

Efficacy of endoscopic removal in the treatment of subglottic ductal cysts

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ABSTRACT

Introduction. Subglottic cyst is a rare cause of airway obstruction, and there is a lack of evidence about the best treatment in the literature. This report describes our experience with endoscopic removal.

Materials and Methods. A retrospective study of all patients undergoing endoscopic removal of subglottic cyst at our healthcare facility between 2014 and 2019 was performed. Clinic and demographic data regarding gestational age, intubation, comorbidities, surgical procedures, first symptoms, endoscopic findings, treatment, and follow-up were collected.

Results. Four patients were identified. Two out of four (50%) were preterm, and all patients were intubated during their neonatal period. Median age and weight at diagnosis were 6.5 months (6-8) and 5.9 kg (3.6-7). Median time between last intubation and diagnosis was 119.5 days (71-171). Biphasic stridor and progressive respiratory distress were the most common clinical symptoms. Airway obstruction of at least 50% was found during upper airway endoscopy in all patients. Endoscopic removal was performed as a first-line treatment in 3 patients, and as a second-line treatment in 1 patient. Median intubation time after treatment and hospital stay were 13.5 hours (0-48) and 7 days (6-9). All four patients are free of disease, with a median follow-up of 17.5 months (6-42).

Conclusion. Subglottic cyst is a rare cause of post-extubation stridor. Endoscopic removal seems to be a feasible and effective treatment with a low recurrence rate.

KEY WORDS: Subglottic Cysts; Treatment; Endoscopic removal; Children.

EFICACIA DE LA ESCISIÓN ENDOSCÓPICA EN EL TRATAMIENTO DE LOS QUISTES DUCTALES SUBGLÓTICOS

RESUMEN

Introducción. Los quistes ductales subglóticos adquiridos (QDSA) son una causa rara de obstrucción de la vía aérea, sin con-

senso en la literatura en cuanto al tratamiento más eficaz. Presentamos nuestra experiencia en el tratamiento de los QDSA.

Material y métodos. Estudio retrospectivo de los pacientes tratados en nuestro centro por QDSA en los últimos 5 años. Se recogieron los antecedentes de intubación, la clínica presentada, el tratamiento realizado, la evolución postquirúrgica, el seguimiento en consulta y los controles endoscópicos.

Resultados. Se identificaron 4 pacientes, con unas medianas de edad y peso de 6,5 meses (6-8) y 5,9 kg (3,6-7), respectivamente. Todos presentaron antecedentes de intubación por intervenciones quirúrgicas, siendo prematuros 2 de ellos. La mediana entre la última intubación y el diagnóstico fue de 119,5 días (71-171). La clínica consistió en estridor bifásico con mejoría postural presentando una ocupación de la luz traqueal de más del 50% en la endoscopia diagnóstica. Se realizó escisión endoscópica (EE) al diagnóstico en 3 de los pacientes y como técnica de rescate en el cuarto por recidiva tras tratamiento con drenaje y dilatación con balón. La mediana de tiempo de intubación postquirúrgica fue de 13,5 horas (0-48) y la estancia hospitalaria de 7 días (6-9). Tras un seguimiento mediano de 17,5 meses (6-42) no se ha presentado ninguna recidiva.

Conclusión. Los QDSA son una causa poco frecuente de estridor post-extubación. La EE permite un tratamiento eficaz con baja tasa de recidiva.

PALABRAS CLAVE: Quistes ductales; Quistes subglóticos; Tratamiento; Escisión endoscópica.

INTRODUCTION

Acquired subglottic ductal cyst (ASDC) is a rare condition which originates from a lesion in the laryngeal mucosa secondary to endotracheal intubation. Typically, it presents, months after extubation, as an intermittent stridor varying with posture and potentially causing complete airway obstruction.

Since it was first described in 1968 by Wigger and Tang⁽¹⁾, case series with increasingly large samples have been published⁽²⁻⁶⁾. This has implied a higher acknowledgment of this pathology, or a real increase in incidence, owing to the higher number of preterms requiring mechanical ventilation in Neonatal Intensive Care Units in the last years.

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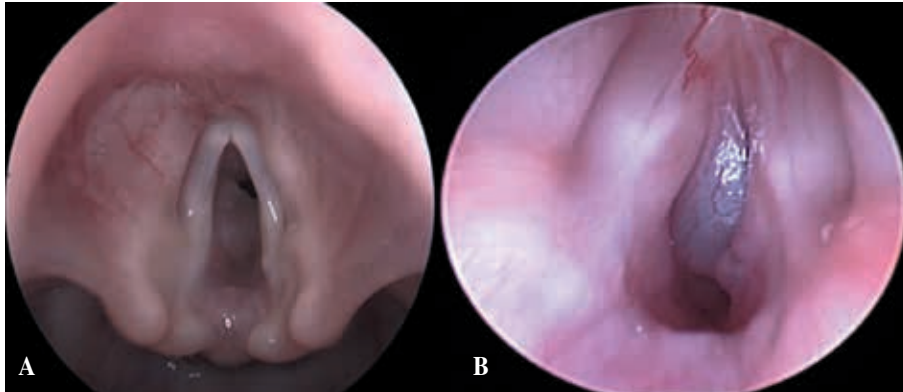


Figure 1. Subglottic ductal cysts before treatment. A) Posterior bilateral ductal cyst in Patient 1. B) Left anterolateral ductal cyst in Patient 3.

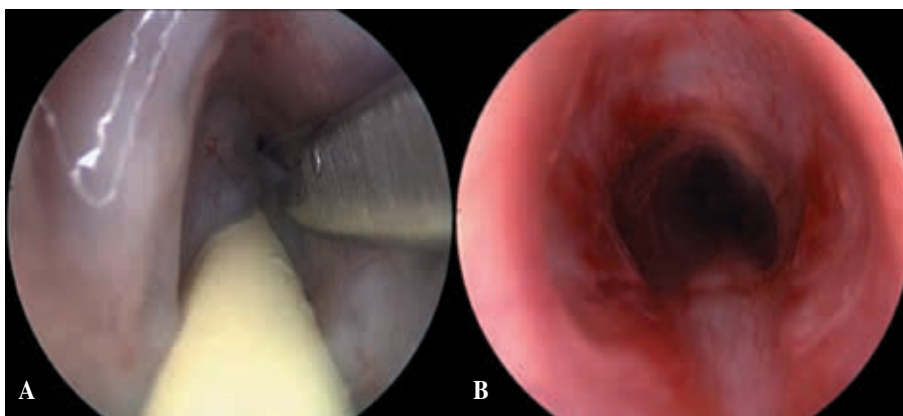


Figure 2. Subglottic ductal cysts. A) Endoscopic removal using microscissors without jet support in Patient 1. B) Immediate postsurgical result in Patient 1.

Even though ASDC needs to be surgically treated, the literature describes various techniques (puncture, balloon dilatation, CO₂ laser treatment, debridement, and removal)⁽⁶⁾, with no consensus on which is the most effective. Therefore, we would like to describe our experience with endoscopic removal.

MATERIALS AND METHODS

A retrospective analysis of ASDC cases treated in our healthcare facility over the last 5 years (2014-2019) was carried out. Patients' histories were analyzed, collecting demographic, gestational, and neonatal characteristics, comorbidities, surgical procedures, intubation history, clinical situation at diagnosis, treatment modality, postsurgical evolution, consultation follow-up, and endoscopic controls.

Endoscopic removal is performed under general anesthesia with direct vision using a 0° rigid scope. After 2% atomized lidocaine instillation in the larynx, the airway is explored under spontaneous ventilation to assess the degree of obstruction (Fig. 1). Once the cyst's origin has been located, the patient is deepened and ventilation is carried out using an intratracheal jet or a thin endotracheal tube. The cyst is removed and unroofed using laryngeal

microsurgery instruments (Fig. 2). If need be, hemostasis is achieved using adrenalin-soaked patties.

RESULTS

Four patients were assessed during the study period, with the characteristics featured in Table 1.

Only one patient was female, and 50% had prematurity history. Median age and weight at diagnosis were 6.5 months (6-8) and 5.9 kg (3.6-7), respectively.

All patients presented at least one intubation episode as a result of a previous surgical procedure in all cases. Median time between the last intubation and diagnosis was 119.5 days (71-171), with a similar initial clinical situation in all patients with biphasic stridor and progressive respiratory difficulty.

Diagnostic fibroscopy allowed the degree of airway obstruction to be assessed. 50% of the cysts were left, and the remaining 50% were posterior bilateral. No findings compatible with concomitant subglottic stenosis were found in any patient. Endoscopic and postsurgical data are featured in Table 2.

In 75% of the patients, endoscopic removal was decided upon as a first-line treatment, de-obstructing the

Table 1. Patient demographic and clinic characteristics.

Patient	Gestational age	Sex	Age at diagnosis	Weight at diagnosis	Previous intubations	Days since last intubation	Cause of last intubation
1	32 weeks	F	6 months	6.3 kg	2	171	Surgery
2	39 weeks	M	6 months	5.5 kg	4	139	Surgery
3	30 weeks	M	7 months	3.6 kg	1	71	Surgery
4	38 weeks	M	8 months	7 kg	1	100	Surgery

Table 2. Endoscopic findings, treatment, evolution, and follow-up.

Patient	Obstruction	Location	Treatment	Intubation	Hospital stay	Recurrence	Follow-up
1	>50%	Posterior bilateral	Removal	0 hours	7 days	No	6 months
2	50%	Posterior bilateral	Dilatation	48 hours	6 days	Yes	1 month
			Removal	3 hours	6 days	No	5 months
3	>75%	Left anterolateral	Removal	24 hours	9 days	No	36 months
4	50%	Posterior left	Removal	24 hours	7 days	No	42 months

airway and allowing for early extubation. In Patient 2, drainage and balloon dilatation had been previously carried out by another department, with cyst recurrence being detected one month following the procedure. Endoscopic removal was decided upon as a salvage therapy, with a good result and early extubation. No recurrence was noted in the remaining patients during the median 17.5-month follow-up (6-42).

DISCUSSION

This study includes a series of 4 ASDC patients treated with endoscopic removal and assessed over a 5-year period. Patients had similar characteristics as in other series published⁽³⁻⁶⁾ regarding prematurity history, intubation, and time to symptom development. Endoscopic removal proved safe and effective, and it had a low recurrence rate during follow-up.

Even though ASDC physiopathology is little known, these cysts seem to have a similar etiology as subglottic stenosis^(5,7). The damage caused by intubation and posterior subepithelial fibrosis in the laryngeal mucosa prevents the correct evacuation of submucosal glands, thus entailing ductal dilatation and cyst formation. This physiopathological mechanism could explain the time lag typically seen between intubation and symptom onset. The location of these lesions is consistent with their iatrogenic origin, given that they occur in the subglottic region (the narrowest area in pediatric airways), mostly on the left side – intubation takes place from the right side.

Diagnostic delay is caused by various factors – physiopathological mechanism, occult clinical course, and symptoms compatible with other pathologies such as bronchiolitis typically occurring in the same age band. Median time from intubation to diagnosis in our series was 119.5 days, consistent with other series such as Lim et al.⁽²⁾ (210 days), Agada et al.⁽⁴⁾ (120 days), Watson et al.⁽³⁾ (127.5 days), and Halimi et al.⁽⁶⁾ (246 days).

In 2003, Lim et al.⁽²⁾ published a retrospective study with 55 cases of ASDC – the largest series published up until then. In their series, they described a recurrence of 43% throughout the median 6-month follow-up, following CO₂ laser treatment or microsurgery. However, data were not stratified according to treatment modality. Watson et al.⁽³⁾ described a recurrence of 50% following marsupialization treatment, with a 4-month follow-up. Agada et al.⁽⁴⁾ described a recurrence of 29% following cyst marsupialization. And Halimi et al.⁽⁶⁾ described a recurrence of 65% following CO₂ laser treatment with a mean 3-year follow-up.

In our study, which had a similar follow-up as that of the aforementioned works, no patient undergoing endoscopic removal presented with ASDC recurrence. This treatment allows cyst content to be freed, de-obstructing the airway and the gland duct, and causing no damage to the underlying mucosa. This could explain the low recurrence rate observed as compared to other established modalities such as CO₂ laser^(2,8), progressively allowing for an earlier extubation thanks to the decrease in the airway edema associated with the procedure.

Our study has some limitations – it is a retrospective study and it features a small number of patients, consid-

ering the pathology's nature and the lack of information regarding previous intubation, given that the 4 patients were intubated in other healthcare facilities.

Even if endoscopic removal seems an effective treatment, comparing it with similar studies proves difficult given its low incidence, the variability of techniques available, and the lack of stratification according to treatment modality. To answer that question, a randomized clinical trial would be required; and considering the pathology's low frequency, it should be a multicenter one. But our results are promising.

Optical coherence tomography allows us to learn more about the evolution of intubation damage in the laryngeal and tracheal mucosa in a non-invasive fashion^(9,10), opening up new research and treatment possibilities regarding this pathology and other airway conditions. Probably, in the near future, we will be able to predict which patients can develop ASDC or ASDC recurrence, and find out which is the most effective treatment in an experimental model.

CONCLUSION

ASDC is a rare cause of airway obstruction which should be included in the differential diagnosis of patients with stridor and intubation history along with subglottic stenosis. Endoscopic removal seems to be an effective treatment with a low recurrence rate and few complications.

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