Case Report

Rare treatment option for a common pediatric airway problem

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Abstract

Laryngomalacia is a common cause of respiratory obstruction with stridor in infants. Although most cases resolve spontaneously, severe laryngomalacia needs surgical intervention. Tracheostomies have been the mainstay of treatment. However, this procedure was associated with high morbidity. At present, newer modalities of treatment are being tried. We discuss successful management of an infant having severe laryngomalacia (who had three failed extubation trials) with glossoepiglottopexy.

Keywords: Airway obstruction, endotracheal intubation, glossoepiglottopexy, laryngomalacia, stridor, tracheostomy

Access this article online

Website: www.ijccm.org

DOI: 10.4103/0972-5229.169355

Quick Response Code:

Introduction

Treatment of laryngomalacia has evolved over the years from a conservative approach awaiting resolution to a myriad of surgical options. In the current clinical practice, patients need not be subjected to tracheostomy (which was associated with high morbidity).[1] This case report is an attempt to increase awareness about the approach to laryngomalacia and a newer surgical option available.

Case Report

A 4-month-old female child was admitted to Pediatric Intensive Care Unit (ICU) with a history of noisy breathing with feeding difficulties since a month, aggravated by fever, and increased respiratory effort since 2 days. There was no cyanosis or variation in symptoms while crying or change in position. On examination, she weighed four kilograms, was febrile with audible inspiratory stridor. There were intercostal, suprasternal, and subcostal retractions (respiratory rate of 64/min) with pectus excavatum. Auscultation revealed bilateral decreased air entry. The oxygen saturation (room air) was 86% (PaO_2-72 mmHg; $PaCO_2$-26 mmHg on an arterial blood gas). In view of respiratory distress, due to probable upper airway obstruction, elective intubation was performed. Laryngoscopy revealed a long curled up epiglottis, and vocal cords were not visible. Only a 3 mm internal diameter endotracheal tube could be inserted with difficulty. In postintubation period, the retractions minimized, and the child was ventilated on pressure synchronous intermittent mandatory ventilation mode on minimal settings. An extubation attempt 5 days later failed, and ventilatory support was continued on continuous positive airway pressure mode. After two further failed extubations (over a period of 12 days), examination under general anesthesia (with preparation for tracheostomy) was considered. General anesthesia was induced with propofol 3 mg/kg while spontaneous respiration was maintained with oxygen, air (50:50), sevoflurane at 1.2 minimum alveolar

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How to cite this article: Dias R, Deshmukh CT, Tullu MS, Divecha C, Karande S. Rare treatment option for a common pediatric airway problem. Indian J Crit Care Med 2015;19:681-3.

concentration with mask on Jackson Rees circuit. After spraying 10% xylocaine over the vocal cords, zero-degree laryngoscopy was performed, which revealed a long floppy epiglottis falling retrogradely on vocal cords (type 4 laryngomalacia). There was dynamic collapse of aryepiglottic folds. The base of tongue, glottis, and subglottis were normal. Based on these findings, a decision to perform a glossoepiglottopexy was made. Muscle relaxation was achieved with atracurium. Two nonabsorbable sutures were taken from epiglottis to base of tongue, one on either side. Neuromuscular blockade was reversed with neostigmine and atropine. She tolerated the extubation well. Her recovery was uneventful, and she was subsequently discharged without any stridor.

Discussion

Laryngomalacia is a common cause of stridor in infants. There is inspiratory collapse of lax supraglottic tissue into the airway causing respiratory obstruction. The stridor worsens with feeding, crying, supine positioning, and agitation.[2] There may be regurgitation, coughing, choking, or failure to thrive.[2] Those with signs and symptoms of severe airway obstruction require urgent airway examination to determine the cause and secure the airway.[3] Hollinger's classification is used for the various types of laryngomalacia–type 1 is anterior prolapse of the arytenoid and corniculate cartilages; type 2 is tubular epiglottis which curls on itself, often associated with type 1; type 3 is anteromedial collapse of the arytenoids; type 4 is posterior prolapse of the epiglottis; and type 5 is short aryepiglottic folds.[4]

Zero-degree laryngoscopy, flexible laryngoscopy, or bronchoscopy is recommended for diagnosis for direct visualization of airway anatomy and dynamics. This should be done by team of pediatric otorhinolaryngologist, intensive care specialist, and anesthetist in an ICU or operation theater setup. It is important to maintain spontaneous ventilation while anesthetizing these children (with a combination of inhalational and intravenous agents) to allow complete dynamic assessment of the airway to confirm diagnosis. Proper identification of patients who require medical and surgical intervention is key to providing successful treatment.[5] The criteria for severe laryngomalacia with surgical indications include those having clinical evidence of severe respiratory obstruction (obstructive sleep apnea, dyspnea at rest, cyanotic episodes with or without supracostal and intercostal retractions), and/or pectum excavatum, and/or those having swallowing disorders (with low weight gain and deficient stature).[1] Children fulfilling these criteria and requiring airway support with weaning failure are ideal candidates for surgical intervention without delay.

Earlier, tracheostomy was the only treatment available to avoid fatal events.[1] At present, better surgical techniques such as tracheal stenting, endoscopic laser correction of lax supraglottic tissue, supraglottoplasty, and epiglottopexy have evolved. Current procedures depend on individual anatomic and functional alterations on a patient-to-patient basis.[1] Supraglottoplasties were first described in 1987 by Zalzal *et al*. in a case series of ten patients having severe laryngomalacia.[6] It has now become the mainstay of surgical management and involves laryngoscopic resection of excess and lax mucosa from epiglottis, aryepiglottic folds, arytenoids, and corniculate cartilages, thus decreasing amount of supraglottic soft tissue collapsing into laryngeal inlet during inspiration. Carbon dioxide laser and microdebrider assisted supraglottoplasty introduced later were the effective first-line treatment for severe type 1, 2, and 3 laryngomalacia. Bilateral supraglottoplasty may also be performed. Failures and complications of supraglottoplasties include need for revision surgery, vocal cord granuloma, edema, webs, and supraglottic stenosis.

Glossoepiglottopexy is another relatively new surgical strategy which is simple, nonexpensive, and effective. It is useful in type 4 laryngomalacia with isolated posterior displacement of epiglottis where the obstruction is due to epiglottis inhalation. Two nonabsorbable sutures are taken bilaterally from the base of the tongue to epiglottis one on each side. Thus, the lingual surface of epiglottis gets attached to the base of the tongue. This prevents collapse of epiglottis over the glottic opening, thus maintaining an adequate laryngeal airway and preserving speech. However, swallowing problems, aspiration, or reflux may occur postoperatively.

The success of these procedures is clinically measured by (postoperative) reduced requirement for airway support, resolution of feeding difficulties, weight, and stature gain.

Thus though laryngomalacia usually resolves spontaneously within the 1st year of life (median time to resolution in isolated laryngomalacia being approximately 36 weeks), surgical intervention may be needed in 15-20% of infants with severe, life-threatening laryngomalacia.[7] These cases should be evaluated by flexible fiberoptic bronchoscopy or zero-degree laryngoscopy to plan further management. With proper selection, glossoepiglottopexy may prove to be a good,

simple, and effective management option, whereby tracheostomy can be avoided.

Learning points

- Children with severe laryngomalacia should be managed by a multidisciplinary team (expert pediatric intensivist, pediatric anesthetist, and pediatric otorhinolaryngologist)
- Surgical intervention should be considered early in severe, life-threatening laryngomalacia requiring prolonged airway support and weaning difficulties
- Glossoepiglottopexy, a newer treatment modality, can be effectively used for treatment of severe type 4 laryngomalacia.

Acknowledgment

The authors thank Dr. Avinash Supe, Dean of Seth G.S. Medical College and K.E.M. Hospital, for granting permission to publish this manuscript.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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