alone. Neonatal stroke as a cause for seizures decreased the likelihood of postoperative dysphagia. There was no association with seizure freedom or postoperative hydrocephalus.

Conclusions. New-onset dysphagia is a frequent and clinically significant consequence of hemispherectomy for intractable epilepsy in pediatric patients. This dysphagia was always self-limited except in those patients in whom preexisting dysphagia was noted.

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Key Words • hemispherectomy • seizures • dysphagia • epilepsy

Functional or disconnective hemispherectomy is a surgical procedure used in intractable, lateralized epilepsy. First conceived as an anatomical resection of the involved hemisphere, it demonstrated acceptable seizure control but was limited by postoperative complications, especially superficial cerebral hemosiderosis and hydrocephalus. In response, Rasmussen developed the functional hemispherectomy, which aimed to provide a complete functional disconnection while minimizing resection of the cerebrum. This trend of functional disconnection without resection has continued in recent years with the development of a number of hemispherotomy techniques. Current literature reports a seizure freedom rate ranging from 61% to 85% after hemispherectomy for medically intractable epilepsy of all causes. Data on other complications of the procedure are more limited and are primarily concerned with the incidence of posthemispherectomy hydrocephalus, which has been reported in 5.3%–23% of patients. Liégeois et al. reported persistent mild dysarthria in pediatric patients undergoing assessment from 1 to 13 years posthemispherectomy; none of these patients was noted to have a swallowing issue. No prior literature exists specifically detailing the incidence and severity of dysphagia after hemispherectomy.

Dysphagia is defined as a clinically evident impairment in swallowing. The swallow process is classically divided into the oral, pharyngeal, and esophageal phases. The oral phase consists of chewing and bolus formation via the tongue and hard palate. It is under voluntary control, and oral-phase dysphagia most frequently manifests as oral residue and difficulty with formation.
and control of the food bolus. The pharyngeal phase is an involuntary process involving coordinated movements in the pharyngeal musculature, vocal cords, and epiglottis that move the food bolus through the pharynx into the esophagus and prevent entry into the trachea. Pharyngeal-phase dysphagia is the most critical and typically results in aspiration and its associated symptoms. The esophageal phase consists of involuntary peristaltic movement of the bolus into the stomach.

Neurological disease resulting in dysphagia most commonly affects the oral and less commonly the pharyngeal phases of swallowing, with approximately one-third of patients demonstrating oropharyngeal dysphagia early after unilateral stroke. The swallow reflex is mediated by both cortical and brainstem input, with lesions of the primary sensorimotor cortex, insula, frontal operculum, anterior cingulate cortex, internal capsule, and supplementary motor area all being associated with dysphagia. Hemispheric dominance of swallowing appears to be present but highly variable between individuals with a suggestion that it shifts between hemispheres at different stages during the swallow.

As prior studies have not specifically addressed these findings, and the cortical control of swallowing would seem to be at risk with the hemispherectomy procedure, we performed a retrospective analysis of our functional hemispherectomy procedure to assess for postoperative dysphagia.

Methods

Patient Selection

We retrospectively identified 39 pediatric patients who underwent functional hemispherectomy at Seattle Children’s Hospital between 1997 and 2012 and who had adequate documentation to assess pre- and postoperative swallowing function. This review was performed in a manner approved by the Seattle Children’s Hospital Institutional Review Board. The majority of procedures were performed by the senior author (J.G.O.). All patients had a diagnosis of intractable epilepsy and were evaluated by an epileptologist and a multidisciplinary epilepsy board prior to hemispherectomy. The majority of procedures were performed using the approach of Schramm et al., namely, resection of the anterior and mesial temporal lobe, corpus callosotomy, frontobasilar and occipital disconnection, and insular resection.

Assessment of Dysphagia

The presence of dysphagia symptoms was determined preoperatively and postoperatively by examining patient records. Dysphagia was defined by documentation of clinical symptoms, initiation of dysphagia diet precautions, and the need for enteral nutrition and/or supplementation. When available, the results of clinical swallow evaluations (CSEs) and videofluoroscopic swallow studies (VFSSs) were used to assess for the presence of dysphagia and to characterize the type and severity of dysphagia present. Clinical documentation was reviewed by a speech pathologist (T.M.). Patients were then stratified using the following classification to characterize the presence of dysphagia after hemispherectomy: new dysphagia, preexisting dysphagia with worsening, preexisting dysphagia without worsening, and no dysphagia. Those patients with new or worsened dysphagia were additionally subdivided into those with evidence of dysphagia on CSE or VFSS and those with clinical evidence only. The duration and severity of symptoms were assessed during the postoperative follow-up period.

Imaging Assessment

Postoperative MR images were examined separately by a neurosurgical resident (R.T.B.) and senior attending neurosurgeon (J.G.O.). Magnetic resonance imaging was used to assess for resection of specific anatomical structures including full cortical disconnection, insular resection, and integrity of the basal ganglia and thalamus before and after hemispherectomy.

Statistical Analysis

Statistical significance was determined using the Fisher’s exact test and the exact Mann-Whitney test using the PASW Statistics 18 (SPSS, Inc.) toolset with a p value < 0.05 deemed significant. Multivariate analysis was additionally performed with a p value of 0.05 deemed significant.

Results

Thirty-nine patients met inclusion criteria. The median age was 5.7 years (range 28 days to 18.9 years). Of the 39 patients the most common indications for surgery were disorders of cortical development (hemimegalencephaly, polymicrogyria, and cortical dysplasia; n = 12, 30.8%), stroke (n = 11, 28.2%), and Rasmussen encephalitis (n = 4, 10.33%), followed by unknown neonatal (n = 3, 7.7%), nonaccidental trauma and Sturge-Weber syndrome (n = 2, 5.1% each) and West syndrome, anaplastic oligodendroglioma, choroid plexus papilloma, encephalitis, and Wolf-Hirschhorn syndrome (n = 1, 2.6% each) (Table 1).

Onset of new dysphagia symptoms was observed in 10 patients (25.6%) after hemispherectomy (Fig. 1A). Of these patients, 8 (80%) demonstrated a CSE or VFSS consistent with dysphagia while the remaining 2 (20%) had only clinical evidence of dysphagia. Preexisting dysphagia symptoms were identified by CSE or VFSS results in 11 patients (28.2%). Multiple assessments via CSE or VFSS were available in two-thirds of patients. Clinical worsening of dysphagia was observed in 6 patients (54.5%) after hemispherectomy with CSE or VFSS confirmation in all 6; there was no evidence of change in preexisting deficits in the 5 (45.5%) remaining patients. The remaining 18 patients (46.2%) did not have clinical evidence of dysphagia prior to or after hemispherectomy.

Of the patients who underwent CSE and/or VFSS for new and/or preexisting dysphagia in the early (< 14 days) postoperative period, dysfunction was identified in the oral phase in 3 (23.1%) and in the oral and pharyngeal (oropharyngeal) phase in 10 (76.9%) (Fig. 1B). For the patients with new dysphagia in the early postoperative pe-
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For patients with new-onset dysphagia, dysfunction in the late postoperative period (> 14 days) was noted in the oral phase in 2 (40%), pharyngeal phase in 2 (40%), and oropharyngeal phase in 1 (20%). For those children with new or worsened dysphagia that persisted longer than 2 weeks, the mean duration of symptoms with oral phase dysphagia only (n = 4) was 32 days compared with a mean duration of 393 days for those with pharyngeal phase dysphagia (n = 5) (Fig. 1C). There was a statistically significant association between pharyngeal swallow...
dysfunction in the late postoperative period and duration of symptoms (p = 0.016). For those patients with preexisting dysphagia, there was no clear association with the type of dysphagia identified and likelihood of clinical worsening after hemispherectomy.

Initiation of dysphagia diet modifications was required in 13 (81.3%) of the 16 children who developed new or worsened dysphagia; for the remaining 3 patients oral intake was initially deemed unsafe. A nasogastric tube was placed for complete or supplemental nutrition in 10 children (62.5%). The mean duration of nasogastric tube placement in all patients was 29 days (range 9–110 days). Additionally, long-term enteric supplementation via placement of a gastric feeding tube was required in 3 children (13.3%).

Dysphagia symptoms persisted for a median 19 days (range 8 days to 4.15 years) in all patients with new or worsened deficits after hemispherectomy as measured by the need for a dysphagia diet or enteral nutrition and/or supplementation. Of the patients with new dysphagia, the median duration of symptoms was 19.5 days (range 8 days to 1.42 years) and for worsened dysphagia the median duration was 19 days (range 12 days to 4.15 years). This postoperative dysphagia resolved in all patients with new or worsened dysphagia and in all except one patient with worsened dysphagia who continued to require enteral supplementation via gastric tube at last follow-up (4.1 years).

Stroke of all types was the most common etiology in patients without dysphagia (n = 11), with only one of those patients developing new dysphagia after hemispherectomy. There was a negative association between stroke and new or worsened dysphagia after surgery (p = 0.014). There was no other clear association with etiology and the incidence of posthemispherectomy dysphagia.

The median age for children with new dysphagia was 6.82 years, for preexisting dysphagia with worsening 2.87 years, for preexisting dysphagia without worsening 1.65 years, and for no dysphagia 6.50 years. The median age at the time of hemispherectomy in patients with new or worsened dysphagia was 4.49 years compared with a median age of 6.12 years for those without (Table 2). There was no significant association between age at the time of hemispherectomy and the occurrence of postoperative dysphagia (p = 0.275).

Of the patients with new or worsened dysphagia 9 (56.3%) underwent a left hemispherectomy and 7 (43.8%) underwent a right hemispherectomy. There was no relationship with new or worsened dysphagia and whether the hemispherectomy was performed on the right or left side (p = 0.748).

Review of postoperative imaging demonstrated that all patients had resection of the insula. There was complete or partial preoperative infarction or absence of the thalamus and basal ganglia in 12 children (30.8%) and of the basal ganglia only in an additional 2 children (5.1%). Of those patients with absence of both (n = 9, 75%), the majority were secondary to neonatal stroke. Preoperative absence of the thalamus and basal ganglia did not influence development of dysphagia after hemispherectomy (p = 0.726). In those children with an intact ipsilateral thalamus prior to surgery, 19 (70.4%) had complete or partial injury as seen on postoperative CT and/or MRI. Of those who did not have injury of the thalamus, 5 (83.3%) developed new dysphagia posthemispherectomy. There was a trend toward association of a structurally preserved thalamus postoperatively and new dysphagia, but this did not achieve statistical significance (p = 0.073). Complete or partial resection or infarction of the basal ganglia was seen in all patients; this did not correlate with postoperative dysphagia.

Postoperative hydrocephalus developed in 6 patients (15.4%), 3 (50%) of whom also had new dysphagia. There was no relationship between postoperative hydrocephalus and dysphagia after hemispherectomy (p = 0.674). Complete and durable absence of clinical seizure events after surgery occurred in 28 patients (71.8%), with a median length of follow-up of 3.54 years (range 0.3–10.6 years). Of the patients who had persistence or late recurrence of seizures, 6 (54.5%) had new or worsened dysphagia after hemispherectomy. There was no relationship between posthemispherectomy clinical seizure absence and the development of dysphagia (p = 0.307).

Discussion

This study represents the first examination of dysphagia after hemispherectomy in pediatric patients. In our series approximately one-quarter of children developed new-onset dysphagia after surgery, the majority of whom had observable pathology on clinical or fluoroscopic evaluation. This finding is in addition to the roughly one-quarter of children who had preexisting swallowing difficulties prior to hemispherectomy. Of these patients with

![Fig. 1. Incidence and characteristics of dysphagia following hemispherectomy. A: Presence or absence of dysphagia in children with and without preexisting deficits after surgery. Values on the y axis are the number of patients. B: Type of dysphagia. The gray bars indicate the early phase (<14 days postoperative) and the black bars indicate the late phase (>14 days postoperative). Values on the y axis are the number of patients. C: Median duration of late-phase dysphagia.](https://example.com/dysphagiafig.png)
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### TABLE 2: Association between clinical and radiological characteristics and presence or absence of posthemispherectomy dysphagia

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>New or Worsened Dysphagia (n = 16)</th>
<th>No or Stable Dysphagia (n = 23)</th>
<th>p Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>median age at op (yrs)</td>
<td>4.49</td>
<td>6.12</td>
<td>0.275</td>
</tr>
<tr>
<td>postop hydrocephalus</td>
<td>3 (18.8%)</td>
<td>3 (13.0%)</td>
<td>0.674</td>
</tr>
<tr>
<td>absence of seizure freedom</td>
<td>6 (37.5%)</td>
<td>5 (21.7%)</td>
<td>0.307</td>
</tr>
<tr>
<td>preop basal ganglia &amp; thalamus damage</td>
<td>4 (25%)</td>
<td>9 (39.1%)</td>
<td>0.726</td>
</tr>
<tr>
<td>postop preservation of thalamus</td>
<td>5 (31.3%)</td>
<td>1 (4.3%)</td>
<td>0.073</td>
</tr>
</tbody>
</table>

Occurrence of dysphagia after hemispherectomy was not dependent on the hemisphere resected, which correlates with the bilateral representation of swallowing and wide individual asymmetry in cortical dominance reported in the literature. In light of this variability, there may be a role for functional imaging assessment of the swallow prior to hemispherectomy as a mechanism to clarify postoperative dysphagia risk. This may be especially relevant in children with preoperative dysphagia for whom the risk of permanent deficit is present. There was no evidence that development of hydrocephalus or seizure freedom posthemispherectomy influenced the presence of dysphagia after surgery.

Based on the observed incidence and severity of dysphagia after hemispherectomy in children, we would not recommend routine preoperative formal swallow evaluation in the absence of clinical dysphagia. This is supported by the fact that in those children with known preexisting dysphagia, the identification of the specific area of the swallow reflex that was impaired did not help in predicting whether symptoms worsened after hemispherectomy. In those children with new or worsened dysphagia after surgery, CSE and/or VFSS are indicated to determine both the severity of symptoms and the specific treatment modality needed. For children with persistent dysphagia in the late postoperative period (> 14 days), repeat CSEs and/or VFSSs are recommended to assess for pharyngeal phase dysphagia; this is significantly associated with a prolongation in symptoms and need for treatment in our cohort.

This study is subject to the inherent limitations of a retrospective analysis. Additionally, preoperative swallow evaluation was unavailable except in patients with previously identified clinical dysphagia. Therefore, we cannot exclude subtle deficits that may not be clinically apparent that predispose children to developing dysphagia after hemispherectomy.

### Conclusions

This study provides evidence that dysphagia is a significant potential complication of hemispherectomy for intractable epilepsy in the pediatric population. This posthemispherectomy dysphagia is clinically significant but usually resolves on its own in the early postoperative period. In patients without identified preexisting deficits this posthemispherectomy dysphagia always resolved completely; permanent issues were only seen in those children with known dysphagia prior to surgery. It is im-

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preexisting deficits, a small majority clinically worsened after surgery.

Clinical symptoms in patients who had new-onset dysphagia were always temporary and typically resolved completely by the end of the 1st postoperative month. The duration of symptoms in patients with worsening of preexisting deficits typically improved to the child’s baseline within the 1st month, but was more variable. The gradual recovery in swallowing function we observed supports the theory proposed by Hamdy et al. that there is a slow reorganization of the swallow control networks into the undamaged contralateral hemisphere after a unilateral hemispheric injury. This may also be influenced by the increased plasticity of the pediatric brain, which is critical for reorganization of cortical swallow networks necessary for recovery after dysphagia. Most children required diet modifications and enteral nutrition during the course of their posthemispherectomy dysphagia. Only one patient with new dysphagia required long-term nutritional supplementation via gastric tube; this was able to be removed by 18 months posthemispherectomy.

Oral-phase dysphagia was seen in all patients with new dysphagia after surgery. This pattern is commonly seen after stroke and with other cortical lesions affecting the primary somatosensory and cingulate cortex. The majority of children also had dysfunction in the pharyngeal phase in the early period after surgery. In those who had persistent pharyngeal-phase dysfunction, there was a significantly longer duration of symptoms. Follow-up clinical or videofluoroscopic swallow assessment in posthemispherectomy patients with persistent dysphagia is important for identifying this pharyngeal dysfunction and can help guide discussion and therapy.

There are data to suggest a role for the basal ganglia and insula in mediating the swallow reflex; however, there was no association with resection of the basal ganglia or insula and dysphagia in our series. There was a strong trend toward an association between posthemispherectomy dysphagia and new complete or partial damage to the ipsilateral thalamus on postoperative imaging. Interestingly, interruption of afferent pharyngeal sensory input to primary somatosensory cortex and other cortical mediators of the swallow reflex has been linked to the presence of dysphagia after neurological injury. This suggests that preservation of the ipsilateral thalamus in the context of the loss of associated inhibitory cortical modulation of brainstem swallow reflex centers after functional disconnection may predispose to dysphagia in these patients.

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portant for neurosurgeons to be aware of the potential risk of dysphagia and resultant need for therapy and to communicate with families preparing for their child to undergo hemispherectomy.

Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author contributions to the study and manuscript preparation include the following. Conception and design: Buckley, Saneto, Ojemann. Acquisition of data: Buckley, Morgan. Analysis and interpretation of data: Buckley, Morgan, Barber, Ojemann. Drafting the article: Buckley, Saneto, Ellenbogen, Ojemann. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Buckley. Statistical analysis: Buckley, Barber. Study supervision: Ojemann.

References

5. Fraser C, Power M, Hamdy S, Rothwell JC, Aziz Q, Thompson DG: Organization of all authors. Approved the final version of the manuscript on behalf of all authors. Reviewed submitted version of manuscript: all authors. Critically revising interpretation of data: Buckley, Morgan, Barber, Ojemann.

16. McKenzie K: The present status of a patient who had the right cerebral hemisphere removed. JAMA 111:168, 1938

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