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Short-term swallowing outcomes following type 1 laryngeal cleft injection

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ABSTRACT

Introduction: Interarytenoid injection augmentation at the time of initial diagnostic endoscopy for aspiration and dysphagia may result in near-immediate improvement in swallowing function, potentially obviating the need for future formal endoscopic repair of type 1 laryngeal cleft. Interarytenoid injection augmentation may also address physiologic aspiration. Early treatment of type 1 laryngeal cleft may allow for expedited liberalization of feedings. The objective of this study was to evaluate the effect of interarytenoid injection augmentation (IIA) for type 1 laryngeal clefts (LC-1) on short-term swallowing function assessed by videofluoroscopic swallowing study (VFSS).

Methods: This was a retrospective cohort study of patients age ≤24 months with dysphagia on preoperative VFSS who underwent IIA with calcium hydroxyapatite for LC-1 during direct laryngoscopy and bronchoscopy from June to October 2017 at a tertiary care academic subspecialty hospital. Exclusion criteria included prior endoscopic or open LC repair (n = 1), gastrostomy tube dependence (n = 1), additional procedures at the time of IIA (supraglottoplasty, frenulectomy, n = 1). Children without postoperative VFSS within 30 days of injection were excluded (n = 2). Fifteen children met inclusion criteria for analysis. The primary endpoint was improvement in safely swallowed consistency as defined by recommendation to liberalize diet by at least a half-consistency (e.g. half-honey to nectar thick liquid). Secondary endpoints included clinical assessment of dysphagia and postoperative respiratory events.

Results: Median [range] age at injection was 15.2 [7.7–24.3] months and 67% of patients were female (n = 10). The majority (13/15) of patients were full-term and 80% of patients (n = 12) had documented gastroesophageal reflux disease (GERD). Median time from injection to VFSS was 16 [9–29] days. Improvement in safely swallowed consistency was noted in 60% (n = 9) of patients. Aspiration completely resolved in two patients. Swallow function was unchanged in 40% of patients (n = 6); no patients experienced worsening dysphagia. No respiratory complications were documented during inpatient observation.

Conclusion: IIA is a safe procedure that may result in immediate improvement in dysphagia in select patients with LC-1. IIA does not address neurologic, developmental, or other anatomic etiologies of dysphagia. Additional studies are required to determine long-term efficacy of IIA on dysphagia and pulmonary complications, as well as the patient- and caregiver-related outcome measures.

1. Introduction

Laryngeal clefts (laryngo-tracheal-esophageal clefts) are abnormal posterior communications between the laryngotracheal complex and the esophagus secondary to incomplete development of the tracheoesophageal septum [1]. Laryngeal cleft severity corresponds to the degree of downward extension; the most commonly used classification scheme is that of Benjamin and Inglis which divides clefts into four types (Fig. 1) [2]. Type 1 laryngeal clefts do not extend below the level of the true vocal folds; children with deep interarytenoid notches are frequently included in this group [3,4]. Type 2 laryngeal clefts extend below the true vocal folds into the cricoid cartilage. Type 3 and type 4 laryngeal clefts extend into the cervical and thoracic trachea, respectively.

Abbreviations: LC-1, Type 1 Laryngeal Cleft; IIA, interarytenoid injection augmentation; VFSS, videofluoroscopic swallowing study

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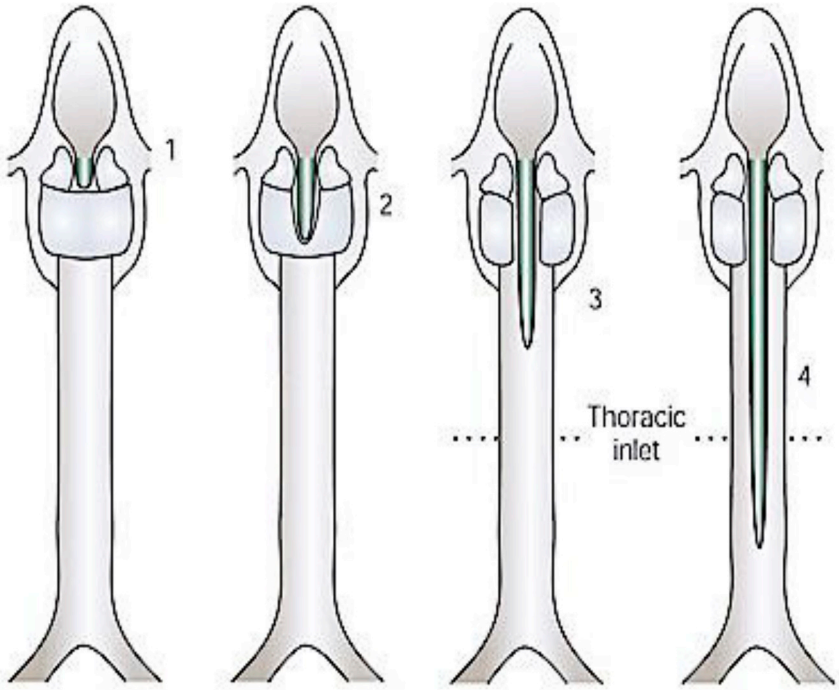

Fig. 1. Benjamin and Inglis' Classification of Laryngeal Clefts. Reprinted with permission from Sage Publishing Co.

Clefts most often occur sporadically, but are also associated with several known syndromes, including VACTERL (vertebral defects, anal atresia, cardiac defects, tracheo-esophageal fistula, renal anomalies, and limb abnormalities) [5]. Major defects and syndrome-associated malformations may be noted prior to or at birth; however, subtler midline abnormalities (e.g. type 1 laryngeal clefts) may not be recognized until patients are symptomatic. Undiagnosed isolated type 1 laryngeal clefts most commonly manifest with difficulty swallowing and/or feeding intolerance, prompting speech and language pathology (SLP) swallow assessment [6]. Laryngeal penetration and aspiration may be diagnosed utilizing fiberoptic endoscopic evaluation of swallowing (FEES) or videofluoroscopic swallowing study (VFSS). Initial management of presumed type 1 laryngeal clefts is often conservative (e.g. thickened feeds, treatment of comorbid GERD), though less than half of children respond completely to these interventions [7–9]. Children who fail conservative management of dysphagia may then require operative endoscopy to determine etiology of swallowing dysfunction.

Formal diagnosis of laryngeal cleft requires direct laryngoscopy with assessment of the interarytenoid area under general anesthesia [10,11]. Following diagnosis, type 1 laryngeal clefts may be treated with formal interarytenoid suture augmentation or interarytenoid injection augmentation (injection laryngoplasty). Interarytenoid suture repair of clefts involves placement of sutures or use of CO2 laser under endoscopic guidance to eliminate the interarytenoid cleft [9,12]. Children may be treated with sedation postoperatively to prevent agitation or crying which may weaken suture repair, which requires overnight observation on the inpatient floor or pediatric intensive care unit [13,14]. Interarytenoid injection augmentation (IIA) continues to increase in popularity as both a diagnostic and therapeutic technique [4,15,16]. Importantly, patients do not require post-procedural sedation. In addition, the patient's laryngeal cleft is managed on the initial endoscopic evaluation, which may obviate the need for repeat anesthetic at a later time point for repair.

Despite increasing popularity of IIA, no consensus exists regarding timing of VFSS surveillance after injection. For patients who have undergone interarytenoid suture augmentation, surveillance VFSS typically occurs at approximately 6–12 weeks [6,13]. During this time, children are maintained on previously thickened diet. For interarytenoid injection augmentation, no formal postoperative recovery time is required at our institution, allowing for earlier assessment of postoperative swallowing function. The purpose of the present study is to review cases of children with documented pharyngeal phase dysphagia on preoperative VFSS who underwent IIA with subsequent assessment of short-term swallowing function.

2. Methods

The study was performed at a tertiary care subspecialty hospital (Massachusetts Eye and Ear, Boston, MA). This study is covered by IRB 09-12-120.

The clinical aerodigestive database was queried for all cases of interarytenoid injection augmentation for type 1 laryngeal cleft performed from June 1, 2017 to October 30, 2017. Retrospective medical record review was performed to obtain the following variables:

- Demographic information (age and sex)
- Medical comorbidities
- Preoperative swallowing function as assessed by VFSS (performance on all consistencies tested)
- Post-surgical monitoring (e.g. desaturations)
- Postoperative swallowing outcomes as assessed by VFSS (performance on all consistencies tested)

Cases were eligible for inclusion if children were between 0 and 24 months at the time of injection and had documented pharyngeal phase dysphagia on preoperative VFSS. Patients were evaluated by pediatric speech and language pathologists and assessed for penetration or aspiration with sampled consistencies. All patients with documented pharyngeal dysphagia underwent suspension laryngoscopy with exposure and palpation of the interarytenoid region for definitive diagnosis (Fig. 2A). A vocal fold distractor was used to separate the vocal folds and a suction probe (not attached to suction) was utilized to palpate the interarytenoid region and confirm presence of a laryngeal cleft as described by Ojha et al. [13] Patients then underwent IIA with Prolaryn Plus (calcium hydroxyapatite within aqueous/glycerin/carboxymethylcellulose gel; Merz North America, Raleigh, NC) (Fig. 2B

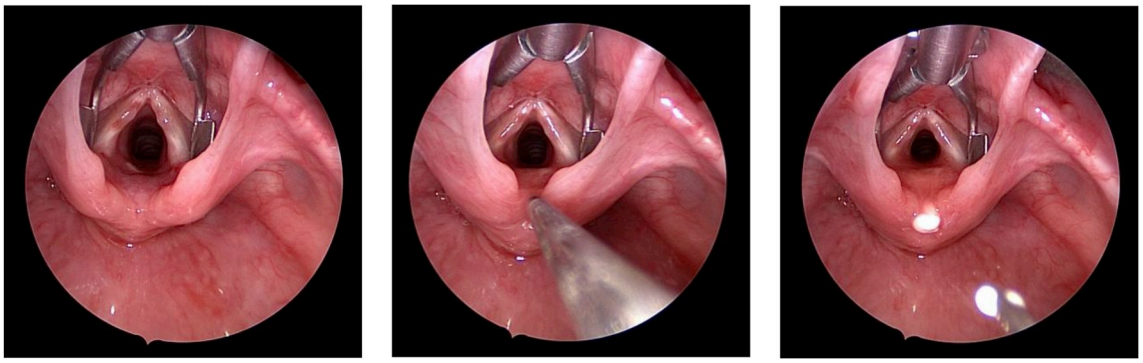

Fig. 2. Interarytenoid injection augmentation. Patient with type 1 laryngeal cleft viewed with suspension microlaryngoscopy before (A), during (B), and after (C) injection of calcium hydroxyapatite within carboxymethylcellulose gel to interarytenoid space.

and C).

Following IIA, patients were admitted to the inpatient floor for monitoring. All patients were placed on continuous oxygen saturation monitoring to evaluate for desaturation events post-procedure. Following discharge, patients were required to have postoperative VFSS documented within 30 days of injection; the majority of studies were performed two weeks after injection. Patients who had previously undergone endoscopic or open repair of laryngeal cleft were excluded from the study. Patients without documented postoperative VFSS were excluded from the study.

We defined *improvement* in safely swallowed consistency as significant decrease in penetration and/or complete resolution of aspiration with a given consistency with subsequent recommendation to liberalize feedings by one-half consistency or greater (e.g. half-honey to nectar thickened liquids). *Resolution* was defined as lack of aspiration or penetration with thin liquids. A postoperative respiratory occurrence was defined as desaturation overnight to below SaO2 of 90% and/or supplemental oxygen requirement prior to discharge.

Descriptive statistics are described using median and ranges. Categorical data are presented as percentages.

3. Results

During the study period, 20 consecutive patients underwent interarytenoid injection augmentation (IIA) for pharyngeal phase dysphagia. All patients had documented preoperative VFSS. The median time from preoperative VFSS to interarytenoid injection augmentation was median [range] 61 [5–270] days. Patients with prior endoscopic or open LC repair (n = 1) and with gastrostomy tube dependence (n = 1) were excluded from this analysis. One child who underwent additional invasive procedures at the time of interarytenoid injection augmentation (supraglottoplasty, frenulectomy) was excluded. Children without postoperative VFSS within 30 days of injection were excluded (n = 2). A total of 15 patients met study inclusion criteria (Fig. 3).

The median [range] age at injection was 15.2 [7.7–24.3] months. Of the 15 patients, 10 were female (67%) and 5 were male (33%). The majority (13/15) of patients were full-term. With regard to comorbidities, 80% of patients (n = 12) had documented GERD and 53% (n = 8) had documented history of bronchiolitis or asthma. Over half (60%, n = 9) had previously been hospitalized for sequelae of aspiration. All children had penetration and/or aspiration with thin liquids on preoperative VFSS consistent with pharyngeal phase dysphagia. Of the included patients, the most common presenting symptoms were recurrent upper respiratory infection (URI) and coughing or choking with feeds. Nine of 15 children (60%) were on nectar-thickened liquid or more restrictive diet at the time of presentation. Patient demographic data and baseline characteristics are detailed in Table 1.

No children (0/15) experienced desaturation or respiratory complications during inpatient observation period. All children were discharged to home with instructions to continue preoperative diet until postoperative VFSS. The median time from injection to VFSS was 16 [9–29] days. We classified success as either improvement in safely swallowed consistency with recommendations to liberalize diet one-half consistency or greater or resolution in symptoms on VFSS (no aspiration or penetration with thin liquids).

At the time of postoperative VFSS, improvement in safely swallowed consistency was noted in 9/15 patients (60%) (Table 2). Complete

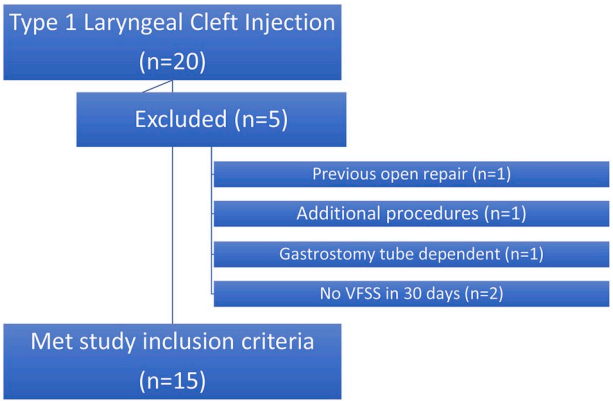

Fig. 3. Study Inclusion Criteria. A total of 20 consecutive children underwent interarytenoid injection augmentation of type 1 laryngeal cleft over study period. Of these, 15 met study inclusion criteria for analysis.

Table 1
Patient characteristics.

Demographics	Median (Range) | No (%)
Age (months)	15.2 (7.7–24.3)
Female sex	10 (67)
Full-term	13 (87)
Comorbidities	**No (%)**
GERD	12 (80)
Asthma and/or Bronchiolitis	8 (53)
Hospitalization for aspiration	9 (60)
Presenting Symptoms	**No (%)**
Aspiration events	15 (100)
Choking on feeds	7 (53)
Chronic cough	3 (20)
Recurrent URI	9 (60)
Preoperative diet (liquid consistency)	**No (%)**
Thin	1 (7)
Half-nectar	5 (33)
Nectar	2 (13)
Half-honey	1 (7)
Honey	6 (40)

Table 2
Postoperative swallowing function.

Postoperative Swallowing Function	No (%)
Improvement in safely swallowed consistency	9 (60)
Resolved	2 (13)
Improved	7 (47)
No change in safely swallowed consistency	6 (40)
Restriction in safely swallowed consistency	0 (0)

resolution of symptoms was noted in 2/15 patients (13%). Swallowing function was unchanged in 6/15 patients (40%). No children experienced worsening dysphagia following laryngeal cleft injection. A comparison of safely swallowed consistency by patient from preoperative to postoperative VFSS is displayed in Fig. 4.

4. Discussion

Interarytenoid injection augmentation is a safe procedure that may improve dysphagia in select patients with type 1 laryngeal cleft. Our goal in the present study was to assess the immediate effect of injection augmentation on postoperative swallowing function. Our study builds on the work of Horn et al. in the assessment of swallowing outcomes within 30 days of injection (range 9–29 days) [16]. Within our study cohort, the majority of patients experienced a degree of immediate improvement in swallowing function as defined by liberalizing diet to a thinner consistency following postoperative VFSS. The overall rate of improvement is consistent with that of Cohen et al., though fewer patients in this study (15 vs 56%) reported complete resolution of aspiration at the time of postoperative VFSS [15]. This difference may be partially attributable to differences in timing of postoperative assessment or patient level variation given sample size. Diet improvement by a single half-consistency may also have variable impact on overall swallowing function depending on starting consistency (e.g. honey vs. nectar-thin). Additionally, in some cases, this degree of modest improvement may not be sufficient to consider the procedure a success in a binary sense.

More widespread use of interarytenoid injection augmentation has the potential to alter the current clinical management algorithm for type 1 laryngeal clefts [6,7]. Before use of interarytenoid injection augmentation, patients did not undergo postoperative VFSS until 6–12 weeks after endoscopic repair [6,13,17]. Patients were maintained on preoperative thickened diet until the time of postoperative swallowing evaluation. While the majority of children did demonstrate some improvement at the two-week time period, reshaping of injectate and reduction of edema may not be complete at the two-week time period in all cases.

Earlier liberalization of diet may have several important implications, including possibly increased breastfeeding duration for the mother-baby dyad, improvement in developmental feeding progression and less secondary impact of thickened feedings. Thickened feedings may result in frustration for both caregivers and children as thickening agents can result in constipation and negative oral experiences possibly leading to oral aversion. Furthermore, the ability to decrease the amount of thickening agent allows for improved nutrient content of feeds. As well, social interaction can be improved with no diet limitations and daycare options for children are additionally expanded.

The videofluoroscopic swallow study is the gold standard for assessing and treating swallow dysfunction and aspiration. Hersh et al. (2017) quantified VFSS radiation exposure over the course of the diagnosis and suture repair treatment of a type 1 laryngeal cleft. Data revealed that on average, a child who received a suture repair underwent 3.24 VFSS radiology exams, which translates to a radiation dose equivalent to 30.6 chest x-rays. The outcomes of IL have the potential to improve quality of life and reduce medical ionizing radiation, an additional detriment of prolonged dysphagia. By reducing detrimental radiation exposure with a reduced total number of VFSS exams over the course of treatment, and improved quality of life by liberalizing diet parameters in a timely manner, care for these patients can truly be optimized. We do acknowledge, however, that the VFSS represents a single point in time and that given the limited duration of effect for IAA, the majority of children will require additional VFSS to assess longer term viability of IAA as a treatment modality for pharyngeal phase dysphagia related to type 1 laryngeal cleft.

Given lack of documented adverse events with interarytenoid injection augmentation in the current study; specifically, the lack of desaturation events or worsening dysphagia following injection, one may argue that the threshold to perform interarytenoid injection augmentation should be lowered. Interarytenoid injection augmentation does not address neurologic, developmental, or other anatomic etiologies of

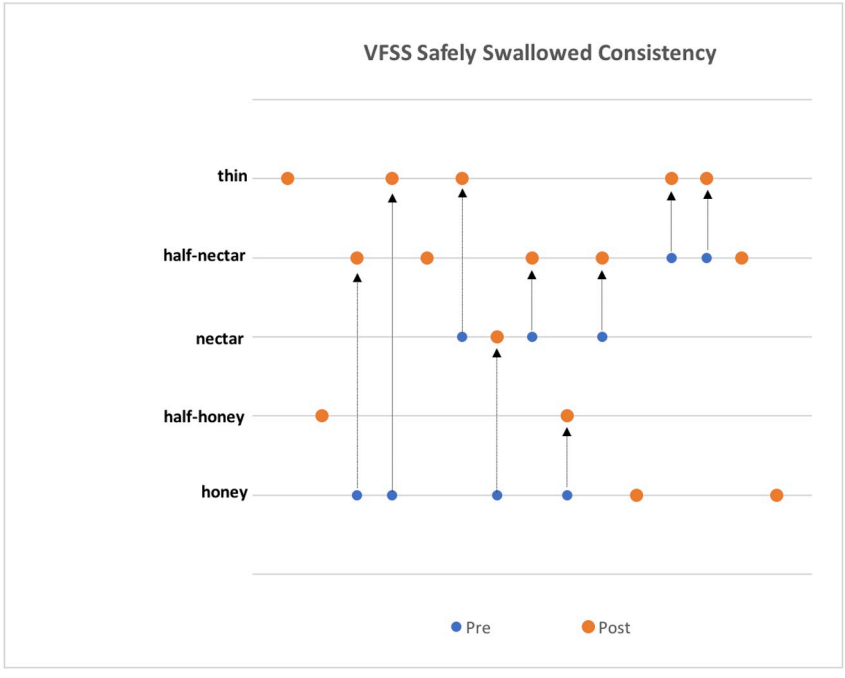

Fig. 4. Improvement in Safely Swallowed Consistency on VFSS. Each point represents single study patient; arrows denote improvement in safely swallowed consistency from preoperative (blue) to postoperative (orange) VFSS. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

dysphagia. This study is also limited by its retrospective nature and single institution design. Additional prospective studies are required to determine long-term efficacy of IL on dysphagia and pulmonary complications.

5. Conclusion

IIA is a safe procedure that may result in immediate improvement in dysphagia in select patients with pharyngeal phase dysphagia. IIA does not address neurologic, developmental, or other anatomic etiologies of dysphagia. Additional studies are required to determine long-term efficacy of IL on dysphagia and pulmonary complications, as well as patient- and caregiver-related outcome measures.

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Conflicts of interest

The listed authors have no conflicts of interest to disclose.

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