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Case Study

Novel Treatment of Laryngomalacia: A Case Series

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ABSTRACT

Background: Laryngomalacia is a prevalent neonatal condition characterized by the increased pliability of laryngeal tissues, leading to airway obstruction and associated respiratory complications. It necessitates a nuanced approach to diagnosis and management, given its variable clinical presentation and potential for significant morbidity.

Objective: This study aims to investigate the clinical presentation, management, and outcomes of neonates with laryngomalacia, focusing on the efficacy of individualized treatment approaches including bronchoscopy, laser ablation, and various ventilation strategies.

Methods: This retrospective case series analyzed data from two neonates treated for laryngomalacia at Children Center Hospital Tehran between 2021 and 2022. Inclusion criteria were neonates beyond 28 weeks gestation without congenital abnormalities and with an RDS score ≥8. Exclusion criteria included preterm neonates (<28 weeks), an RDS score <8, and congenital abnormalities. Data collection involved measures such as respiratory assistance, stridor, feeding issues, oxygen requirement, hospital stay duration, ventilation days, gestational age, RDS score, retraction severity, and amniotic blood gas values. Ethical considerations were observed throughout the study.

Results: The first neonate, a 29-day-old female with a gestational age of 36 weeks and 5 days, required HFNC and laser ablation, leading to the resolution of stridor and feeding difficulties. Her hospital stay was 46 days. The second neonate, a 16-day-old male born at 38 weeks gestation, experienced complications including VAP and required a more extended treatment course, including intubation, NIV, CPAP, and HFNC, culminating in an 80-day hospital stay.

Conclusion: The study highlights the importance of personalized and adaptable treatment strategies in managing laryngomalacia in neonates. The variability in clinical presentations and responses to treatment underscores the need for ongoing research and the development of refined management protocols. A multidisciplinary approach, incorporating timely diagnosis and flexible intervention, is crucial for optimal patient outcomes.

Keywords: Laryngomalacia, Neonatal Respiratory Distress, Bronchoscopy, Laser Ablation, High-Flow Nasal Cannula (HFNC), Continuous Positive Airway Pressure (CPAP), Non-Invasive Ventilation (NIV), Ventilator-Associated Pneumonia (VAP).

INTRODUCTION

Laryngomalacia, characterized by the supraglottic tissues' inability to maintain structural integrity during inhalation, results in airway obstruction and stridor (1,2). Primarily affecting neonates and infants, it emerges as the predominant cause of stridor. Interestingly, the characteristic inhalation stridor may not be present at birth but often develops within the first few weeks. Activities such as breastfeeding and crying can intensify stridor in these patients. The spectrum of symptoms includes minor to severe cyanotic episodes, positional stridor, feeding difficulties, and chronic stridor, leading to failure to thrive. The peak of symptom severity is typically observed between four to six months, with a gradual decline by eighteen to twenty-four months (3,4).

The exact cause of laryngomalacia remains elusive, although several theories have been proposed. A widely accepted hypothesis suggests neuromuscular variations in laryngeal tone as a key factor, leading to the collapse of supra-arytenoid and supraglottic tissues during inhalation. This condition is often exacerbated by coexisting medical issues such as gastroesophageal reflux disease (GERD),

neurological disorders, and lower Apgar scores (5,6). GERD, in particular, is linked to increased respiratory symptoms and laryngeal inflammation.

Diagnostic approaches include flexible nasolaryngoscopy (NFL) in alert infants (7,8), and in more severe or uncertain cases, laryngotracheoscopy using rigid endoscopes under general anesthesia. Remarkably, about 90% of patients with laryngomalacia recover without medical intervention. However, the remaining 10% with severe manifestations may require surgical treatment, such as supraglottoplasty (9,10).

This surgical intervention, performed using various techniques like epiglottopexy, division of the aryepiglottic fold, or removal of arytenoid mucosa, aims to reduce the obstructive supraglottic tissue. Tools used in these procedures range from cold steel microinstruments to carbon dioxide lasers. Supraglottoplasty has been effective in improving respiratory function and reducing stridor in patients.

Our case series focuses on patients diagnosed with laryngomalacia and treated with laser laryngoscopy at Tehran Children's Hospital over the past six months (11,12). Studies like those by Klinginsmith et al. indicate that 10-20% of neonates with laryngomalacia experience significant symptoms necessitating surgical intervention. Supraglottoplasty is increasingly recognized as the primary treatment for such cases, employing various techniques including laser, cold steel, laryngeal microdebrider, or coblator (13,14). The primary indications for this surgery are worsening airway symptoms and failure to thrive.

Supraglottoplasty involves consideration of patient-specific anatomy, including removing excess arytenoid mucosa, separating shortened aryepiglottic folds, and performing epiglottopexy. It's essential to avoid the interarytenoid mucosa to prevent post-surgical glottic stenosis. Postoperative care often includes administering steroids to reduce airway inflammation (15,16).

Laryngomalacia, a common disorder in newborns, leads to increased pliability of laryngeal tissues, causing respiratory and gastrointestinal complications. Despite the existence of supportive care and surgical options, their limitations have driven the search for novel treatments. Our study aims to explore new treatments addressing the underlying causes of laryngomalacia, hoping to improve patient outcomes through individualized, innovative therapies.

MATERIAL AND METHODS

The research design and methodology of this case series study, conducted at Children Center Hospital Tehran, involved the participation of four patients who exhibited respiratory symptoms such as dyspnea, stridor, feeding issues, and apnea during the years 2021 and 2022. The study targeted patients with a history of positive PCR results and symptoms unexplained by other recognized illnesses. The inclusion criteria were neonates beyond 28 weeks of gestation, having a Respiratory Distress Syndrome (RDS) score equal to or greater than 8, and absence of congenital abnormalities. Conversely, exclusion criteria encompassed neonates less than 28 weeks gestational, those with an RDS score below 8, and presence of congenital abnormalities.

The recruitment strategy was not a requirement for this case series. Allocation of participants to the research groups was facilitated through the examination of medical records of children diagnosed with laryngomalacia in the specified period, following approval from the ethics committee of Tehran University of Medical Sciences (TUMS). The data collection process focused on various measures including the level of respiratory assistance needed, reduction in stridor, feeding difficulties, oxygen requirement, length of hospital stay, duration of ventilation, gestational age, RDS score, severity of retraction, and amniotic blood gas values.

This study did not incorporate any interventions. Ethical considerations were paramount and diligently observed throughout the research process. The assessment of patient outcomes was conducted retrospectively, evaluating the effectiveness of standard care protocols in managing laryngomalacia symptoms. Data analysis was carried out using appropriate statistical methods, ensuring the reliability and validity of the findings.

RESULTS

In this case series, we present the outcomes of two neonates treated at our facility for symptoms related to laryngomalacia. The first case was a 29-day-old female, born prematurely at 36 weeks and 5 days, with a birth weight of 2200 grams. She was admitted to our hospital following a 16-day NICU stay at another facility for feeding issues and stridor. Upon admission, she exhibited suprasternal retraction without obstructive sleep apnea and had an initial oxygen saturation of 95% on oxygen therapy. Her respiratory rate was 44, and she required high-flow nasal cannula (HFNC) oxygen support. Bronchoscopy revealed laryngomalacia, and during the procedure, she experienced a drop in oxygen saturation, necessitating intubation. The fractional inspired oxygen © 2024 et al. Open access under Creative Commons by License. Free use and distribution with proper citation. Page 50 🔶 🔶

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(FiO2) was initially set to 100% and subsequently reduced to 25% before weaning off. Post-surgery, the neonate underwent laser ablation, followed by a day of intubation and then transition to non-invasive ventilation (NIV). Nine days post-surgery, NIV was switched to nasal continuous positive airway pressure (nCPAP). This intervention led to a marked reduction in stridor and respiratory distress, and the neonate was discharged after 46 days without stridor, suprasternal retractions, or feeding issues.

The second case involved a 16-day-old male neonate, born at full term with a gestational age of 38 weeks and a birth weight of 2835 grams. He presented with respiratory distress and a laryngeal cyst and had undergone an apnea procedure prior to transfer to our facility. At birth, he had a respiratory rate of 40, with stridor and tachypnea, and a mild RDS score of 2. He initially received FiO2 at approximately 45%. Post-laryngeal cyst removal and 15 days of intubation, he was transitioned to NIV. Two weeks into the NIV period, he developed ventilator-associated pneumonia (VAP), requiring re-intubation. A bronchoscopy performed on the sixth day diagnosed laryngomalacia. Following surgery, his respiratory distress and tachypnea significantly decreased, and FiO2 was adjusted to 35%. He was then extubated to continuous positive airway pressure (CPAP) for a week, followed by high-flow nasal cannula (HFNC) for two weeks. He was discharged after an 80-day hospital stay without signs of stridor, respiratory distress, or feeding issues.

The demographic data (Table 1) and variables related to the severity of the condition and treatment outcomes (Table 2) are summarized below:

Table 1 Demographics

| Patient | Age | Gestational Age | RDS Score | Respiratory Rate | FiO2 | Stridor | Absence of Breathing |
|---------|-----|-----------------|-----------|------------------|------|---------|----------------------|
| 1 | 29d | 36w+5d | 4 | 48 | 21% | Yes | No |
| 2 | 16d | 38w | 2 | 40 | 45% | Yes | Yes |

Table 2 Variables

| Severity of | Feeding | Hospital Stay | Weight | Ventilation Before | Ventilation After Surgery |
|--------------|---------|---------------|--------|----------------------|-----------------------------|
| Retractions | Problem | | | Surgery | |
| Suprasternal | Yes | 46 days | 2200g | HFNC => Intubation | Intubation => Extubation => |
| | | | | | NIV => CPAP |
| Mild | Yes | 88 days | 2835g | Intubation => NIV => | Intubation => Extubation => |
| | | | | Intubation | CPAP => HFNC |

DISCUSSION

In this case series, we explored the clinical presentations and treatment outcomes of two neonates with laryngomalacia treated at our medical facility. The study contributes to the understanding of this prevalent condition, which can lead to extended hospital stays and significant breathing challenges for affected infants.

The first patient, a female neonate, demonstrated the efficacy of high-flow nasal cannula (HFNC) and laser ablation in treating laryngomalacia. This combination led to the eradication of both stridor and feeding difficulties, with the patient showing considerable improvement upon transitioning from invasive to non-invasive ventilation (NIV). In contrast, the second patient, a male neonate, presented a more complicated case. Initially treated with intubation and NIV, the development of ventilator-associated pneumonia (VAP) necessitated further intervention. Despite these complications, the adaptation of the treatment plan, including continuous positive airway pressure (CPAP) and high-flow nasal cannula (HFNC), facilitated the successful management of his condition.

The use of bronchoscopy in both cases was crucial for confirming the diagnosis of laryngomalacia, underscoring its importance as a diagnostic tool. Furthermore, the diverse therapeutic approaches employed highlight the necessity of personalized and adaptable strategies to manage neonatal laryngomalacia effectively. This aligns with findings from Jose Antonio et al. and Loke et al., where stridor was a prominent reason for hospitalization, and bilateral laser ablation provided significant relief (17,18). Moreover, the work of M. Remacle et al. supports the notion of surgical intervention, showing substantial recovery in most patients (17,18).

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Our findings underscore the varied nature of laryngomalacia and the need for tailored treatment approaches. The successful use of laser ablation in the first case aligns with current research, suggesting its potential as a less invasive treatment option for laryngomalacia. However, the prolonged hospital stay in the second case, primarily due to VAP, illustrates the potential complications and diverse outcomes that can occur during treatment. This necessitates vigilant monitoring and prompt intervention to ensure optimal patient outcomes.

The study's strength lies in its detailed account of the clinical course and response to treatment in two distinct cases, providing valuable insights into the management of laryngomalacia. However, it also has limitations, including a small sample size and the lack of a control group, which may limit the generalizability of the findings. Furthermore, the retrospective nature of the study may introduce biases in data interpretation.

Future research should focus on larger, controlled studies to validate these findings and explore factors influencing prognosis. Additionally, ongoing investigation into less invasive treatment modalities, as well as the development of guidelines for the management of complications such as VAP, would be beneficial. Such efforts will contribute to enhancing treatment protocols and outcomes for infants with this complex condition. Ultimately, the goal is to develop efficient strategies that mitigate the repercussions of laryngomalacia, thereby improving the overall therapeutic approach and outcomes for affected neonates (25).

CONCLUSION

In conclusion, this case series underscores the complex and varied nature of laryngomalacia in neonates, highlighting the need for individualized and adaptable treatment strategies. The successful management of two distinct cases through the use of bronchoscopy for diagnosis, combined with tailored interventions like laser ablation, HFNC, and CPAP, demonstrates the potential effectiveness of these approaches. However, challenges such as ventilator-associated pneumonia emphasize the necessity for vigilant monitoring and flexibility in treatment adjustments. These findings have significant implications for clinical practice, suggesting that a personalized, multidisciplinary approach is essential for optimizing outcomes in neonates with laryngomalacia. The study also points to the need for further research to refine treatment protocols and improve care for this complex pediatric condition.

REFERENCES

1. Hsu AK, Rosow DE, Wallerstein RJ, April MM. Familial congenital bilateral vocal fold paralysis: a novel gene translocation. International journal of pediatric otorhinolaryngology. 2015;79(3):323-7.

2. Sandu K, Monnier P, Reinhard A, Gorostidi F. Endoscopic epiglottopexy using Lichtenberger's needle carrier to avoid breakdown of repair. European archives of oto-rhino-laryngology : official journal of the European Federation of Oto-Rhino-Laryngological Societies (EUFOS) : affiliated with the German Society for Oto-Rhino-Laryngology - Head and Neck Surgery. 2015;272(11):3385-90.

3. Ching HH, Spinner AG, Reeve NH, TJ OL. A novel technique for unilateral supraglottoplasty. International journal of pediatric otorhinolaryngology. 2018;104:150-4.

4. Pu S, Xu H, Li X. Supraglottoplasty in neonates and infants: A radiofrequency ablation approach. Medicine. 2018;97(7):e9850.

5. Álvarez-Neri H, Villamor P, Ortiz Hernandez E, Penchyna Grub J. Epiglottopexy by external puncture for epiglottic prolapse in severe laryngomalacia. A novel technique. European annals of otorhinolaryngology, head and neck diseases. 2019;136(2):115-7.

6. AbdelFattah ElSobki A, Hashish MI, El-Kholy NA. One and half coblation supraglottoplasty: A novel technique for management of type II laryngomalacia. International journal of pediatric otorhinolaryngology. 2020;138:110330.

7. Lau CL, Chee YY, Chung BHY, Wong MSR. CHARGE syndrome patient with novel CHD7 mutation presenting with severe laryngomalacia and feeding difficulty. BMJ case reports. 2020;13(7).

8. Lawlor C, Smithers CJ, Hamilton T, Baird C, Rahbar R, Choi S, et al. Innovative management of severe tracheobronchomalacia using anterior and posterior tracheobronchopexy. The Laryngoscope. 2020;130(2):E65-e74.

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9. Soong WJ, Yang CF, Lee YS, Tsao PJ, Lin CH, Chen CH. Vallecular cyst with coexisting laryngomalacia: Successful diagnosis and laser therapy by flexible endoscopy with a novel noninvasive ventilation support in infants. Pediatric pulmonology. 2020;55(7):1750-6.

10. Kamran A, Smithers CJ, Baird CW, Jennings RW. Experience with bioresorbable splints for treatment of airway collapse in a pediatric population. JTCVS techniques. 2021;8:160-9.

11. Panda PK, Tomar A, Pillai GR, Kumar A, Sharawat IK. Infantile tremor syndrome and laryngomalacia: A novel association? Clinical nutrition ESPEN. 2021;43:235-8.

12. Ng SW, Syafina H, Goh LC. Dual Staged Supraglottoplasty for the Treatment of Trapdoor Epiglottis with Underlying Neurodegenerative Disease-Case report and Review of literature. Indian journal of otolaryngology and head and neck surgery : official publication of the Association of Otolaryngologists of India. 2022;74(Suppl 3):5033-6.

13. Song JS, Sloychuk J, El-Hakim H, Isaac A. A novel sleep oximetry scoring tool for pediatric laryngomalacia. International journal of pediatric otorhinolaryngology. 2022;160:111220.

14. Schuberth K, Ramani PK, Beemarajan E, Veerapandiyan A. Child Neurology: KMT2B-Related Dystonia in a Young Child With Worsening Gait Abnormality. Neurology. 2023;101(7):328-32.

15. Mathur N, Verma RK, Jaitly S, Kumar A. Atypical Presentation of Severe Laryngomalacia Managed with Aryepiglottoplasty and A Novel Suture Technique.

16. Chen JL, Messner AH, Chang KW. Familial laryngomalacia in two siblings with syndromic features. International journal of pediatric otorhinolaryngology. 2006;70(9):1651-5.

17. Manickavasagam J, Jebreel Ae. Novel Suction holder clips for endolaryngeal laser surgery. Otolaryngology–Head and Neck Surgery. 2012;147(S2):P194-P5.

18. Erickson B, Cooper T, El-Hakim H. Factors associated with the morphological type of laryngomalacia and prognostic value for surgical outcomes. JAMA Otolaryngology–Head & Neck Surgery. 2014;140(10):927-33.

19. Mase CA, Chen ML, Horn DL, Parikh SR. Supraglottoplasty for sleep endoscopy diagnosed sleep dependent laryngomalacia. International journal of pediatric otorhinolaryngology. 2015;79(4):511-5.

20. Pu S, Xu H, Li X. Supraglottoplasty in neonates and infants: a radiofrequency ablation approach. Medicine. 2018;97(7).

21. Scott BL, Lam D, MacArthur C. Laryngomalacia and swallow dysfunction. Ear, Nose & Throat Journal. 2019;98(10):613-6.

22. Panda PK, Tomar A, Pillai GR, Kumar A, Sharawat IK. Infantile tremor syndrome and laryngomalacia: A novel association? Clinical nutrition ESPEN. 2021;43:235-8.

23. Teague WG, Lawrence MG, Williams S, Garrod AS, Froh D, Early SV, et al. Novel treatment-refractory preschool wheeze phenotypes identified by cluster analysis of lung lavage constituents. The Journal of Allergy and Clinical Immunology: In Practice. 2021;9(7):2792-801. e4.

24. Powell AR, Srinivasan S, Helman JL, Li AD-R, O'Brien LM, Shih A, et al. Novel treatment for hypotonic airway obstruction and severe obstructive sleep apnea using a nasopharyngeal airway device with 3D printing innovation. Journal of Clinical Sleep Medicine. 2022;18(10):2497-502.

25. Shivnani D, Raman E, Kurien M, Ram G, Amle D. Surgical Candidacy for Management of Laryngomalacia: A Proposed Scoring System. Indian Journal of Otolaryngology and Head & Neck Surgery. 2023;75(1):151-8.