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A STRIDULOUS INFANT WITH FEEDING DIFFICULTY: A CASE REPORT OF CONGENITAL LARYNGEAL SACCULAR CYST.

Chun Tau NG[1,2], Siti Hajar SANUDIN[2], Halimuddin SAWALI[2], Aun Wee CHONG[1]

[1] Department of Otorhinolaryngology, University Malaya, Kuala Lumpur, Malaysia.
[2] Department of Otorhinolaryngology, Hospital Queen Elizabeth, Sabah, Malaysia.

ABSTRACT

Laryngeal cyst in pediatric age group is an infrequent condition. It frequently presents with laboured respiration, respiratory distress during feeding, hoarseness, feeble or absent cry. Herein, we report a case of congenital laryngeal saccular cyst, which was successfully treated with transoral endoscopic cold instrumental excision. This case highlights the significance of suspicion in stridulous infants with feeding anomalies which warrants an early referral to otolaryngologist.

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Laryngeal cyst in pediatric age group is an infrequent condition. It frequently presents with laboured respiration, respiratory distress during feeding, hoarseness, feeble or absent cry. Herein, we report a case of congenital laryngeal saccular cyst, which was successfully treated with transoral endoscopic cold instrumental excision. This case highlights the significance of suspicion in stridulous infants with feeding anomalies which warrants an early referral to otolaryngologist.

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INTRODUCTION

Congenital laryngeal cyst is an unusual laryngeal pathology that occurs during early infancy and neonatal period. The incidence of congenital laryngeal cyst range from 1.4 to 1.8 in 100,000 newborns.[1,2]

Congenital anomalies of the larynx often constituents a wide variety of pathological conditions. Among the variety of congenital anomalies of the larynx, laryngomalacia is most commonly found, followed by subglottic stenosis and vocal cord paralysis.[1,3] Congenital laryngeal cyst is considered rare, which only represents 1.5% of all congenital laryngeal anomalies.[4] We described a case of laryngeal saccular cyst in an infant successfully treated with endoscopic cold instrumental excisionwith good outcome.

Corresponding author: Chun Tau NG (MBBS, MRCSEd (ENT)), Hospital Queen Elizabeth, 13a, Jalan Penampang, 88200 Kota Kinabalu, Sabah.
Email: ronn_90@hotmail.com;
Telephone: +60167988802

CASE REPORT

A 1 year 3 months old baby girl, born preterm at 30 weeks was referred to our centre for intermittent noisy breathing since the age of 6 months old which was aggravated by activity. Antenatal history was uneventful. There was no history of intubation since birth.

She was noted to have aspiration symptoms and cyanosis during feeding on several episodes. Her cry was otherwise strong and weight was striving well.

Upon examination, she was well under room air with soft inspiratory stridor. Flexible nasopharygolaryngoscopy noted a cyst-like mass between the right false cord and vocal cord, crossing midline and partially obstructing the airway. Other laryngeal structures were normal. There was no palpable mass over the neck.

Contrasted Computed Tomography (CT Scan) of Neck showed a fairly well-defined hypodense lesion with faint perceptible peripheral enhancement of lesion at the supraglottic region, measuring 0.8 x 1.0 x 0.7cm (AP x W x CC) (Figure 1a and 1b). This lesion caused displacement of right true cord medially and airway narrowing at the level of C2. The rest of the visualized tracheal was normal.

The patient subsequently underwent endoscopic excision under general anaesthesia. Intraoperatively, there was a cystic mass over the right laryngeal ventricular saccule, lateral to the right true cord (Figure 2a). Hydro dissection was done with saline and cystic wall ruptured intraoperatively. The entire cystic wall was excised in total (Figure 2b). Postoperatively, she was kept intubated for a day in Paediatrics Intensive Care Unit and completed IV Dexamethasone 0.15mg/kg TDS for 3 days. She was well post-op without signs of respiratory distress and discharged on day 4 post-operation. There is no recurrence noted upon 2 years of follow-up to date.

DISCUSSION

Congenital laryngeal cysts are generally categorized into 3 categories: ductal, saccular and thyroid-cartilage foramen cyst.[5] They are commonly situated at lateral supraglottic, distinctly beneath normal laryngeal mucosa. Extralaryngeal extension of the cyst is also possible, extending to the anterior cervical triangle via foramen of superior laryngeal bundle in the thryrohyoid membrane.[5]

Ductal cysts can be found at any site within larynx due to retention of mucin in the collecting ducts of submucosal glands. It accounts for the majority of cases, most com-

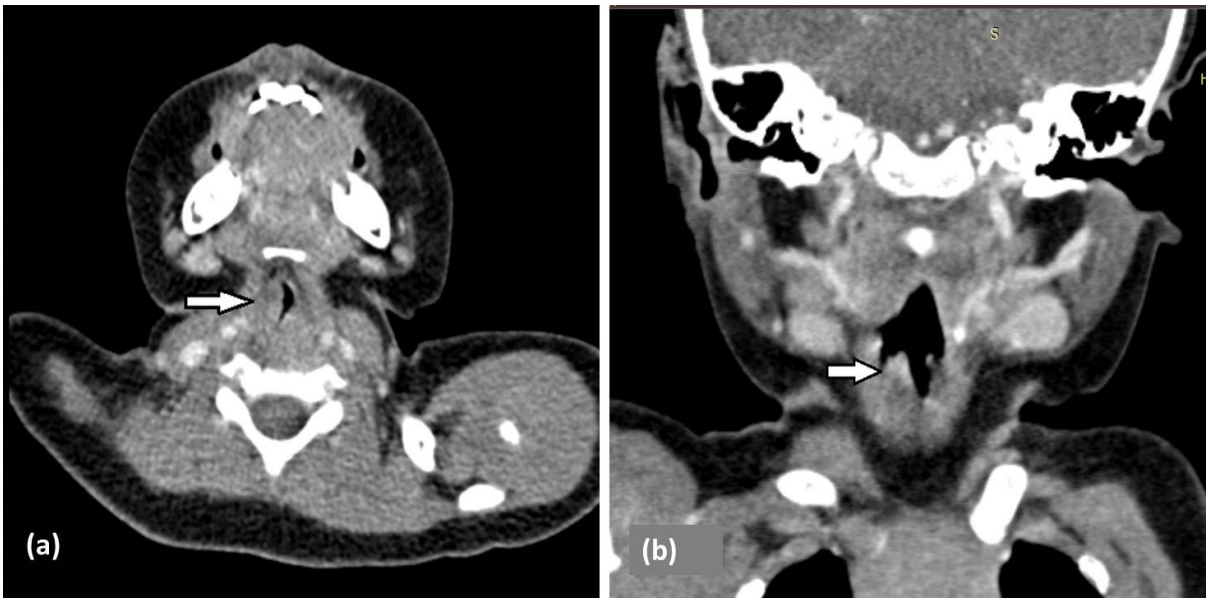

Figure 1: CT scan, transverse section (a) and coronal section (b), show fairly well-defined hypodense lesion with faint perceptible peripheral enhancement of lesion at the supraglottic region, causing narrowing of airway (white arrows).

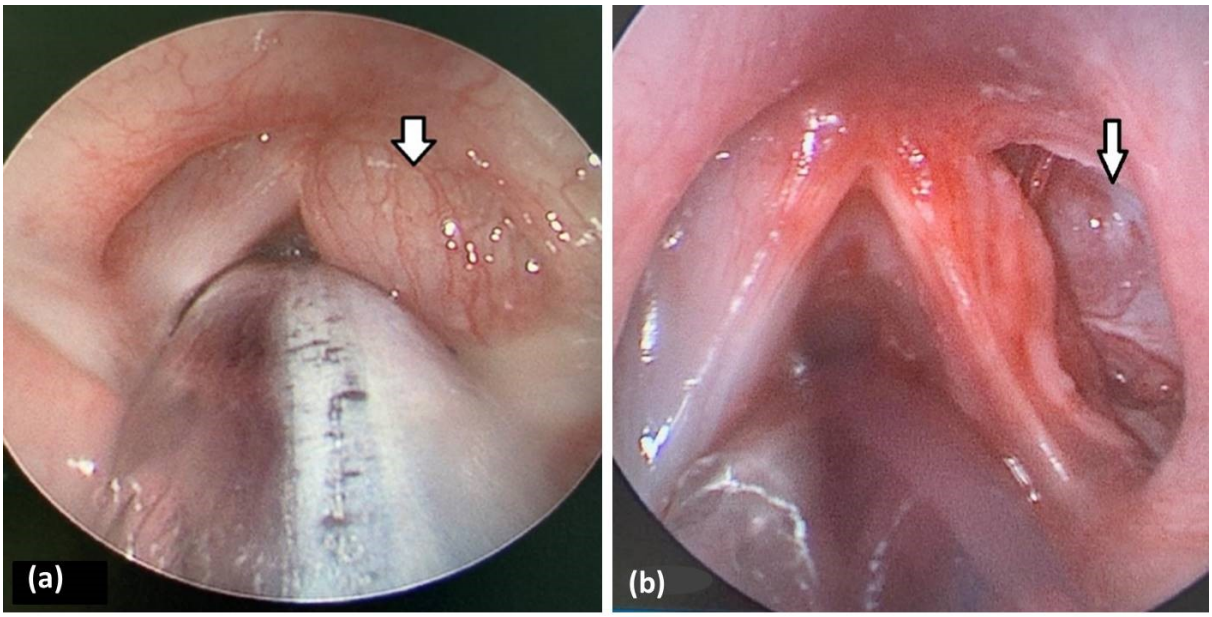

Figure 2: (a) shows a bulging cystic mass over right laryngeal ventricle (White arrow), (b) shows a cavity over right laryngeal ventricle and saccule post excision (White arrow).

monly at vallecula, originating from the obstruction of laryngeal epithelial mucus gland.[6] Saccular cysts are those supraglottic lesions located at the anatomical plane of saccule, a structure that produces mucus and lubricates vocal cord.[7] It originates from the blockage or atresia of laryngeal ventricular orifice, causing retention of mucus, often due to chronic inflammation.[8] They can be distinguished from laryngocele, which is a dilatation of laryngeal saccule containing air, while saccular cyst contains mucus. The third group is thyroid-cartilage foraminal cyst, which is a rare and distinct category of the entity.[5,6]

Congenital saccular cyst which often presents early after birth, are dangerous for newborns. It frequently presents with laboured respiration, respiratory distress during feeding, hoarseness, feeble or absent cry, or a cystic mass in the neck.[5] Pak MW et al suggested a majority of onset of symptoms within the first 2 weeks of life, with most patients (56%) presented on the first day of life.[1] However, in our case, symptoms were not apparent before the 6th month of life. Thus, late onset of similar symptoms should not exclude the suspicion of laryngeal cyst as the cyst may not exhibit any symptoms before growing to a significant size which would have caused significant upper airway narrowing. It is reported that the size of cyst has to reach 1.5 cm to be clinically symptomatic.[1] Larger laryngeal cysts pose more serious consequences if left untreated as they may lead to airway obstruction and eventually death. Smaller cysts may present later after birth as seen in this case, with feeding difficulty, muffled cry, hoarseness and cyanotic episode.

The initial detection of saccular cyst is mainly based on clinical suspicion. Traditionally, infants with such airway and feeding anomalies are offered a series of investigations which includes lateral radiograph of neck, Cincinnati's view of the trachea, barium swallow followed by direct laryngoscopy under general anaesthesia.[1] From our experience, bedside flexible nasopharyngolaryngoscopy is the best tool to provide a rapid and definite diagnosis in working up for a stridulous child. It offers a prompt diagnosis without exposure to radiation and the requirement of general anaesthesia. It was reported

that 35.7% of these cases were mistaken as laryngomalacia as they often share common clinical presentations.[9] Thus, all infants who exhibit stridor, feeding anomalies or cyanosis should raise alarm and suspicion among all disciplines especially primary care providers. Our case highlights the importance of initiating an early referral to otolaryngologist for assessment and transnasal endoscopic examination. This would encourage early diagnosis and prompt management before severe complications occur. Generally, diagnostic CT scan or MRI are not necessary in most simple cases. However, they do provide a clearer assessment and definition of the lesion by offering information regarding its size, location, extension, anatomical relationship and airway assessment.[9]

There are a variety of treatment strategies reported in the literature to manage congenital saccular cysts. Generally, there are 2 main types of operative approaches: open-neck excision and endoscopic management. Endoscopic management includes endoscopic aspiration, marsupialization with cold instruments, CO2 laser ablation and extended ventriculotomy. Due to the rarity of such cases, the consensus on the best treatment strategy remains challenging to achieve.[6,10] The decision on the suitable treatment modality should be based on the expertise and availability of instruments in respective centres. Some authors suggested an open excisional approach is required for definitive treatment as there is evidence of high recurrence rate for endoscopic treatment.[11,12,13] Civantos et al suggested multiple repeated endoscopic procedures were required in their study (up to an average of 6 procedures per patient) as the recurrence rate was high. The details of surgical techniques were not mentioned in the article. It was reported that repeated endoscopic procedures and endotracheal intubation further led to scarring, subglottic stenosis and requirement for tracheostomy.[13] On the other hand, there is conflicting evidence suggesting endoscopic treatment is adequate to achieve effective cures without recurrence.[1,6,14] Moreover, Pak et al suggested endoscopic deroofing is as effective as endoscopic excision. It is technically simpler, thus should be the treatment of choice.[1] Kirse DJ et al suggested endoscopic ventriculotomy with the excision of false cord and ventricular mucosa as definitive treatment.[14]

Our case was successfully managed with endoscopic excision using cold instruments without recurrence after 2 years of follow-up. Before excision, we injected saline surrounding the cyst as a form of hydro dissection. This greatly eased the process of excision. We believe that endoscopic excision with cold instruments helps to reduce the risk of collateral thermal damage to surrounding anatomical structures and minimise postoperative laryngeal oedema. It offers an advantage over endoscopic marsupialization with CO2 Laser, considering the latter's limited availability in hospitals due to cost; has higher risk of thermal injury and airway combustion.[6] Compared to open approaches, our approach of endoscopic excision has shorter anaesthetic duration, significantly lower risk of superior laryngeal nerve injury and is cosmetically more appealing.[10,14] It is advisable to keep patients intubated with secured airway postoperatively in infants due to potential airway oedema secondary to intraoperative manipulation. Needle aspiration remains a temporary measure in life-threatening upper airway obstruction as the recurrence is high after this procedure. The mucous glands in the epithelial lining of the cyst are believed to continue to secrete mucus following needle aspiration, causing recurrence.[6,14] We suggest needle aspiration to be performed only as an emergency measure to reduce the size of cyst for intubation, but not as a definite treatment.

CONCLUSION

Though rare, congenital saccular cyst should be considered in infants presenting with stridor and feeding difficulties. All stridulous infants warrant early referral to otolaryngologist for flexible nasopharyngolaryngoscopy for assessment. Endoscopic excision with cold instrumentation should be considered as one of the treatment choices as it potentially provides advantages over endoscopic laser CO2 ablation and external approaches.

CONFLICT OF INTEREST DISCLOSURES

There is no conflict of interest to be disclosed by all authors.

CONSENT

We have acquired consent for the above images to be used for publication purpose.

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