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Laryngomalacia and Swallowing Function in Children

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Abstract

Objectives—1) To determine the prevalence of dysphagia in children with laryngomalacia; 2) To ascertain whether severity of laryngomalacia influences the presence of swallowing dysfunction; 3) To examine whether patients with medical comorbidities and laryngomalacia have a higher prevalence of swallowing dysfunction.

Study Design—Retrospective cohort study.

Methods—All patients seen in the Aerodigestive Center at our institution between January 2007 and December 2012 with the diagnosis of laryngomalacia were included. Swallowing function was assessed by symptoms, clinical swallowing evaluations (CSE) performed by speech pathologists, modified barium swallow studies (MBS), and fiberoptic endoscopic evaluations of swallowing (FEES).

Results—There were 324 patients with laryngomalacia identified (41.4% female; 58.6% male). Severity of laryngomalacia was categorized in 279 patients, with 62.7% mild, 28.7% moderate, and 8.6% severe. Gastroesophageal reflux disease (GERD) was diagnosed in 69.8% of patients. Other medical comorbidities included Down syndrome (3.1%), neurologic impairment (6.5%), and congenital heart disease (0.9%). Symptoms of dysphagia or feeding difficulty were present in 163/324 (50.3%) and failure to thrive was present in 31/324 patients (9.6%). At least one abnormal swallowing assessment was present in 97/120 (80.8%) patients presenting with subjective dysphagia and 43/65 (66.2%) patients without subjective dysphagia. A total of 140/185 (75.7%) patients had at least one abnormal baseline swallowing assessment.

There was no significant relationship between severity of laryngomalacia and presence of abnormal swallowing function based on symptoms, CSE, MBS, or FEES. However, patients with greater severity were more likely to have failure to thrive.

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There was not a significant association between the presence of swallowing dysfunction or disease severity and medical comorbidities such as Down syndrome, neurologic impairment, or congenital heart disease. However, GERD was more likely to be present in patients with moderate and severe laryngomalacia than in patients with mild disease.

Conclusion—Swallowing dysfunction is common in children with laryngomalacia regardless of disease severity or other medical comorbidities. Swallowing studies are frequently abnormal in laryngomalacia patients presenting both with and without subjective symptoms of dysphagia. Dysphagia assessment should be considered as part of the evaluation of infants with laryngomalacia.

Level of evidence—4

Keywords

laryngomalacia; swallowing; dysphagia; supraglottoplasty; pediatric

Introduction

Laryngomalacia is the most common cause of stridor in infants and the most common congenital anomaly of the larynx.^{1,2} Laryngomalacia affects 50% to 75% of infants with stridor.²⁻⁴ Patients typically present with inspiratory stridor during the first few weeks of life, which usually worsens over the first 6 months of life and peaks in severity at about 6 months of age, followed by gradual improvement in the symptoms, with most patients being symptom free by age 18 to 24 months.^{4,5} The stridor is typically worse with agitation, crying, feeding, and supine positioning. In addition to stridor, patients with laryngomalacia can have feeding difficulty, failure to thrive, dysphagia, aspiration, apnea, cyanosis, reflux, obstructive sleep apnea, and pulmonary hypertension in severe cases.^{4,6,7} Gastroesophageal reflux disease (GERD) is a well-established comorbidity of laryngomalacia and many patients with laryngomalacia have symptoms of and are treated for reflux.^{4,8-12}

The diagnosis of laryngomalacia is typically made with flexible fiberoptic laryngoscopy. Findings include collapse of the supraglottic structures during inspiration, leading to inspiratory stridor and airway obstruction. Common endoscopic features include inspiratory prolapse of the arytenoid cartilages, redundant arytenoid mucosa, shortened aryepiglottic folds, and an “omega”-shaped or tubular epiglottis.^{13,14}

The etiology of laryngomalacia is likely multifactorial, with anatomic, inflammatory, and neurologic factors all contributing to the disease process.^{4,7,15,16} Anatomic factors include abnormal prolapse of laryngeal tissue, sometimes associated with tissue redundancy, leading to supraglottic obstruction.^{4,15-19} The cartilaginous theory suggests that immaturity or weakness of the laryngeal cartilage contributes to the obstruction.^{4,20,21} GERD is intimately associated with laryngomalacia and may play a role in the etiology of the disease, although a clear causal mechanism has not been established.^{4,8,10} The literature best supports neurologic mechanisms as the principal cause for laryngomalacia, including neuromuscular hypotonia and impaired neuromuscular control.^{4,18,22} Thompson demonstrated that

abnormal sensorimotor integrative function and laryngeal tone play an important role in the etiology of laryngomalacia.⁴

Given that swallowing interrupts breathing, infants with airway compromise or respiratory distress may not be able to safely coordinate sucking, swallowing, and breathing, leading to dysphagia and aspiration.^{4,6} Indeed, patients with laryngomalacia can have coughing and choking during feeding, feeding difficulty, dysphagia, aspiration, failure to thrive, or worsening of stridor during feeding.^{6,23,24} Swallowing function can be assessed based on symptoms, clinical swallowing evaluations (CSE) performed by a speech pathologist, and objective instrumental swallowing studies including modified barium swallow studies (MBS) and fiberoptic endoscopic evaluation of swallowing (FEES) examinations. The true incidence of dysphagia and aspiration in infants with laryngomalacia is unknown.

A previous study has revealed that patients with more severe disease are more likely to have symptoms of feeding problems.⁴ In addition, there is evidence that patients with medical comorbidities such as neurologic impairment or congenital heart disease tend to have a greater severity of laryngomalacia.^{4,7,25}

The primary goal of this study was to evaluate swallowing function in children diagnosed with laryngomalacia. The specific objectives included the following: 1) To determine the prevalence of dysphagia in children with laryngomalacia based on symptoms, clinical swallowing evaluations (CSEs) by speech pathologists, and objective instrumental swallowing studies (MBS and FEES); 2) To ascertain whether severity of disease influences the presence of swallowing dysfunction in patients with laryngomalacia; and 3) To examine whether patients with medical comorbidities such as neurologic impairment, congenital heart disease, or Down syndrome have a higher prevalence of swallowing dysfunction compared with patients without such comorbidities.

We proposed the following hypotheses: 1) Dysphagia and feeding difficulties would be common in patients with laryngomalacia; 2) Patients with more severe laryngomalacia would be more likely to have swallowing dysfunction; and 3) Patients with comorbidities such as neurologic impairment, congenital heart disease, or Down syndrome would have a greater severity of laryngomalacia and a higher prevalence of swallowing dysfunction compared with patients without these comorbidities.

Materials and Methods

This retrospective cohort study was reviewed by the Institutional Review Board at our institution and approved by expedited review. A retrospective chart review was performed for all patients seen in the Aerodigestive Center with laryngomalacia between January 1, 2007 and December 31, 2012 (6 year period). A search was performed for patients with an ICD-9 diagnosis code of 748.3 (other anomalies of larynx, trachea, and bronchus). Patients with this diagnosis code that did not have laryngomalacia were excluded. All of these patients were seen and treated by one of three pediatric otolaryngologists working in the Aerodigestive Center. Data were collected on de-identified data sheets and compiled and sorted on an Excel spreadsheet (Microsoft Corporation, Redmond, WA).

Data collected included age, gender, treating physician, gestational age, age at diagnosis, comorbidities (such as syndromes, gastroesophageal reflux disease (GERD) and laryngopharyngeal reflux (LPR), neurologic disease, congenital heart disease, and history of intubation or tracheotomy), presenting symptoms, other airway abnormalities (such as TVF immobility, laryngeal cleft, tracheomalacia, and subglottic stenosis), severity of laryngomalacia, and treatment modalities. Presenting symptoms evaluated included stridor, feeding difficulty, failure to thrive, dysphagia, aspiration, apnea, cyanosis, gastroesophageal reflux, obstructive sleep apnea, and pulmonary hypertension. Symptoms of dysphagia and feeding difficulty were grouped together, including coughing and choking with feeds, regurgitation, and caregiver report of aspiration.

GERD was diagnosed based on symptoms and findings during flexible laryngoscopy or upper aerodigestive tract endoscopy in the operating room, including esophageal biopsies. Findings at endoscopy suggestive of reflux included the following: erythema or edema of the true vocal folds, arytenoids, and interarytenoid or postcricoid areas; cobblestoning, erythema, or edema of the tracheobronchial mucosa; and erythema, edema, friability, or vertical furrowing of the esophageal mucosa. Esophageal biopsy findings that histologically revealed esophagitis (including at least 2 of the following three features: basal cell hyperplasia, increased papillary height, and epithelial inflammation) were also considered diagnostic of GERD. Only one patient in this study underwent pH probe testing.

Severity of laryngomalacia was determined by the pediatric otolaryngologist in the Aerodigestive Center based on presenting symptoms, physical examination, and findings on flexible laryngoscopy, using the criteria established in the laryngomalacia severity scoring system designed by Thompson.⁴ Patients diagnosed with *mild laryngomalacia* had symptoms of inspiratory stridor with or without coughing during feeding and patients with *moderate laryngomalacia* had inspiratory stridor with the presence of any of the following symptoms: choking or gasping during feeding, frequent regurgitation, brief inconsequential apneas or cyanotic episodes, or intermittent dyspnea with retractions that did not require medical attention.⁴ Patients with *severe laryngomalacia* had inspiratory stridor with life-threatening complications such as failure to thrive, apnea, cyanosis, or dyspnea requiring medical intervention, pectus excavatum, pulmonary hypertension, and cor pulmonale.⁴

Information about swallowing function was obtained from clinical symptoms, clinical swallowing evaluations (CSE) performed by speech pathologists in the Aerodigestive Center, modified barium swallow studies (MBS), and fiberoptic endoscopic evaluation of swallowing (FEES) examinations. CSE, MBS, and FEES studies were performed at the discretion of the evaluating pediatric otolaryngologist and speech pathologist in the Aerodigestive Center. For the clinical swallowing evaluations (CSE), the presence of congestion, coughing, choking, oxygen desaturation, or signs of respiratory distress were noted with various consistencies of feeds. The consistencies given for the CSE examinations as well as the MBS and FEES studies included thin liquids, nectar-consistency liquids (one ounce of rice cereal per two tablespoons of liquid), honey-consistency liquids (one ounce of rice cereal per one tablespoon of liquid), pureed, and solids when appropriate based on age.

The MBS studies were performed in the radiology department, with a radiologist and speech pathologist present. The patients were positioned in an age-appropriate tumbleform chair and fed various consistencies mixed with barium. Swallowing was assessed with fluoroscopy in the anterior-posterior and lateral planes. For each study, fluoroscopy times were minimized and kept to a total of 5 minutes or less. For MBS studies, the presence of penetration, aspiration, premature spillage, abnormal coordination, and other abnormalities were recorded across various consistencies of feedings.

The FEES examinations were performed in the endoscopy room in the Aerodigestive Center, with a speech pathologist and attending pediatric otolaryngologist present. Children were typically positioned on a caregiver's lap in an upright or semi-reclined position with gentle restraint. For infants under age one, no topical anesthesia was usually given; for older children, a small amount of topical 2% lidocaine was instilled in the nasal cavity. A 2.5 mm or 4 mm diameter flexible scope was advanced through the nasal cavity. During the flexible laryngoscopy, the nasal cavity, nasopharynx, oropharynx, hypopharynx, and larynx were assessed. Vocal fold mobility was evaluated and the presence and degree of laryngomalacia was assessed. The patients were then fed an age appropriate diet across various consistencies. For most infants, formula or breast milk was given via a bottle. However, there were a few instances in which infants were breast fed only and would not take a bottle; in these cases, FEES examinations were performed during breast feeding. For the FEES studies, the presence of aspiration, penetration, and premature spillage across various consistencies was noted.

Data was analyzed with IBM SPSS (IBM Corporation, Armonk, NY). Comparisons of categorical data between groups were performed with the Chi-square test, Fisher's exact test, or logistic regression as appropriate. Comparisons of continuous variable data were performed with rank sum tests and Kruskal-Wallis tests. Dunn's procedure with a Bonferroni correction was done for multiple comparison testing. For all tests, statistical significance was defined as a P value less than 0.05.

Results

Demographics, Presentation, Symptoms, Comorbidities, and Disease Severity

During the study, 554 patients were seen in the Aerodigestive Center with the ICD-9 diagnosis code of 748.3 (other anomalies of larynx, trachea, and bronchus). Of these patients, 324 had laryngomalacia. 134 patients were female (41.4%) and 190 were male (58.6%). The median age at first development of symptoms was 0.0 months (SD 3.0 months; range 0-37 months). The median age at diagnosis of laryngomalacia was 3.0 months (SD 11.9 months; range 0-115 months).

Of the 324 patients in the study, 268 patients (82.7%) were born full term (≥ 37 weeks gestation) and 56 patients (17.3%) were born prematurely (< 37 weeks gestation). The median gestational age was 40.0 weeks (SD 3.0 weeks; range 24-41 weeks). Forty-nine patients (15.1%) had a history of a NICU admission at birth and for these patients, the median length of stay was 18.0 days (SD 22.4 days; range 1-120 days). Presenting

symptoms included stridor (n=301, 92.9%), apnea (N=61, 18.8%), cyanotic episodes (n=42, 13.0%), and retractions (n=58, 17.9%).

Severity of laryngomalacia was categorized in 279 patients, and of these patients, 175 (62.7%) were mild, 80 (28.7%) were moderate, and 24 (8.6%) were severe. Severity of laryngomalacia was not reported in 45 subjects. Table I summarizes demographics, medical comorbidities, and symptoms in patients with mild, moderate, and severe laryngomalacia. Severity of laryngomalacia was not affected by gender, age at onset of symptoms, or medical comorbidities. We did find that patients with severe laryngomalacia had a significantly younger age at diagnosis compared with patients with moderate laryngomalacia ($P=0.02$). Patients with more severe laryngomalacia had a significantly greater prevalence of some symptoms, including apnea, cyanosis, failure to thrive, and retractions. There was no significant difference between males and females with respect to age at onset of symptoms, age at diagnosis, medical comorbidities, or presenting symptoms.

Most patients with laryngomalacia were also diagnosed with GERD (n=226, 69.8%). Medical comorbidities also included Down syndrome (n=10, 3.1%), neurologic impairment (n=21, 6.5%), and congenital heart disease (n=3, 0.9%). When combining moderate and severe disease, patients in these groups were more likely to be diagnosed with GERD (82/104, 78.8%) than patients with mild disease (118/175, 67.4%) ($P=0.04$). There was no significant difference in the prevalence of any other medical comorbidities (other than GERD) based on severity of laryngomalacia.

Prevalence of Dysphagia

Symptoms of dysphagia or feeding difficulty were present in 163/324 patients (50.3%) and failure to thrive or poor weight gain was present in 31/324 patients (9.6%).

Swallowing evaluations included CSE in 53 patients (16.3%), MBS in 72 patients (22.2%), and FEES examinations in 130 patients (40.1%). There was a statistically significant association between the presence of subjective feeding difficulty or dysphagia and the presence of at least one abnormal pre-treatment swallowing study (CSE, MBS, or FEES) ($P=0.04$).

Table II compares the CSE, MBS, and FEES results in patients who presented with subjective dysphagia with those who presented without subjective dysphagia. There were no statistically significant differences in the rates of abnormal individual studies for all three of these evaluations between these two groups. However, when combining all swallowing studies, including CSE, MBS, and FEES, more patients presenting with subjective dysphagia had at least one abnormal swallowing assessment (97/120, 80.8%) compared with patients presenting without subjective dysphagia (43/65, 66.2%) (Table II, $P=0.03$). A total of 140/185 (75.7%) patients in the study had the presence of aspiration, penetration, premature spillage, or other signs of abnormal swallowing function on at least one pre-treatment study.

There was no independent effect of age at onset of symptoms, age at time of diagnosis, gender, or gestational age on swallowing function.

Swallowing Function and Disease Severity

There was no significant difference in the subjective report of feeding difficulty or dysphagia, nor in the results of objective swallowing tests, based on severity of laryngomalacia (Table I, $P=0.26$). When swallowing studies were combined (including CSE, MBS, and FEES), at least one abnormal study was found in 78/102 patients (76.5%) with mild laryngomalacia, 34/45 patients (75.6%) with moderate laryngomalacia, and 12/18 patients (66.7%) with severe laryngomalacia (Table III).

There was also no significant difference in the proportion of patients with stridor between the three severity groups ($P=0.36$). However, patients with more severe disease were more likely to have failure to thrive (Table I, $P=0.002$).

Swallowing Function and Comorbidities

There was no significant difference in subjective swallowing dysfunction reported in patients with or without Down syndrome ($P=0.50$), neurologic impairment ($P=0.34$), or congenital heart disease ($P=0.51$) (Table 1). There was also no significant difference in percentage of abnormal MBS or FEES exams (aspiration, penetration, or premature spillage) in patients with or without Down syndrome ($P=0.48$), neurologic impairment ($P=0.35$), or congenital heart disease ($P=0.70$). In addition, there was not a significant difference in the percentage of patients with abnormal CSE assessments in patients with or without Down syndrome ($P=0.45$), neurologic impairment ($P=0.62$), or congenital heart disease ($P=0.55$).

Furthermore, we did not find a significant relationship between the severity of laryngomalacia and the presence of Down syndrome ($P=0.55$), neurologic impairment ($P=0.59$), or congenital heart disease ($P=0.26$) (Table I).

Synchronous Airway Abnormalities

Other synchronous airway abnormalities in addition to laryngomalacia were present in 94 patients (29.0%), and included vocal fold paresis or paralysis (1.8%, $n=6$), type 1 laryngeal cleft (4.9%, $n=16$), tracheomalacia (3.1%, $n=10$), and subglottic stenosis (17.6%, $n=57$). Of the patients with subglottic stenosis, 52 (91.2%) were grade 1, four (7.0 %) were grade 2, and one (2.8 %) was grade 3. Of the six patients with vocal fold paresis or paralysis, three were unilateral and three were bilateral. There was not a significant independent effect of the presence of synchronous airway abnormalities on swallowing function.

Discussion

Patients with laryngomalacia can have difficulty coordinating breathing and swallowing, with resultant dysphagia, feeding difficulties, and aspiration.^{4,6,23,24} Possible etiologies of dysphagia in patients with laryngomalacia include airway obstruction leading to difficulty coordinating sucking, swallowing, and breathing, decreased laryngeal sensation secondary to GERD, and altered sensorimotor integrative function of the larynx.^{4,6} This study evaluated swallowing function in a large series of patients with laryngomalacia, assessing dysphagia based on symptoms, CSE, MBS, and FEES studies.

Our first hypothesis was that dysphagia and feeding difficulties would be common in patients with laryngomalacia. Our study supports this hypothesis, with 50.3% of patients having symptoms of dysphagia or feeding difficulty on initial presentation and 9.6% of patients having failure to thrive or poor weight gain. Thompson also found that feeding problems such as coughing, choking, and regurgitation are common in patients with laryngomalacia, second in prevalence only to stridor. (4)

In addition, in our study, 75.7% of patients had at least one abnormal pretreatment swallowing assessment (CSE, MBS, or FEES). More patients had positive FEES and MBS results than clinical symptoms of dysphagia, suggesting that these tests may have identified some cases of clinically occult aspiration. Alternatively, FEES and MBS may have yielded some false positive results, and there may have been patient selection bias, with tests being performed more commonly in patients with subjective swallowing concerns. FEES studies have previously been shown to be valuable and feasible in evaluating children with dysphagia^{26,27} and to correlate well with MBS study results.²⁸

In this study, since patients were seen in a dedicated Aerodigestive Center where clinical data collection is prioritized, an attempt was made to perform swallowing studies as often as feasible in patients with laryngomalacia. Still, due to selection bias and the retrospective nature of this project, more dysphagia studies were performed in laryngomalacia patients with symptoms of dysphagia than in those without such symptoms. To minimize the effect of this selection bias on the results, we divided the patients with laryngomalacia into two study groups: 1) the 163 patients with symptoms of dysphagia (of whom 120 underwent at least one swallowing study), and 2) the 161 patients without symptoms of dysphagia (of whom 65 underwent at least one swallowing study). The study results were then evaluated separately in each group. The majority of patients in both groups did have at least one positive dysphagia study, although the percentage of having at least one positive study was higher in the symptomatic group (80.8%) compared to the asymptomatic group (60.2%).

Our second hypothesis was that patients with more severe laryngomalacia would be more likely to have swallowing dysfunction, since increased infant airway obstruction is thought to be able to lead to more difficulty coordinating sucking, swallowing, and breathing. In addition, the presence and degree of symptoms including dysphagia play an important role in defining the category of severity of laryngomalacia (mild, moderate, or severe).^{4,7} However, in this study, we did not find a significant effect of severity of laryngomalacia on swallowing dysfunction, based on symptoms and results of CSE, MBS, and FEES studies. We did find that patients with more severe disease were more likely to have failure to thrive.

In contrast to our study, Thompson found that feeding symptoms such as coughing, choking, and regurgitation were more common in patients with moderate and severe disease than in patients with mild disease.⁴ Reasons for our study's inability to demonstrate a correlation between disease severity and dysphagia prevalence may include: 1) possible variation in categorization of disease severity between individual clinicians in the Aerodigestive Center, and 2) incomplete data, with 45 patients (13.9%) not having been assigned a disease severity level. The possibility also exists that the data are correct, with dysphagia prevalence being high even in patients with mild laryngomalacia. Future similar studies from a larger number

of institutions would help clarify the true association between disease severity and laryngomalacia prevalence.

Our final hypothesis was that patients with comorbidities (such as neurological impairment, congenital heart disease, and Down syndrome) would have a greater severity of laryngomalacia and a higher prevalence of swallowing dysfunction compared with patients without these comorbidities, as respiratory problems in children with these disorders have been previously reported, due to factors such as decreased laryngeal tone and poor sensorimotor coordination, leading to apnea, cyanosis, stridor, hypoxia, and increased work of breathing.^{4,6,24,25,29,30,31} However, we did not find a significant relationship between the severity of laryngomalacia and the presence of these comorbidities. In addition, we found that the presence of these comorbidities was not associated with any differences among the percentage of patients with symptoms of dysphagia, nor the percentage of patients with abnormal results on CSE, MBS, or FEES examinations. In our study, we also found that GERD was more likely to be present in patients in the combined moderate and severe disease severity groups than in patients in the mild disease severity group.

Similar to the results of our study, Thompson found that GERD is more common in patients with moderate and severe disease compared with mild disease. (4) However, Thompson also found that neurologic disease and congenital heart disease were more commonly seen in patients with severe laryngomalacia than in those with mild or moderate disease. (4) It is possible that the small numbers of patients with each of these comorbidities in our study did not allow us to detect a significant relationship with disease severity or the presence of swallowing dysfunction.

Most patients in our study had mild disease (62.7%), followed by moderate (28.7%) and severe (8.6%) disease. The presenting symptoms in our patients were similar to those in the literature, including stridor, dysphagia or feeding difficulty, apnea, cyanotic episodes, failure to thrive, and retractions.^{4,7,13,32} GERD was also quite common in our cohort of patients with laryngomalacia. We diagnosed GERD based on symptoms and findings during flexible laryngoscopy or upper aerodigestive tract endoscopy in the operating room, including esophageal biopsies. Only one patient in our study underwent pH probe testing, as this technique is not routinely used at our institution for the diagnosis of reflux in children. The heterogeneity in the diagnosis of GERD in this study may have caused some patients to be incorrectly diagnosed. In the literature, there is also significant heterogeneity in the methods of diagnosis of GERD in patients with laryngomalacia.¹⁰

We found synchronous airway lesions in 29.0% of the patients, which included vocal fold paresis or paralysis, type 1 laryngeal cleft, tracheomalacia, and subglottic stenosis. Many of these synchronous airway lesions were found incidentally during operative airway endoscopy (often at the time of supraglottoplasty) and may not have been clinically significant. The incidence of synchronous airway lesions in the literature ranges from 7.5% to 64%; the large range is likely explained by the methods used to diagnose these secondary airway lesions.^{7,33,34} We did not find a significant effect of the presence of other airway abnormalities (including type 1 laryngeal clefts) on swallowing function.

Given the high prevalence of dysphagia in children with laryngomalacia, we feel that it is important to assess swallowing function in children with this disease. In our Aerodigestive Center, we plan to implement a clinical care pathway, in which caregivers of all patients with laryngomalacia have specific questions asked about swallowing symptoms, a CSE by a speech pathologist, and at least one baseline objective instrumental swallowing assessment (MBS or FEES).

In our paper, we characterized the severity of laryngomalacia according to the criteria provided in the 2007 Triological Thesis *Laryngoscope* publication by Dana Thompson.⁴ This categorization system does allow for some inherent subjectivity, but was the most thorough and well-established system we could find. If there were any inaccuracies in our disease severity categorization, they most likely arose not from using the Thompson system per se, but from us grading disease severity in a retrospective fashion, thus allowing for the possibility that not all data was available for accurate grading to take place. Still, the patient records we reviewed did provide a large amount of data, and we did the best we could, given the retrospective nature of the study, to assign disease severity properly. We are instituting a prospective data collection and disease severity categorization process to help further enhance accuracy in future studies.

Limitations of this study include its retrospective design and the fact that some patients did not have follow up and that there was variability in the length of follow up. Reporting of symptoms may have been biased by the retrospective nature of the study. In addition, not all of the patients had all of the same swallowing assessments (CSE, MBS, and FEES). Another potential limitation is that this study did not specifically evaluate the correlation between CSE, MBS, and FEES results, although previous studies have demonstrated a good correlation between CSE and MBS studies when looking for aspiration or penetration of liquids³⁵ and between FEES and MBS studies.³⁶ Furthermore, the number of patients with medical comorbidities such as neurologic impairment, congenital heart disease, and Down syndrome were relatively small, which could have prevented detecting an association between these comorbidities and disease severity or the presence of swallowing dysfunction.

From the outset, the goal of this paper was to characterize the association between laryngomalacia and dysphagia and to look at the prevalence of dysphagia in patients with laryngomalacia. The next logical issue to address would be the impact of medical and surgical treatment of laryngomalacia on swallowing function. We have been collecting data on this topic and are planning to analyze and present this information in another manuscript. Furthermore, a future prospective study in which all patients with laryngomalacia receive CSE, FEES, and MBS studies before and after treatment and caregivers receive a standardized questionnaire about dysphagia and feeding difficulties will help better elucidate the relationship between laryngomalacia and swallowing function in children.

Conclusion

Swallowing dysfunction is very common in children with laryngomalacia. It is thus important to assess swallowing function in patients with laryngomalacia before and after treatment. Swallowing studies are frequently abnormal in laryngomalacia patients presenting

both with and without subjective symptoms of dysphagia or feeding difficulty. This study did not find a significant effect of severity of laryngomalacia on swallowing dysfunction. We also did not find a significant relationship between the presence of comorbidities such as neurologic impairment, congenital heart disease, and Down syndrome and severity of laryngomalacia or presence of swallowing dysfunction. GERD was the only medical comorbidity that we found to be associated with greater severity of laryngomalacia. Future prospective studies are needed to further elucidate the relationship between treatment and swallowing function in patients with laryngomalacia.

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References

1. Cotton RT, Richardson MA. Congenital laryngeal anomalies. *Otolaryngol Clin North Am.* 1982; 14:203–218.
2. Holinger LD. Etiology of stridor in the neonate, infant and child. *Ann Otol Rhinol Laryngol.* 1980; 89:397–400. [PubMed: 7436240]
3. Holinger P, Johnson KC, Schiller F. Congenital anomalies of the larynx. *Ann Otol Rhinol Laryngol.* 1954; 63:581–606. [PubMed: 13208076]
4. Thompson DM. Abnormal sensorimotor integrative function of the larynx in congenital laryngomalacia: A new theory of etiology. *Laryngoscope.* 2007; 117(Suppl. 114):1–33.
5. Holinger P, Brown W. Congenital webs, cysts, laryngoceles, and other anomalies of the larynx. *Ann Otol Rhinol Laryngol.* 1967; 76:744–752. [PubMed: 6059212]
6. Richter GT, Wooten CT, Rutter MJ, Thompson DM. Impact of supraglottoplasty on aspiration in severe laryngomalacia. *Ann Otol Rhinol Laryngol.* 2009; 118:259–266. [PubMed: 19462845]
7. Thompson DM. Laryngomalacia: factors that influence disease severity and outcomes of management. *Curr Opin Otolaryngol Head Neck Surg.* 2010; 18:564–570. [PubMed: 20962644]
8. Giannoni C, Sulek M, Friedman EM, Duncan NO III. Gastroesophageal reflux association with laryngomalacia: a prospective study. *Int J Pediatr Otorhinolaryngol.* 1998; 43:11–20. [PubMed: 9596365]
9. Halstead LA. Role of gastroesophageal reflux in pediatric upper airway disorders. *Otolaryngol Head Neck Surg.* 1999; 120:208–214. [PubMed: 9949354]
10. Hartl TT, Chadha NK. A systematic review of laryngomalacia and acid reflux. *Otolaryngol Head Neck Surg.* 2012; 147:619–626. [PubMed: 22745201]
11. Matthews BL, Little JP, McGuirt WF, Koufman JA. Reflux in infants with laryngomalacia: results of 24-hour double-probe pH monitoring. *Otolaryngol Head Neck Surg.* 1999; 120:860–864. [PubMed: 10352440]
12. Yellon RF, Goldberg H. Update on gastroesophageal reflux disease in pediatric airway disorders. *Am J Med.* 2001; 111(Suppl 8A):78S–84S. [PubMed: 11749930]
13. Olney DR, Greinwald HJ Jr, Smith RJ, Bauman NM. Laryngomalacia and its treatment. *Laryngoscope.* 1999; 109:1770–1175. [PubMed: 10569405]
14. Shah UK, Wetmore RF. Laryngomalacia: a proposed classification form. *Int J Pediatr Otorhinolaryngol.* 1998; 46:21–26. [PubMed: 10190701]
15. Holinger LD, Konior RJ. Surgical management of severe laryngomalacia. *Laryngoscope.* 1989; 99:136–142. [PubMed: 2913424]

16. Senders CW, Navarrete EG. Laser supraglottoplasty for laryngomalacia: are specific anatomical defects more influential than associated anomalies on outcome? *Int J Pediatr Otorhinolaryngol*. 2001; 57:235–244. [PubMed: 11223456]
17. Hill FT. Congenital laryngeal stridor. *Laryngoscope*. 1930; 40:44–64.
18. McSwiney PF, Cavanagh NP, Languth P. Outcome in congenital stridor (laryngomalacia). *Arch Dis Child*. 1977; 52:215–218. [PubMed: 849000]
19. Sutherland GA, Lack HL. Congenital laryngeal obstruction. *Lancet*. 1897; 2:653–655.
20. Kelemen G. Congenital laryngeal stridor. *Arch Otolaryngol*. 1953; 58:245–268.
21. Lane RW, Weider DJ, Steinem C, Marin-Padilla M. Laryngomalacia: a review and case report of surgical treatment with resolution of pectus excavatum. *Arch Otolaryngol*. 1984; 110:546–551. [PubMed: 6743107]
22. Belmont JR, Grundfast K. Congenital laryngeal stridor (laryngomalacia): etiological factors and associated disorders. *Ann Otol Rhinol Laryngol*. 1984; 93:430–437. [PubMed: 6497235]
23. Rastatter JC, Schroeder JW, Hoff SR, Holinger LD. Aspiration before and after supraglottoplasty regardless of technique. *Int J Otolaryngol*. 2010;91284.
24. Schroeder JW, Thakkar KH, Poznanovic SA, Holinger LD. Aspiration following CO₂ laser-assisted supraglottoplasty. *Int J Pediatr Otorhinolaryngol*. 2008; 72:985–990. [PubMed: 18448173]
25. Hoff SR, Schroeder JW, Rastatter JC, Holinger LD. Supraglottoplasty outcomes in relation to age and comorbid conditions. *Int J Pediatr Otorhinolaryngol*. 2010; 74:245–249. [PubMed: 20022388]
26. Richter GT, Thompson DM. The surgical management of laryngomalacia. *Otolaryngol Clin North Am*. 2008; 41:837–864. [PubMed: 18775337]
27. Hartnick CJ, Hartley BE, Miller C, Willging JP. Pediatric fiberoptic endoscopic evaluation of swallowing. *Ann Otol Rhinol Laryngol*. 2000; 109:996–999. [PubMed: 11089988]
28. Willging JP. Endoscopic evaluation of swallowing in children. *Int J Pediatr Otorhinolaryngol*. 1995; 32(Suppl):S107–S108.28.
29. Eustaquio M, Lee EN, Digoy GP. Feeding outcomes in infants after supraglottoplasty. *Otolaryngol Head Neck Surg*. 2011; 145:818–822. [PubMed: 21746842]
30. Preciado D, Zalzal G. A systematic review of supraglottoplasty outcomes. *Arch Otolaryngol Head Neck Surg*. 2012; 138:718–721. [PubMed: 22801660]
31. Denoyelle F, Mondain M, Grésillon N, Roger G, Chaudré F, Garabédian EN. Failures and complications of supraglottoplasty in children. *Arch Otolaryngol Head Neck Surg*. 2003; 129:1077–1080. [PubMed: 14568790]
32. Roger G, Denoyelle F, Triglia JM, Garabédian EN. Severe laryngomalacia: surgical indications and results in 115 patients. *Laryngoscope*. 1995; 105:1111–1117. [PubMed: 7564844]
33. Dickson JM, Richter GT, Meinzen-Derr J, et al. Secondary airway lesions in infants with laryngomalacia. *Ann Otol Rhinol Laryngol*. 2009; 118:37–43. [PubMed: 19244962]
34. Schroeder JW, Bhandarkar ND, Holinger LD. Synchronous airway lesions and outcomes in infants with severe laryngomalacia requiring supraglottoplasty. *Arch Otolaryngol Head Neck Surg*. 2009; 135:647–651. [PubMed: 19620584]
35. DeMatteo C, Matovich D, Hjartarson A. Comparison of clinical and videofluoroscopic evaluation of children with feeding and swallowing difficulties. *Dev Med Child Neurol*. 2005; 47:149–157. [PubMed: 15739718]
36. Leder SB, Karas DE. Fiberoptic endoscopic evaluation of swallowing in the pediatric population. *Laryngoscope*. 2000; 110:1132–1136. [PubMed: 10892683]

Table 1
Summary of demographics, medical comorbidities, and symptoms in patients with mild, moderate, and severe laryngomalacia

	Mild N=175	Moderate N=80	Severe N=24	Total N=324	Significance
Median age in months at time symptoms began	0.0 (sd 2.6, range 0-19)	0.0 (sd 4.5, range 0-37)	0.0 (sd 0.9, range 0-4)	0.0 (sd 3.0, range 0-37)	P=0.29
Median age in months at time of diagnosis	3.0 (sd 11.7 range 0-110)	3.5 (sd 9.7, range 0-57)	2.0 (sd 5.1 range 0-21)	3.0 (sd 11.9, range 0-115)	P=0.01
Gender:					
Male	103 (58.9%)	47 (58.8%)	10 (41.7%)	190 (58.6%)	P=0.27
Female	72 (41.1%)	33 (41.2%)	14 (58.3%)	134 (41.4%)	
Comorbidities:					
None (other than GERD)	108 (61.7%)	53 (66.2%)	18 (75.0%)	210 (64.8%)	P=0.40
GERD/Reflux	118 (67.4%)	61 (76.2%)	21 (87.5%)	226 (69.8%)	P=0.07 [†]
Down Syndrome	5 (2.8%)	4 (5.0%)	0 (0%)	10 (3.1%)	P=0.55
Neurologic Impairment	9 (5.1%)	7 (8.8%)	1 (4.2%)	21 (6.5%)	P=0.59
Congenital Heart Disease	2 (1.1%)	0 (0%)	1 (4.2%)	3 (0.9%)	P=0.26
Symptoms:					
Stridor	159 (90.8%)	75 (93.8%)	24 (100.0%)	301 (92.9%)	P=0.36
Feeding difficulty/dysphagia	86 (49.1%)	39 (48.8%)	16 (66.7%)	163 (50.3%)	P=0.26
Apnea	24 (13.7%)	19 (23.8%)	7 (29.2%)	61 (18.8%)	P=0.05 [*]
Cyanosis	16 (9.1%)	11 (13.8%)	6 (25.0%)	42 (13.0%)	P=0.07 [#]
Failure to thrive	8 (4.6%)	12 (15.0%)	5 (20.8%)	31 (9.6%)	P=0.002 [@]
Retractions	22 (12.6%)	20 (25.0%)	10 (41.7%)	58 (17.9%)	P=0.001 [^]

Note that the severity of laryngomalacia was not reported in 45 subjects. Therefore, the numbers in the mild, moderate, and severe columns do not sum up to the total numbers for gender, comorbidities, and symptoms.

~ significant for severe versus moderate groups (P=0.02)

[†] significant for mild versus combined moderate/severe groups (P=0.04)

^{*} significant for mild versus moderate groups (P=0.04), mild versus severe groups (P=0.05), but not moderate versus severe groups (P=0.59)

[#] significant for mild versus severe (P=0.02) and combined mild/moderate versus severe (0.037), but not mild versus moderate (P=0.74) or moderate versus severe (P=0.19)

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significant for mild versus moderate ($P=0.004$), mild versus severe ($P=0.01$), and combined mild/moderate versus severe ($P=0.005$), but not moderate versus severe ($P=0.53$)
 significant for mild versus moderate ($P=0.014$), mild versus severe ($P=0.001$), and combined mild/moderate versus severe ($P=0.003$), but not moderate versus severe ($P=0.114$)

Table II
Comparison of Swallowing Study Results in Patients With and Without Subjective Dysphagia

Type of Swallowing Assessment	Subjective Dysphagia N=163	No Subjective Dysphagia N=161	Total N=324	Significance
Clinical Swallowing Evaluation (CSE)	20/31 (64.5%)	9/22 (40.9%)	29/53 (54.7%)	P=0.10
Modified Barium Swallow Study (MBS)	38/54 (70.4%)	11/18 (61.1%)	49/72 (68.1%)	P=0.56
Fiberoptic Endoscopic Evaluation of Swallowing (FEES)	60/82 (73.2%)	32/48 (66.7%)	92/130 (70.8%)	P=0.43
At Least One Abnormal Swallowing Assessment	97/120 (80.8%)	43/65 (66.2%)	140/185 (75.7%)	P=0.03

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Table III
Summary of Swallowing Dysfunction based on Severity of Laryngomalacia

	Mild	Moderate	Severe	Total	Significance
Any aspiration or penetration noted (FEES or MBS)	34/97 (35.1%)	19/44 (43.2%)	10/18 (55.6%)	69/179 (38.5%)	P=0.27
Any aspiration, penetration, or premature spillage (FEES or MBS)	71/97 (73.2%)	28/44 (63.6%)	12/18 (66.7%)	126/179 (70.4%)	P=0.41
Any aspiration, penetration, or premature spillage (FEES, MBS, or CSE)	78/102 (76.5%)	34/45 (75.6%)	12/18 (66.7%)	140/185 (75.7%)	P=0.62

Note that the severity of laryngomalacia was not reported in 45 subjects. Therefore, the numbers in the mild, moderate, and severe columns do not sum up to the total numbers for the various swallowing assessment results.