



Case Report

Modified approach for pediatric external cricopharyngeal myotomy

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ABSTRACT

Pediatric cricopharyngeal achalasia is an uncommon but important cause of oropharyngeal dysphagia. Failure of upper esophageal sphincter relaxation is the currently understood pathophysiology. Therapies include balloon dilation, botulinum toxin injection, and endoscopic or open cricopharyngeal myotomy (CPM). Open CPM is usually performed at the posterior midline of the cricopharyngeus and can be a risky procedure given concern for esophageal perforation and damage to the recurrent laryngeal nerve. Here, we present a novel modified technique for open CPM using a superficial anterolateral transection approach in the case of a young male with refractory cricopharyngeal achalasia.

1. Introduction

First described in an infant in 1969, cricopharyngeal achalasia (CPA) is an uncommon, but well-established cause of pediatric oropharyngeal dysphagia [1]. Children typically present with pharyngeal phase dysphagia and coughing, choking, gagging, or nasal regurgitation with repeated attempts. Liquids typically cause more severe symptoms due to their proclivity for laryngeal penetration [2]. The cricopharyngeus is tonically contracted and opens intermittently to allow passage of fluid or gas. Neuromuscular dysfunction, specifically the failure of the upper esophageal sphincter (UES) to appropriately relax upon presentation of a food bolus, is the working understanding of CPA [3].

Early diagnosis is important due to the potential for surgical correction and to ensure swallowing is established during the appropriate developmental stage. Videofluoroscopic swallow study (VFSS) remains the most common modality to identify oropharyngeal dysfunction, and in CPA, can reveal inadequate passage of contrast into the esophagus due to obstruction at the level of the cricopharyngeus (CP) muscle. In order to prove failure of UES relaxation in the strictest sense, esophageal manometry is required. Manometry can also be used for confirmation or in cases when the VFSS is indeterminate or inconsistent with clinical findings. In adults, manometry is a standardized diagnostic tool [4]. However, a lack of pediatric normative data makes analysis challenging in children. Abnormal patterns in adults may be normal in children and therefore all pediatric manometry results, regardless of diagnosis, still need to be interpreted in the context of the VFSS and clinical scenario [5,6].

Historically, transcervical open cricopharyngeal myotomy (CPM)

has been the mainstay of treatment, though recent reports have suggested less-invasive approaches, including balloon dilation [7–9], endoscopic CPM [10,11], and botulinum toxin injections [12,13]. However, randomized studies comparing these techniques have not been performed to date. The vast majority of case series describe open or external CPM performed using a posterior midline transection [14–17]. Here, we present a novel, technically simpler and less morbid variation of the prevailing transcervical open cricopharyngeal myotomy in the case of a young male with chronic aspiration refractory to balloon dilation and botulinum toxin injection.

2. Methods

According to the Massachusetts Eye and Ear Infirmary Human Studies Committee guidelines, this single patient report was not deemed research and as such did not require Institutional Review Board approval.

A six-year-old male, former 35-week-old twin with a past medical history of oromotor apraxia, recurrent croup, and a long-standing history of feeding and swallowing problems since shortly after birth presented to our clinic for pharyngeal dysphagia with coughing on both liquids and solids. Notably, he also had a history of recurrent pneumonia secondary to intractable aspiration of liquids. Initial VFSS was difficult to interpret secondary to compliance and suggested no frank aspiration with varying amounts of laryngeal penetration even to thin liquids. Clinically, he appeared to cough and sputter up to honey consistency which he could not tolerate enough to maintain adequate hydration. He had persistent reflux symptoms as well and

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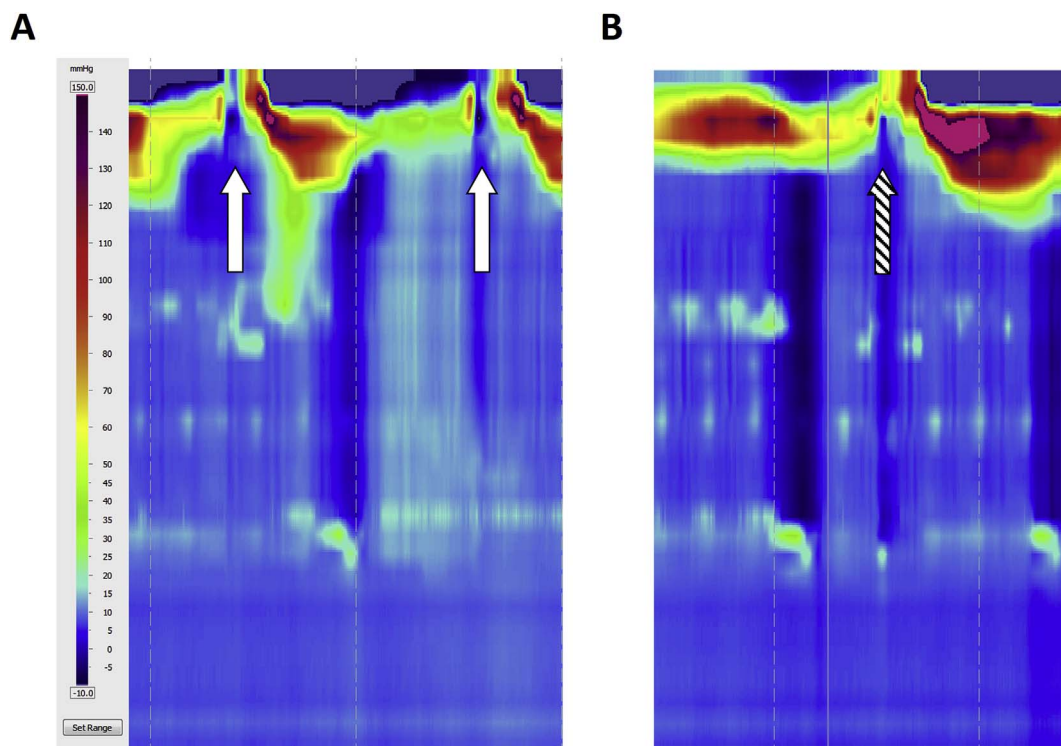


Fig. 1. Esophageal Manometry of Upper Esophageal Sphincter (UES) A) Example of normal UES relaxation, B) Incomplete UES relaxation in patient with slightly elevated residual pressure concerning for achalasia. (White arrows denote relaxation of UES; Striped arrow notes area of incomplete relaxation).

esophagogastroduodenoscopy revealed reflux changes along with abnormal impedance testing. Eventually due to worsening clinical symptoms (recurrent infections) and poor hydration, he underwent gastrostomy tube placement. Repeat VFSS, again difficult due to patient cooperation, suggested a potential restrictive defect at the level of the CP. Subsequent esophageal manometry revealed trace elevated UES pressures (Fig. 1). In the experience of the senior author, CPA management has been successful using an algorithmic approach beginning with balloon esophageal dilation, followed by concurrent CP botulinum toxin injection as needed, and reserving CPM for refractory cases. In this case, clinical symptoms improved after CP dilation, with decreased episodes of laryngeal penetration on increasingly advanced diet consistencies. Unfortunately, the patient's symptoms always recurred after several weeks. Therefore, botulinum toxin injection to the CP was used in conjunction with dilation and the patient had relief for nearly six weeks before symptoms returned. Given the refractory nature of the clinical scenario, the decision was made to pursue CPM as a definitive treatment.

The patient was brought to the operating room for external cricopharyngeal myotomy. A Savary dilator was placed into the esophagus as a stent and secured to the lip. A midline curvilinear horizontal incision was made extending below the cricoid. After the strap muscles were divided vertically in the anatomic midline, the trachea was palpated and the thyroid isthmus was identified inferiorly. This was carefully divided with monopolar cautery to allow for better exposure of the trachea. Next, 4.0 prolene sutures were placed through the cricoid lateral to midline and used as retraction sutures. The cricoid was retracted laterally to the left side, and the cricothyroid muscle and cricopharyngeus muscles were identified as shown in Fig. 2. The cricopharyngeus was transected vertically at its anterolateral attachment to the cricoid using bipolar cautery. The same technique was then completed on the right side. Hemostasis was verified, the wound was closed in layers, and a rubber band drain was left in the wound bed. He was restarted on his home diet of honey-thickened liquids post-operatively.

3. Results

The post-operative course was unremarkable; the drain was removed on the first post-operative day, and an immediate VFSS showed dramatic improvement in the timeliness of pharyngeal swallow onset with marked reduction in laryngeal penetration. The patient was discharged on a thin liquid diet. At the two week follow up visit, the incision was well-healed and the patient continued to do well, tolerating thin liquids. Repeat swallow evaluation at eight weeks post-operatively showed normal pharyngeal onset timing and no documented episodes of laryngeal penetration on any consistency (Fig. 3); the gastrostomy tube was removed and the patient continues to do well six months after the surgery. Repeat manometry was deferred given lack of clinical indication.

4. Discussion

The benefits of minimally invasive procedures explain their draw. Decreased morbidity, decreased pain, shorter hospital stay, quicker return to work, and improved cosmesis are attractive targets that minimally invasive surgery offers [18]. However, with pediatric cases, there are important considerations. Endoscopic procedures in the pediatric airway can be challenging due to narrow access and developing structures, although a recent article reported a successful endoscopic myotomy in an 8-month-old child [11]. A systematic review in adults showed a statistically significant higher success rate with endoscopic CP myotomy as compared to open myotomy [19]. Though the review findings support a minimally invasive approach, they importantly do not account for the pediatric population, and also do not reveal the specific types of open surgical approaches to CP myotomy—we are left to suspect that the majority were likely posterior midline transections, the traditional approach. Known complications from myotomy (either open or endoscopic) include hemorrhage, neck or retropharyngeal hematoma, wound infection, inadequate myotomy, recurrent laryngeal nerve (RLN) palsy or transection, salivary leak, esophageal perforation,

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