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Prevalence and Risk Factors for Swallowing Dysfunction in Patients With Laryngomalacia Treated Medically and Surgically

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Abstract

Objective. To identify the prevalence, pattern, and nature of swallowing dysfunction (SwD) in a consecutive cohort of patients with laryngomalacia (LM), and to determine factors associated with a higher burden of SwD.

Study Design. This was a retrospective review of consecutive patients diagnosed with LM by 2 pediatric otolaryngologists between 2013 and 2022 and a minimum of 3-month follow-up.

Setting. Tertiary care pediatric otolaryngology referral center.

Methods. Consecutive cohort of patients less than 3 years old with LM diagnosed on flexible laryngoscopy were reviewed. Patients with incomplete follow-up, lack of swallowing assessment, and genetic conditions or syndromes were excluded. All patients underwent at minimum a systematic clinical swallowing evaluation by a speech-language pathologist specialized in pediatric dysphagia. Patients with concerning clinical exams underwent instrumental swallow evaluation (Videofluoroscopic Swallow Study [VFSS] or Flexible Endoscopic Evaluation of Swallowing [FEES]). The prevalence of abnormalities of clinical swallowing evaluation, instrumental swallow evaluation data, and details of management were collected.

Results. Two hundred and twelve patients met criteria and were included in the final analysis. One hundred and fifteen patients (54%) had an instrumental assessment (VFSS or FEES). Of the instrumental assessments performed, 96 (69%) were abnormal. Of the total patient cohort, 55 (26%) had laryngeal penetration and/or aspiration. One hundred and seventeen (55%) had clinical or instrumental indications for intervention, with 18 (8%) requiring tube feeding. Patients with severe LM and those treated surgically had a statistically significant higher rate of penetration and aspiration.

Conclusion. Patients with LM have a high burden of dysphagia requiring medical intervention. The authors advocate for routine and systematic assessment of all patients with LM for swallowing dysfunction.

Keywords

dysphagia, fiberoptic endoscopic evaluation of swallowing, laryngomalacia, pediatric, swallowing dysfunction, videofluoroscopic swallowing study

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Laryngomalacia (LM) is one of the most common diagnosed pathologies of the upper airway in infants and children. Supraglottic collapse on inspiration often leads to the primary features of the condition—stridor and work of breathing.[1,2] While considerable research has focused on the respiratory presentations of LM, recently studies have started to report on feeding difficulties and swallowing dysfunction (SwD) experienced by patients with LM. Feeding and swallowing difficulties may present as choking, gagging, coughing, emesis, regurgitation, and inefficient feeding. SwD can occur in patients with LM due to several factors including impaired suck-swallow-breathe coordination,

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negative supraglottic pressure promoting aspiration, or abnormal sensorimotor integrative function.[2-4] To evaluate swallow physiology in these children, both clinical and instrumental assessments are utilized. Instrumental assessments, including Videofluoroscopic Swallow Study (VFSS) and Flexible Endoscopic Evaluation of Swallowing (FEES), allow for real-time visualization of the swallowing process and assessment of airway protection during swallowing. The prevalence of SwD and aspiration in patients with LM is not well studied. To date, only a minority of studies have reported a systematic use of instrumental assessment for categorizing the severity of swallowing difficulties in children with LM, with more studies focussing on the effects of surgery.[5,6] From a recent systematic review of swallowing dysfunction in children with LM, only 4 studies have attempted to quantify SwD in association with LM while using either FEES, VFSS or both.[5]

The majority of patients with LM will experience spontaneous resolution of the condition by age 12 to 24 months, while severe cases require surgical intervention. However, as infancy represents a time of critical growth and development, it is important to identify and treat pediatric SwD early in order for children to reach their full neurological and developmental potential.[7] Additionally, the burden of disease and parental quality of life impact of LM may be higher on those who experience SwD.[8] The prevalence, associations, and natural history of SwD are thus not fully understood despite the growing attention this problem is receiving in the literature. The objective of this research work was to define the prevalence, nature, severity, and associations of SwD in a consecutive cohort of patients with LM, and to compare those treated surgically versus nonsurgically. The ultimate goal is to establish a role for systematic swallowing evaluation in patients presenting with LM.

Materials and Methods

This retrospective cohort study was approved by the University of Alberta Health Research Ethics Board prior to study commencement (Pro00100650). A retrospective chart review of pediatric patients diagnosed with LM by 2 pediatric otolaryngologists at a tertiary pediatric referral center between 2013 and 2022 was conducted. The electronic medical records (EMRs) were screened for patients with a visit diagnosis or problem list that included a diagnostic code consistent with LM. The continuity of electronically available data during his period, the fact that all are assessed at a single clinic or inpatient center, and the consistency of EMR use by the practitioners, ensured a consecutive cohort of all patients with an LM diagnosis. Deidentified data were collected, compiled and sorted on an Excel spreadsheet (Microsoft Corp).

Pediatric patients (under 3 years of age) with an endoscopic diagnosis of LM based on flexible nasopharyngoscopy by a staff pediatric otolaryngologist, a clinical swallowing evaluation by a speech-language pathologist (SLP), and a minimum 3-month follow-up, were eligible. In addition to the clinical swallowing evaluation, SwD symptoms were assessed using a previously validated pediatric dysphagia questionnaire.[9] Any patient who exhibited clinical concerns for aspiration based on clinical swallow evaluation and questionnaires underwent an instrumental assessment. We excluded patients with incomplete follow-up, no available objective swallowing data, those who had airway surgery prior to assessment, and genetic or neurological syndromes. Demographic and past medical history were collected including age, gender, clinical severity of LM, comorbidities, prematurity (<36 weeks gestation), twin or triplet status, clinical gastroesophageal reflux disease (GERD), and surgical interventions. LM clinical severity of LM was graded using the Thompson grouping system.[4] We collected symptoms and signs of SwD including choking/coughing, congestion after feeds, apneas, or cyanosis with feeding and recurrent pneumonias.

All instrumental SwD data was collected by a pediatric SLP using either a FEES or a VFSS. The decision between the 2 tests was made based on developmental age and stage, likelihood of patient cooperation, and route of feeding (eg, breast or bottle fed infants). Variables collected from the instrumental swallowing evaluations included swallow onset, laryngeal penetration, aspiration (silent or overt), postswallow residue, and pharyngonasal backflow. The VFSS was performed in the radiology department, with the SLP and a radiologist present. The patients were positioned in an age-appropriate tumble form chair and fed different consistencies of fluids mixed with barium. Swallowing was assessed with fluoroscopy for a short interval and images were captured in the lateral plane, across multiple consistencies as needed. The FEES examinations were performed collaboratively by SLP and a pediatric otolaryngologist.

Decisions regarding treatment (medical or surgical) were made in collaboration between the treating surgeon, SLP, and shared decision-making with the patient's family and/or caregivers, taking into account clinical severity and quality of life impact.

The data was analyzed using Statistical Package for the Social Sciences 23. Descriptive statistics were used with frequencies and percentages for categorical data and with mean/median and standard deviations for continuous data. The primary outcome variable was the prevalence of abnormalities (deep penetration or aspiration) on instrumental swallow assessment. The association between SwD measures and various predictor variables was assessed using univariate analysis (χ^2 or Fischer's exact for categorical variables, Pearson's correlation for continuous variables). The association between LM severity by Thompson group and SwD was assessed using Fisher's Exact for a 3×2 contingency table. Patients were then divided into surgical and nonsurgical cohorts. The prevalence of various SwD measures was compared among surgical and nonsurgical

groups using Fischer's exact or χ^2 tests. The level of statistical significance was set at $P < .05$.

Results

A total of 273 patients with LM were eligible for inclusion. Of these, 61 were excluded for various reasons including age (22), incomplete follow-up information (24), and major genetic or neurological diagnoses (15). In total 212 patients met the inclusion criteria and were included in the final analysis. The patient demographics and comorbidities are summarized in **Table 1**. The median age was 4 months, with a slight male predominance of 1.3:1. The mean follow-up was 8 months, and the majority of patients (117, 55%) had mild disease (Thompson Group 1). The most common caregiver-reported symptom was choking with liquids, which was present in 58% of patients. 10% of patients presented with recurrent pneumonias and 15% reported apneas or cyanosis with feeding (**Table 2**).

Following the clinical evaluation, 115 patients (54%) had an instrumental swallowing assessment: 63 (30%) had FEES only, 27 (13%) had VFSS only, and 25 (12%) had both FEES and VFSS. In total there were 140 instrumental assessments performed (88 FEES and 52 VFSS). Of the 140 instrumental assessments performed, 96 (69%) were abnormal (**Table 3**). The types of abnormalities seen on instrumental swallow assessment are summarized in **Table 4**. Half of patients had laryngeal penetration whereas one quarter had aspiration, the majority of which was silent. When patients were stratified by Thompson severity grouping, the severe group had a significantly higher prevalence of silent aspiration than milder patients (**Table 4**). When patients were subdivided into surgically treated and nonsurgically treated cohorts, the surgical group had a higher burden of SwD on preoperative instrumental assessment, including a higher prevalence of penetration and aspiration (**Table 5**).

When considering all 212 patients as a single cohort, 79 patients (37%) had an abnormal instrumental assessment, with 55 patients (26%) showing laryngeal penetration or aspiration.

Details of SwD management are summarized in **Table 6**. Nearly half of patients had abnormalities requiring intervention, including feeding modification (changes to rate, position, type of feeding) and/or thickening of feeds. 8% required tube feeding. No associations were found between SwD measures on instrumental assessment and age, GERD, prematurity, or other comorbidities.

Table 1. Patient Demographics and Comorbidities

Variable	N (total = 212)
Mean age in months (SD; range)	6 (6; 0-36)
Gender (male:female)	1.3:1
Mean follow-up in months (SD; range)	8 (4; 3-24)
GERD (N, %)	99 (47)
Prematurity (N, %)	19 (9)
Previous intubation (N, %)	13 (6)
Failure to thrive (N, %)	18 (8)
Secondary airway lesion (N, %)	21 (10)
Other comorbidities (N, %)	39 (18)
Thompson severity score (I:II:III)	117:83:12

Table 2. Summary of SwD Symptoms in Patient Cohort

Symptom/sign	N (%); total = 212
Choking with liquids	123 (58)
Choking with solids	42 (20)
Recurrent pneumonia/chest infection	21 (10)
Cyanosis with feeding	18 (8)
Apnea with feeding	23 (11)
Stridor primarily with feeding	136 (64)

Abbreviation: SwD, swallowing dysfunction.

Table 3. Summary of Swallow Assessment Abnormalities

Type of study	Normal (%)	Abnormal (%)
Clinical swallow assessment (n = 212)	97 (46)	115 (54)
FEES (n = 88)	36 (41)	52 (59)
VFSS (n = 52)	8 (15)	44 (85)

Abbreviations: FEES, Flexible Endoscopic Evaluation of Swallowing; VFSS, Videofluoroscopic Swallow Study.

Discussion

There is a growing body of literature that aims to establish and further clarify the association between LM and SwD. The findings of the current study help to demonstrate the prevalence of SwD in patients with LM as well as to clarify certain correlations regarding SwD in LM patients that will help to guide clinicians in the diagnostic workup and risk stratification of these patients.

Our results showed a high burden of symptoms as well as a high prevalence of objective signs of SwD in patients with LM. This is in keeping with other literature that examined a similar cohort of patients. Simons et al found that 80% of patients with LM and SwD symptoms had an abnormal instrumental swallow assessment.[3] Similarly Scott et al found that 86% of their cohort presented with SwD, with 57% demonstrating penetration or aspiration on VFSS, however, their cohort had a high proportion (one third) with genetic or neuromuscular comorbidities.[10] In 2019, Irace et al found that 90% of patients presenting with LM and SwD symptoms had abnormalities on VFSS, with 42% having aspiration.[2] The current cohort adds weight to the notion that LM is partially a disease of swallowing physiology and that the 2 are intimately related.

In the current study, there was interestingly a lack of association between GERD and SwD, despite some studies

Table 4. Instrumental Swallow Evaluation Findings According to Laryngomalacia Severity

		Thompson severity score			
Instrumental finding	All instrumentals (n = 115)	I (n = 117)	II (n = 83)	III (n = 12)	*P* value
Laryngeal penetration	59 (50%)	16 (14%)	39 (47%)	4 (33%)	<.001
Nonsilent aspiration	10 (9%)	5 (4%)	3 (4%)	2 (17%)	.158
Silent aspiration	17 (15%)	2 (2%)	9 (11%)	6 (50%)	<.001

Table 5. Instrumental Swallow Evaluation Findings in Surgical and Nonsurgical Cohorts

Instrumental finding	All instrumentals (n = 115)	Surgical (n = 98)	Nonsurgical (n = 114)	*P* value
Laryngeal penetration	59 (50%)	40 (69%)	18 (16%)	<.001
Nonsilent aspiration	10 (9%)	8 (8%)	2 (2%)	.046
Silent aspiration	17 (15%)	14 (14%)	3 (3%)	.002

Table 6. Summary of Management of SwD in Surgical and Nonsurgical Cohorts

Management	All patients (n = 212)	Surgical (n = 98)	Nonsurgical (n = 114)	*P* value
Feeding modification	85 (40%)	45 (46%)	40 (35%)	.086
Thickened feeds	14 (7%)	9 (9%)	5 (4%)	.150
Tube feeding/alternate route	18 (8%)	12 (12%)	6 (5%)	.063

Abbreviation: SwD, swallowing dysfunction.

showing that treating GERD and its symptoms can help improve swallowing and aspiration.[11] Indeed other authors have failed to show an association with SwD and GERD in infants with LM. There may be a number of reasons for this, such as unclear criteria for diagnoses of GERD. Reflux symptoms may also be caused by a different mechanism when it coexists with LM such as secondary to negative supraglottic inspiratory pressure. This may, in fact, shed some light onto the lack of evidence to support the efficacy of proton pump inhibitors in improving the symptoms of LM, and the wide variation in practice around their use.[12]

Our results also did not show an association between SwD and prematurity among infants with LM, in contrast with De Moreno and Matt as well as Irace et al.[2,13] This may be due a difference in patient populations, as De Moreno studied only patients who underwent supraglottoplasty, and Irace only included data on patients who had clinical SwD symptoms in addition to LM.

The relation between LM severity and SwD is interesting, given that most scales that aimed to grade LM severity such as the one devised by Thompson, include swallowing symptoms and their severity as part of the method of classification.[4] Despite this, other studies have failed to demonstrate a clear association between severe LM and SwD.[3,8] In addition to the lack of clear and strict criteria of what constitutes "severe," this may also be a product of low numbers, as most studies have a small proportion of patients that fall into this category. Nevertheless, the current study did find an association between LM severity and abnormal instrumental assessment, supporting the notion that patients with more severe LM are more likely to have SwD. Moreover, a sizeable number of patients who fell in to the "mild" category had objective signs of SwD, thus strengthening the case for a formal assessment or at least screening of swallowing in all patients with LM.

Our cohort demonstrated a higher burden of SwD in patients who were managed surgically. Although some of this is explained by the fact that surgeons may consider SwD as part of their surgical decision-making, the literature surrounding SwD and supraglottoplasty is far from clear-cut. Whereas some authors report an almost entirely positive effect of supraglottoplasty on swallowing,[6,14] others have shown a high burden of aspiration both before and after supraglottoplasty,[15,16] and others demonstrated relatively high rates of SwD in otherwise healthy infants after supraglottoplasty.[17] Of course many of these studies suffer from either small numbers, lack of a control group, and the fact that more severely affected infants who are more likely to aspirate, are also more likely to undergo surgery. The literature regarding the use of SwD as a surgical decision-making factor in LM is still evolving.

This study had important limitations, namely its retrospective design, single center experience, and under-representation of certain subgroups, as well as the fact that not every patient underwent an instrumental swallowing assessment. Due to the availability of data, an independent review of FESS and VFSS findings was not possible and the authors relied on reports and clinic notes from the instrumental assessments. The selection criteria and decision-making regarding treatment (medical or surgical) may have also been biased by SwD data, however, this is inherent in the retrospective design. Comparing medically and surgically treated patients longitudinally was beyond the scope of the current study. Despite these, the strengths of the study include the inclusion of consecutive patients as opposed to a convenience sample, the robust and consistent use of a systematic approach to SwD evaluation, and the high rate of complete follow-up information.

Conclusion

Patients with LM have a high burden of SwD which is clinically significant. Patients with more severe LM and those treated surgically may have a higher risk of SwD and abnormal instrumental assessment. The authors advocate for a systematic approach to SwD evaluation and workup in patients with LM, with a low threshold for instrumental assessment. The role of SwD in surgical decision-making requires further research and evaluation.

Author Contributions

Janelle Sloychuk, performed data collection, helped analyze the data, wrote the first draft of the manuscript, and edited and approved the final manuscript; **Amy Callaghan**, participated in study design, data collection, helped analyze the data, and edited and approved the final manuscript; **Amanda Adsett**, participated in study design, assisted with data collection, edited and approved the final manuscript; **Daniela M. Isaac**, helped conceive the study, participated in study design, co-supervised data collection, and edited and approved the final manuscript; **Hamdy El-Hakim**, helped conceive the study, participated in study design, and edited and approved the final manuscript; **Andre Isaac**, principal investigator in study conception and design, co-supervised data analysis, guided initial drafting of the manuscript, and edited and approved the final manuscript.

Disclosures

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References

1. Cooper T, Benoit M, Erickson B, El-Hakim H. Primary presentations of laryngomalacia. *JAMA Otolaryngol Head Neck Surg*. 2014;140:521-526. doi:10.1001/jamaoto.2014.626
2. Irace AL, Dombrowski ND, Kawai K, et al. Evaluation of aspiration in infants with laryngomalacia and recurrent respiratory and feeding difficulties. *JAMA Otolaryngol Head Neck Surg*. 2019;145(2):146-151. doi:10.1001/JAMAOTO.2018.3642
3. Simons JP, Greenberg LL, Mehta DK, Fabio A, Maguire RC, Mandell DL. Laryngomalacia and swallowing function in children. *Laryngoscope*. 2015;126(February):478-484. doi:10.1002/lary.25440
4. Thompson DM. Abnormal sensorimotor integrative function of the larynx in congenital laryngomalacia: a new theory of etiology. *Laryngoscope*. 2007;117:1-33. doi:10.1097/MLG.0b013e31804a5750
5. Jaffal H, Isaac A, Johannsen W, Campbell S, El-Hakim HG. The prevalence of swallowing dysfunction in children with laryngomalacia: a systematic review. *Int J Pediatr Otorhinolaryngol*. 2020;139:110464. doi:10.1016/j.ijporl.2020.110464
6. Rossoni EP, Miranda VSG, Barbosa LDR. The prevalence of dysphagia in children with laryngomalacia pre and postsupraglottoplasty: a systematic review with meta-analysis. *Int Arch Otorhinolaryngol*. 2024;28(1):e170-e176. doi:10.1055/s-0042-1755309
7. Dodrill P, Gosa MM. Pediatric dysphagia: physiology, assessment, and management. *Ann Nutr Metab*. 2015;66(suppl 5):24-31. doi:10.1159/000381372
8. Thottam PJ, Simons JP, Choi S, Maguire R, Mehta DK. Clinical relevance of quality of life in laryngomalacia. *Laryngoscope*. 2016;126(5):1232-1235. doi:10.1002/LARY.25491
9. Baqays A, Johannsen W, Rashid M, et al. Parent-reported outcome questionnaire for swallowing dysfunction in healthy infants and toddlers: construction and content validation. *Otolaryngol Head Neck Surg*. 2021;165(1):197-205. doi:10.1177/0194599820970950
10. Scott BL, Lam D, MacArthur C. Laryngomalacia and swallow dysfunction. *Ear Nose Throat J*. 2019;98(10):613-616. doi:10.1177/0145561319847459
11. Suskind DL, Thompson DM, Gulati M, Huddleston P, Liu DC, Baroody FM. Improved infant swallowing after gastroesophageal reflux disease treatment: a function of improved laryngeal sensation? *Laryngoscope*. 2006;116(8):1397-1403. doi:10.1097/01.MLG.0000225942.33102.9B
12. Duncan DR, Larson K, Davidson K, et al. Acid suppression does not improve laryngomalacia outcomes but treatment for oropharyngeal dysphagia might be protective. *J Pediatr*. 2021;238:42-49.e2. doi:10.1016/J.JPEDS.2021.06.051
13. Anderson de Moreno LC, Matt BH. The effects of prematurity on incidence of aspiration following supraglottoplasty for laryngomalacia. *Laryngoscope*. 2014;124(3):777-780. doi:10.1002/lary.21855
14. Richter GT, Wootten CT, Rutter MJ, Thompson DM. Impact of supraglottoplasty on aspiration in severe laryngomalacia. *Ann Otol Rhinol Laryngol*. 2009;118(4):259-266. doi:10.1177/000348940911800404
15. Schroeder JW, Thakkar KH, Poznanovic SA, Holinger LD. Aspiration following CO_2 laser-assisted supraglottoplasty.

Int J Pediatr Otorhinolaryngol. 2008;72(7):985-990. doi:10.1016/j.ijporl.2008.03.007

16. Rastatter JC, Schroeder JW, Hoff SR, Holinger LD. Aspiration before and after supraglottoplasty regardless of technique. *Int J Otolaryngol*. 2010;2010:1-5. doi:10.1155/2010/912814
17. Chun RH, Wittkopf M, Sulman C, Arvedson J. Transient swallowing dysfunction in typically developing children following supraglottoplasty for laryngomalacia. *Int J Pediatr Otorhinolaryngol*. 2014;78(11):1883-1885. doi:10.1016/j.ijporl.2014.08.017