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Injection laryngoplasty in children with cystic fibrosis and abnormal swallow

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ABSTRACT

Objective: Children with cystic fibrosis (CF) are uniquely vulnerable to the pulmonary complications of chronic aspiration. We present a case series of children with CF and evidence of chronic aspiration who underwent injection laryngoplasty to improve the safety and efficacy of their swallow.
Study design: Retrospective chart review.
Setting: Tertiary care children's hospital.
Methods/Subjects: A chart review was performed on three consecutive cases of children with CF and evidence of aspiration on modified barium swallow (MBS) evaluated at a tertiary care, academic children's medical center.
Results: Three patients with CF underwent injection laryngoplasty for evidence of aspiration or laryngeal penetration on MBS evaluation. Normal laryngeal anatomy was identified intraoperatively in each case. At the time of the procedure, patients were 22 months, 70 months, and 24 months old, and follow-up information was available for 7 months, 11 months, and 12 months post-procedure, respectively. Presenting symptoms included chronic cough, cough with oral liquids, and recurrent pneumonia. Each patient underwent successful injection into the interarytenoid space. Post-operatively, modified barium swallow demonstrated resolution of aspiration or penetration in all patients. No procedure-related complications were encountered.
Conclusion: Patients with CF are highly susceptible to pulmonary infections, and aggressive treatment of chronic aspiration is often necessary. Injection laryngoplasty may be effective in normalizing swallowing in these children. Future study will elucidate the duration of effect and if this technique improves long-term pulmonary outcomes in CF patients.

1. Introduction

Cystic fibrosis (CF) is the most common autosomal recessive disease that shortens the lifespan in the Caucasian population. CF is a multisystem disease that affects the sinuses, pancreas, liver, lungs, and reproductive organs, though the respiratory system is the greatest source of morbidity and mortality in these patients [1]. Children with cystic fibrosis (CF) demonstrate gastroesophageal reflux (GER) at a rate 6–8 times higher than unaffected children, and the resulting aspiration of gastric contents contributes significantly to the progression of pulmonary disease [2–6].

Most authors advocate for aggressive and early treatment of GER in children with CF in order to limit progression of pulmonary disease [2–5]. Conservative measures include feeding modifications and anti-reflux medication including proton pump inhibitors or H2-blockers. In children with persistent failure to thrive, pulmonary infections, or decline in pulmonary function, surgical intervention such as a Nissen fundoplication are considered [2]. Fundoplication has proven effective in the treatment of GER in this population [2,3]. However, in the vulnerable CF population, this surgery is associated with a 16% rate of major complication including intraoperative morbidity, gastric perforation, peritonitis, hiatal hernia, esophageal stricture, and surgical failure. Forty eight percent had minor complications including persistent retching and emesis [2].

Injection laryngoplasty (IL) has become routine in the diagnosis and management of the mild laryngeal anomaly, a type I laryngeal

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clefts (LC-I). Multiple substances including gelfoam, carboxymethylcellulose aqueous gel, and hyaluronic acid derivatives have been successfully placed in the interarytenoid space during IL to diagnose or treat LC-I [7,8]. In those children who fail conservative management, IL offers rates of symptom resolution that ranges from 56% to 72%. Though repeat injections may be necessary, the benefits from IL appear to persist beyond the half-life of the injected substance [7,8]. The long-term efficacy of IL may be a consequence of the child to learning to compensate for the defect slowly as the injected substance resorbs.

Recently, Horn et al. demonstrated the efficacy of IL in children with chronic aspiration and an anatomically normal larynx [9]. The authors suggested that the interarytenoid space may represent a point of vulnerability in the immature larynx and that temporary support in this region by IL may be appropriate in children with aspiration despite an anatomically normal larynx. This assertion is sensible considering the relatively minor and subtle distinction between an anatomically normal interarytenoid notch and the pathologic LC-I that extends to the level of the true vocal cords. In the Horn study, 6 of the 28 children who were injected had an LC-I identified by palpation on microdirect laryngoscopy. 5/6 children with LC-I improved after IL and were able to tolerate a less restrictive diet while 11/22 children with normal anatomy had improvements in their ability to swallow without aspirating.

At the time of diagnosis of CF, 20% of infants and 25–55% of children above the age of 1 year have abnormal GER [3]. They incur significant pulmonary morbidity due to chronic reflux and aspiration of stomach contents. IL in CF patients, even in children with normal laryngeal anatomy, may prove to be a useful means of controlling chronic aspiration. We reviewed the charts of three children with CF who had clinical and radiographic evidence of chronic aspiration. Each child had undergone IL to augment a deep interarytenoid notch which was felt to be a source of weakness in their growing larynx.

2. Methods

After institutional review board (IRB) approval was obtained, we reviewed the charts of 3 children with CF who had undergone IL. Each child had clinical evidence of chronic aspiration and a modified barium swallow (MBS) study which demonstrated laryngeal penetration or aspiration. The patients' families were consented for microdirect laryngoscopy and injection laryngoplasty and underwent an extensive discussion regarding the risks of the procedure including, but not limited to, failure to improve symptoms, failure to reduce aspiration, and post-operative airway compromise.

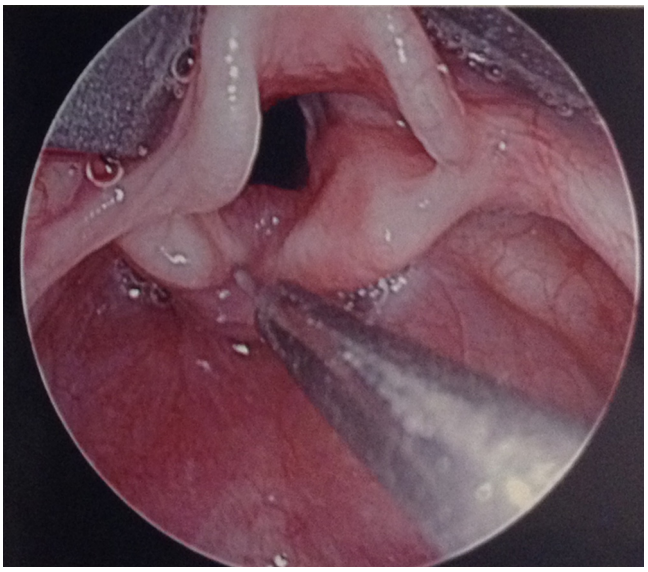

Fig. 2.

2.1. Surgical technique

All procedures were performed under general anesthesia in the operating theater. Spontaneous ventilation was maintained throughout without the use of endotracheal intubation. A standard microdirect laryngoscopy and bronchoscopy was performed first using a 4 mm telescope (Storz, Germany). The laryngoscope was placed into suspension (Fig. 1). Palpation of the interarytenoid was performed using a microlaryngeal blunt probe or spatula. Even if the standard definition of a type 1 laryngeal cleft was not identified, the interarytenoid area was injected with a total of 0.3 mL–0.5 mL of Radiesse Voice Gel® (San Mateo, CA) (Fig. 2). The results after injection of the interarytenoid space can be found in Fig. 3.

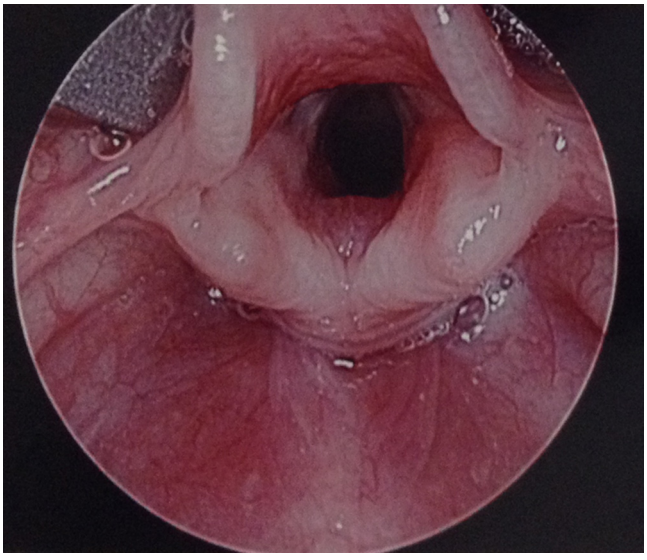

Fig. 1.

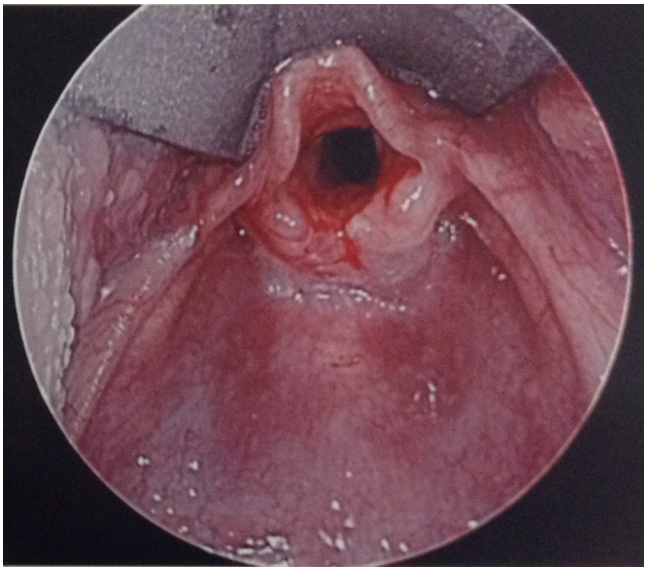

Fig. 3.