

Clinical Relevance of Quality of Life in Laryngomalacia

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Objectives/Hypothesis: To examine aspects of laryngomalacia and correlate findings with quality of life (QOL).

Study Design: Prospective cross-sectional study.

Methods: Seventy-two patients with laryngomalacia were examined; the mean age was 8.8 weeks. Parents answered questions from the Infant and Toddler Quality of Life Questionnaire-47 (ITQOL-SF47). Fiberoptic laryngoscopy and endoscopic examination of swallowing (FEES) were performed. The presence of laryngomalacia-associated characteristics and swallowing status were recorded. Patient age, sex, presence of reflux, clinical severity, anatomical findings, and swallowing results were evaluated through logistic regression. Independent sample *t* tests were used to compare responses on the ITQOL-SF47. Overall laryngomalacia ITQOL-SF47 scores were compared to the scores of a large healthy sample population.

Results: Forty-three (60%) patients had mild laryngomalacia, and 61 (85%) patients had findings suggesting gastroesophageal reflux disease. The most common abnormality was shortened aryepiglottic folds. Ten patients failed FEES. Patients with moderate laryngomalacia ($\chi^2 = 7.62$; $P = .006$) or prolapsing cuneiforms ($\chi^2 = 4.79$; $P = .029$) were more likely to fail FEES. Laryngomalacia severity impacted parental perception of their child's health ($P < .05$). Parents of children who demonstrated aspiration or penetration reported significant emotional impact (mean = 56.9; $t = 2.74$; $P = .008$). The mean ITQOL-SF47 scores of patients were significantly lower in certain sections than the reported general sample population.

Conclusions: Epiglottal prolapse correlated with severity of laryngomalacia and cuneiform prolapse with swallowing dysfunction. Perceptions of worsening health and physical ability were related to severity of disease. Swallowing dysfunction had a significant emotional impact on parental daily life. Infants with laryngomalacia have a lower QOL.

Key Words: Airway, aspiration, flexible endoscopic evaluation of swallowing, laryngomalacia, pediatrics, swallowing, quality of life.

Level of Evidence: 3

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INTRODUCTION

Laryngomalacia is a common condition of newborns and is the most common cause of stridor in infants.¹ Laryngomalacia-associated stridor is often described as high pitched and frequently occurs during the inspiratory phase of respiration, during which the lack of neuromuscular or structural support allows the supraglottic structures to prolapse.^{1,2} The majority of laryngomalacia cases are self-limited and resolve spontaneously by the age of 12 to 24 months, but an estimated 10% to 30% are severe enough to require surgical intervention.¹⁻⁴

Disease severity is generally classified as mild, moderate, or severe based on associated feeding and obstructive symptoms.² Mild disease is described as inconsequential intermittent stridor and sporadic feeding difficulties, and moderate disease presents with dyspnea and consistent feeding difficulties.^{2,5} Severe

laryngomalacia is present in approximately 20% of infants with stridor and is characterized by recurrent cyanosis, apneic events, and difficulty with feeding, often with associated penetration/aspiration and failure to thrive.^{2,5} Clinical examination is occasionally coupled with fiberoptic anatomical assessment of the larynx and fiberoptic endoscopic evaluations of swallowing (FEES).

It is estimated that 65% to 100% of infants with laryngomalacia also suffer from gastroesophageal reflux disease (GERD).⁴ This association with GERD often results in acid spillage onto the supraglottic structures and propagation of disease through associated edema and desensitization.⁴ Currently, it is recommended that most or all laryngomalacia patients with feeding difficulties be treated for GERD with acid suppression therapy.⁴

Laryngomalacia affects the aerodigestive system and can be a significant factor in early life development for both patient and caregiver perception of patient health. A retrospective study found that 84% of the parents contacted reported anxiety and fear associated with their children's breathing.⁶ In 2014, Kilpatrick et al. prospectively examined 26 families of infants with laryngomalacia and utilized a nonvalidated quality of life (QOL) survey they designed.⁵ Their study demonstrated that patients with more severe disease requiring surgical intervention had higher parental burden of disease.⁵

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TABLE I.
Fiberoptic Laryngoscopy Findings.

Anatomical Finding	Present, No. (%)	Absent, No. (%)
Omega epiglottis	24 (33)	48 (67)
Cuneiform prolapse	42 (58)	30 (42)
Arytenoid prolapse	43 (60)	29 (40)
Epiglottal prolapse	14 (19)	58 (81)
Shortened AE folds	59 (82)	13 (18)
Overly acute epiglottis at laryngeal inlet	12 (17)	60 (83)
GERD changes	61 (85)	11 (15)

Presence or absence of six variances of supraglottic laryngeal anatomy in patients with diagnosed laryngomalacia and GERD changes (arytenoid, posterior glottis, or vocal fold erythema and/or edema).^{7,9,10}

AE = aryepiglottic; GERD = gastroesophageal reflux disease.

Few data exist examining the association of clinical and anatomical findings of laryngomalacia or the effect of laryngomalacia on parental and child QOL. No laryngomalacia studies to date have used validated surveys. The objective of this study was to examine how the severity of laryngomalacia and FEES findings related to each other and to QOL using a validated instrument. We hypothesized that severity of laryngomalacia and certain anatomical findings would correlate with a higher rate of penetration/aspiration on FEES and lower Infant and Toddler Quality of Life Questionnaire-47 (ITQOL-SF47) scores.

MATERIALS AND METHODS

The University of Pittsburgh Institutional Review Board approved the protocol for collection, analysis, and reporting of data prior to the start of this project. Infants and toddlers (1–52 weeks old) were recruited for this prospective cohort study from the Children’s Hospital of Pittsburgh of the University of Pittsburgh Medical Center Aerodigestive Center between July 2013 and January 2015. Subjects enlisted were absent of neurodevelopmental delay and void of medical comorbidities besides prematurity and GERD. All were originally evaluated for respiratory or feeding complaints and noted to have laryngomalacia both clinically and on fiberoptic laryngoscopy by a pediatric otolaryngologist.^{2,4,7} During initial examination, patients’ guardians answered questions from the ITQOL-SF47.⁸ All patients also underwent FEES conducted by a team of an otolaryngologist and a speech–language pathologist (SLP).

Patients were examined and categorized as having mild, moderate, or severe laryngomalacia based on history of presenting illness and physical examination. To ensure accurate and consistent diagnoses, all examiners categorized patients into groups based on findings described by Thompson in 2007.⁷ Group 1 included patients with mild disease and was reserved for those with inspiratory stridor with or without coughing during feeding.⁷ Subjects in group 2 had moderate disease, defined as inspiratory stridor accompanied by dysphagia with or without weight loss, coughing and choking while feeding with regurgitation, gasping events, minor apneic events, brief cyanotic events, and/or shortness of breath with retractions.⁷ Group 2 excluded any patients requiring aggressive medical intervention.⁷ Group 3 was comprised of patients with severe laryngomalacia, characterized by stridor with life-threatening complications such as failure to thrive, cyanosis resulting in

medical intervention, dyspnea with associated retraction requiring medical intervention, severe hypoxia, prolonged apneic pauses, pectus excavatum, pulmonary hypertension, or cor pulmonale.⁷

Flexible fiberoptic laryngoscopy and FEES were performed on all patients, and the presence or absence of six defined laryngomalacia-associated anatomical characteristics were recorded (Table I).⁷ There were an attending pediatric otolaryngologist and an SLP present for the FEES examinations. Patients were defined as failing their FEES examination if penetration or aspiration was observed with either thin or thickened consistencies. All fiberoptic and FEES information was recorded and deidentified.

All patients were evaluated for the presence of GERD through medical history and flexible fiberoptic laryngoscopy findings suggesting disease-associated changes (Table I). All patients diagnosed with GERD demonstrated at least one presenting symptom described by the American Academy of Pediatrics clinical report on gastroesophageal reflux, as well as arytenoid, posterior glottis, or vocal fold erythema and/or edema on endoscopic examination.^{9,10}

Perceived health status and health-related QOL was measured using the ITQOL-SF47.⁸ All guardians were asked to complete the ITQOL-SF47 at the time of their initial visit. The questionnaire was comprised of 47 items in the following areas: overall health, physical abilities, growth and development, pain, temperament and moods, behavior, general health, parental emotional impact, parental time impact, and family cohesion.^{8,11} Once completed, this validated questionnaire was scored and recorded by an independent researcher using the HealthActChq confidential scoring rules.¹¹ For each concept, item responses were scored, summed, and transformed on a scale from 0 (worst health) to 100 (best health).¹¹

Patient age, sex, presence of reflux, clinical severity of laryngomalacia, anatomical findings, and FEES results were examined through logistic regression. Independent samples *t* tests were used to compare responses on the ITQOL-SF47 between guardians of patients with mild and moderate laryngomalacia. Overall laryngomalacia ITQOL-SF47 scores were also compared to the reported scores of a large (*n* = 410) healthy sample population that was utilized in the original validation of the questionnaire.¹² A probability value of <.05 was used to define statistical significance.

RESULTS

Seventy-two patients with laryngomalacia were included in this study. The patients’ mean age was 8.8 weeks (standard deviation = 7.42; range = 1–52 weeks), and 34 (47%) were male. Of the 72 patients, 43 (60%) were diagnosed with mild laryngomalacia and 29 (40%) with moderate disease. No patients demonstrated severe disease.

Sixty-one (85%) of the patients had clinical and physical findings suggesting GERD (Table I). Female patients demonstrated a significantly higher likelihood of having GERD ($\chi^2 = 6.24$; *P* = .013).

Table I includes flexible fiberoptic laryngoscopy examination findings. The most common laryngomalacia abnormality was shortened aryepiglottic folds, which was seen in 59 (82%) of the patients examined. An overly acute epiglottal angle was the least commonly identified abnormality (*n* = 12; 17%). The odds of patients with moderate laryngomalacia having epiglottal prolapse were 5 times higher than those with mild

TABLE II.
Mean ITQOL Scores (Standard Deviation) for General Population (n = 410)¹⁰ and Laryngomalacia Population (n = 72).

ITQOL Scale Scores	General Population ¹⁰	Laryngomalacia	<i>t</i>	<i>P</i>
Physical function	97.1 (9.8)	81.8 (18.2)	5.99	<.001
Growth and development	86.5 (10.6)	85.1 (11.9)	1.01	.31
Bodily pain	83.8 (16.8)	63.4 (24.6)	6.54	<.001
Temperament and moods	77.2 (10.5)	78.0 (15.1)	-0.29	.98
General health perceptions	79.0 (14.5)	72.6 (14.9)	10.58	<.001
Parental emotional impact	92.1 (10.5)	73.7 (20.5)	7.4	<.001
Parental time impact	93.0 (11.0)	88.4 (19.1)	1.93	.60
Family cohesion	75.3 (18.8)	83.8 (20.1)	-3.22	<.001

ITQOL = Infant and Toddler Quality of Life Questionnaire.

laryngomalacia ($\chi^2 = 7.01$; $P = .008$; odds ratio = 5.13; 95% confidence interval = 1.42-18.51).

When FEES was performed, 10 (14%) patients failed the examination, with eight (11%) patients demonstrating penetration and two (3%) patients demonstrating aspiration with thin liquids. Patients with moderate laryngomalacia ($\chi^2 = 7.62$; $P = .006$) or prolapsing cuneiforms ($\chi^2 = 4.79$; $P = .029$) were significantly more likely to have aspiration or penetration on FEES. Among patients who failed the FEES examination, 9 (90%) had prolapsing cuneiforms, compared to only one (10%) of those who did not fail the FEES examination.

Laryngomalacia severity significantly impacted parental perception of their child. Parents of patients with moderate laryngomalacia rated their child's physical abilities (mild mean score = 86.9; moderate mean score = 76.2; $t = 2.19$; $P = .033$) and health (mild mean score = 75.6; moderate mean score = 67.9; $t = 2.19$; $P = .032$) lower when compared to children with mild disease. Parents of children who failed the FEES examination reported higher levels of emotional impact from their child's disease (mean = 56.9) than parents of children who did not fail the FEES examination (mean = 76.2; $t = 2.74$; $P = .008$). When comparing the mean ITQOL-SF47 section scores of subjects in this study to the reported general sample population, statistically significant lower scores were demonstrated in all categories except growth and development, temperament and mood, and parental time impact (Table II).¹²

DISCUSSION

Laryngomalacia is a common disorder that affects the supraglottic structures. An estimated 10% to 20% of these patients present with signs of upper airway obstruction.¹³ There have been numerous studies examining the presentation, cause, associated symptoms, and management of this disorder, but few have examined its influence on the QOL of patients and their families or stratified disease characteristics.^{1,2,5-7} In our study, we prospectively examined laryngomalacia in detail focusing on both clinical and physical diagnosis as well as its effect on swallowing and QOL.

Laryngomalacia symptoms usually begin within 10 days of life and peak at 6 to 8 months.⁴ Although in

some studies males have demonstrated higher disease prevalence, sex has not been shown to influence severity.^{4,14,15} In our current study, patients were diagnosed upon clinical investigation at an average of 8.8 weeks of age and male sex was not shown to predispose children to laryngomalacia or to impact disease severity.

Gastroesophageal reflux was identified in 85% of patients in this study, and females with laryngomalacia demonstrated higher odds of having this comorbidity. Similarly, Thompson reported that GERD was present in 65.7% to 80.0% of patients with laryngomalacia.⁷ Contrary to our findings, previous studies have identified a higher prevalence of GERD in male children (1-18 years of age), but few data exist examining sex and GERD among infants with laryngomalacia.^{16,17}

The association of anatomical findings with clinical severity of disease has been underinvestigated.¹⁸ In 1999, a study described various types of anatomical findings associated with laryngomalacia to direct treatment, but how and whether these findings related to clinical severity was not described.¹⁴ Meanwhile, Manning et al. demonstrated that clinically severe laryngomalacia was associated with shortened aryepiglottic folds.¹⁸ However, they did not examine other aspects of the supraglottic anatomy. All patients in the present study underwent both fiberoptic laryngoscopy and FEES. Shortened aryepiglottic folds were the most common supraglottic anatomical finding in patients with laryngomalacia, and the presence of epiglottal prolapse was associated with more severe clinical disease (Table I). When examining swallowing, clinically moderate laryngomalacia and the anatomical finding of prolapsing cuneiforms were associated with increased odds of aspiration or penetration on FEES. These findings indicate that clinical severity is related to supraglottic obstructive disease. They also support the belief that certain obstructive supraglottic anatomical aspects like redundant and prolapsing overlying cuneiform tissue can further propagate reflux and associated swallowing dysfunction.

Previous studies on the burden of laryngomalacia have been limited, with little focus on clinical severity and associated anatomy. In our current study, we demonstrated that caregivers' perceptions of worsening infant health and physical ability were directly

associated with clinical severity of laryngomalacia using the validated ITQOL-SF47. These findings are similar to those of a recent study that demonstrated worsening perceived general health scores in patients requiring supraglottoplasty compared to those who did not undergo surgery.⁵ Our study also revealed that the emotional impact of this disease on parents is significantly affected by the swallowing status of their child, as parents of infants with failed FEES reported worse scores. The overall impact of laryngomalacia on both the patient and caregiver is further illustrated with the significantly lower ITQOL-SF47 scores demonstrated by our patient population when compared to those of a previously reported general population (Table II).¹²

This study is not without limitations. This study was conducted in an outpatient setting and thus was limited to clinically mild or moderate disease. This is possibly secondary to our institution's tertiary status and thus severe disease being addressed in an inpatient setting. Limiting this study to mild or moderate disease may have also factored into a lower number of patients demonstrating aspiration or penetration of FEES when compared to all laryngomalacia or severe disease. The ITQOL-SF47 is limited, as it relies on parental perception and not actual patient reported or objective information. The time at home with each patient, individual parental stability, and experience with childcare was also not taken into account in this study. In addition, our mean patient ITQOL-SF47 scores were compared to those of a previously reported large general population and not to our own control group.

Despite the noted limitations, this study represents a prospective examination of laryngomalacia as it pertains to clinical and physical findings as well as QOL. By utilizing strict clinical categorization of laryngomalacia severity and specific anatomical findings, we were able to find relationships between certain anatomical abnormalities and severity. Through the utilization of FEES, we were also able to examine how these aspects affected swallowing status. Using a validated survey, we were able to gain insight on how the caregiver perceived laryngomalacia, how this perception correlated with various aspects of this disease, and how it compared to the reported general population.¹²

CONCLUSION

Laryngomalacia is a common disorder that can cause significant airway and feeding problems. Epiglottal prolapse was associated with increased severity of disease, and patients with prolapsing cuneiforms demonstrated a higher rate of swallowing dysfunction.

Perceptions of worsening health and physical ability were directly related to the clinical severity of disease, and caregivers of children who demonstrated aspiration or penetration reported a significant emotional impact on their daily life. When compared to the reported general population, infants with laryngomalacia demonstrated a significantly lower QOL in many aspects examined.

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