Laser supraglottoplasty for laryngomalacia: A pediatric case series

Berat Demir¹, Adem Binnetoglu²

¹Department of Otorhinolaryngology Head and Neck Surgery, Marmara University Faculty of Medicine, Istanbul Turkey

ABSTRACT

We retrospectively reviewed the medical records of a total of nine patients who underwent laser SGP for severe laryngomalacia at our hospital between 2014 and 2015. All patients had respiratory distress as soon as birth. All patients had one or more comorbidities. Only one patient who had vocal cord paralysis remained tracheostomy-dependent at three years of follow-up. In conclusion, supraglottoplasty is an effective and safe method in the treatment of severe laryngomalacia.

Keywords: Laryngomalacia, laser, supraglottoplasty, pediatric airway.

Laryngomalacia (LM) is the most common cause of congenital stridor in infants, affecting 50 to 75% of all cases with congenital laryngeal anomalies.[1] It occurs due to dynamic collapse of the supraglottic structures in the course of inspiration and the severity of symptoms depends on the degree of airway obstruction. Although the exact pathophysiology is unclear, several theories have been advanced to explain the etiology of the supraglottic airway collapse. The diagnosis is substantially based on officebased flexible laryngoscopy, which confirms LM and rules out other causes of supraglottic obstruction. The majority of patients present with inspiratory stridor shortly after birth, which becomes worse in the first several months of life.

About 20% of patients with LM require a surgical intervention due to a severe clinical presentation including recurrent cyanosis, apneic pauses, feeding difficulty, aspiration, and failure to thrive or worsening of symptoms, despite conservative treatment.[2] Laryngomalacia can be accompanied by several comorbidities, including gastroesophageal reflux disease, neurological deficits, the presence of an additional airway lesion, congenital heart disease, and the presence of a syndrome or genetic disorder.[2] Supraglottoplasty (SGP) is currently the most common surgical treatment for severe LM.[3] It represents the surgical mainstay of severe LM. Carbon dioxide (CO₂) laser or cold instruments can be used for surgery by trimming bilateral

Received: March 01, 2019 Accepted: June 09, 2019 Published online: November 08, 2019

Correspondence: Berat Demir, MD. Marmara Üniversitesi Pendik Eğitim ve Araştırma Hastanesi, Kulak Burun Boğaz Hastalıkları Anabilim Dalı, 34899 Pendik, İstanbul, Turkey. e-mail: drberatdemir80@hotmail.com

Doi: http://dx.doi.org/10.5606/Tr-ENT.2019.41636

Citation:

Demir B, Binnetoglu A. Laser supraglottoplasty for laryngomalacia: A pediatric case series. Tr-ENT 2019;29(3):150-154.

²Department of Otorhinolaryngology Head and Neck Surgery, Saint Elizabeth Medical Center, Massachusetts, USA

arytenoids and dividing the aryepiglottic to improve the supraglottic airway.

In this review, we discuss the clinical manifestations of the patients with severe LM who underwent SGP at our institution.

CASE SERIES

This retrospective, clinical study included medical data of a total of nine patients (6 males, 3 females; mean age 4.7 months; range, 3 months to 7 months) who underwent laser SGP (LSGP) for symptomatic LM between January 2014 and January 2015. The diagnosis of LM was based on medical history and clinical findings and was confirmed via transnasal fiberoptic laryngoscopy (TNFL) under general anesthesia, which allows the patient to breath spontaneously while evaluating the dynamic upper airway.

The supraglottic obstruction was established in accordance with the Olney's classification to state the morphological type of LM.[4] Severe LM with inspiratory stridor, dyspnea with chest retractions, oxygen desaturations, cyanosis, obstructive apnea, feeding difficulties with or without failure to thrive, and the presence of a tracheostomy on account of severe LM were the indications for LSGP (Table 1). Full anti-acid suppression with a proton pump inhibitor and/or H2 receptor antagonist and dietary measures were applied to all patients before surgery. A written informed consent was obtained from each parent. The study protocol was approved by the Acıbadem University Ethics Committee (No: 2019-9/6). The study was conducted in accordance with the principles of the Declaration of Helsinki.

Table 1. Indications for laser supraglottoplasty

Indications for laser supraglottoplasty Severe laryngomalacia with inspiratory stridor Dyspnea with chest retractions

Oxygen desaturation

Cyanosis

Obstructive apnea

Feeding difficulties with or without failure to thrive

Presence of a tracheostomy on account of severe laryngomalacia

The procedure was performed using a CO₂ laser (Velas II DIODE CO₂ laser, Wuhan, China) connected to a microscope (ZEISS OPMI Vario ModelS88; Zeiss Meditec AG, Jena, Germany). The laser parameters used were as follows: ultrapulse mode, 125 mJ/cm², and 250 microspot. In type 1 LM, resection and/or vaporization of the redundant mucosa over the arytenoid, corniculate, and cuneiform cartilage were performed. In type 2 LM, the shortened aryepiglottic folds were sectioned and the lateral edges of the epiglottis were resected. In addition, excessive supraglottic tissues were partially resected and/or evaporated. All patients stayed overnight in the pediatric intensive care unit (ICU) and were extubated one day after surgery. Epiglottopexy was performed by passing two or three trans-oral resorbable sutures (3.0 Vicryl) to the tongue base.

Case 1- Patient had respiratory distress as soon as birth. The age at the time of diagnosis was seven months. According to the classification of the disease at the time of diagnosis, the patient was classified as type 1 (Table 2). Gastroesophageal reflux disease was present (Table 3). The patient had complete resolution of the initial symptoms after LSGP.

Case 2- Had respiratory distress as soon as birth, but symptoms gradually worsened. Gastroesophageal reflux disease was present. The patient was three months at the time of diagnosis and had type 1 laryngomalacia. A revision LSGP was not required. The patient had complete resolution of the initial symptoms after LSGP.

Case 3- Had respiratory distress as soon as birth. The patient was four months at the time of diagnosis. Type 1 laryngomalacia was identified on direct laryngoscopy. Gastroesophageal reflux disease was present. A revision LSGP was not required. The patient had complete resolution of the initial symptoms after LSGP.

Table 2. Types of laryngomalacia

	Number of patients
Type 1	5
Type 2	3
Type 3	1

152 Tr-ENT

Table 3. Comorbidities accompanied to the laryngomalacia	Table 3. Comorbidities accor-	npanied to the lary	ngomalacia
--	-------------------------------	---------------------	------------

Comorbidities	Number of patients
Gastro-esophageal reflux disease	7
Neurological disease	2
Genito-urinary disease	1
Cardiovascular disease	1
Born prematurely	1

Case 4- Had respiratory distress as soon as birth. The patient was 6.5 months at the time of diagnosis. A revision LSGP was not required. The patient had complete resolution of the initial symptoms after LSGP.

Case 5- Had respiratory distress as soon as birth. The patient was 5.5 months at the time of diagnosis. A revision LSGP was not required. The patient had complete resolution of the initial symptoms after LSGP.

Case 6- Had respiratory distress as soon as birth. The patient was four months at the time of diagnosis. Gastroesophageal reflux disease was present. Type 1 laryngomalacia was identified on direct laryngoscopy. A revision LSGP was not required. The patient had complete resolution of the initial symptoms after LSGP.

Case 7- Had respiratory distress as soon as birth. The patient was 5.5 months at the time of diagnosis. Type 2 laryngomalacia was identified on direct laryngoscopy. Tracheostomy was performed preoperatively. After the operation, tracheostomy was decannulated at one year of follow-up. The patient had complete resolution of the initial symptoms after LSGP.

Case 8- Had respiratory distress as soon as birth. The patient was 4.5 months at the time of diagnosis. Type 2 laryngomalacia was identified on direct laryngoscopy. Gastroesophageal reflux disease was present with neurological disease. Tracheostomy was performed preoperatively. Tracheostomy was decannulated at three years of follow-up.

Case 9- Had respiratory distress as soon as birth. The patient was born prematurely and was three months at the time of diagnosis. Type 3 laryngomalacia was identified on direct

laryngoscopy. Tracheostomy was performed preoperatively. The patient had comorbidities such as neurological disease, genitourinary disease, cardiovascular disease, and gastroesophageal reflux disease. The patient died from recurrent pneumonia and comorbidities (neurological deficits).

DISCUSSION

Laryngomalacia is the most common congenital laryngeal anomaly during childhood. The presenting symptoms include stridor, dyspnea on exertion, and possibly obstructive sleep apnea and dysphonia, which start a few weeks after birth and reach its peak in the first four to eight months of life with typically resolution by the age of two years. [5,6] In the present study, our patients were admitted to our clinic when their symptoms were at peak levels (mean: 4.2 months), consistent with previous studies.^[5,6] Gastroesophageal or laryngopharyngeal reflux is the most common comorbidity of LM at a rate ranging between 70 and 80%.[7] Consistent with the literature, seven of nine patients (78%) had gastroesophageal reflux in our study. On the other hand, previous studies have not shown a causal relationship between the two entities, vet.[8]

The certain etiology of LM is still unknown and sustains to be an area of great attention and examination. Possible theories of etiology include the anatomic, cartilaginous, and neurological components. The neurological theory is the most optimal elucidation of laryngeal malfunction and, consequently, is the widely accepted etiological theory.[9] Rathi and Rathi[10] reported a new theory attributed to the relative imbalance of requirement for the supply of air, proposing the rise in demand, leading to a turbulent airflow, boosting vacuum pressure and leading to collapse of the laryngeal structures. It cannot be predicted as to which child would require surgery. However, the aforementioned authors proposed that the requirement for surgery could be predicted based on this theory. In another study, Sacca et al.[11] reported that many different respiratory tract disorders could be present in patients with 22q11 deletion syndrome, such as subglottic stenosis and laryngo- or tracheomalacia.

With airway anomalies, particularly when accompanied by congenital heart disease, 22q11.2 deletion syndrome should be considered. Several studies have reported that synchronous airway lesions can accompany LM. [12-14] Tracheomalacia, subglottic stenosis, and vocal cord paralysis are particularly leading pathologies in LM. In our study, comorbid laryngeal anomalies were found in six patients, vocal cord paralysis in one patient, and subglottic stenosis in five patients.

authors have suggested synchronous airway pathologies are associated with the severity of LM and increased the need for surgery, [12,13] whereas others have claimed that synchronous airway pathologies have no impact on time to healing and no connection between the severity of LM and synchronous airway pathologies.[4,14] In the majority of cases, high success rates can be achieved in case of subglottic stenosis and vocal cord paralysis.. Most cases (80%) are classified as mild to moderate. [15] Fifteen percent of cases are categorized as severe and needing SGP. Severe stridor, obstructive apnea, severe suprasternal or intercostal retractions, feeding difficulties, and failure to thrive are surgical indications.

Furthermore, LM may manifest itself with several comorbidities, which affect the symptoms and prognosis of the disease. Although all children presented with comorbidities in our study, we obtained an overall 95% success rate after SGP. Previous studies have also documented that SGP is an effective and safe method in the treatment of LM.[16,17] Through a definitive surgical technique with minor collateral tissue damage, very little laryngeal edema, and a bloodless surgical field, LSGP has advantages over cold instruments or microdebriders. Furthermore, LSGP provides rapid recovery without significant comorbidities. Ching et al.[18] used bipolar radiofrequency ablation (coblation) and showed that this technique was safe and could lead to substantial improvement in the apnea-hypopnea index in patients with obstructive sleep apnea. In addition, type of LM determines which type of surgery to perform.

Until now, three main LM types have been identified. $^{[15]}$ CO₂ LLSGP is effective in 95% of type 1 and 2 LMs with parameters set to

ultra-pulse mode, 125 mJ/cm², 250 m spot size, and 10 Hz repetition rate. However, for type 3 LM, in addition to the CO₂ laser with parameters set to the CW mode, 3 W output power and 500 μ spot size epiglottopexy is suggested. The poorer power density leads to more coagulation necrosis in type 3 LM. Laser SGP can be further improved with the progress of technology. Instead of the type of LM, comorbid condition of the patient determines the surgical success. Comorbid conditions including neurological deficits, congenital heart disease, or multiple levels of airway obstruction have worse consequences of surgery. $^{\rm [3]}$

In conclusion, LM is the most common cause of stridor during childhood. About 15% of children have symptoms severe enough to require a surgical intervention. Supraglottoplasty is currently the most common surgical treatment for severe LM. Of note, comorbidities decrease the success rate of SGP and may eventually require tracheotomy.

Declaration of conflicting interests

The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

Funding

The authors received no financial support for the research and/or authorship of this article.

REFERENCES

- 1. Holinger LD. Etiology of stridor in the neonate, infant and child. Ann Otol Rhinol Laryngol 1980;89:397-400.
- 2. Walner DL, Neumann DB, Hamming KK, Miller RP. Supraglottoplasty in Infants: A Staged Approach. Ann Otol Rhinol Laryngol 2015;124:803-7.
- 3. Richter GT, Thompson DM. The surgical management of laryngomalacia. Otolaryngol Clin North Am 2008;41:837-64.
- 4. Olney DR, Greinwald JH Jr, Smith RJ, Bauman NM. Laryngomalacia and its treatment. Laryngoscope 1999;109:1770-5.
- 5. Landry A, Thompson DM. Congenital Laryngomalacia: Disease spectrum and management. In: Lioy J, Sobol SE, editors. Disorders of the neonatal airway. New York: Springer; 2015. p. 51-9.
- Thorne MC, Garetz SL. Laryngomalacia: Review and Summary of Current Clinical Practice in 2015. Paediatr Respir Rev 2016;17:3-8.
- 7. Bibi H, Khvolis E, Shoseyov D, Ohaly M, Ben Dor D, London D, et al. The prevalence of gastroesophageal reflux in children with tracheomalacia and laryngomalacia. Chest 2001;119:409-13.

154 Tr-ENT

8. Hartl TT, Chadha NK. A systematic review of laryngomalacia and acid reflux. Otolaryngol Head Neck Surg 2012;147:619-26.

- 9. Thompson DM. Abnormal sensorimotor integrative function of the larynx in congenital laryngomalacia: a new theory of etiology. Laryngoscope 2007;117(Suppl)1-33.
- Rathi A, Rathi S. Relative imbalance as etiology of laryngomalacia - A new theory. Med Hypotheses 2017;98:38-41.
- 11. Sacca R, Zur KB, Crowley TB, Zackai EH, Valverde KD, McDonald-McGinn DM. Association of airway abnormalities with 22q11.2 deletion syndrome. Int J Pediatr Otorhinolaryngol 2017;96:11-14.
- Schroeder JW, Bhandarkar ND, Holinger LD. Synchronous airways lesions and outcomes in infants with severe laryngomalacia requiring supraglottoplasty. Arch Otolaryngol Head Neck Surg 2009;135:647-51.
- 13. 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.
- 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.
- 15. Monnier P, editor. Pediatric Airway Surgery: Management of Laryngotracheal Stenosis in Infants and Children. New York: Springer Science & Business Media, 2010.
- 16. Lee KS, Chen BN, Yang CC, Chen YC. CO2 laser supraglottoplasty for severe laryngomalacia: a study of symptomatic improvement. Int J Pediatr Otorhinolaryngol 2007;71:889-95.
- 17. Reinhard A, Gorostidi F, Leishman C, Monnier P, Sandu K. Laser supraglottoplasty for laryngomalacia; a 14 year experience of a tertiary referral center. Eur Arch Otorhinolaryngol 2017;274:367-74.
- 18. Ching HH, Spinner AG, Reeve NH, O-Lee TJ. A novel technique for unilateral supraglottoplasty. Int J Pediatr Otorhinolaryngol 2018;104:150-4.