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CASE REPORT

Saccular cyst – Cause of intermittent stridor in a child: A case report

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Abstract Saccular cyst is a rare lesion of larynx causing respiratory obstruction and stridor in neonates and infants. Herein we present a case of a 4 year old who presented to us with intermittent respiratory distress and stridor. Fibreoptic bronchoscopy showed the presence of saccular cyst overhanging the anterior part of right vocal cord. Endoscopic marsupialization of cyst is done using laryngeal microdebrider with stripping of cyst lining. Saccular cyst should be considered in the differential diagnosis of children presenting with intermittent stridor.

1. Introduction

Stridor is a high pitched, musical or harsh sound produced as a result of turbulent air flow due to obstruction at one or more levels of the airway. Saccular cysts, although rare, are an important cause for stridor, particularly in the paediatric age group. They are abnormal dilatations of the saccule, which is the antero-superior portion of the laryngeal ventricle. These dilatations if large invade the supraglottis leading to stridor, sometimes so severe as to amounting for an urgent tracheostomy. They may be either congenital or acquired. Congenital cases present shortly after birth. Their incidence is 1.82 cases per 1,00,000 live births.[1] Acquired cases may result as a cause of prolonged intubation, neoplasm, trauma or laryngeal surgery.[2] They are usually symptomatic at birth or infancy but 50% saccular cyst may be asymptomatic and may detected later in life or may be found only during autopsy.[3]

We report herein, a case of a 4 year old child who presented with a history of intermittent respiratory distress and stridor, diagnosed as saccular cyst of larynx arising from the anterior part of right ventricle and managed by endoscopic marsupialization. This case report aims to show the relevance of saccular cysts in the differential diagnosis of children presenting with a history of intermittent stridor.

2. Case report

A 4 year old male child presented to the paediatric emergency of our institution with complaints of noisy breathing and hoarseness of voice for 3 months. It used to resolve on its own intermittently but the symptom had increased in severity for last 1 week after the episode of upper respiratory tract infection. There was no history of feeding difficulty, fever, sore throat, positional change. Child was maintaining a saturation

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of 92–95% on oxygen. The antenatal and birth history was uneventful.

Examination revealed that child had biphasic stridor at the time of presentation. There was severe suprasternal and intercoastal and substernal retractions. Pectus excavatum was noted on general physical examination. Lateral view radiograph showed a faint shadow at the level of glottis (Fig. 1a. An urgent contrast enhanced computed tomography of the neck was ordered which showed an enhancing lesion in the area of the aryepiglottic fold with the airway being grossly normal (Fig. 1b).

Child was shifted to the operating room for direct laryngoscopic examination under general anaesthesia. However, we were unable to intubate the child and in view of dipping saturation an emergency tracheostomy had to be done. Awake fibreoptic laryngoscopy revealed a globular, smooth mucosa covered bulge in the supraglottis at the level of false cords compromising more than 90% of the airway. Lesion appeared to be arising from the right ventricle (Fig. 2). The cyst was marsupialized under general anaesthesia using laryngeal microdebrider followed by ventriculotomy using rigid laryngoscopy (Fig. 3). Cyst wall was removed till its base in the ventricle. Post-operative radiograph showed no lesion. Child was decannulated on the 2nd post op day and discharged with normal breathing and voice.

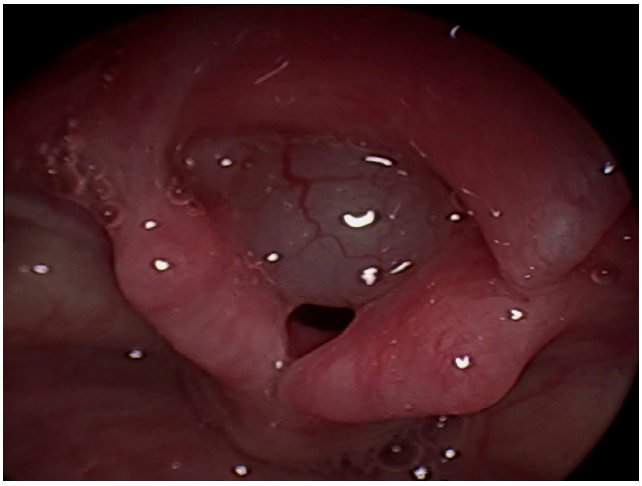

Figure 2 Fibreoptic laryngoscopy showing cyst arising from anterior part of right ventricle protruding into laryngeal lumen.

3. Discussion

Saccular cysts are mucus-filled dilatations of the laryngeal saccule. The function of the saccule is to provide lubrication to the vibrating vocal cords with mucus produced by its epithelium. Continuous secretion of mucus in a blocked orifice of the laryngeal ventricle is the basic mechanism of saccular cyst formation.[3] Saccular cyst may be congenital or acquired. Congenital saccular cyst occurs due to atresia of saccular orifice or because of abnormal migration of fourth branchial arch which forms sequestrated cyst.[4] They may be *anteriorly* placed (anterior saccular cyst), arising from anterior ventricle and extending medially into the laryngeal lumen between the true and false vocal cords as seen in the present case. Lateral saccular cyst are *laterally* placed and extend from the false cord and expands posterosuperiorly into the aryepiglottic fold and may bulge into pyriform sinus.[5]

The usual presentation is an infant or a young child with weak cry, hoarseness of voice, noisy breathing, occasionally throat pain and feeding difficulties. Important differential diagnoses include laryngomalacia, laryngocele, hemangiomas, hamartomas, ductal cysts, etc.[1] Although there are similarities between laryngocele and saccular cyst, they are differentiated

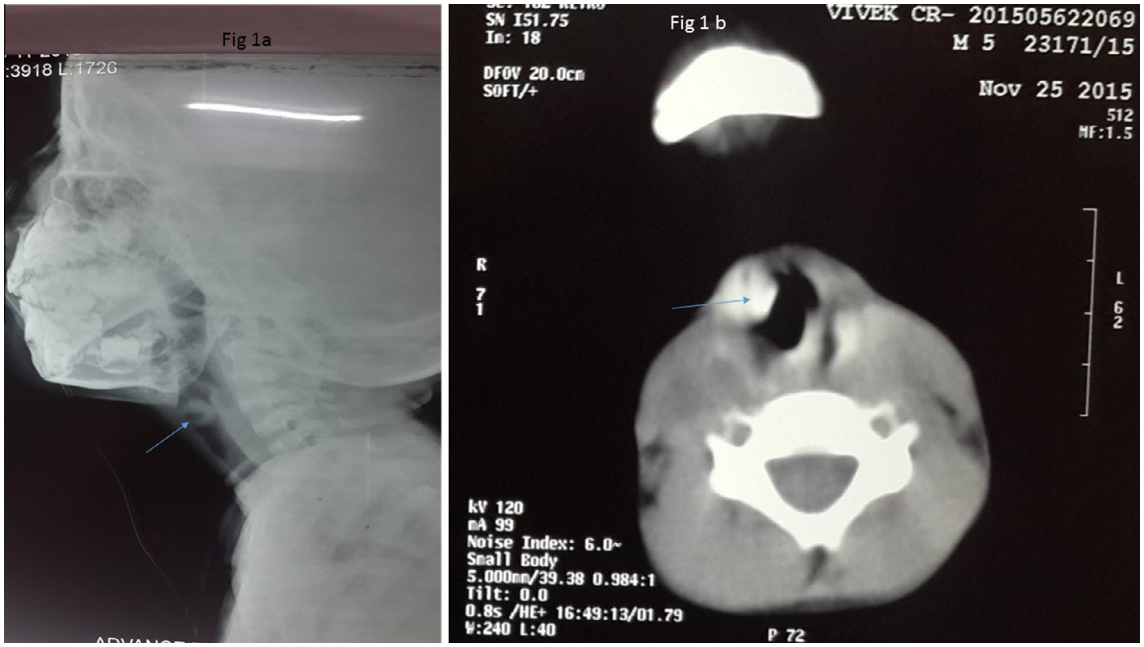

Figure 1 (a) Lateral radiograph showing faint hypodensity at the level of glottis. (b) CECT neck showing enhancing lesion in the area of the aryepiglottic fold.

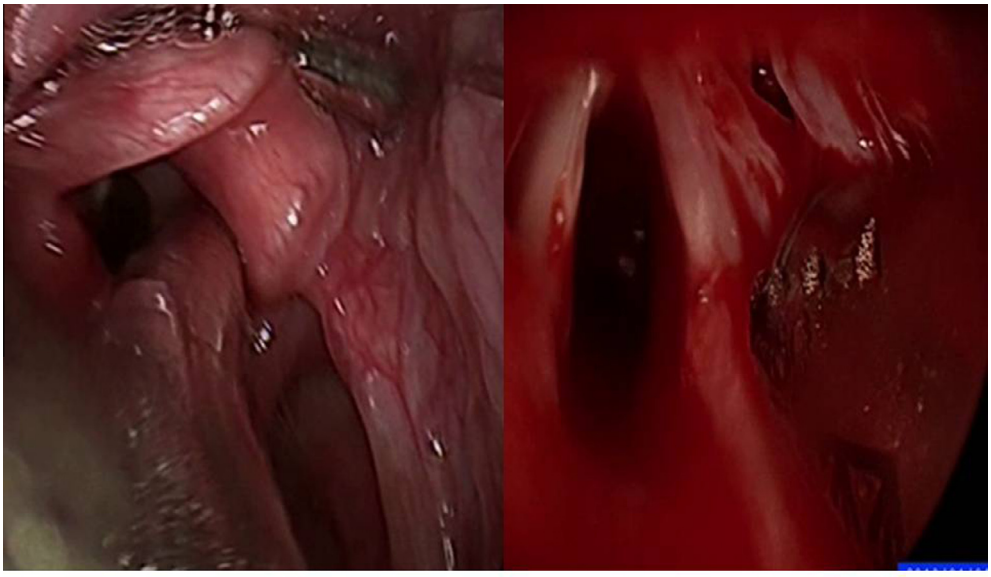

Figure 3 surgical procedure – marsupialization of cyst wall using laryngeal microdebrider.

by the absence of an opening into the larynx in the latter. Additionally, laryngoceles are usually air-filled rather than mucus-filled. Also, laryngoceles tend to cause episodic symptoms, with respiratory distress typically becoming worse on crying.

Diagnosis is confirmed by direct visualization of the larynx by fibreoptic laryngoscopy or direct laryngoscopy under general anaesthesia. Saccular cysts typically are seen in the supraglottic larynx as a smooth mucosa covered bulge, occluding the airway. The aryepiglottic folds, ventricle, false cords, arytenoids may be distorted. A preoperative CT scan can also be done for better surgical planning. Treatment options include cyst aspiration, endoscopic CO2 laser ablation, marsupialization and external ventriculotomy.[6] In 1978, Hollinger et al.[7] suggested cyst aspiration as a treatment for this condition, however due to high recurrence rates, this was superseded by other methods. Laser ablation of the cyst wall and marsupialization are commonly used procedures of choice in primary cases. However, in recurrent cases, external approaches via laryngofissure or lateral cervical approach might have to be used for complete removal.[8]

In conclusion, saccular cysts, although uncommon, are important causes of stridor in the paediatric age group. Even deaths due to sudden asphyxiation have been reported. A thorough history and physical examination with prompt investigations and early surgical intervention need to be undertaken to prevent morbidity and possible mortality as a result of saccular cysts.

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