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**Endoscopic Surgical Management for Laryngomalacia
Case Report and Review of the Literature**

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ENDOSCOPIC SURGICAL MANAGEMENT FOR LARYNGOMALACIA CASE REPORT AND REVIEW OF THE LITERATURE

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Laryngomalacia is the most common of the many causes of respiratory stridor in the newborn. It may be identified by fiberoptic nasopharyngoscopy in the nursery or office. Several anatomic mechanisms of supraglottic collapse have been reported in the literature. The most common is a narrowing of the supraglottic airway with blockage of the glottic opening by the redundant tissue of the aryepiglottic folds. Although surgery rarely is indicated, severe airway obstruction, necessitating surgical intervention, can occur. Resection of supraglottic tissue should be performed only as an alternative to tracheotomy. Surgical procedures ranging from tracheotomy to epiglottidectomy have been advocated. Direct visualization of the obstructing tissue by nasopharyngoscopy allows the planning of an appropriate surgical procedure. In a patient with lateral supraglottic collapse, deep resection of the epiglottis would be expected to weaken the support of the aryepiglottic folds and aggravate the airway condition. Similarly, resection of tissue along the aryepiglottic folds will be useful only if preoperative evaluation demonstrates the obstruction to be at this location.

KEY WORDS - carbon dioxide laser, endoscopy, laryngomalacia, supraglottic collapse.

INTRODUCTION

Airway obstruction in the infant can be caused by a variety of disorders of the upper respiratory tract. One of the most common causes of obstruction is the collapse of the supraglottic area during inspiration. This cause was first described by Lees¹ in 1883. The term congenital laryngeal obstruction was coined to describe this entity in 1897 by Sutherland and Lack.² Other synonyms for this condition have included congenital stridor, congenital laryngeal stridor, inspiratory laryngeal collapse,³ and laryngomalacia.⁴ This terminology is confusing, since some authors consider the term congenital laryngeal stridor to encompass many causes⁵ and reserve the term laryngomalacia for stridor caused by supraglottic collapse.⁴

Laryngomalacia is usually, a benign condition presenting soon after birth,⁶ with resolution in the majority of children by 2 years of age;^{3,7-9} however, the stridor can persist in children past 5 years of age.^{6,8,10} The stridor often will worsen in infants up to 9 months of age before resolution begins. Despite the usual benign course, deaths have been reported.⁸ Pectus excavatum is a common associated finding,^{7,8,11} and resolution of the chest deformity has been associated with relief of the airway obstruction.⁷ Feeding difficulties are common,^{3,7,8,11} and failure to thrive has been reported by several authors.^{7,8,12} Gastroesophageal reflux has been reported in up to 80 % of cases,¹² and severe respiratory compromise with the development of cor pulmonale can occur.

The exact cause of laryngomalacia is unknown; however, two groups of theories have been pro-

posed. A neuromuscular cause was proposed first by Thomson and Turner.¹³ Advocates of this group of theories believe the supraglottic narrowing is secondary to a muscular coordination disorder and represents either the effects of ill-coordinated respiration and abnormal airflow on the soft, pliable supraglottic structures^{13,14} or repetitive supraglottic contractions that ultimately produce a persistent supraglottic narrowing.^{15,16}

The second group of theories state that the supraglottic narrowing is passive and due to a primary structural defect.^{17,18} Sutherland and Lack² proposed that the obstruction was caused by a combination of a congenital malformation and the normal flaccidity of the infantile larynx. Ferguson¹⁹ stated that the supraglottic area is abnormally flaccid in this condition because of an immaturity and developmental delay of this structure. Schulman et al²⁰ reported three cases of inspiratory stridor in a single family. Histologic study of the children's tracheal cartilage in this cohort revealed abnormalities in the cartilage matrix. This finding was not duplicated, however, by Kelemen⁵ who did an autopsy of four children with laryngomalacia. Other authors have also reported normal histology of the epiglottic cartilage.^{2,21} Schwartz³ noted a high incidence of micrognathia in patients with laryngomalacia and theorized that a loss of external supraglottic support was the underlying cause. Other authors also have reported micrognathia associated with this condition.^{5,8}

CASE REPORT

The patient was a 9-month-old boy who pre-

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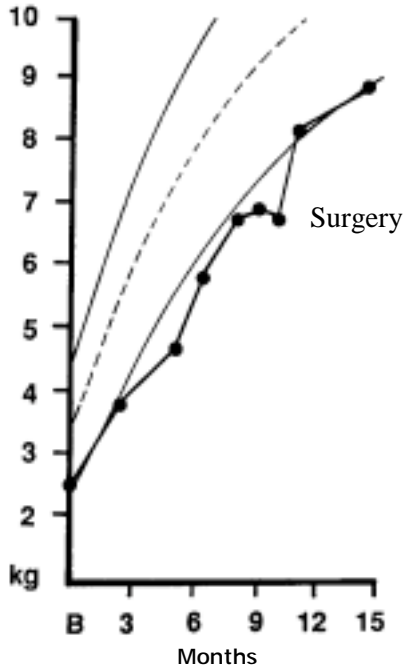


Fig 1. Weight gain of patient in case report. Weight gain ceased for 2-month period prior to surgery. B -birth, dashed line -mean weight for age, area between solid lines - 95th percentile.

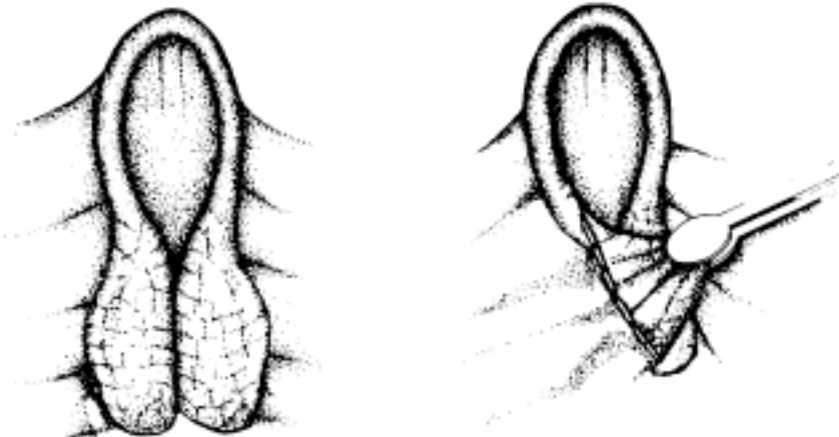
sented with stridor that had been present since birth. The stridor was both inspiratory and expiratory, most severe on inspiration, and somewhat relieved when the patient was in the supine position. He also had difficulty eating, and weight gain had ceased for a 2-month period prior to definitive treatment (Fig 1). On physical examination, the infant had marked inspiratory and moderate expiratory stridor, along with moderate supraclavicular and mild sternal and substernal retractions. His larynx (seen with a nasopharyngoscope) was collapsed in the lateral dimension, and redundant tissue, occluding the glottis, extended from the arytenoid eminences to the epiglottis (Fig 2A). This tissue was sucked into the glottis during inspiration, and air could be seen escaping between redundant mucosa and soft tissue on the aryepiglottic folds during ex-

piration. The true vocal cords, which were covered by the overhanging floppy tissue, could not be seen. The epiglottis was the typical omega shape seen in children with this condition.

The evaluation of the infant's airway included a barium swallow study, chest x-ray, and soft tissue lateral x-ray films of the neck. There was no evidence of cor pulmonale. The infant was taken to the operating room, where a direct laryngoscopy and tracheoscopy were performed. A suspension laryngoscope was inserted and positioned, so that the glottis was opened and the tissue extending from the right arytenoid to the epiglottis was seen. Jet anesthesia was used to ventilate the patient. A cup forceps was used to grasp the midportion of the redundant aryepiglottic fold and to provide medial traction. The redundant mucosa, soft tissue, and accessory cartilages were then transected, using a carbon dioxide laser, from the arytenoid apex to the epiglottis. The arytenoid and epiglottic cartilages were not exposed. The laryngoscope was then repositioned, and the same procedure was performed on the opposite side. The patient was allowed to breathe spontaneously, and complete relief of airway obstruction and stridor was obtained. The total tissue removed measured 0.7 x 0.5 x 0.3 cm, and contained mucosa with accessory cartilages. No muscle was identified histologically. The deep aryepiglottic tissue was nicked on one side; the resulting bleeding point required cautery with a defocused laser beam. Perioperative steroids and antibiotics were used.

Postoperatively, the patient continued to have complete relief of his airway symptoms. He had mild difficulty eating for a 2-week period, with mild symptoms of aspiration (intermittent coughing while drinking). These symptoms resolved, and a rapid weight gain soon followed. By 6 weeks after the procedure, the infant had gained 1.4 kg and was back on the normal growth curve. The parents also reported a marked increase in vocalization and physical activity. Repeated fiberoptic endoscopy showed a fully healed supraglottic area with a good

Fig 2. Artist's depiction of laryngeal appearance. A) Preoperatively, lateral borders of inferior epiglottis were compressed medially, with redundant tissue on aryepiglottic fold. From superior view, epiglottis would appear omega shaped. B) Endoscopic resection of redundant tissue on aryepiglottic folds.



A

B

airway and mobile true vocal cords during inspiration and expiration. Three months later, the infant had no stridor, weighed 8.9 kg, and continued to show a steady weight gain.

DISCUSSION

Because the vast majority of children with laryngomalacia have a benign course, surgery rarely is indicated. Severe airway obstruction associated with apnea and/or failure to thrive, however, are strong indicators for surgical intervention. Holinger¹¹ has treated these severely affected children with tracheotomy. On the other hand, a curative procedure often can be performed if the obstructing tissue can be accurately identified.

Three anatomic mechanisms (regardless of the primary or secondary causes) have been proposed:^{6,13,14}

1) epiglottic prolapse, in which the epiglottis is drawn down like a lid; 2) arytenoid prolapse, in which the arytenoid mucosa meets in the midline and prolapses anteriorly; and 3) lateral supraglottic collapse, in which the supraglottic area is compressed laterally. These mechanisms may be found alone or in combination in individual patients.

The first cause has been reported by several authors to cause stridor in newborns. Iglauer²¹ reported that the removal of the epiglottic tip (using a nasal snare in an awake 5-month-old infant) resulted in immediate disappearance of spells of asphyxia and the cessation of stridor over the subsequent 12 days. Histologic examination of the epiglottis was normal. Schwartz³ reported a case successfully treated by resection of the epiglottis. Hill¹⁷ and Headly²² also described cases in which the epiglottis was the cause of the obstruction.

The latter two theories (arytenoid prolapse and lateral supraglottic collapse) have been proposed as common causes of stridor in newborns.²³ Crooks²³ reported that in patients with arytenoid prolapse, the epiglottis was not folded. He also described curing the stridor in one infant by removal of the soft tissue flaps on the top of the arytenoid eminences. Lane et al⁷ reported a child whose stridor was caused by arytenoid and epiglottic prolapse. In this child, the anteroposterior instead of the lateral laryngeal dimension was compromised and produced the obstruction. This condition was treated successfully by resecting the redundant tissue over the arytenoid eminences and epiglottis.

A commonly described anatomic correlate is collapse of the lateral walls of the supraglottic area. Variot²⁴ reported a child with this condition who died of inadvertent decannulation. Postmortem examination showed that the obstruction was relieved by the phalanges of the tube holding apart the arytenoid eminences and aryepiglottic folds. He proposed cutting away a portion of the aryepiglottic

folds as definitive treatment.²⁴ Readon²⁵ also reported a case of obstruction due to lateral supraglottic collapse, and advocated resection of the aryepiglottic folds for treating this condition. Kelemen⁵ studied the supraglottic histology of three infants with congenital inspiratory stridor. He found the aryepiglottic folds to contain scant muscle; in one infant, it contained "many folds"; in another it "showed loose mucosal wrinkles." Angyal¹⁸ reported the endoscopic and histologic findings from a child with lateral supraglottic collapse. He found soft tissue folds originating from the region of the ventricular bands that invaginated inward with inspiration. On autopsy, the aryepiglottic folds were devoid of muscle and were seen to be formed by a duplication of a thickened mucosal membrane. Many other authors also have reported lateral supraglottic collapse causing stridor in infants.^{1,2,19,26-29}

In spite of the ease of diagnosis and the outlining of treatment principles as early as 1898,²⁴ few reports of aryepiglottic fold resection for the treatment of this disorder can be found in the literature. In 1928, Hasslinger²⁸ reported two children in whom immediate relief of stridor was obtained after resecting redundant mucosa of the aryepiglottic folds using cup forceps. Histologic examination of the tissue revealed a flattened epithelium surrounding a loose, edematous, connective tissue with scant cellularity. Zalzal et al³⁰ recently reported a series of ten patients treated by this procedure. All patients were reported to have good results. In our reported case, the larynx was compressed in the lateral dimensions. The lateral edges of the epiglottis also were displaced medially, which caused it to assume the typical omega shape. Treatment consisted of removing only the loose soft tissue on top of the aryepiglottic folds. This technique is similar to that used in upper lid blepharoplasty. In the larynx, the excess tissue is grasped by cup forceps, tented medially, and cut at its base with a CO₂ laser parallel to the aryepiglottic fibromuscular tissue (Fig 2B). Neither the arytenoid nor epiglottis cartilages are exposed.

In order to identify the obstructing tissue, preoperative fiberoptic examination on an awake patient is optimal. Examination under general anesthesia can be done on a spontaneously breathing patient; however, this cannot be done under deep general anesthesia, since all tissues are lax and are distended readily by a direct endoscope. Proper identification of the cause of the obstruction is essential. Some cases of epiglottic prolapse may be cured by resecting the epiglottic tip; however, many children with laryngomalacia will have lateral supraglottic collapse, which will be aggravated, not helped, by such a procedure. We must agree with Zalzal et al³⁰ that surgery should be used only as an alternative to tracheotomy, since it does have the potential com

plications of aspiration and supraglottic stenosis.

A recent study³¹ has shown synchronous airway lesions in up to 27% of infants with laryngomalacia. Associated abnormalities include subglottic stenosis, tracheomalacia, and vocal cord paralysis. We agree with Zalzal et al³⁰ that a preoperative evaluation of the lower airway with soft tissue airway films, chest x-ray films, and a barium swallow

study is very helpful. Routine bronchoscopy also should be considered strongly and is mandatory if resection of the obstructing supraglottic tissue does not completely relieve the respiratory symptoms during the operation. We agree with Gonzalez et al³¹ that further prospective studies using both endoscopy and radiographic examination will help clarify the optimal workup for these patients.

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