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INTRODUCTION

Laryngeal clefts are rare congenital anomalies resulting from failed fusion of the tracheoesophageal septum early in fetal development. Failure of this process leads to a myriad of congenital defects ranging from esophageal atresia to laryngeal cleft formation. The earlier in development failure of fusion occurs, the more devastating the resultant defect tends to be. Although Laryngeal clefts are most commonly felt to be inherited sporadically, there have been some case series identifying familial associations of the defect. In these families, it was proposed that the defect may actually have been inherited in an autosomal dominant fashion. Sporadic instances of laryngeal cleft are rare, occurring only in approximately one out of every 10,000 to 20,000 live births.

Laryngeal clefts are most commonly classified based on the degree of fissuring. This classification scheme, first proposed by Benjamin and Inglis, groups laryngeal clefts based on the extent of the fissure in the craniocaudal aspect. Type I laryngeal clefts extend only into the arytenoid mucosa, often found on endoscopic examination as a deep interarytenoid notch. Type II laryngeal clefts lead to a notch within the posterior cricoid itself. Type III clefts extend fully through the posterior cricoid with possible extension into the proximal trachea, and Type IV clefts extend more distally into the trachea, occasionally extending into the carina.

Symptoms of a type I laryngeal cleft are typically nonspecific and include chronic cough, choking, and aspiration. Patients may present with recurrent aspiration pneumonia. Videofluoroscopic swallow study (VFSS) is a useful tool in diagnosis of a type I cleft, as it can show evidence of posterior laryngeal penetration with oral intake. Diagnosis typically is confirmed with direct laryngoscopy and palpation of the interarytenoid space. Management of type I laryngeal clefts is varied, with accepted interventions ranging from conservative measures such as use of thickened liquids and acid suppression to minimally invasive surgical intervention. Most centers propose treating initially with conservative measure followed by surgery only for those patients who do not improve with conservative measures.

Although endoscopic closure with use of CO₂ laser has been commonly used in repair of laryngeal clefts, injection laryngoplasty is being implemented with increasing frequency. This technique has been shown to decrease symptoms in patients with type I laryngeal cleft, and it is commonly used as an initial surgical intervention for patients with this condition at our institution. Complications from this procedure are infrequent, and there are essentially no reports of complication rates within the currently available literature. This case report serves to detail a previously unknown complication following injection laryngoplasty for a type I laryngeal cleft.

CASE REPORT

A 12-month-old male presented to the pediatric otolaryngology clinic for evaluation of dysphagia and concern for laryngomalacia. He had poor oral intake and was largely dependent on nasogastric feeds. He additionally had intermittent stridor, worsened with oral feeding or supine positioning. The patient was taken for gastrostomy (G) tube placement and underwent simultaneous direct laryngoscopy and bronchoscopy. In addition to moderate laryngomalacia, the patient was noted to have a type I laryngeal cleft. He was subsequently taken for supraglottoplasty and simultaneous injection laryngoplasty of the laryngeal cleft. Supraglottoplasty was performed first and involved releasing the aryepiglottic folds and excision of redundant arytenoid mucosa. Following this, 0.2 mL of Surgifoam (Ethicon, Somerville, NJ) was injected into the posterior larynx. This was an area of concern and the patient subsequently developed a large postcricoid mucocele requiring revision surgery and marsupialization of the cyst just 2 weeks following the initial surgery.

Key Words: Injection laryngoplasty, mucocele, dysphagia, aspiration, complication.

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A 12-month-old male underwent injection laryngoplasty for dysphagia associated with a deep interarytenoid notch. He subsequently developed a large postcricoid mucocele requiring revision surgery and marsupialization of the cyst just 2 weeks following the initial surgery.

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injected under visualization with a 4.0 mm 0° Hopkins rod directly into the laryngeal cleft. Inspection with the telescope showed adequate bulking in the area of clefting. The patient was admitted to the pediatric intensive care unit for close airway monitoring overnight and was discharged home the following day with instructions to continue tube feeds and trials of oral feeding.

Within 1 week postoperatively, the patient developed stridor that was worse than his preoperative state. He also developed pauses with sleep that had not been present prior to surgery. He was prescribed Augmentin and a steroid taper, but the symptoms progressed. As such, the patient was brought into clinic for reevaluation on postoperative day 13. A flexible scope examination was performed to evaluate the patient’s larynx. He was noted to have a large mass concerning for a granuloma arising from the left postcricoid mucosa, which had not been present preoperatively. He was prescribed a longer steroid taper and continued antibiotic course with instruction to return for repeat evaluation in 1 week.

On postoperative day 15, the patient presented to the emergency department due to further worsening of his stridor, which was now associated with sternal retraction and tracheal tugging. The patient was admitted to the hospital and subsequently taken to the operating room for direct laryngoscopy. The mass was again noted to be arising from the postcricoid mucosa on the left side (Fig. 1). Palpation of the mass revealed it to be largely cystic in nature. Needle aspiration of the cyst was performed initially without significant decompression. A sickle knife was used to incise the mucosa overlying the cyst, which led to expression of trapped mucous. As the cyst was probed further, a material with appearance consistent with the Surgifoam was encountered and removed (Fig. 2). The cystic space was probed until all visualized Surgifoam was removed. The area probed included the left postcricoid region, inferiorly to the level of the esophageal inlet. Following conclusion of the procedure, the patient was extubated without difficulty. Pathologic evaluation of the cyst contents revealed a gelatinous, nonbiologic material consistent with Surgifoam.

The patient was observed overnight and had bedside swallow assessment performed the following day. This revealed no evidence of aspiration, and the patient was cleared to resume small volume oral feeding trials in addition to G-tube feeds. He was discharged home in stable condition later that day. At the most recent follow-up, the patient was continuing outpatient feeding therapy with some improvement in oral intake. Flexible scope exam was repeated 2 months following marsupialization of the postcricoid cyst. The exam at that time revealed a normal appearing supraglottis with no evidence of recurrence of the cyst. Scope exam 5 months following the procedure revealed no evidence of recurrence.

Fig. 1. Direct laryngoscopy on postoperative day 16 revealing a large cystic mass emanating from the postcricoid mucosa. [Color figure can be viewed in the online issue, which is available at www.laryngoscope.com.]

Fig. 2. (A) View of the postcricoid mucocele following incision and expression of mucous. (B) Closer evaluation showing inferiorly tracking cyst cavity. Surgifoam demarcated by the asterisk. [Color figure can be viewed in the online issue, which is available at www.laryngoscope.com.]
DISCUSSION

To our knowledge, this report describes a previously unknown complication of laryngeal cleft injection. One potential mechanism for this complication was improper location of the Surgifoam injection. The material could have been injected too far posteriorly and to the left of midline, allowing the substance to migrate between the mucosa and the arytenoid and cricoid cartilages into a potential space created by the injected material. Subsequently, this would have led to obstruction of one or more mucous glands causing formation of a mucocele. Alternatively, expansion of the Surgifoam following injection could have led to progressive swelling, as has been described in anterior cervical fusion. Finally, although Surgifoam is a commonly used substance for injection laryngoplasty in adults, no study has specifically evaluated the use of this particular injectate in children. Given the lack of data in this population, the possibility of local inflammatory response or foreign body reaction to the Surgifoam cannot be discounted as a possible contributing factor to this complication. No matter the initial mechanism, the continued accumulation of mucous led to worsening inspiratory stridor due to glottic obstruction and worsened dysphagia from baseline due to obstruction of the esophageal inlet. Fortunately, simple marsupialization of the cyst and removal of the Surgifoam led to complete resolution without further recurrence.

As noted previously, injection laryngoplasty for type I laryngeal clefts is accepted as an effective means of reducing aspiration noted on VFSS. One series noted complete resolution of aspiration in 56% of patients following injection alone. However, five patients in this series went on to require a formal endoscopic repair. A more recently published series examined injection laryngoplasty for chronic aspiration in patients with or without a laryngeal cleft. Five out of six patients with laryngeal cleft had improvement in aspiration. Interestingly, 50% of patients who had chronic aspiration without laryngeal cleft showed improvement following interarytenoid injection laryngoplasty. This raises the question as to whether there is some contribution from incompetence of the arytenoid complex leading to aspiration in some instances. It has been noted that patients who show aspiration of thin liquids are more likely to have benefit from interarytenoid injection.

Although retention cysts have been reported within the postcrioid mucosa, they are much less common than retention cysts appearing either within the vallecula or other supraglottic subites. Any cyst arising from supraglottic structures tend to present with similar symptoms if they are greater than 1 cm, with common symptoms including dysphagia and respiratory issues such as stridor. The present literature on laryngeal cleft injection has limited information regarding complications.

Although this may suggest a relative lack of complications using injection laryngoplasty, an alternative possibility is that the use of this particular injectate in children is accepted as an effective means of limiting aspiration. One reported case of mucocele following injection alone. However, this may suggest a relative lack of complications using injection laryngoplasty, an alternative possibility is that the use of this particular injectate in children is accepted as an effective means of limiting aspiration.

CONCLUSION

Future studies on injection laryngoplasty should focus on complications. Though this is likely an uncommon complication of injection laryngoplasty, it led to significant morbidity for this patient, as the patient required repeat operation and hospitalization. As such, practitioners should bear this in mind whenever discussing injection laryngoplasty for laryngeal cleft with patients and their families.

BIBLIOGRAPHY