Vallecular Cyst as a Cause of Obstructive Sleep Apnea in an Infant

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CASE REPORTS

A 3-month-old baby was diagnosed with obstructive sleep apnea (OSA) on polysomnography (PSG) with a high apnea hypopnea index (AHI). On further investigations he was found to have a vallecular cyst that was successfully treated. We discuss the clinical presentation of vallecular cysts and the importance of polysomnography in identifying

this rare condition.

Keywords: Vallecular cysts, laryngeal cysts, obstructive sleep apnea in infants, cleft lip and sleep apnea

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OSA can present in infancy and is usually caused by craniofacial abnormalities or upper airway obstruction. The pathophysiology is multifactorial and determined by anatomical and neuromuscular factors that influence upper airway size, tone, and function. Laryngeal cysts can potentiate upper airway obstruction but have rarely presented as OSA in the pediatric population.

REPORT OF CASE

A 3-month-old baby with a history of incomplete right cleft lip and alveolus was referred by the Craniofacial Surgery team for PSG for evaluation prior to surgical closure. Patient was born full term via C-section for fetal decelerations with an uneventful perinatal course. He had stridor, snoring, and gasping during sleep since birth. A PSG was performed during daytime nap. He slept for 64 minutes with an arousal/awakening index of 5.6 per hour. He was found to have 45 obstructive apneas, 2 obstructive hypopneas, and 4 mixed apneas (AHI 64.8/h, baseline oxygen saturation 94% to 96%, desaturation index 89/h, SpO₂ nadir 75%, P_{ET}CO₂ highest 47 torr). Due to OSA severity, he was admitted for further investigation.

On physical exam, he was well developed, noticeably stridulous, with high arched palate and right incomplete lip with alveolar cleft. He had a pectus excavatum deformity with mild subcostal retractions but good bilateral air entry and clear breath sounds.

Chest x-ray incidentally showed a right middle lobe infiltrate, and he was found to be positive for metapneumovirus. He was treated for presumed aspiration pneumonia, and reflux management was initiated for clinical evidence of reflux. He was noted to have laryngomalacia on bedside laryngoscopy performed by the otorhinolaryngologist. He was discharged home on oxygen supplementation at half liter per minute (Lpm) via nasal cannula. He had a subsequent hospitalization for RSV bronchiolitis within two weeks of discharge.

Subsequently, on outpatient follow-up with the otorhinolaryngologist, full office laryngoscopy revealed a vallecular cyst (**Figure 1**) in the larynx. Surgical excision was planned, and the

cyst was successfully marsupialized. Six weeks later, a repeat PSG during daytime nap was performed; it showed marked improvement of his obstructive apneas, and reduction of AHI from 64.8 to 7.2 (baseline oxygen saturation 96% to 98%; desaturation index 12/h; SpO₂ nadir 76%, P_{ET}CO₂ highest 44 torr). His need for supplemental oxygen was reduced to 0.25 Lpm. He had his cleft lip successfully repaired at 9 months of age, and a follow-up PSG during daytime nap was completely normal (AHI 0.2/h; baseline oxygen saturation 95% to 97%; desaturation index 1.2/h; SpO₂ nadir 91%, P_{ET}CO₂ highest 45 torr).

DISCUSSION

Laryngeal cysts are a rare cause of stridor in infants. They are more commonly seen in adults.^{1,2} They have been variously classified based on location, histology, size, and contents.¹ The vallecula is the depression behind the root of the tongue between the median and lateral epiglottic folds on each side. Vallecular cysts belong to the category of ductal cysts described in DeSanto's early classification system of laryngeal cysts.¹ This system classified laryngeal cysts into ductal, saccular, and thyroid foraminal subtypes. Ductal cysts are caused by mucus retention in the submucosal collecting ducts. Vallecular cysts can contribute to upper airway obstruction by posterior displacement of the supraglottis.

Clinical presentations of vallecular cysts include stridor, feeding problems, failure to thrive, gastroesophageal reflux, apnea/cyanosis, chest retractions, hoarse cry, as well as respiratory distress and life threatening events.^{3,4} Laryngomalacia can coexist with vallecular cysts.⁵ Diagnosis is by direct laryngoscopy and bronchoscopy, though imaging studies, particularly magnetic resonance imaging, can provide valuable information. Management is by trans-oral endoscopic surgical removal or marsupialization.

Our patient was diagnosed with OSA on PSG. It should be noted that PSG performed during daytime nap may underestimate the severity of OSA. OSA has often been the initial presentation in adults with laryngeal cysts but rarely in children.

Figure 1—Vallecular cyst seen during surgical resection

One report⁶ mentions PSG as part of diagnostic evaluation of an infant, but no details were provided.

The mechanism of upper airway obstruction in infants is multifactorial. The presence of an obstructing lesion in the hypopharynx has the potential of causing severe upper airway obstruction in the immature infant airway. Infants with cleft lip and/or palate have a higher incidence of airway obstruction and sleep-disordered breathing.⁷ Even in the presence of a single underlying anatomic defect as cleft lip, it is prudent to look for possible coexistent etiologies contributing to OSA which could be treatable. Our patient also had laryngomalacia and recurrent respiratory infections. This case highlights the importance of maintaining a heightened index of suspicion and performing a complete upper airway exam, including an endoscopic examination to identify structural and anatomical lesions in an infant with OSA and a high AHI. Our patient had residual OSA on PSG after removal of the vallecular cyst possibly due to postoperative changes, though his cleft lip was still unrepaired. With time

and after successful closure of cleft lip, his OSA completely resolved. This case is an example of coexistent anatomic and structural lesions culminating in severe OSA in infancy.

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