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Case report

Epiglottopexy by external puncture for epiglottic prolapse in severe laryngomalacia. A novel technique

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ABSTRACT

Introduction: Laryngomalacia (LM) is the first cause of stridor in infants. 10 to 20% of patients with LM may require surgery due to the development of severe symptoms. Supraglottoplasty is the most commonly performed surgery for severe LM. However, it is insufficient for the rostrocaudal displacement of the epiglottis against the posterior pharyngeal wall.

Case summary: We report a case of a 2-month-old infant with severe laryngomalacia with a remarkable collapse of the epiglottis towards the glottis with secondary obstruction of the airway, alteration in swallowing and failure to thrive. The patient was treated satisfactorily through epiglottopexy by an external puncture. During a follow-up of 2 years, the patient has been asymptomatic, without any adverse event.

Discussion: Glottic obstruction from posterior epiglottic collapse is the most severe type of laryngomalacia, generating severe respiratory symptoms and failure to thrive. Epiglottopexy by external puncture is a new technique, certainly affordable since it does not require special instruments and it can be performed in medical centers through suspension laryngoscopy. It can be achieved alone or in combination with traditional supraglottoplasty.

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1. Introduction

Laryngomalacia (LM) is the first cause of stridor in infants and the most frequent congenital alteration of the larynx [1]. Stridor results from the collapse of supraglottic tissues into the airway. Although frequently LM is a self-limited condition at the age of 12 to 24 months, 10 to 20% of patients may require surgery due to the development of severe symptoms including progressive inspiratory stridor, respiratory distress, cyanosis, feeding disorders, failure to thrive, desaturation, and apneas [1,2].

While there are different types of laryngeal collapse, retroflexed epiglottis that touches the posterior pharyngeal wall represents the severest form of LM [3]. It is present in combination with other laryngeal changes in 24.8% of patients with severe LM, whereas, it is the only finding in 9.4% of the cases [1,4].

Supraglottoplasty is the most commonly performed surgery for symptomatic LM. However, this surgery is insufficient for the rostrocaudal displacement of epiglottis against the posterior pharyngeal wall.

The objective of this study is to present a case of severe LM with serious epiglottic collapse, satisfactorily managed by a novel technique of epiglottopexy by an external puncture.

2. Case report

A 2-month-old male was referred to our department with a month history of inspiratory stridor, accompanied by nocturnal apneas, cyanosis, alteration in swallowing and failure to thrive.

Respiratory distress was rapidly progressive, requiring intubation in the emergency room under rapid sequence sedation with difficulty for intubation, requiring three attempts to achieve it. He was extubated two days later, persisting with respiratory distress and apneas. A diagnostic laryngoscopy was performed evidencing prolapse of mucosa overlying the arytenoids, shortened aryepiglottic folds, and the epiglottis was projected inside the subglottic space during inspiration in a remarkable way (Fig. 1).

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Fig. 1. Diagnostic laryngoscopy. Prolapse of mucosa overlying the arytenoids and shortened aryepiglottic folds. Epiglottis was projected inside the subglottic space during inspiration.

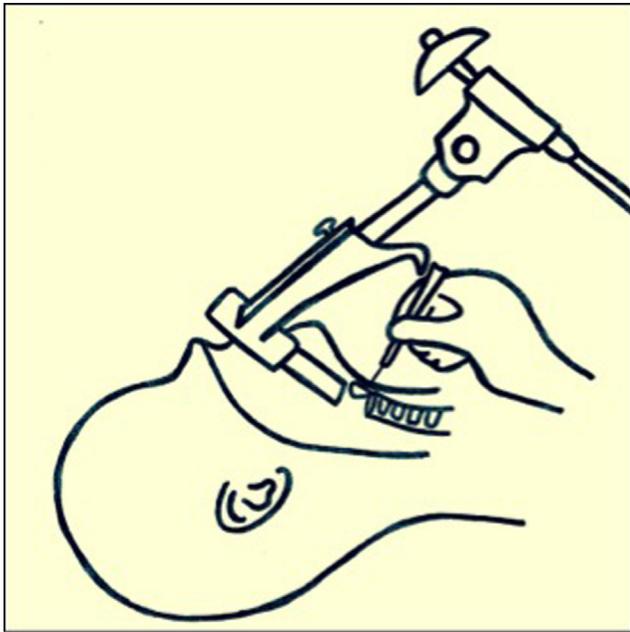


Fig. 2. Transcervical external punctures in the midline at the level of the vallecula with Prolene 2.0 through a 16 Fr metal catheter.

Informed and written consent of the parents of the patient was obtained for supraglottoplasty plus epiglottopexy. Initially, after the anesthetic induction, when trying to introduce the endotracheal tube, the rostrocaudal collapse of the epiglottis did not allow intubation in two tries. In a third attempt, video-assisted endotracheal intubation was achieved.

Suspension laryngoscopy was performed, leaving enough space to approach the anterior portion of the neck. Two surgeons performed the procedure. The first surgeon was in charge of holding the 0°–4 mm endoscopy for visualization and transillumination of the vallecula. The second surgeon performed transcervical external punctures. The first puncture was made in the midline at the level of the vallecula, through a 16 Fr metal catheter, a prolene 2.0 thread looped, crossed the base of the epiglottis (Fig. 2). The second puncture was also performed in the midline, crossing the vallecula, 0.4 inches above the first puncture, and through the catheter, a prolene 2.0 thread not looped, crossed the apex of the epiglottis (Fig. 3a). Then, the end of the unlooped thread was passed through the loop of the lower suture (Fig. 3b). Externally, the two ends of the lower puncture were pulled, so that the end of the upper thread came out to the skin. Subsequently, a U-shaped epiglottopexy suture was left, controlling the epiglottis to avoid deformation or mechanical collapse, enough to control its post-projection (Fig. 3c). The suture was tied externally in a subdermal plane by a 2-mm incision that was

sutured with Monocryl 5.0. After epiglottopexy, traditional supraglottoplasty by cold steel technique was performed.

The patient was transferred to the intensive care unit and remained intubated for 48 hours. After his extubation, he received CPAP assistance for 24 hours, with saturations above 94–99%, with a frank improvement of its respiratory status, without stridor and with an early tolerance of feeding, without aspiration events.

He was discharged after 4 days and was followed periodically by external consultation for two years, continuing asymptomatic without respiratory distress, with adequate growth and a normal neurological status.

3. Discussion

Different types of anatomical features and supraglottic collapse in children with laryngomalacia (LM) have been described. However, the two most relevant classification systems involve the static and dynamic laryngeal changes. Olney classification [1] takes into account the static findings. According to this classification, during endoscopy it is possible to find: prolapse of mucosa overlying the arytenoid cartilages (type I), foreshortened aryepiglottic folds (type II), or posterior displacement of the epiglottis (type III). Commonly, patients with severe LM may present a combination of findings of two or even all the types of LM described [2,5]. On the other hand, the dynamic changes of the larynx during inspiration that can be found according to the Groningen [4] classification are: Inward collapse of arytenoids cartilages (type 1), medial displacement of aryepiglottic folds (type 2), and rostrocaudal displacement of epiglottis against the posterior pharyngeal wall (type 3).

Retroflexed epiglottis that touches the posterior pharyngeal wall represents the severest form of LM [3]. It is present in combination with other laryngeal changes in 24.8% of patients with severe LM, whereas, it is the only finding in 9.4% of the cases [1,4].

Traditionally, severe LM is treated surgically by supraglottoplasty (SG), correcting the short aryepiglottic folds and reducing the volume of the arytenoid mucosa. SG has been shown to be a safe and effective surgery in otherwise healthy children [3,5,6]. However, in cases of type 3 LM, SG may be insufficient, while epiglottopexy (EP) is an option for the management of severe epiglottic rostrocaudal collapse.

Some surgical techniques have been described for the exclusive management of epiglottic collapse.

Traditionally, management includes a transoral suture that anchors the epiglottis to the tongue base [1,5,7] or an epiglottoplasty, excising redundant mucosa over the lateral edges of the epiglottis [8]. Fajdiga et al. [7] described the epiglottic suture to correct the pathological omega-shape epiglottis. This approach allows the modification of the epiglottis shape, although the downfall of the epiglottis towards the posterior wall is not really improved.

Sandu et al. [9] describe a modified technique of epiglottopexy using a Lichtenberger's needle carrier, a successful technique.

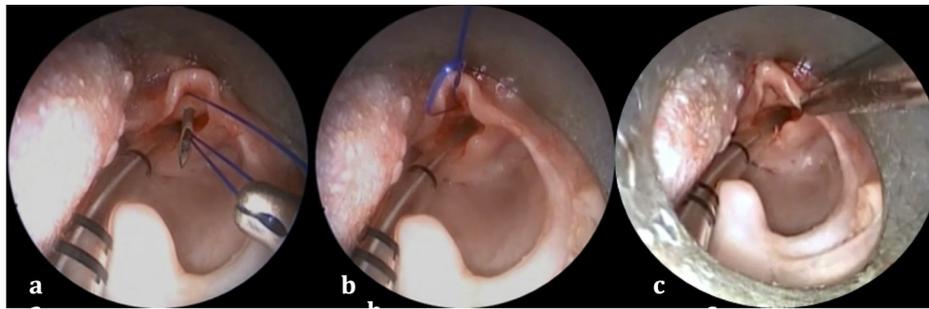


Fig. 3. First puncture with the thread looped, crossed the base of the epiglottis. The second puncture crossed the apex of the epiglottis (a). The end of the unlooped thread is passed between the loop of the lower suture (b). Externally, the two ends of the lower puncture were pulled, so that the end of the upper thread came out to the skin. U-shaped epiglottopexy suture controls the post-projection of the epiglottis, also avoiding deformation and mechanical collapse (c).

However, it requires the availability of special equipment for its achievement. EP by an external puncture is a new technique, since it does not require special instruments and can be performed in medical centers with suspension laryngoscopy.

Taking into account that less than 24.8% of patients have a type 3 LM [1,4], it is imperative to perform a diagnostic pre-surgical endoscopy, since the present surgical technique will be beneficial only in cases of epiglottic collapse. On the other hand, patients with LM usually have varied presentations and sometimes a combination of collapse types can be seen in the same patient. Therefore, EP by external puncture is a procedure that can be performed alone or in combination with SG.

Regarding anesthetic management, glottic obstruction from epiglottic collapse makes intubation difficult, as was the situation in the present case. Therefore, our recommendation is to be prepared for a difficult airway in patients with severe LM, especially in type 3 collapse [10].

Our patient did not present postoperative swallowing disorders and he did not require postoperative rehabilitation. However, as previously reported in EP, in cases of swallowing disorders, patients may require swallowing therapy with a recovery of normal feeding within a couple of days [3].

Prospective studies would be very useful not only to evaluate the effectiveness and safety of this new technique in severe type 3 LM but also its effectiveness as a treatment for concurrent obstructive sleep apnea.

4. Conclusions

Although there is no consensus regarding gold standard treatment for type 3 Laryngomalacia, the present case report shows that

epiglottopexy by external puncture is a surgery easy to carry out with good long-term results.

Disclosure of interest

The authors declare that they have no competing interest.

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