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Pediatric laryngeal cleft repair and dysphagia

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ABSTRACT

Objective: To describe changes in diet and swallow function in patients with a laryngeal cleft after surgical repair of the laryngeal cleft.**Methods:** Retrospective case series performed using chart review. Primary outcomes were diet and swallow function before and after laryngeal cleft repair. Clinical evaluation and video fluoroscopic swallow studies (VFSS) were used to assess pre- and post intervention swallowing.**Results:** 16 pediatric patients were included in this study. Preoperatively, 14 (88%) patients had diet restrictions. Postoperatively, 12 (75%) patients tolerated a regular diet without limitation. 4 (25%) patients had no reduction in diet restrictions over the course of this study. For the 10 patients who transitioned to a regular diet postoperatively, a median of 300 days (range: 26 days - 3 years) passed to document achieving a regular diet. This was corroborated by an increase in normal oral and pharyngeal phase swallow function on VFSS postoperatively when compared with preoperative VFSS results.**Conclusion:** Dysphagia improves in most patients after laryngeal cleft repair. The range in time to a normal diet was wide. This may facilitate improved preoperative counseling and preparation of families' expectations.

1. Introduction

Laryngeal cleft is a relatively rare congenital abnormality in which there is incomplete separation of the larynx, and possibly trachea, from the pharynx and esophagus due to midline defect of the interarytenoid muscle, cricoid cartilage, and/or tracheoesophageal septum, depending on severity of the cleft [1,2]. The most frequent presenting symptoms are aerodigestive in nature with severity ranging from cough with drinking, to frank aspiration with respiratory distress [3–5]. Cleft severity and comorbidities contribute to symptom severity [1]. Low severity, mildly symptomatic laryngeal clefts may be managed conservatively with feeding modifications, while more severe clefts require surgical intervention [1]. Dysphagia persisting despite medical optimization and feeding modifications is a major indication for surgical intervention.

Our group aimed to better understand the effect of surgical repair on dysphagia. Postoperative diet or swallow function has been reported in several studies, but their primary focus was the surgical method and frequency with which revision surgery was required [3,6,7]. In 2014,

Osborn et al. looked at postoperative swallow function using objective swallow function assessments and found improvement in dysphagia over time for most, but not all, patients. They found that it took over 3 months for more than half of their cohort to achieve a regular diet postoperatively. Osborn et al. focused on postoperative swallow function rather than change from preoperative to postoperative swallow function; although they did report this change for a subset of their patients [8].

The objective of our study was to compare preoperative and postoperative diet and swallow function in patients with a laryngeal cleft to better understand the expected change in dysphagia following surgical intervention and resolution periods.

2. Materials and methods

We obtained approval from the University of Michigan Institutional Review Board to perform a retrospective chart review of pediatric patients undergoing laryngeal cleft repair from 1992 through 2015 (HUM00110934).

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The Electronic Medical Record Search Engine (EMERSE) [9] was used to identify patients with the keywords “laryngeal cleft”. Once all patient medical records were identified, their charts were hand-reviewed to determine eligibility. Exclusion criteria included conservative management of the laryngeal cleft or if the repair was performed at a different institution. Surgical interventions included open and endoscopic approaches, which were performed at the discretion of the surgeon.

The primary outcomes were change in diet and change in dysphagia. Diet was determined by guardian reports in concert with VFSS reports when VFSS was performed. Dysphagia was determined based on data from video fluoroscopic swallow studies (VFSS) only or 3 phase swallow studies. All VFSS were performed by a speech and language pathologist (SLP) or occupational therapist. Dysphagia was categorized as mild, moderate, or severe by SLP primarily based on the degree to which dysphagia impacted the patient’s ability to meet their nutritional needs orally. A mild impairment typically required some degree of diet or positioning modification, a moderate impairment often required supplemental tube feedings, and a severe impairment required the patient to be nil per oral (NPO) with strict tube feeding or with therapeutic tastes only.

Laryngeal cleft grade was classified using the classification system published by Benjamin and Inglis [2]. All grade 1 clefts included in the study went at least to the level of the true vocal cords. Grade II was defined as including a portion of the cricoid lamina. Grade III clefts extended through the entire posterior cricoid lamina. Grade IV clefts extended into the thoracic trachea.

Statistical analyses were performed using Statistical Packages for Social Sciences (SPSS) version 14 (New York, USA). T test, chi square, Fisher’s exact and ANOVA test were utilized. Multiple linear logistic regression and survival analysis were used for multi variable analysis. A p value of 0.05 was considered significant.

3. Results

EMERSE identified 306 patients meeting our search criteria. After chart review, 16 patients were included in this study. See the flow chart in Fig. 1 for inclusion criteria.

There were 3 (19%) females and 13 (81%) males with a median age

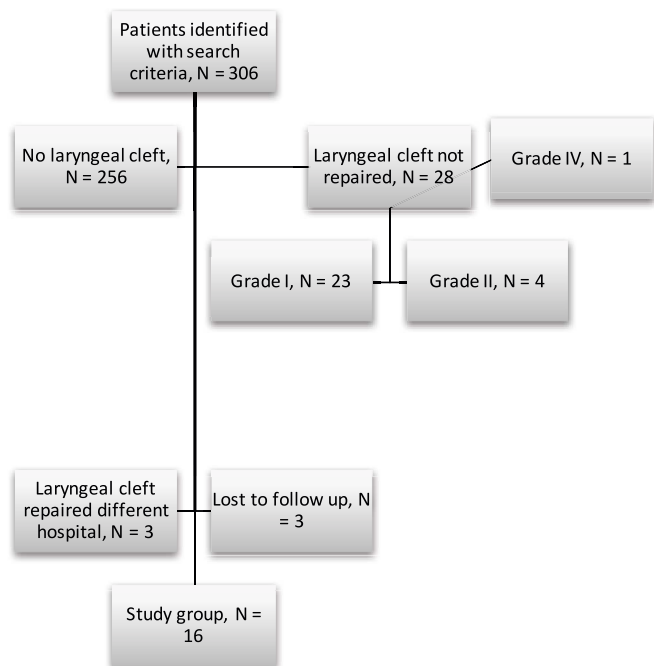


Fig. 1. Flow chart showing patients excluded and included in the current study.

Table 1

Laryngeal cleft grade and surgical intervention for 16 patients included in the study. All injections were of carboxymethylcellulose gel. Only 1 patient had a second injection. Values shown as: N (%).

Grade	Injection 8 (50)	Endoscopic suture 3 (19)	Open repair 5 (31)
I 7 (43.8)	7 (100)	0 (0)	0 (0)
II 2 (12.5)	1 (50)	1 (50)	0 (0)
III 6 (37.5)	0 (0)	2 (33)	4 (67)
IV 1 (6.3)	0 (0)	0 (0)	1 (100)

at surgical repair of 20 months old (range: 1 month–7 years). Laryngeal cleft grade and surgical intervention performed are detailed in Table 1. Two patients, both with grade III clefts, initially were managed with endoscopic intervention, but required revision open repair. Only the open repair is listed in Table 1 for these patients.

Feeding difficulty (ranging from cough with drinking to aspiration with all oral intake) was the primary presenting symptom for all but 1 patient, who presented with isolated respiratory symptoms. The majority (n = 14, 88%) of patients had a modified pre-operative diet. Eleven (68%) patients had a feeding tube preoperatively, 10 were gastrostomy tubes and 1 was a nasogastric tube. Of these, 7 (63%) were removed following laryngeal cleft repair. Of the four patients who still had a feeding tube at last follow up, two patients were dependent on tube feeding and two patients were taking a regular diet, but had no documented feeding tube removal. See Table 2 for additional information regarding patient medical and surgical comorbidities.

Fig. 2 illustrates preoperative diet as a function of laryngeal cleft grade. Fig. 3 demonstrates pre- and final post-operative diets for all patients. Most (n = 12, 75%) patients could tolerate a regular diet postoperatively by last follow up. Fig. 4 depicts change in diet comparing pre- and post-operative diets. No patient had an increase in their diet restrictions postoperatively. Four (25%) patients had no change in their diet restrictions postoperatively. Their laryngeal cleft grades were I-IV. All four patients had postoperative direct laryngoscopy ruling out persistent laryngeal cleft. The patients with grade I and IV clefts remained on purees and NPO, respectively. The patients with grade II and III clefts remained on honey thick diets.

Importantly, for those with preoperative diet modifications, it took a median of 10 months (range from 26 days to 3 years) for patients to take a regular diet without restriction. Only 6 patients had both pre- and post-operative VFSS to assess change in dysphagia pre- and post-operatively. No significant change in pharyngeal dysphagia was observed as 1 patient had improvement and 1 patient had worsening of their pharyngeal phase dysphagia (Fig. 5). 2/6 (33%) of patients had improvement in their oral phase dysphagia after laryngeal cleft repair (Fig. 6).

There was no statistically significant association between the

Table 2

Patient medical and surgical comorbidities. All preoperative airway surgeries were tracheostomies. Diagnostic direct laryngoscopy and bronchoscopy was not included. Concomitant airway diagnoses included tracheobronchomalacia, subglottic stenosis, and glossoptosis. GERD = gastroesophageal reflux.

Comorbidity	N (%)
Any comorbidity (except GERD)	13 (81)
Preoperative GERD medication	5 (31)
Postoperative GERD medication	7 (44)
Preoperative airway surgery	5 (31)
Concomitant airway diagnosis	7 (44)
Syndrome or genetic abnormality	6 (38)
Cardiovascular comorbidity	5 (31)
Neurocognitive comorbidity	3 (19)
Oral aversion	2 (13)
Tracheoesophageal fistula (repaired)	1 (6)

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