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Congenital Vallecular Cyst in a Two-Month-Old Boy: A Case Report

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ABSTRACT

Vallecular cysts are an uncommon cause of airway obstruction in children. It consists of glandular respiratory epithelium lined by squamous epithelium. The diagnosis of this rare entity is often difficult to establish due to its symptomatologic overlap with various pediatric conditions. Here, I report the first case of vallecular cyst in Qassim, Saudi Arabia diagnosed in an infant with upper airway symptoms and failure to thrive. He was diagnosed by Flexible fiberoptic laryngoscopy and underwent successful surgical excision. Postoperative course was uneventful, and a one-month follow-up revealed complete recovery.

Key words: Vallecular, Stridor, Laryngeal, Cyst, Infant

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INTRODUCTION

Vallecular cysts are an uncommon cause of airway obstruction in children, with an incidence of approximately 1.87 to 3.49 per 100,000 births [1]. Moreover, approximately 0.9 to 2 percent of infants born with stridor have vallecular cysts [2]. Laryngeal cysts usually develop within the first week of life, however, the diagnosis is often established later [3]. Vallecular cysts are ductal variants of laryngeal cysts that develop through the obstruction of the salivary gland ducts and mucinous glands at the vallecular and tongue base causing airway compromise [4,5].

From a histological perspective, the vallecular cysts consist of glandular respiratory epithelium lined by squamous epithelium. The preponderance of glandular and lymphoid tissue in the area facilitates the development of these cysts usually due to mucocele infection and inflammation following upper respiratory tract infection [6-8]. The diagnosis of this rare entity is often difficult to establish due to its symptomatologic overlap with various pediatric conditions. Here, I report the first case of vallecular cyst in Qassim, Saudi Arabia diagnosed in an infant with upper airway symptoms and failure to thrive.

CASE REPORT

A 2-month-old boy born full-term via spontaneous vaginal delivery with a birth weight of 3 kg presented to the pediatric emergency department for severe respiratory distress, progressive feeding difficulties, failure to thrive, stridor, and apneic episodes. The patient had a history of perioral cyanosis and choking during feeding for one week. There was no documented fever, vomiting, diarrhea, or urinary symptoms. In another hospital, congenital laryngomalacia was suspected. On physical examination, the patient was alert, afebrile, tachypneic, tachycardiac, and Spo2 96% on 2 L 02 via nasal cannula. Throat examination revealed a clear cystic lesion at the base of the tongue during coughing. Chest examination showed inspiratory stridor, pectus excavatum, and intercostal/subcostal retraction. The rest of the findings were unremarkable. A partial workup for sepsis was normal. Chest radiography showed bilateral haziness. Radiography of the post-nasal space revealed a soft tissue intensity in the nasopharynx, causing airway narrowing (Figure 1). Barium swallow test showed gastroesophageal reflux (Figure 2A and 2B).

Contrast-enhanced cervical computed tomography (CT) revealed a 1.5 x 1.9 cm well-defined cystic nasopharyngeal lesion, closely abutting the posterior aspect of the soft palate and the posterolateral aspect of the left tongue and causing significant airway narrowing (Figure 3). Nasopharyngeal branchial cyst was considered a differential diagnosis. Flexible fiberoptic laryngoscopy revealed a supraglottic soft tissue lesion at the vallecular level that triggered the collapse of the supraglottic. Complete excision of the vallecular cyst and redundant tissue was then performed. Histopathology revealed a 1.3 x 1.3 x 0.2 cm cyst lined by stratified squamous epithelium with a focal area of chronic inflammation in the absence of malignancy. These findings are consistent with a benign vallecular cyst.

The patient was discharged in a good condition and was prescribed domperidone and ranitidine. One-month follow-up showed that the patient completely recovered from the condition with no complaints.

DISCUSSION AND CONCLUSION

Persistent neonatal stridor requires thorough evaluation to determine the etiology, which can

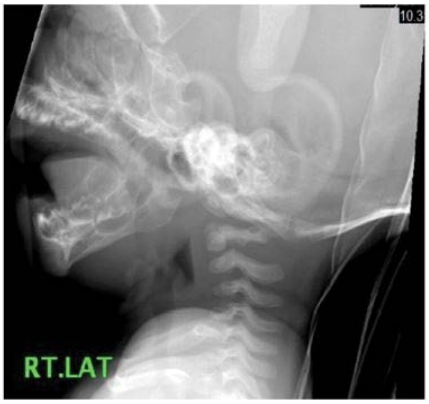

Figure 1: Showed a soft tissue intensity in the nasopharynx, causing airway narrowing.

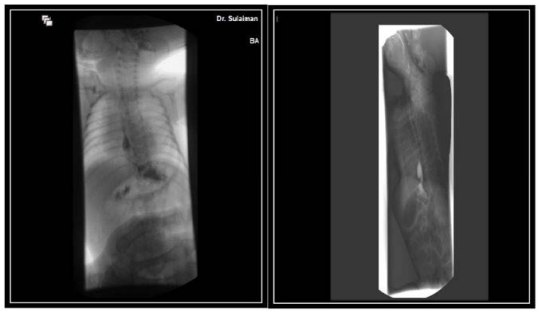

Figure 2A and 2B: Barium Swallow: Showed gastroesophageal reflux during the study, normal anatomy and esophageal motility.

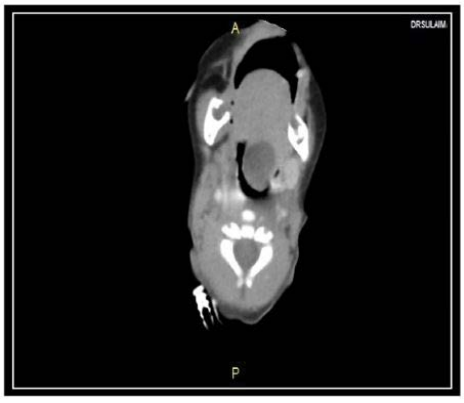

Figure 3: CT Neck: Showed a 1.5 x 1.9 cm well-defined cystic nasopharyngeal lesion, closely abutting the posterior aspect of the soft palate and the posterolateral aspect of the left tongue and causing significant airway narrowing.

impact the decision for critical intervention. Neonates have narrower airways such that slight variations in diameter can substantially increase resistance. The nature of the stridor in relation to respiration may provide clues to the obstruction site. The inspiratory stridor may involve an extra-thoracic lesion in both glottic and supraglottic areas, whereas expiratory stridor may indicate intrathoracic origin [9]. An infant with a prolonged inspiratory phase or neck hyperextension may have an extrinsic obstruction above or at the larynx [10]. Extra-thoracic stridor may be due to congenital structural abnormalities such as vocal cord paralysis, laryngomalacia, and infections such as epiglottitis [11,12].

Laryngomalacia is a common cause of neonatal inspiratory stridor that worsens with lying supine, agitation, and feeding [9]. It usually occurs within the first few weeks after birth.

Vocal cord paralysis is an acquired or congenital cause of inspiratory stridor that is usually biphasic and occurs during feeding. Unilateral involvement can lead to a weak cry and dysphonia while bilateral involvement may result in cyanosis, apnea, and distress [10,11] . Unilateral paralysis may be due to thoracic surgery and birth trauma, whereas bilateral cord paralysis may result from hydrocephalus, perinatal asphyxia, and Arnold-Chiari malformation [10].

First described by Abercrombie in 1881 [4], vallecular cyst is a benign laryngeal cyst, which rarely causes congenital stridor. It is one of the various causes of congenital laryngeal malfunction (i.e., laryngeal webs, clefts, stenosis, and cysts). According to the classification by De Santo for a laryngeal cyst [4], this has been described as base-of-the-tongue cysts, epiglottic cysts, mucous retention cysts, and ductal cysts. Moreover, vallecular cysts are secondary cysts that develop from either mucous gland obstruction or cystic tongue lesions generated from the foregut's misplaced embryonic remnants [9, 13]. Moreover, aryepiglottic cysts are also common forms of pediatric laryngeal cysts that are often unilocular cysts consisting of clear sterile fluid generated from the lingular surface of the epiglottis [14]. Vallecular cysts are lined by the respiratory or non-keratinizing squamous epithelium [15]. A British-Pakistani population study over 10 years provided an incidence of more than 3.49 cases of vallecular cysts per 100,000 live births [16].

Infants with vallecular cysts may present with failure to thrive, feeding issues, stridor, and cyanosis. The severity and timing of vallecular cysts are based on the size and extent of airway involvement. The median age of symptom onset and diagnosis is three and 40 days, respectively [1].

There should be an increased clinical suspicion for vallecular cysts in infants with stridor a few hours after birth. The diagnosis of a vallecular cyst is usually made directly through laryngoscopy. Endoscopy should be performed in a controlled environment to assess the degree of airway compromise. X-ray of the lateral neck may highlight the possibility of vallecular cysts, as explained in the present case. Clinically, it can mimic laryngomalacia because it causes posterior displacement of the epiglottis. Vallecular cysts may present as a low-density attenuation on neck CT and a non-contrast enhancing lesion, hyperintense on T2, and hypointense on T1 on MRI [17, 18].Thyroid imaging and biochemical tests may be performed to rule out thyroglossal duct cysts, which may mimic these imaging findings [4]. Various diagnostic techniques are crucial to establish a diagnosis of vallecular cyst.

Surgical removal of vallecular cysts (e.g., excision, marsupialization, or aspiration) is the treatment of choice [18]. Because of the increased risk of recurrence, simple aspiration of the cyst is not recommended [19]. In conclusion vallecular cyst is an unusual cause of neonatal stridor, which can be life-threatening. This case emphasizes the significance of a thorough assessment involving physical examination, imaging, and histopathology in making an initial diagnosis and in deciding the intervention suited for the patient. An interdisciplinary approach involving not only the pediatricians but also the otorhinolaryngologists is tantamount to its management. Nevertheless, surgical excision remains to be the treatment of choice.

REFERENCES

1. Tsai YT, Lee LA, Fang TJ, et al. Treatment of vallecular cysts in infants with and without coexisting laryngomalacia using endoscopic laser marsupialization: Fifteen-year experience at a single center. Int J Pediatr Otorhinolaryngol 2013; 77:424–428.

2. Suzuki J, Hashimoto S, Watanabe K, et al. Congenital vallecular cyst in an infant: Case report and review of 52 recent cases. J Laryngol Otol 2011; 125:1199-283.
3. Khashu M , Osiovich H , Kozak F, et al. Congenital epiglottic cyst presenting with severe airway obstruction at birth. J Perinatol 2006; 26:71-72.
4. DeSanto L W, Devine K D, Weiland LH. Cyst of the larynx classification. Laryngoscope 1970; 80: 145-76.
5. Gogia S, Agarwal SK, Agarwal A. Vallecular cyst in neonates: Case series: A clinicosurgical insight. Case Rep Otolaryngol 2014; 2014: 64868.
6. Parelkar SV, Patel JL, Sanghvi BV, et al. An unusual presentation of vallecular cyst with near fatal respiratory distress and management using conventional laparoscopic instruments. J Surg Tech Case Rep 2012; 4:118-20.
7. Stack JR, Ridley MB. Epiglottic abscess. Head Neck 1995; 17:263-5.
8. Jason M L, Smith LP, Cohen MA, et al. Diagnosis and treatment of pediatric vallecular cysts and pseudocysts. Int J Pediatr Otorhinolaryngol 2011; 75:899-984.
9. C. A. Daniel M. Neonatal stridor. J Pediatr 2010; 2012:201.
10. Leung AK, Cho H. Diagnosis of stridor in children. Am Fam Physician 1999; 60:2289-96.
11. Claes J, Boudewyns A, Deron P, et al. Management of stridor in neonates and infants. B-ENT 2005; 113-25.
12. Mathew JL, Singh M, Gupta R, et al. Vallecular cyst-revisited. Indian J Pediatr 2008; 75:1081-102.
13. Lahiri AK, Somashekar KK, Wittkop B, et al. Large vallecular masses; differential diagnosis and imaging features. J Clin Imaging Sci 2018; 8:26.
14. Cuillier F, Samperiz S, Testud R, et al. Antenatal diagnosis, and management of a vallecular cyst. Ultrasound Obstet Gynecol 2002; 20:623-626.
15. Youjin L, Alexandria L I, Dombrowski ND, et al. Vallecular cyst in the pediatric population: evaluation and management. Int J Pediatr Otorhinolaryngol 2018; 113:198-203.
16. Prowse S, Knight L. Congenital cysts of the infant larynx. Int J Pediatr Otorhinolaryngol 2012; 76:708-711.
17. Abd Aziz A, Fadzil Abdullah A, Ahmad AR. Two cases of congenital vallecular cyst: A reminder of the potentially fatal cause of upper airway obstruction in infants. Malays J Med Sci 2010; 17:68-73.
18. Abdulla AF. Congenital vallecular cyst causing airway compromise in a 2- month-old girl. Case Rep Med 2015; 975859:2015.
19. Raftopulos M, Soma M, Lowinger D, et al. Vallecular cysts: A differential diagnosis to consider for neonatal stridor and failure to thrive. JRSM Short Rep 2013; 4:29.