



CASE STUDY

Surgical Management of Acquired Subglottic Cysts[☆]

Tratamiento quirúrgico de los quistes subglóticos adquiridos

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Introduction

Laryngeal cysts are congenital or acquired lesions located at supraglottic, glottic or subglottic levels. They are characterised by producing obstruction of the airway and alteration in crying or in the voice.^{1,2} The incidence of congenital laryngeal cysts is 1.82 per 100 000 live births,³ while that of acquired cysts is unknown. This pathology, in spite of its low frequency, enters in the differential diagnosis of high dyspnoea in the paediatric patient.⁴

Subglottic location of laryngeal cysts is closely related with a history of neonatal endotracheal intubation, especially in preterm infants but also in full term newborns.^{5,6} There are a few cases of congenital subglottic cyst lacking antecedents of endotracheal intubation described in the literature.⁷

The aetiopathogenic hypothesis of acquired subglottic cysts is the obstruction of the drainage duct of 1 or various mucous glands induced by the process of tissue repair, with granulation, fibrosis and metaplasia, after a focal lesion

of the subglottic mucosa.⁸ In some cases, they can also be associated with other pathologies of the paediatric upper airway, such as subglottic stenosis.⁹

Despite the close relationship with endotracheal intubation, no clear association between the period of intubation and the risk of developing this pathology has been found. Cases have appeared following hours, days and weeks of intubation. The estimated time frame from the aggression to the mucosa to the growth of the subglottic cyst is normally months.⁴

The number of neonatal intensive care units has grown, bringing a consequent increase in the survival of preterm infants. According to some authors, this has led to a slight growth in the incidence of subglottic cysts.⁸

Treatment is based on transoral marsupialization of the lesion using laryngeal microsurgery instrumentation, CO₂ laser or, most recently, laryngeal microdebriders. The success rate of surgical management is high regardless of the instruments used.¹⁰

Clinical Cases

Case 1: preterm male, birth at 30 weeks, with history of meconium aspiration syndrome that required 4 days of endotracheal intubation during the neonatal period, with the symptoms being resolved completely. When the patient was 6 months old, he developed symptoms of progressive respiratory difficulty with stridor. He consequently underwent

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Figure 1 Subglottic cyst depending on the left lateral wall with obstruction of 95% of the subglottic space.

fibroscopic examination of the upper airway under sedation, at which time a cystic lesion was seen in the subglottic area, pedunculated in the left lateral wall, which obstructed the airway lumen by approximately 85% (Fig. 1).

After the diagnosis of subglottic cyst, the patient is taken to surgery, where a direct laryngoscopy is performed along with removal of the cyst including its base of implantation, using laryngeal microsurgery instrumentation (Fig. 2). Extubation is carried out in the operating room. Postoperative treatment with corticoids, aerosols with racemic adrenaline and antibiotics is instituted and the patient is discharged from hospital 3 days after the operation. The patient remains asymptomatic over the months and the check-up laryngeal endoscope exams carried out at 2 and 6 months show a normal airway.

Case 2: 2-month-old female infant with history of 42-week twin pregnancy, who presents an episode of sudden dyspnoea that required 24h of endotracheal intubation, without being able to establish a diagnosis for her pathology. When she is 7 months old, she is admitted to another hospital centre for a clinical picture of acute dyspnoea with biphasic stridor and cough. The diagnosis of severe (Cotton grade III) subglottic stenosis is given and she is treated through 2 endoscopic dilatations with balloon. Postoperative endoscopic check-ups are normal.

The symptoms of the patient reappear when she is 10 months old; she was examined in our centre for suspicion of reoccurrence of the subglottic stenosis. She undergoes endoscopic exploration of the upper airway under general anaesthesia, showing subglottic oedema and the presence of a pedunculated subglottic cystic lesion in the left lateral wall with obstruction of 80% of the laryngeal lumen. The cyst is first punctured and then resected with laryngeal microsurgery instrumentation; the same postoperative treatment is established as in the previous case and she is discharged from hospital 10 days after surgery. Two months later she undergoes endoscopic control under general anaesthesia, the presence of minimal remains of the lesion is found and the remains are only punctured. In the 8-month endoscopic control, the airway shows a normal aspect. The patient is asymptomatic at all times.

Discussion

The low frequency of subglottic cysts makes it almost impossible to know their true incidence and a great range of



Figure 2 Surgical aspects of puncture, aspiration and resection of the base of implantation of the subglottic cyst.

figures appear in the literature. Bauman and Benjamin,⁹ in a series of 19 patients with laryngeal cysts, found that 78% were multiple subglottic cysts; Lim et al.,² in a retrospective analysis of 2055 patients who underwent respiratory endoscopy, found 55 cases, making it the most extensive series published to date.

Agada et al.⁷ found a frequency of only 0.05% (7 patients), of which 8% were premature, in a cohort of 12 740 patients admitted to 2 neonatal intensive care units in the United Kingdom during a 10-year period. Watson et al.⁶ reported 14 cases of subglottic cysts in 206 patients who underwent direct laryngoscope for stridor in a 2-year period, classifying them as the fourth cause of obstruction in the paediatric airway in their experience.

Treatment of the subglottic cyst will depend on its location, size, degree of obstruction, being single or multiple, and whether it is concomitant with other pathologies of the airway.⁶ Direct laryngoscope and resection of the lesion is the treatment of choice in the majority of the cases. Ventilation control is the greatest obstacle in carrying out the procedure, with it sometimes being necessary to perform an emergency tracheotomy in some cases.⁴ As indicated earlier, the success of the procedure is independent of the type of material used, whether laryngeal microsurgery instrumentation, CO₂ laser or microdebrider. However, some authors report that the laryngeal microdebrider is superior due to its quickness and comfort when removing not only the cyst but also its base of implantation.¹⁰ In our experience, the use of laryngeal microsurgery instrumentation made it possible for us to resect the lesions, including their base of implantation, comfortably and with success. We feel that the high recurrence rate after surgical treatment found in the literature (25%–40%)^{2,6,7,10} is conditioned not by the instrumentation used, but by incomplete resection of the lesion.

Conflict of Interest

The authors have no conflicts of interest to declare.

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