Laryngomalacia: Our Clinical Experience

Original Investigation

Yüksel Olgun, Hüseyin Özay, Aslı Çakır, Taner Kemal Erdağ
Department of Otorhinolaryngology, Dokuz Eylül University School of Medicine, İzmir, Turkey

Abstract

Objective: The aim of this study is to analyse the clinical symptoms, follow-up and treatment properties of the laryngomalacia patients that we encountered between 2009 and 2014.

Methods: Records of 81 laryngomalacia patients who were followed up in our clinic between 2009 and 2014 were retrospectively analysed. Patients' gender, age, time of onset of the symptoms, chief complaints, other co-existing congenital laryngeal anomalies and treatment and follow-up properties were evaluated.

Results: Of the 81 patients, 48 were male and 33 were female, and the mean age was 4.9 months. The average period of follow-up was 12.1 months. The chief complaints at the time of admission were stridor (100%) and episodic cyanosis with feeding (27.16%). Symptoms of 75 patients were resolved at an average of 8.2 months with conservative treatment. Three patients underwent supraglottoplasty. Tracheotomy and posterior cordotomy was performed for a patient with co-existing vocal cord paralysis. Additional tracheotomy was necessary for a patient with pulmonary co-morbidities and for another with co-existing subglottic stenosis.

Conclusion: Laryngomalacia is the most common cause of stridor in infants. The majority of laryngomalacia patients can be managed conservatively by close follow-up. For patients in whom respiratory and feeding problems persist or growth retardation develops, surgical treatment is performed. Tracheotomy may be necessary for a small group of patients with additional diseases.

Keywords: Airway, laryngomalacia, supraglottoplasty, stridor

Introduction

Laryngomalacia is the most common congenital laryngeal anomaly during childhood. The diagnosis, treatment, and follow-up processes of laryngomalacia, which is the most frequent cause of stridor in newborns and infants, are important for families and physicians (1-3). Infants with laryngomalacia mostly present with complaints such as wheezing, feeding problems, and coarse voice. In a few cases that are not appropriately treated, important problems such as serious growth retardation, recurrent aspiration pneumonia, pulmonary hypertension, and cor pulmonale can occur (1, 3).

Symptoms develop because of the collapse of the supraglottic structures in the laryngeal lumen during inspirium. Although the cause of this collapse may be attributed to the short aryepiglottic folds, anatomical factors such as the structure of the epiglottis, and easily flexible characteristics of immature cartilage tissues, the widely accepted cause currently is the neurological theory (1-5). It is suggested that decreased laryngeal tonus associated with sensorimotor dysfunction and particularly elevated thresholds of the laryngeal adductor reflex lead to the development of laryngomalacia (2, 6, 7).

This study aimed to present the symptoms, treatments, and follow-ups of patients with laryngomalacia who were undergoing treatment at our clinic for a 6-year period and to discuss the case with respect to the current literature.

Methods

The study was initiated after receiving ethical approval from the noninvasive research ethics board of Dokuz Eylül University (file no, GOA-233 and

This study was presented at the 13th Congress of the European Society of Pediatric Otorhinolaryngology, 18-21 June 2016, Lisbon, Portugal.

Address for Correspondence: Yüksel Olgun
E-mail: yuksel.olgun@deu.edu.tr

Received Date: 14.08.2016
Accepted Date: 25.09.2016

DOI: 10.5152/tao.2016.1852

decision no, 2015/24-17). Medical records of patients who visited the Department of Otorhinolaryngology in Dokuz Eylül University between January 2009 and December 2014 and who were diagnosed with laryngomalacia after a fiberoptic flexible endoscopic examination were retrospectively analyzed. Patients were evaluated in terms of gender, age, symptoms, onset and cessation times of symptoms, accompanying congenital laryngeal anomalies, other coexisting non-laryngeal anomalies or diseases, and durations of follow-up and treatment.

Patients diagnosed with laryngomalacia were asked to visit our outpatient clinic for routine control examinations with fiberoptic flexible endoscopy at 3-month intervals. They were referred to the Department of Pediatric Gastroenterology for evaluation in terms of reflux and growth. The frequency of follow-up was increased in patients who had severe laryngomalacia in the first examination or who had deteriorated respiratory distress during follow-ups and who had apparent growth retardation. These patients were followed up at 1-month intervals. Patients whose minimum follow-up was shorter than 3 months were excluded.

Results

Between January 2009 and December 2014, 88 patients were diagnosed with laryngomalacia at our clinic. We included 81 patients who were followed up for at least 3 months (Table 1).

Laryngeal pathologies along with laryngomalacia were observed in three (3.7%) of 81 patients. While one patient had unilateral vocal cord paralysis and another had bilateral vocal cord paralysis, subglottic stenosis coexisting with laryngomalacia was observed in one patient having a history of prolonged intubation. Moreover, two patients had simultaneous bilateral choanal atresia.

Seventy-five of 81 patients were followed up with recommendations, and it was observed that their symptoms regressed in approximately 8.2 months of follow-up from the first admission. Two patients, who were evaluated in this group and who did not undergo any laryngeal intervention, underwent surgery because of simultaneous bilateral choanal atresia. Supraglottoplasty was performed in three patients who were followed up with a conservative approach but who had growth retardation and progression in their symptoms. The ages of these patients were 1.5, 2, and 14 months, respectively. The common finding of these three patients was short aryepiglottic folds. During surgeries of these patients, after administering muscle relaxants, the subglottic region and trachea were evaluated using a 2.7-cm rigid endoscope without intubation. After ensuring the absence of additional laryngeal anomalies, aryepiglottic folds that were present at the base of the epiglottis were cut using microscissors. No increase in aspiration and respiratory distress was observed in any patient during the postoperative period. Few days after the surgery, a dramatic improvement in symptoms was observed. Treatment for reflux was initiated in the three patients by the Department of Pediatric Gastroenterology before the surgery. These patients continued to receive this treatment for 3 months after the surgery. After follow-up with tracheotomy, unilateral posterior cordotomy was performed in a 2-year-old patient having bilateral vocal cord paralysis and laryngomalacia for a year. In the follow-up of the patient who was decanulated without any problem on the 30th day after the surgery, symptoms were observed to have regressed, and there was no complaint associated with aspiration. Supraglottoplasty was not performed during posterior cordotomy in this patient whose laryngomalacia signs were regressed during the time in which the patient was followed up through tracheotomy. Conversely, tracheotomy was performed in two patients of our patient group. The first was a patient having dextrocardia and pulmonary hypertension along with laryngomalacia. This patient, who continues to need a mechanical ventilator and has underlying pulmonary problems, is still being followed up with tracheotomy. The other patient had subglottic stenosis with laryngomalacia. After performing tracheotomy in this patient, surgery for stenosis was recommended; however, the parents did not accept the recommendation, and the patient was excluded from our follow-up process.

Table 1. Demographic and follow-up features of the patients

Gender	48 males / 33 females
Age at first admission, mean±standard deviation	4.9±3.67 months
Duration of follow-up, mean±standard deviation	12.1±8.95 months
Main complaints at admission	-Inspiratory stridor: 81 patients (100%) - Respiratory distress during feeding, cyanosis attacks: 22 patients (27.16%) - Having recurrent lower respiratory tract infections: two patients (2.46%)

Discussion

Laryngomalacia is the most frequent cause of stridor in newborns and infants and is the underlying reason in 60 to 75% of cases with stridor (3, 8). Its incidence is reported to be 1.6 times higher in boys than in girls (1, 9). The symptoms that start a few weeks after the birth reach its peak in the first 4–8 months, and they are relieved at the age of approximately 1 year and completely disappear in 24 months in most cases (1, 3, 10, 11). Consistent with the literature, we observed that our patients were admitted to our clinic when their symptoms were at the peak level (average, 4.9 months), and the complaints of the patients, who were conservatively followed up, either regressed to a great extent in approximately 8.2 months after the first admission or completely healed.

Gastroesophageal or laryngopharyngeal reflux is the most common comorbidity accompanying laryngomalacia, and its incidence varies between 65% and 100% (1, 3). It has been reported that reflux directly contributes to the development of laryngomalacia, and reflex thresholds of laryngeal adductor increase with the exposure of chemoreceptors in the laryngeal mucosa to chronic acid (2). Moreover, it is considered that inflammation and mucosal edema associated with reflux result in airway

obstruction (12). According to another study, a great negative intrathoracic pressure occurs because of upper airway obstruction in patients with laryngomalacia (11). Reflux-induced feeding problems can develop in a wide range, including cough during feeding, aspiration pneumonias recurring because of regurgitation, and growth and developmental retardation (1, 3). All patients who were diagnosed with laryngomalacia at our clinic were referred to the Department of Pediatric Gastroenterology for monitoring their growth and development and planning their treatments for reflux. The parents of patients were informed about some suggestions such as breastfeeding in the vertical position, thickening the baby formula, and feeding slowly, which would reduce reflux.

All our patients had feeding problems at various levels, but this situation was particularly apparent in 24 patients. Twenty-two patients (27.16%) were followed up for cyanosis attacks that sometimes occurred, and two patients (2.46%) were followed up for recurrent aspiration pneumonias, and their treatments were planned. Supraglottoplasty was performed in three patients whose symptoms did not improve despite conservative precautions and who developed growth retardation. The complaints of other patients were relieved with conservative approaches and medical treatment.

It should be noted that laryngomalacia is the most common congenital laryngeal anomaly during childhood, but there may be other coexisting airway problems. Some studies have reported that other airway pathologies can accompany laryngomalacia at rates varying from 7.5% to 64% (13-19). In particular, tracheomalacia, subglottic stenosis, and vocal cord paralysis are leading pathologies. In our series, comorbid laryngeal anomalies were found in only three patients (3.7%); vocal cord paralysis in two patients and subglottic stenosis in one. This rate is lower than that reported in the literature. This might have resulted from the fact that direct laryngoscopic examination was performed only in patients with serious laryngomalacia symptoms. The diagnoses of some minor airway problems, including mild tracheomalacia or early stage subglottic stenosis, might not have been established because direct laryngoscopy was not performed in every patient. However, the need for direct laryngoscopy even in patients who have mild-to-moderate laryngomalacia and whose symptoms improved with positive response to conservative treatment is controversial (3). Moreover, although the incidence of coexisting airway problems was reported to be high in the literature, Mancuso et al. (13) specified that additional intervention was required only in 4.7% of these patients.

In most of patients, symptoms associated with laryngomalacia regress within the first year with the growth of the child and conservative follow-up treatment, and they disappear in most patients in 2 years. However, surgical treatment is considered for patients at a rate varying between 4.2% and 20% (10, 12). The probability of the need for surgical treatment is stated to be higher in patients with a history of prematurity, early onset of symptoms, and admission to the emergency unit with serious respiratory distress compared with other patients with laryngomalacia (10, 20). Garritano and Carr (12), who performed supraglottoplasty in 17 patients, reported that aryepiglottic folds were short in 16 patients, the situation of the epiglottis was turned toward the posterior, and the need for supraglottoplasty could be high in patients with short aryepiglottic folds.

Thompson (21) stated that severe laryngomalacia could be mentioned in the presence of inspiratory stridor accompanied with cyanosis and apnea, oxygen saturation level of 86% and lower at rest, recurrent aspirations, and growth–developmental retardation. Furthermore, they emphasized that these patients were candidates for surgery. While conservative follow-up was sufficient for 75 (92.59%) of 81 patients with laryngomalacia, supraglottoplasty was performed in three patients (3.7%) because of progressed symptoms during follow-up, development of cyanosis attacks while feeding, and occurrence of growth–developmental retardation.

Tracheotomy is an appropriate treatment choice in patients with no improvement in their symptoms despite supraglottoplasty or with other comorbid airway problems (3). Of our patients, three (3.7%) underwent tracheotomy. In one patient, bilateral vocal cord paralysis with laryngomalacia was the cause. While posterior cordotomy was performed in the patient whose vocal cord paralysis persisted during the 1-year follow-up, decanulation was subsequently possible. Tracheotomy was performed because of coexisting pulmonary disease in one patient and coexisting subglottic stenosis in another patient.

In the national literature review, no original study, except one reporting the results of supraglottoplasty, was found about laryngomalacia (5). Considering this point, our study is one of the first ones in our country on the follow-up and treatment processes in a large series of patients with laryngomalacia.

Conclusion

Laryngomalacia is the most common cause of stridor in the neonatal period and during infancy. All patients should be closely followed up in terms of both the severity of symptoms associated with reflux and respiration and growth. Conservative approaches and close follow-up are adequate in most patients. Patients with increased respiratory symptoms and developed growth retardation during follow-up are appropriate candidates for supraglottoplasty. Conversely, tracheotomy may be required in patients with other airway problems accompanying laryngomalacia and with systemic comorbidities.

Ethics Committee Approval: Ethics committee approval was received for this study from the ethics committee of Dokuz Eylül University Noninvasive Research Ethics Board (GOA-233, 2015/24-17).

Informed Consent: Informed consent was not received due to the retrospective nature of the study.

Peer-review: Externally peer-reviewed.

Author contributions: Concept - Y.O., T.K.E.; Design - Y.O.; Supervision - T.K.E; Resource - Y.O.; Materials - Y.O., H.Ö., A.Ç.; Data

Collection and/or Processing - Y.O., H.Ö., A.Ç.; Analysis and/or Interpretation - Y.O., T.K.E; Literature Search - Y.O., H.Ö., A.Ç.; Writing - Y.O.; Critical Reviews - T.K.E.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study has received no financial support.

References

1. Landry A, Thompson DM. Congenital Laryngomalacia: Disease spectrum and management. In: Lioy J, Sobol SE. Disorders of the neonatal airway. Newyork: Springer; 2015. p. 51-9. [CrossRef]
2. Thompson DM. Abnormal sensorimotor integrative disfunction of the larynx in congenital laryngomalacia: a new theory of etiology. Laryngoscope 2007; 117: 1-33. [CrossRef]
3. Thorne MC, Garetz SL. Laryngomalacia: Review and summary of current clinical practice in 2015. Paediatr Respir Rev 2016; 17: 3-8. [CrossRef]
4. Manning SC, Inglis AF, Mouzakes J, Carron J, Perkins JA. Laryngeal anatomic differences in pediatric patients with severe laryngomalacia. Arch Otolaryngol Head Neck Surg 2005; 131: 340-3. [CrossRef]
5. Pamuk AE, Süslü N, Günaydın RO, Atay G, Akyol U. Laryngomalacia: patient outcomes following aryepiglottoplasty at a tertiary care center. Turk J Pediatr 2013; 55: 524-8.
6. Munson PD, Saad AG, El-Jamaal SM, Dai Y, Bower CM, Richter GT. Submucosal nerve hypertrophy in congenital laryngomalacia. Laryngoscope 2011; 121: 627-9. [CrossRef]
7. Ünal ÖF. Larinksin konjenital anomalileri ve edinsel larinks stenozu. Türkiye Klinikleri J E.N.T. Special Topics 2009; 2: 21-31.
8. Escher A, Probst R, Gysin C. Management of laryngomalacia in children with congenital syndrome: The role of supraglottoplasty. J Pediatr Surg 2015; 50: 519-23. [CrossRef]
9. Zoulmalan R, Maddalozzo J, Holinger LD. Etiology of stridor in infants. Ann Otol Rhinol Laryngol 2007; 116: 329-34. [CrossRef]
10. Wright CT, Goudy SL. Congenital laryngomalacia: Symptom duration and need for surgical intervention. Ann Otol Rhinol Laryngol 2012; 121: 57-60. [CrossRef]
11. Olney DR, Greinwald RJ, Smith RJ, Bauman NM. Laryngomalacia and its treatment. Laryngoscope 1999; 109: 1770-5. [CrossRef]
12. Garritano FG, Carr MM. Characteristics of patients undergoing supraglottoplasty for larygomalacia. Int J Pediatr Otorhinolaryngol 2014; 78: 1095-100. [CrossRef]
13. Mancuso RF, Choi SS, Zalzal GH, Grundfast KM. Laryngomalacia. The search for the second lesion. Arch Otolaryngol 1996; 122: 302-6. [CrossRef]
14. Yuen HW, Tan HK, Balakrishnan A. Synchronous airway lesions and associated anomalies in children with laryngomalacia evaluated with rigid endoscopy. Int J Pediatr Otorhinolaryngol 2006; 70: 1779-84. [CrossRef]
15. Rifai HA, Benoit M, El-Hakim H. Secondary airway lesions in laryngomalacia: a different perspective. Otolaryngol Head and Neck Surg 2011; 144: 268-73. [CrossRef]
16. Dickson JM, Richter GT, Meinzen-Derr J, Rutter MJ, Thompson DM. Secondary airway lesions in infants with laryngomalacia. Ann Otol Rhinol Laryngol 2009, 118: 37-43. [CrossRef]
17. Schroeder JW, Bhankdar ND, Hollinfer LD. Synchronous airway lesions and outcomes in infants with severe laryngomalacia requiring supraglottoplasty. Arch Otolaryngol Head Neck Surg 2009; 135: 647-51. [CrossRef]
18. Krashin E, Ben-Ari J, Springer C, DeRowe A, Avital A, Sivan Y. Synchronous airway lesions in laryngomalacia. Int J Pediatr Otorhinolaryngol 2008; 72: 501-7. [CrossRef]
19. Dursun G. Konjenital Larenks Anomalileri. Türkiye Klinikleri J E.N.T. Special Topics 2011; 4: 92-7.
20. Day KE, Discolo CM, Meier JD, Wolf BJ, Halstead LA, White DR. Risk factors for supraglottoplasty failure. Otolaryngol Head Neck Surg 2012; 146: 298-301. [CrossRef]
21. Thompson DM. Laryngomalacia: factors that influence severity and outcomes of management. Curr Opin Otolaryngol Head Neck Surg 2010; 18: 564-70. [CrossRef]