International Journal of Pediatric Otorhinolaryngology 115 (2018) 10–18

Contents lists available at ScienceDirect

International Journal of Pediatric Otorhinolaryngology

journal homepage: www.elsevier.com/locate/ijporl

Outcomes and swallowing evaluations after injection laryngoplasty for type I laryngeal cleft: Does age matter?

Elisabeth Cole[a], Alexandra Dreyzin[a], Amber D. Shaffer[b], Allison B.J. Tobey[b,d], David H. Chi[b,d], Tony Tarchichi[c,d,*]

[a] Department of Pediatrics, UPMC Children's Hospital of Pittsburgh, USA
[b] Division of Pediatric Otolaryngology, UPMC Children's Hospital of Pittsburgh, USA
[c] Paul C. Gaffney Division of Pediatric Hospital Medicine, UPMC Children's Hospital of Pittsburgh, USA
[d] University of Pittsburgh School of Medicine, USA

ARTICLE INFO

Presented at the 2018 Annual Scientific Meeting of the American Society of Pediatric Otolaryngology, National Harbor, MD, April 21, 2018.

Keywords:
Laryngeal cleft
Injection laryngoplasty
Prolaryn gel
Aspiration
Dysphagia

ABSTRACT

Objectives: To improve the recognition of differences in presentation amongst patients with type 1 laryngeal clefts of various ages and better understand the age dependent outcomes of injection laryngoplasty. A second aim was to analyze the discrepancies between swallow assessment modalities in various age groups with type I laryngeal clefts undergoing injection laryngoplasty.

Methods: A retrospective review of electronic medical records of patients who underwent injection laryngoplasty from 2009 through 2015 at a tertiary care children's hospital. Data extracted included: Demographics, histories and physical exam findings, diagnostic studies, and medical and surgical treatments.

Results: Most (72/102, 70.6%) patients were male with a median gestational age at birth of 37 weeks (range 24–41 weeks). Formula thickening and GERD medications were used in 94/102 (92.2%) and 97/102 (95.1%) patients, respectively. Comorbid GERD, laryngomalacia, tracheomalacia, and subglottic stenosis were present in 98/102 (96.1%), 40/102 (39.2%), 9/102 (8.8%), and 14/102 (13.7%) patients, respectively. There was no significant difference in demographics, comorbidities or medical therapy between age groups. Symptoms at presentation differed between age groups with stridor ($\chi^2(1) = 11.6$, $p = 0.002$) and cyanosis ($\chi^2(1) = 8.13$, $p = 0.012$) being more common in the 0–3-month group compared to the 12–36 month group. Symptom resolution and the odds of undergoing additional surgery (second injection or suture repair) over time, however, did not differ. There was a significant reduction in aspiration with thins during FEES (McNemar $\chi^2(1) = 10.7$, $p = 0.002$) and aspiration with nectar during MBS (McNemar $\chi^2(1) = 5.26$, $p = 0.035$) post-injection. After injection, there was significant agreement in aspiration with thins between FEES and MBS (kappa = 0.308 ± SE 0.170, $p = 0.035$). However, finding aspiration with thins was more common during MBS than during FEES (McNemar $\chi^2(1) = 7.00$, $p = 0.016$). There were no differences in swallow evaluation findings between the age groups.

Conclusions: Symptoms of type I laryngeal clefts may differ by age. However, there was no impact of age on the safety and efficacy of surgical intervention.

1. Introduction

Though considered a rare congenital anomaly, laryngeal clefts affect approximately 1 in 10,000 to 20,000 live births [1]. Laryngeal cleft can result in laryngeal penetration and aspiration, leading to feeding difficulties and respiratory disorders in young children [2–4]. Laryngeal clefts are classified by depth of defect, types 1 through 4, ranging from least to most anatomically disruptive [5]. Classically, a type I laryngeal cleft is characterized as a supraglottic interarytenoid defect that extends no deeper than the level of the true vocal folds and does not involve the cricoid cartilage [3,5]. Though there is not a significant malformation of the aerodigestive tract, type I laryngeal clefts are associated with both respiratory and feeding morbidity, including aspiration, pneumonia, failure to thrive, and respiratory distress [4,6].

The clinical presentation of type I clefts is subtler than types 2–4, often resulting in delayed diagnosis [7]. Moreover, since the aerodigestive defect is minimal, some may argue a type 1 cleft is a variant of normal and thus only needs to be addressed when clinically

* Corresponding author. Children's Hospital of Pittsburgh, 4401 Penn Avenue Pittsburgh, PA, 15224, USA.
E-mail address: tony.tarchichi@chp.edu (T. Tarchichi).

https://doi.org/10.1016/j.ijporl.2018.09.006
Received 5 July 2018; Received in revised form 21 August 2018; Accepted 12 September 2018
Available online 13 September 2018
0165-5876/

symptomatic, therefore multiple diagnostic tools are used to assess for dysphagia resulting from the cleft. Useful studies include clinical feeding assessments, modified barium swallow (MBS) and fiberoptic endoscopic evaluation of swallow (FEES) [3]. All diagnostic swallow studies in infants require patient and family participation, which is influenced by multiple confounding factors; environment, fatigue, hunger level, presence of URIs. Clinical feeding assessments allow the patient to feed in the most natural manor, however, is the most subjective and can miss silent aspiration. MBS is considered the gold standard for diagnosis of aspiration, as it is the least subjective. It does require the child to be feeding from a bottle/cup/straw, often in an upright, rigid position, thus not allow for assessment of breast feeding or feeding in non-upright positions. The fluorography equipment needed for MBS is cumbersome and therefore cannot not easily move with the child making imaging at appropriate angles that can be useful to determine site of aerodigestive anomalies difficult. Lastly, fluorography exposes the child to ionizing radiation, which accumulative doses can increase the risk of neoplasms and thyroid or thymus dysfunction. FEES, on the other hand requires no radiation, can be performed while breast feeding and can be performed in multiple positions. A FEES requires instrumentation with a fiberoptic laryngoscope. This can result in slight velar insufficiency and can be uncomfortable, which may upset the child, both of which can affect swallow. Lastly, there is “white out” phase during the actual swallow where the glottic view is obscured secondary to epiglottic inversion on a FEES. Therefore, evidence of spillage, penetration and aspiration during the pre and post swallow phase is actually what is being assessed in a FEES. Over the course of management of type I laryngeal clefts, patients undergo multiple evaluations, which incurs costs for the medical system and exposure to ionizing radiation for the patient thus the type of study and timing of study need to be carefully considered [8].

Once diagnosed, multiple options for management of laryngeal clefts are available [9]. Conservative measures include thickening feeds, modifying pacing and positioning during a feed, and addressing comorbidities including gastroesophageal reflux, eosinophilic esophagitis, food allergies, and reactive airway disease, which may contribute to airway dysfunction. Should these less invasive interventions fail, surgical interventions such as endoscopic repair, transoral robotic surgery, and injection laryngoplasty are available. Selection of the appropriate method of management is dependent on several factors, including age of the patient, type of cleft, and associated comorbidities [10]. While endoscopic repair has been considered the gold standard treatment for the correction of laryngeal clefts, injection laryngoplasty is a minimally invasive alternative that has proven to be favorable in children with type I laryngeal clefts [11].

Injection laryngoplasty was first reported in 2000 by Kennedy et al. [10] and has been performed in children ranging in age from 2 weeks to 14 years [12] with a mean age at injection of 9–11 months [11,13]. In most hands, injection laryngoplasty is faster and easier to perform and caries less risk compared to surgical repair. Numerous injectable materials are available for injection laryngoplasty, and the duration **these** materials remain present in the tissue is estimated to range from 6 weeks to 2 years depending on the material used. One potential drawback of a temporary material is the effects on symptoms may only be temporary as well, yet studies have shown this not to be the case in some patients [11]. Injectable materials also aid in “diagnosis” as they allow one to assess the degree the cleft plays in a patient's overall swallow dysfunction.

Studies have shown that early identification and surgical correction of laryngeal clefts are associated with improved outcomes in children, including decreased rates of hospitalizations due to improved respiratory status post-operatively [4,14]. A previous study demonstrated that patients successfully managed conservatively were significantly younger at diagnosis than those who were managed surgically (mean ages 10.3 vs 22.2 months) [15], but there are currently no studies comparing the management of laryngeal clefts in patients three months of age or less to older counterparts. Furthermore, no studies exist which illustrate the impact of injection laryngoplasty in these very young patients.

It is not well understood what the role of MBS or FEES is in evaluating success of cleft repair in modifying symptoms. While the current literature uses MBS and FEES routinely, and occasionally interchangeably, to qualitatively evaluate improvement in swallowing function following type I laryngeal cleft repair, no studies exist that evaluate the validity of either modality at accurately assessing improvement when compared to each other or to clinical swallowing evaluations (CSE) performed by a licensed speech pathologist.

To better understand the clinical practices surrounding laryngeal cleft injection and post-injection monitoring, we performed a retrospective record review at a large pediatric tertiary care facility to specifically compare the outcomes of injection laryngoplasty in patients less than and greater than four months of age.

2. Methods

2.1. Subjects

Following approval from the University of Pittsburgh Institutional Review Board, a list of patients who underwent injection laryngoplasty from 2009 through 2015 at a tertiary care children's hospital was assembled using current procedural terminology codes for direct microlaryngoscopy with injection (31,570 and 31,571). All consecutive patients with injection laryngoplasty during this time frame were included. Patients without injection laryngoplasty during this time frame, and those without at least one follow-up visit occurring ≥ 1 month following injection were excluded. At our institution, injection laryngoplasty is performed in patients with signs or symptoms of penetration or aspiration of thin liquids and deep interarytenoid notch on palpation. Aspiration was diagnosed either by clinical signs such as coughing or choking, history of aspiration pneumonia, or evidence of aspiration on swallowing evaluation (including clinical evaluation, MBS, or FEES). Injection laryngoplasty was performed with aqueous/glycerin/carboxymethylcellulose gel (Radiesse/Prolaryn™ Gel, Merz North America, Raleigh, NC) via suspension laryngoscopy under general anesthesia with spontaneous ventilation as previously described [11].

2.2. Data collection

A retrospective review of the electronic medical record was conducted. Data extracted included:

- Demographics and birth history including gestational age at birth and time spent in the neonatal intensive care unit (NICU). Patients were grouped according to age (0–3 months, 4–11 months, and 12–36 months).
- Recommendation for thickened feeds and diagnosis of or treatment for gastroesophageal reflux disease (GERD)
  - o Patients with poor weight gain, dysphagia, abdominal pain, esophagitis, chronic cough, reflux, throat pain, or vomiting in the absence of other causes were diagnosed with GERD. Invasive pH probe or impedance monitoring was not performed in these children due to their age (< 3 years old).
- Airway comorbidities (laryngomalacia, tracheomalacia, and subglottic stenosis) and interventions (supraglottoplasty)
- Symptoms (stridor, choking, coughing, dysphagia, failure to thrive, apnea, aspiration, retraction, and cyanosis) reported during clinic visits prior to first injection and at any subsequent visits until the end of the study period.
  - o A symptom was included as a presenting symptom/occurring at first injection if it was described in the documentation for the clinic visit immediately prior to the first injection.