International Journal of Pediatric Otorhinolaryngology 78 (2014) 583–587

Contents lists available at ScienceDirect

International Journal of Pediatric Otorhinolaryngology

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

Prospective quality of life assessment in congenital laryngomalacia

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ARTICLE INFO

Article history:
Received 31 October 2013
Received in revised form 3 January 2014
Accepted 4 January 2014
Available online 14 January 2014

Keywords:
Laryngomalacia
Quality of life

ABSTRACT

Purpose of study: Disturbances in breathing or feeding may profoundly affect parental perceptions of a newborn's health. Previous research into quality of life for patients with laryngomalacia is limited to retrospective analysis. The purpose of this study is to prospectively evaluate the quality of life of families of infants with laryngomalacia and the impact of surgical and non-surgical treatments.
Design and method: Pilot prospective analysis using the laryngomalacia quality of life (QOL) survey in families of infants with newly diagnosed laryngomalacia under age one year. A 29-question survey regarding severity of symptoms related to overall health, airway, and swallowing is completed at initial and post-treatment visits. Responses are quantified over a range from 1 to 5 (1 never to 5 always).
Results: Twenty-six families were enrolled in the study. Eleven patients were managed medically and fifteen underwent supraglottoplasty. The overall mean QOL score for patients treated medically was 2.57 (standard error, SE 0.16) on initial visit and 1.67 (SE 0.16) post-treatment (mean 3.9 months). Patients undergoing supraglottoplasty had an overall mean QOL score of 3.59 (SE 0.14) on initial visit and 2.22 (SE 0.22) post-treatment (mean 3.5 months). Analysis of variance (ANOVA) and post hoc testing revealed significant improvement between initial and follow-up visits in both treatment groups ($p < 0.01$). Patients who underwent supraglottoplasty had significantly higher scores at initial visit ($p < 0.01$). No statistically significant difference was noted between patient groups post-treatment ($p > 0.05$).
Conclusions: Prospective QOL assessment of children with laryngomalacia and their families reveals a significant burden of disease. Quality of life improves in all patients but may improve more significantly in patients managed surgically.

1. Introduction

Congenital laryngomalacia is the most common cause of stridor in infants, found in 45–75% of neonates with this presenting symptom [1]. It is clinically defined by inspiratory stridor due to prolapse of supraglottic structures into the airway. Patients initially present within the first weeks of life, and stridor often worsens with feeding, agitation, and supine positioning. While historically attributed to cartilage immaturity or anatomic abnormality, neuromuscular immaturity of the larynx is the most accepted etiologic theory for development of laryngomalacia in the infant [2,3].

Clinical symptoms are used to stratify patients according to disease severity. Mild laryngomalacia is stridor without increased work of breathing and intermittent feeding difficulty. Patients with moderate laryngomalacia exhibit intermittent dyspnea which self-resolves but have more feeding difficulty, including prolonged feeding time, choking, cough, or emesis. While many patients with mild or moderate disease have resolution of symptoms by age twelve to twenty-four months, severe laryngomalacia can result in recurrent cyanosis, hypoxia, apnea, retractions, aspiration, weight loss, and failure to thrive [4,5].

Early intervention for patients with severe laryngomalacia is warranted and typically requires surgical management via supraglottoplasty and acid suppression therapy [4]. Surgery involves a rigid microlaryngoscopy and bronchoscopy (MLB) for full airway evaluation followed by release of the aryepiglottic folds and removal of the redundant supraarytenoid tissue. Supraglottoplasty is successful in over 90% of patients with severe laryngomalacia and has a low complication rate [1].

With its significant impact on both airway and feeding, patients with laryngomalacia often require daily medication, feeding modifications, and close follow-up. Patients progress to surgical intervention if symptoms worsen or cannot be controlled with medical management alone. Given the spectrum of the disease, one may infer that quality of life (QOL) is negatively affected for both patients with laryngomalacia and their caregivers. Milczuk and

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http://dx.doi.org/10.1016/j.ijporl.2014.01.001

Johnson performed a retrospective QOL study for patients with laryngomalacia in 2000. Forty-four patients were involved in the study in which parents answered questions via telephone survey. Interestingly, the majority of survey respondents reported little effect on QOL due to laryngomalacia; however, 84% endorsed frequent feelings of anxiety or fear about their child's breathing. As a retrospective study, recall bias and conflicting results may underestimate the impact of laryngomalacia on QOL for patients and caregivers [6].

This study aims to prospectively evaluate the quality of life of families of infants with laryngomalacia with respect to the diagnosis of laryngomalacia and the impact of medical and surgical management. We hypothesize that QOL is significantly affected by laryngomalacia and that QOL improves significantly after intervention.

2. Methods

Prior to initiation of the study, approval from the Institutional Review Board (IRB) at Arkansas Children's Hospital (ACH) was obtained for the study protocol. Patients were recruited from a tertiary pediatric institution over an eighteen month period from 2011 to 2012. Patients with a new diagnosis of laryngomalacia under age one year were included; patients with complex medical comorbidities including cardiac or neurological disease were excluded. All patients underwent a thorough history and physical examination, including flexible fiberoptic laryngoscopy by an experienced pediatric otolaryngologist to confirm the diagnosis. Patients with significant dysphagia symptoms were also assessed by a speech language pathologist who assisted in management.

The laryngomalacia quality of life survey (Fig. 1) was administered to the patient's primary caregiver at the initial visit, a one month follow-up visit, and a late follow-up visit (minimum 3 months from time of initial visit). Survey questions were created by our team with intent to quantify a caregiver's level of anxiety and the effect on daily function. The newly developed survey tool included 29 questions; 8 questions assess general health, 12 questions assess respiratory status, and 9 questions assess feeding status. Responses are scored over a range of 1–5 (1 – never, 2 – rarely, 3 – sometimes, 4 – often, 5 – always).

Question	Never	Rarely	Sometimes	Often	Always
How often is your child sick?	1	2	3	4	5
How much does your child's health impact your daily life?	1	2	3	4	5
Do you feel your child is different from other children?	1	2	3	4	5
Do you make special arrangements for your child due his/her health condition?	1	2	3	4	5
Does your child's condition negatively impact your spousal or family relationships?	1	2	3	4	5
How often does your child appear excessively fussy or uncomfortable?	1	2	3	4	5
Does your child require more attention and care than other kids of the same age?	1	2	3	4	5
How often do you worry that your child's condition affects his/her normal growth and development?	1	2	3	4	5
	Never	**Rarely**	**Sometimes**	**Often**	**Always**
How often does your child have difficulty breathing?	1	2	3	4	5
How often do you stay up at night to watch your child's breathing?	1	2	3	4	5
How often are you worried about your child's oxygen level?	1	2	3	4	5
Is your child's breathing	1	2	3	4	5

Fig. 1. Laryngomalacia quality of life survey.