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Case Report

A Rare Case of Neonatal Laryngeal Cyst Presenting with Stridor

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Abstract

Laryngeal cysts are a rare cause of stridor. Only a few cases have been reported in the literature. This is a case of 19 days old infant presenting with stridor and feeding difficulty, successfully treated with direct laryngoscopic marsupialization of the cyst. The role of flexible laryngoscopy and CECT neck is pivotal. The child had an uneventful post-operative course and breathing and feeding improved.

Keywords: Stridor; Laryngeal Cyst; Laryngomalacia

Introduction

Neonatal laryngeal cyst is an extremely rare occurrence, which is a fluid-filled cyst present in the larynx. Pediatricians and otolaryngologists should be aware of this problem, as nearly 50% of reported cases are diagnosed at autopsy [1]. It can cause stridor, respiratory distress and obstruction. Severe narrowing of the infant's airway requires urgent intervention. Laryngeal cysts are usually located at the level of supraglottis. Congenital laryngeal cysts of the vocal cords and subglottis region are extremely rare with very few cases reported in the literature. Congenital laryngeal cysts may cause upper airway obstruction in neonates that can lead to serious morbidity and mortality if diagnosed and treated late. The annual rate of this condition is 1.82 per 100,000 live births [2].

Santo., *et al*. in 1970 distinguished 2 types of congenital laryngeal cysts – ductal and saccular. Laryngeal duct cysts result from obstruction of the laryngeal epithelial mucosal glands and account for 75% of congenital laryngeal cysts. A saccular cyst is a retention of mucus in the larynx due to obstruction or narrowing of the laryngeal opening of the ventricles [3]. The most common symptoms caused by laryngeal cysts are stridor, dysphagia, and cyanotic spells. Diagnostic modality of choice is direct laryngoscopy. The treatment is always emergent and includes marsupialization of the cyst.

The most common cause of stridor in infants is laryngomalacia. Because of similar symptoms, congenital laryngeal cysts are often misdiagnosed as laryngomalacia [4]. This leads to serious morbidity and mortality if there is delay in diagnosis and treatment [5]. In cases of laryngomalacia, the problem resolves on its own within the first 2 years of life, whereas laryngeal cysts will require early surgical intervention.

Case Report

A 19 day old female infant was admitted to the neonatal intensive care unit (NICU) with feeding difficulty and respiratory distress (intercostal retraction). She was born via normal and spontaneous vaginal delivery at 33 weeks of gestation after an uneventful pregnancy. Birth weight was 2.5 kg, with APGAR score 7/10. She weighted 2.1 kg, heart rate was regular without added sounds, abdomen was soft and non-tender, bilateral air entry was reduced, upper chest retraction was present, anterior fontanelle was at level, cry was normal and neurological examination was also normal. Routine blood workup was sent and the child was started on 10 lit oxygen via face mask.

At 21 days of life, flexible fibreoptic laryngoscopy was carried out revealing a cyst on left aryepiglottic fold pushing the epiglottis on right side. The scope couldn't be negotiated further down into the glottis. The cyst was mobile on respiration and thus some airway was present. In this view tracheostomy was delayed and for further evaluation contrast enhanced computed tomography of neck (CECT NECK) was performed, confirming a well-defined fluid filled cystic lesion approx. 8x12x9 mm seen at level of glottis, causing significant airway narrowing. No other congenital malformation was elucidated. Otolaryngological referral was done and child was planned for surgery the next morning.

At 22 days of life the child was shifted to the operation theatre and videolaryngoscopic intubation was done under general anaesthesia. Direct laryngoscopy was carried out revealing a cyst occupying the supraglottis. It was seen involving the left aryepiglottic fold and false vocal cord (Figure 1). It extended medially into the glottis and posteriorly in the left pyriform sinus and post cricoid area. The cyst was soft in consistency and with gentle probing right true vocal cord could be visualised. The subglottic area appeared normal.

The cyst was grabbed with straight cup forceps and marsupialization was done with left curved scissors. Clear fluid was expressed and suctioned. The redundant flimsy cyst wall was excised till the left aryepiglottic fold. The child was shifted to NICU with endotracheal tube and Ryle's tube. Extubation was done the next day and the recovery was uneventful. Respiratory distress and feeding problem improved after excision of cyst.

Figure 1: Blue arrow shows direct laryngoscopic view of left laryngeal cyst occupying the supraglottis and completely involving the epiglottis and left aryepiglottic folds.

Discussion

Congenital laryngeal cysts are a rare cause of neonatal airway obstruction, although the majority of neonates with laryngeal cysts have been reported to have concomitant laryngeal molluscum contagiosum [5]. Santo in 1970 distinguished two types of congenital laryngeal cysts - tubular and saccular. Laryngeal cysts develop either as embryological malformations or as a result of mucinous duct obstruction [3]. In a retrospective study done by Sakakura., *et al*. laryngomalacia was found to be the most common congenital laryngeal anomaly. It was followed by subglottic stenosis and vocal cord paralysis [6]. Congenital laryngeal cysts are very rare with an annual incidence of only 1.8 per 100,000 live births.

Severity of symptoms and timing of laryngeal cysts are related to location and size of the cyst [7]. A larger cyst can present at birth with an airway obstruction, while smaller cysts can cause more subtle breathing problems, while eating and talking. The most frequent symptom is stridor that mimics laryngomalacia. It may be associated with muffled crying, hoarseness, cyanosis and difficulty in feeding. In contrast, laryngomalacia almost never causes voice change [8]. In 1961, Birch studied 200 randomised infants and children with stridor at the London Hospital and found only 4 infants with laryngeal cysts [2]. It should be noted that symptoms such as poor feeding, cyanosis at suckling and growth retardation may be the only symptoms that are easily overlooked [9,10].

All infants should be investigated for wheezing upon birth. Classically, a series of investigations including lateral neck

radiographs, followed by direct laryngoscopy under general anesthesia have been advocated as the gold standard [11,12]. Diagnosis of congenital vocal cysts is based on symptoms and clinical course. To make a quick and sure diagnosis of neonatal wheezing, flexible laryngoscopy should be performed. Imaging tests may also suggest the initial diagnosis. Although lateral neck X-rays show most cysts, CT and magnetic resonance imaging are preferable because they provide a better definition of the lesion in terms of size, location, extent, and involvement of neighbouring structures as well as airway compromise. Needle aspiration can confirm diagnosis and more a definitive treatment should be marsupialization of the cyst. However, the cyst can easily reappear immediately after this procedure, but in our study, no recurrence was found after needle aspiration.

Immediately after diagnosis, all patients should be carefully examined and operated under general anesthesia. If difficult intubation is anticipated, access to the laryngeal inlet is facilitated by initial decompression of the cysts with a large-bore needle or by retrograde percutaneous intubation [10,11]. Tracheostomy should be avoided as far as possible for the risk of tracheal stenosis in future. Choice of definitive surgical treatment remains controversial. In general, the cysts can be removed via endoscopic or an external approach. Endoscopic procedure including marsupialization, excision and de- roofing have been practiced as primary treatment while excision through laryngo-fissure or via a lateral cervical approach with or without tracheostomy is reserved for recurrent or deep seated ventricular cysts [12]. Endoscopic drainage is a safe and effective primary treatment for most neonatal laryngeal cysts [2].

Marsupilization and excision are equally effective in relieving obstruction and neither approach leads to recurrence. The time for cyst recurrence after aspiration varies from 5 to 10 days. De-roofing led to cyst recurrence within 1 to 10 months [13]. There was no recurrence after complete excision of the cyst wall with CO_2 laser and endoscopic micro suturing as was reported by Mitchell., *et al*. The most common symptom of congenital laryngeal cyst is stridor. The CO_2 laser was first used in 1984 to ablate or vaporize the wall of the cyst. As the cyst grows, the stridor may increase and be accompanied by changes in voice, periods of cyanosis, retractions, and difficulty feeding [11,14].

Leading cause of stridor in infants, neonates and young children is laryngomalacia, which account for 60-70% of cases. However, laryngomalacia never leads to a change in voice quality.8 Another difference is that in laryngomalacia stridor reduces in the prone position, whereas with laryngeal cysts, the symptoms are reduced when the patient is lying on the affected side [15].

Vito forte., *et al*. 2004 introduced a new classification system to better assess the origin of these cysts and guide successful surgical management. Simple supraglottic and subglottic mucosal cysts pose no diagnostic or therapeutic challenge and have been successfully treated with a single endoscopic resection or marsupialization. The remaining congenital cysts in the study were considered more complex and all required open surgical approach [16].

Conclusion

Congenital laryngeal cysts are rare lesions occurring during the neonatal period. They present with stridor and respiratory distress. Physical examination, such as palpation of the neck as well as imaging with CT or MRI, is necessary for diagnosis, grading, and planning for surgical resection. Endoscopic marsupilization is a simple, effective and adequate treatment for this disease. Special emphasis is laid upon the role of flexible laryngoscopy in neonatal period to diagnose the cause of stridor.

Ethical Clearance

Obtained from institutional ethical committee.

Informed Consent

Obtained from the patients parents.

Financial Disclosure

NIL.

Conflict of Interest

NIL.

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