

Hypopharyngeal Pharyngoplasty in the Treatment of Severe Aspiration Following Skull Base Tumor Removal

Experience in Pediatric Patients

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Objective: To report the surgical treatment of severe swallowing disorders associated with skull base surgery resulting in unilateral pharyngolaryngeal paralysis in pediatric patients.

Design: Retrospective case review.

Setting: Tertiary referral center for pediatric otolaryngology.

Patients: Five infants undergoing swallowing rehabilitation surgery for severe dysphagia and aspiration resulting from skull base or brainstem surgery.

Intervention: A hypopharyngeal pharyngoplasty, consisting of the partial resection of the inferior constrictor and cricopharyngeal muscles, was performed for the treatment of severe swallowing disorders. A thyroplasty was also performed if clinically significant glottic incompetence was present.

Main Outcome Measures: Functional outcomes after surgery were evaluated with a videoendoscopic swallowing study and videofluoroscopy. Postoperative clinical

evaluation included respiratory, swallowing, and nutritional outcomes.

Results: A hypopharyngeal pharyngoplasty was performed following a mean period of 6 weeks (range, 1-10 weeks) after skull base surgery. In 3 patients a thyroplasty and a temporary tracheotomy were performed. Oral feeding was reintroduced after a mean period of 6 days (range, 4-20 days). Complete oral feeding autonomy was obtained after 13 days (range, 7-25 days). Postoperative swallowing assessment revealed the disappearance of pharyngeal stasis and aspiration in all patients. Three infants died because of tumor recurrence. Neither dysphagia or bronchopulmonary infections were observed after a mean follow-up period of 33 months (range, 6-61 months).

Conclusions: Pharyngolaryngeal paralysis represents a severe consequence of skull base and brainstem surgery. This condition leads to high morbidity, particularly in the pediatric population. The hypopharyngeal pharyngoplasty, with a possible thyroplasty, may be considered to treat patients with severe pharyngolaryngeal paralysis after skull base or brainstem surgery.

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THE AGGRESSIVE TREATMENT of skull base and brainstem lesions can result in injury to the lower cranial nerves (CNs). Deficits of the vagus (X), glossopharyngeal (IX), or hypoglossal (XII) CNs can adversely affect swallowing, voice production, and airway maintenance. This can have a major impact on the patient's quality of life.¹⁻³ In the pediatric population, skull base and brainstem tumors are typically characterized by an infiltrative pattern leading to a high risk of neurological deficits. Moreover, these lesions often require adjuvant radiotherapy that may further injure the lower CNs and/or chemotherapy.⁴⁻⁶ In severe cases, pharyngolaryngeal paralysis may occur and results in dysphagia and aspiration pneumonia and increases sig-

nificantly the morbidity of adjuvant therapy. Therefore, an aggressive rehabilitative approach of lower CN deficits is often required.

Numerous surgical procedures have been proposed in adult patients to improve airway protection, ranging from vocal fold injection to laryngectomy.^{1,3,7-13} In patients with lower CN deficits, the restoration of glottic competence alone seems to be ineffective. Additional problems occur as a result of pharyngeal stasis resulting in dysphagia and aspiration. The reduction of the paralyzed pharyngeal cavity has been proposed^{14,15} to decrease pharyngeal retention. Herein, we report our experience of the surgical treatment of unilateral pharyngolaryngeal paralysis using hypopharyngeal pharyngoplasty associated with thyroplasty in pediatric patients.

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METHODS

All surgical procedures were approved by our institutional review board. A retrospective medical chart review was performed that included all infants referred to our department for severe dysphagia and aspiration related to unilateral laryngeal and pharyngeal palsy secondary to brainstem or skull base surgery from November 1998 to December 2008.

FUNCTIONAL ASSESSMENT

Swallowing evaluation included a videoendoscopic swallowing study with sensory testing and a videofluoroscopy with oral administration of flavored barium. Prior to surgical planning, a laryngeal electromyography (EMG) was performed to assess the severity of the nerve deficit when medical history and swallowing evaluation were not consistent. Laryngeal EMG was performed under general anesthesia with spontaneous breathing.

SURGICAL CANDIDACY

Surgical treatment for dysphagia was proposed in selected cases of lower CN deficit as a result of brainstem or skull base tumor removal. Clinically severe dysphagia or aspiration and poor prognosis of recovery (intraoperative nerve section or denervation pattern on electromyography) were used to identify candidates for surgery.

THYROPLASTY PROCEDURE

In cases of clinically significant laryngeal incompetence, a thyroplasty was performed as the first step of the procedure. To allow perioperative endoscopic control of the glottis, a tracheotomy was performed at the beginning of the procedure. According to the procedure described by Link et al,¹⁶ the true vocal cords were localized by insertion of a needle through the thyroid cartilage into the endolarynx under endoscopic control. As described by Isshiki et al,¹⁷ the medialization was performed using a cartilage graft, and the vocal cord placement was assessed endoscopically.

HYPOPHARYNGEAL PHARYNGOPLASTY AND CRICOPHARYNGEAL MUSCLE SECTION

Patients who did not require a thyroplasty were intubated via the orotracheal route. The larynx was then dissected laterally, and the superior laryngeal nerve was identified. After rotating the larynx, the pharyngeal constrictor and cricopharyngeal muscles were exposed. The inferior constrictor muscle was sectioned vertically 2 mm behind the posterior border of the thyroid cartilage lamina. Inferiorly the muscular section was extended to the cricopharyngeal muscle fibers, distant to the inferior horn to preserve the recurrent laryngeal nerve. Superiorly the section reached the upper edge of the superior cornu of the thyroid cartilage. A fusiform section was performed, allowing the removal of about 50% of both the inferior constrictor muscle and the membranous pharyngeal wall (**Figure 1**). The pharyngeal edges were approximated in 2 layers (mucosal and muscular).

POSTOPERATIVE CARE

Extubation was performed at the end of the procedure. In patients undergoing a thyroplasty, a tracheotomy remained temporarily in place to secure the airway during the postoperative period. Decannulation was performed after the glottis lumen

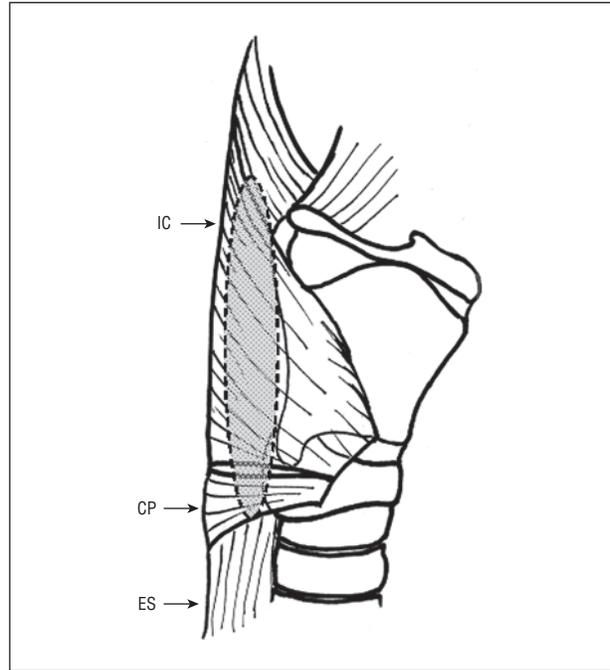


Figure 1. Schema of hypopharynx resection. The inferior constrictor (IC) was sectioned vertically behind the posterior border of the thyroid cartilage alar. The muscular section was extended inferiorly to the cricopharyngeal muscle fiber (CP) and to the upper edge of the superior cornu of the thyroid cartilage. After pharyngeal mucosa section, more than 50% of the inferior constrictor and membranous pharyngeal wall was resected (gray area). ES indicates the esophagus.

was evaluated with flexible laryngoscopy. Oral feeding was reintroduced after videoendoscopic and videofluoroscopic examinations demonstrated pharyngeal healing and effective swallowing. Patient data are listed in the **Table**.

RESULTS

PATIENTS AND FOLLOW-UP

Five infants, 2 boys and 3 girls, were referred for surgical treatment of a severe swallowing disorder after skull base surgery. Four patients were treated with surgical resection of a brainstem ependymoma, and 1 patient underwent removal of a large vagal ganglioneuroma involving the jugular foramen. In 2 cases the swallowing disorder was present prior to skull base surgery. The mean age was 23 months (range, 11-52 months). The mean follow-up period was 33 months (range, 6-61 months). During the follow-up period 3 infants died because of brainstem tumor recurrence at 6, 22, and 46 months after the hypopharyngeal pharyngoplasty procedure.

SWALLOWING EVALUATION

All patients demonstrated severe postoperative swallowing deficits associated with recurrent aspiration and dysphonia. They all required enteral feeding as a consequence of altered pharyngolaryngeal function. A deficit of the glossopharyngeal and vagus nerves were observed in all 5 patients. Three patients had a facial nerve deficit. The spinal accessory nerve and hypoglossal nerve

Table. Patient Data

Patient No./ Age, mo	Diagnosis	Cranial Nerve Deficit	Preoperative Symptoms	Surgical Procedure	Postoperative Evolution (d)
1/16	Brainstem ependymoma	VII, IX, X	Dysphagia NGT dep Aspiration Dysphonia	HP + TP after 6 wk Temporary tracheotomy	Decannulation (3) Ablation NGT (25) Unrestricted diet Tumor recurrence Died 22 mo AP
2/11	Brainstem ependymoma	IX, X, XI, XII	Dysphagia Gastrostomy tube dep Aspiration Dysphonia	HP + TP after 8 wk Temporary tracheotomy	Decannulation (10) Ablation NGT (21) Unrestricted diet Rare liquid aspiration Tumor recurrence Died 46 mo AP
3/52	Jugular foramen ganglioneuroma	IX, X	Dysphagia NGT dep Aspiration Dysphonia	HP + TP after 10 wk Temporary tracheotomy	Decannulation (3) Ablation NGT (17) Unrestricted diet Rare liquid aspiration Alive 61 mo AP
4/17	Brainstem ependymoma	VII, IX, X, XI	Dysphagia NGT dep Aspiration	HP after 5 wk	Ablation NGT (5) Unrestricted diet Alive 31 mo AP
5/21	Brainstem ependymoma	VII, IX, X, XI, XII	Dysphagia NGT dep Aspiration Dysphonia	HP after 1 wk	Ablation NGT (8) Unrestricted diet Tumor recurrence Died 6 mo AP

Abbreviations: AP, after procedure; dep, dependent; HP, hypopharyngoplasty; NGT, nasogastric tube; TP, thyroplasty.

were affected in 3 and 2 patients, respectively. Intraoperative section of the vagus nerve occurred in 2 cases. In the 3 other cases, the severity of the deficit was evaluated with a laryngeal EMG between the fifth and the seventh postoperative week. Laryngeal EMG confirmed a severe denervation pattern in all 3 cases.

Initial videoendoscopic swallowing assessment revealed in all cases a pharyngeal stasis related to a failure of contraction and elevation of the paralyzed hemipharynx. During swallowing, asymmetric contraction led to rotation of the normal pharyngeal wall. This resulted in misdirected propulsion of the bolus to the weakened hemipharynx rather than progression through the superior esophagus sphincter. It is noteworthy that all patients presented with hypopharyngeal salivary stasis and aspiration of salivary during breathing (**Figure 2**). Videofluoroscopy confirmed the inefficiency of pharyngeal contraction with permanent residual stasis and aspirations in all cases. Nasal reflux was noted in 4 cases.

SWALLOWING REHABILITATION SURGERY

A hypopharyngeal pharyngoplasty was performed at mean of 6 weeks (range, 1-10 weeks) after skull base surgery. The procedure was performed between the first 2 cycles of chemotherapy in 3 cases. In 1 patient with a poor oncologic prognosis, the parents rejected adjunctive oncologic treatment. Therefore, the hypopharyngoplasty was performed 1 week after brainstem surgery for palliation (eg, nutritional support) and allowed early discharge.

Three patients presented with vocal fold paralysis and clinically significant glottic incompetence requiring a thyroplasty. In the 2 other patients, the paralyzed vocal fold was diagnosed in a paramedian position, and a thyroplasty was not performed.

No complications were observed. In patients requiring a tracheotomy, decannulation was achieved between postoperative days 3 and 10. Infants were discharged home or to the pediatric oncologic department in a mean of 8 days (range, 7-11 days).

SWALLOWING OUTCOMES AND FEEDING

Postoperative videoendoscopy showed disappearance of pharyngeal stasis and efficient pharyngeal propulsion in all patients. Postoperative videofluoroscopy confirmed the efficiency of pharyngeal propulsion and the absence of pharyngeal stasis or laryngeal aspiration in all patients. In 4 patients, nasal reflux was observed during videofluoroscopy.

Oral feeding was introduced after a mean time of 6 days (range, 4-20 days). In infants receiving chemotherapy, normal oral feeding was proposed after the second cycle to reduce the feeding problems related to nausea and vomiting. Finally, complete oral feeding autonomy was obtained after a mean period of 13 days (range, 7-25 days), allowing the removal of the nasogastric tube after a mean period of 15 days (range, 8-25 days) and unrestricted diet within the first postoperative month. Postoperative weight gain was observed in all patients.

Feeding autonomy and airway protection remained stable over time in all patients, even in those with tumor recurrence. No patient developed recurrent dysphagia or bronchopulmonary infection. Nevertheless, some intermittent but uncommon aspirations during swallowing liquids were reported in 2 cases. In all 4 patients with initial nasal reflux, no symptoms were reported after the second month. The 3 patients treated with associated thyroplasty demonstrated a notable improvement in voice quality.

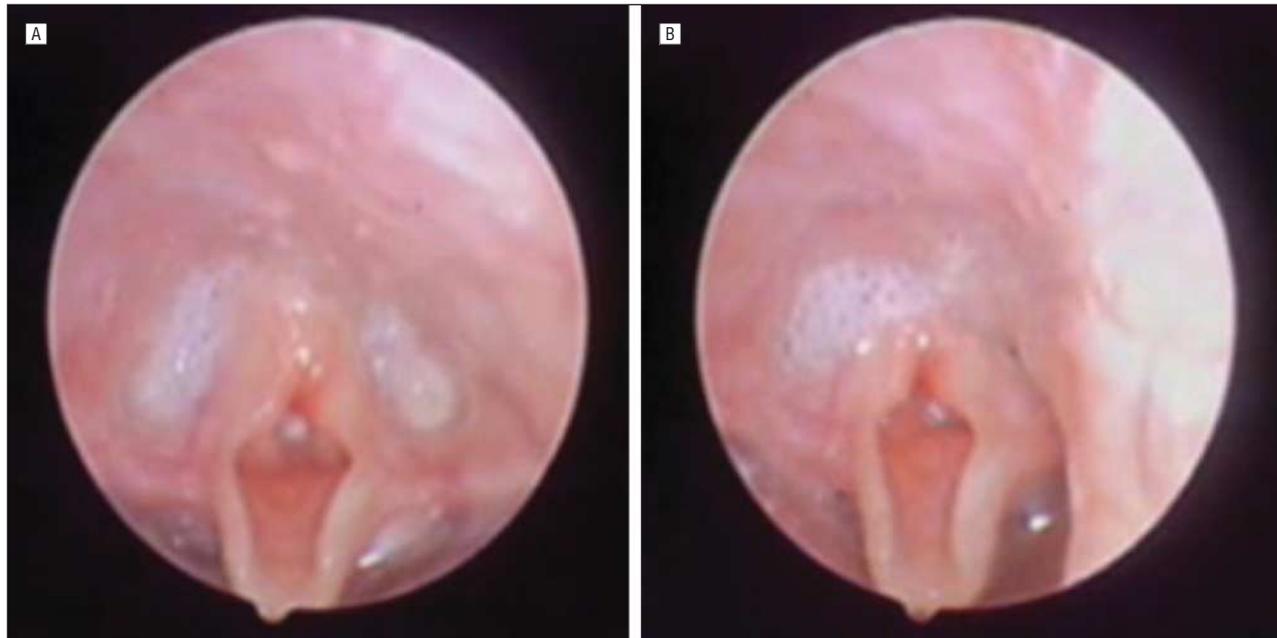


Figure 2. Endoscopic view of paralyzed pharyngolarynx during swallowing. A, The pharyngeal phase shows the contraction of the left constrictor muscle associated with a dilated right pharyngeal lumen, inducing a rotation of the pharyngeal lumen with a displacement of the median line to the left. B, The default of posterior constrictor support induced decreases the propulsion efficiency with a residual stasis in the right piriform sinus.

COMMENT

Swallowing disorders after skull base surgery may occur due to injury to the lower CNs. They remain a major contributor to postoperative morbidity¹⁻³ leading to a high rate of mortality.³ In the pediatric population, adjuvant chemotherapy is generally required, increasing the morbidity of postoperative swallowing disorders.⁴⁻⁶ During chemotherapy, a higher risk of vomiting associated with aspiration and pulmonary infection is observed and might be considered even higher during the myelosuppression phase.

The pathophysiologic characteristics of swallowing disorders after unilateral lower CN paralysis have been previously described.^{7,8,11,13,18} The unilateral deficit of the glossopharyngeal nerve and the vagus nerve results in a loss of sensory afferent feedback from the tongue, the lateral pharyngeal wall, and the supraglottic larynx. This results in palatopharyngolaryngeal paralysis, glottal incompetence, inadequate pharyngeal elevation, and failure of cricopharyngeal relaxation during swallowing. The main cause of aspiration is pharyngeal stasis resulting in the propulsion of the bolus to the dilated paralyzed hemipharynx rather than down the esophagus during pharyngeal contraction.^{8,10,14,18} Pharyngeal dysfunction, not just glottal incompetence, may result in problems with aspiration. Aspiration may occur during the pharyngeal phase of swallowing or in the post-swallowing period owing to pooling and overspill.

In most cases, the lower CN deficit is partial and/or temporary and can be treated medically with changes in diet, thickened liquids, speech therapy, salivary reduction, respiratory physiotherapy, or temporary enteral feeding. An early and accurate treatment is required in the rare cases of severe swallowing disorders associated with complete paralysis of the lower CNs.

Treatment of swallowing disorders requires a precise functional assessment of the sensory and motor functions of the larynx and pharynx. In pediatric and adult patients, functional assessment is based on a videoendoscopic swallowing study associated with sensory testing and videofluoroscopy.^{13,18-21} The upper esophageal sphincter and pharyngoesophageal coordination can be evaluated by manometry. However, in the case of unilateral dysfunction, manometry could be difficult to interpret and inconsistent with fluoroscopic assessment.¹⁸ Laryngeal EMG can be performed to assess the status of the vagus nerve.¹³ Although its interpretation is subject to debate,²² we believe a severe denervation pattern is associated with a poor prognosis of recovery. In this situation, early surgical treatment should be considered, particularly when adjuvant chemotherapy is needed.¹³

Various surgical procedures have been described to treat unilateral pharyngolaryngeal paralysis in adults, but to our knowledge no study reports treatment in pediatric population. The severe morbidity associated with pharyngolaryngeal paralysis led some authors to propose early tracheotomy^{1,3,7,11,12} to reduce aspiration and facilitate pulmonary toilet. Severe cases may require a laryngotracheal separation or laryngectomy.^{7,11} Associated dysphagia can be treated by gastrostomy allowing nutritional support. In the pediatric population, tracheotomy and gastrostomy increase the morbidity of the adjuvant chemotherapy, as well as hospitalization duration and quality of life.

Surgical procedures proposed to address the laryngeal component only, such as medialization thyroplasty, vocal cord augmentation,^{1,7-9,11,13} or arytenoid adduction,¹² may be ineffective, with residual dysphagia and aspiration reported in 17% to 28% of cases.^{9,10,20,23} To reduce the pharyngeal stasis, section of the cricopharyngeal muscle has been proposed alone or in association with a laryngeal pro-

cedure.^{1,2,7,9-11,13} The effectiveness of such a combined procedure seems to be inconstant,^{1,2,10} with residual dysphagia requiring gastrostomy or residual aspiration requiring tracheotomy. As described in the second paragraph of this section, dysphagia and aspiration are principally attributed to bolus propulsion failure associated with constructor paralysis and stasis in the dilated and insensate pharyngeal wall. The reduction of this nonfunctional pharyngeal cavity seems to be the most effective procedure. A similar procedure was previously described by Mok et al¹⁴ reporting the combination of hypopharyngeal pharyngoplasty and medialization laryngoplasty in 8 adults. An additional palatoplasty was performed in 5 patients.

Sato et al¹⁵ proposed reduction of the paralyzed pharyngeal cavity by autologous fat injection in the medial wall of the pyriform sinus, aryepiglottic fold, false vocal fold, and thyroarytenoid muscle. This endoscopic procedure is less invasive than the external approach, but the reduction of the pharyngeal cavity seems to be less effective. Moreover, the injection of the lateral laryngeal margin could lead to respiratory obstruction in small infants. In this population, the long-term effect of fat injection remains uncertain due to fat resorption and declining fibrovascular support of fat graft during homogenization.²⁴

On the one hand, hypopharyngeal pharyngoplasty is more invasive than previously cited procedures and might be proposed in patients who had failed treatment with other, less invasive procedures. On the other hand, in pediatric patients the risk of aspiration is considerable with the vomiting related to chemotherapy. This potential increase in aspiration combined with myelosuppression argues for early and aggressive treatment of swallowing disorders in this population. The advantage of the proposed procedure is to treat in a single stage the 3 major causes implicated in dysphagia and aspiration: pharyngeal dilatation, failure of cricopharyngeal relaxation, and failure of laryngeal closure.

Our experience is limited, but the results of hypopharyngeal pharyngoplasty seem satisfactory, with rapid feeding autonomy, significant reduction of aspiration, and the avoidance of a tracheotomy and gastrostomy in some patients. Infants can often be discharged home or to the medical oncologic department postoperatively in good condition and within a short time. The short postoperative hospitalization allows performing this procedure between 2 chemotherapy cycles without interfering with the chemotherapy protocol. Long-term follow up in a larger group is necessary to evaluate the results over time, particularly when the procedure is performed in young infants.

In conclusion, in pediatric patients as in adults, pharyngolaryngeal paralysis represents a severe consequence of skull base and brainstem surgery, with a high morbidity and negative impact on quality of life. In most severe cases, early surgical treatment may be considered to reduce the disability and shorten hospitalization. The hypopharyngeal pharyngoplasty, with a possible thyroplasty, may be considered to treat patients with extremely severe pharyngolaryngeal paralysis after skull base or brainstem surgery.

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