



VALLECULAR CYST IN NEONATES: A CASE REPORT

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ABSTRACT

Introduction: Congenital valvular cysts are rare and can present with acute airway obstruction with wheezing as a chief complaint in neonates and young children. In absent of early detection and appropriate treatment, airway obstruction can cause severe morbidity and mortality. Symptoms appear in most affected infants within the first week of life. Even with case reports in the literature, symptoms must be identified and recognized for early detection of congenital trough cysts.

Case report: A 12-days-old term male baby was referred from pediatric department to ENT department with chief complaint of shortness of breath. The patient always coughs when suckling milk, and vomits. The patient had marked inspiratory stridor with suprasternal and epigastrium retraction. At the oropharynx there was a rounded and translucent mass (cyst) found at oropharynx region with size 2x2x2 cm. Neck soft tissue X-ray examination revealed compressed air column, caused by opaque soft tissue density on the posterior part of the tongue, suggestive of a vallecular cyst. Chest x-ray examination on this patient shown bronchopneumonia bilateral. The patient diagnosed with upper airway obstruction grade 2 due to cyst at region vallecula with suspected aspiration pneumonia was made. The next examination for this patient was tracheotomy with local anaesthesia for securing airway and planning to do flexible laryngoscope. On flexible laryngoscope examination, it was revealed that cystic mass was found attached to the left side of the vallecula and epiglottis measuring 2x2x1 cm and the arytenoid cartilage was edema. We plan to do cyst extirpation approach microlaryngoscopy but it was delayed because of the restrictions on elective surgery, caused by pandemic Covid-19 situation, at Hasan Sadikin Hospital. After given maximum medicamentosa such as infusion of D5 NS 21 ml/hour, Cefotaxime 4 x 150 mg IV, Metronidazole 3 x 30 mg IV, Methylprednisolone 3 x 6 mg IV, Paracetamol 3 x 50 mg IV drip, the cyst was found reduced in size. The follow up treatment planning was cyst marsupialization but it was postponed due to good response during therapy.

Conclusion: Vallecular cysts are rare upper aerodigestive tract congenital diseases. A complete medical history, ENT examination, imaging, flexible laryngoscope, and clinical investigations are all recommended for diagnosis. Management therapy for vallecular cyst usually with aspiration and marsupialization. Antibiotics can be used for therapy but do not guarantee recurrence.

Keywords: Cyst, extirpation, management, vallecular

INTRODUCTION

Neonatal laryngeal cysts are a relatively uncommon yet recognizable cause of inspiratory stridor and respiratory distress in infants. While smaller cysts may not exhibit any symptoms, larger cysts can pose a significant risk by obstructing the airway due to their location and the relatively small size of an infant's respiratory tract.¹ This obstruction can lead to acute episodes of airway blockage. Typically, affected newborns experience symptoms such as inspiratory stridor, respiratory discomfort, and difficulties with feeding within the first week after birth. Mucus retention cysts, also known as congenital vallecular cysts, have the potential to develop on the mucosal surface of various structures within the larynx, including the true vocal fold, epiglottis, or vallecula.² Among these, vallecular cysts are particularly concerning as they can displace the posterior supraglottic region, resulting in the collapse of the airway and compromising the ability to breathe during inspiration. This combination of factors can significantly impair the respiratory function of affected infants.³

Vallecular cysts are believed to develop due to either the blockage of ducts or the presence of developmental abnormalities in the mucous glands. To diagnose these cysts, a flexible nasopharyngolaryngoscopy is typically performed. However, the most reliable diagnostic tool is direct laryngoscopy, which is

considered the gold standard. In terms of treatment, there are several definitive options available. These include aspiration, marsupialization, or complete excision of the cyst. Aspiration involves removing the fluid or contents of the cyst using a needle and syringe. Marsupialization is a surgical procedure where an opening is created in the cyst to promote drainage and prevent recurrence. Complete excision involves removing the entire cyst surgically. For infants with a favorable long-term prognosis but complicated by aspiration pneumonia, an endoscopic marsupialization surgical approach is recommended. This procedure aims to alleviate symptoms, improve breathing, and reduce the risk of further respiratory complications. By creating a controlled opening in the cyst, the trapped contents can be drained, reducing the risk of aspiration and subsequent pneumonia.⁴⁻⁶

We present the case of a 12-day-old infant who was brought to our hospital and referred to ENT surgeon for further management due to vallecular cyst.

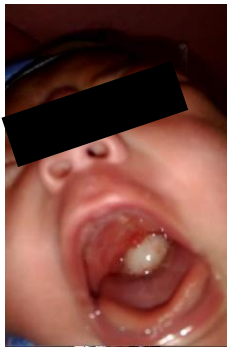
1. CASE REPORT

A 12-days-old term, male infant was referred to us with chief complaint of shortness of breath since 12 days ago, feels worse for the last 3 days. The baby was born to mother P4A1 38 weeks of gestation, born in midwife, crying immediately, history of cyanosis,

history of NICU hospitalization, and use of ventilation was denied. The patient always coughs when drinking milk, and comes out of the nose and mouth. History of fever, travelling history, history of contact with confirmed COVID-19 patients was denied. Because of his complaint the patient was treated at Majalaya Hospital for 2 days, laboratory, thorax and soft tissue x-ray examinations were carried out and a lump in the mouth was found, then he went to Hasan Sadikin Hospital for further treatment.

There weren't any disturbances during his pregnancy, no history of drug consumption, bleeding from the birth canal, and premature rupture of membranes was also denied.

On the physical examination, general condition of the baby looks moderately sick, and compos mentis. The vital sign shown pulse rate 140x/minutes, temperature 36,6C, but had a very high respiration rate, as shown 60x/minutes. Measured SpO₂ shown 86% free air and corrected until reach 99% with simple mask 5 lpm. The baby's weight was 2900 grams. She had marked inspiratory stridor with suprasternal and epigastrium retraction and tachypnoea. Auscultation on pulmonary shown there was a decreased vesicular sound on right lung, ronchi was present on both lung and there was no wheezing. Examination on ear, nose, maxillofacial and neck also considered no abnormality. On nasopharynx-orpharynx, tonsil T1 and normal, but there was a whitish mass (cyst) found at oropharynx region with size 2x2x2 cm and there is pus.

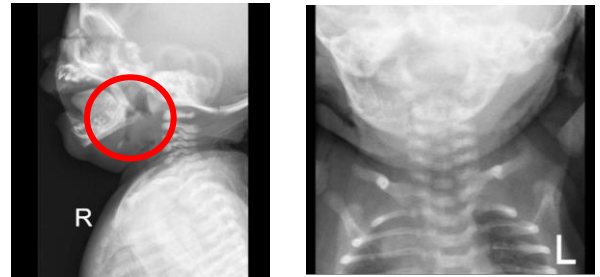


Picture 1. Clinical Picture Found Whitish Mass at Oropharynx

Laboratory findings shown there was a markedly increase of APTT with a value of 38.70. Urea and creatinine level shown an abnormal value, with ratio 12/0.53. pCO₂ level also considered as very high, which is 74.5. pO₂ value was 28.5. The baby has undergone TCM SARS-COV-2 examination and the result was negative. Neck soft tissue x-ray examination shown compressed air column, there was opaque soft tissue density on the posterior part of the tongue, which suggest a vallecular cyst. Chest x-ray examination on this patient shown bronchopneumonia bilateral, no intrapulmonary metastases, and no cardiomegaly.



Picture 2. Thorax X-Ray Results



Picture 3. Neck Soft Tissue X-Ray Results

A patient diagnosis upper airway obstruction grade 2 due to cyst at oropharynx region with suspected aspiration pneumonia was made. Patient planned tracheotomy with local anaesthesia on indications of upper airway obstruction grade 2 due to cyst at region oropharynx and also installed orogastric tube.



Picture 4. Clinical Picture Baby After Tracheotomy

After procedure tracheotomy, the management plan of this patient was observation of vital signs, air passage, emphysema subcutis, infusion of D5 NS 21 ml/hour suction and humidification and medical treatment with cefotaxime 4x150mg IV, paracetamol 3x50mg (IV drip), change gauze stoma daily. From pediatric department they suggest to give enteral feeding of breast milk or formula and periodic suction. LFO endoscopy and cyst extirpation plan in general anesthesia approach microlaryngoscopy was planned.

Flexible laryngoscope was performed. The result shown there was a septum deviation towards left side hypertrophy inferior conchae and narrowing 2/3 choana. On epiglottis, cystic mass was found attached to the left side of the vallecula attached to the epiglottis measuring 2x2x1 cm. Arytenoid cartilage was edema. We diagnosed patient with upper airway obstruction grade 2 due to at region cyst vallecula with bronchopneumonia after tracheotomy post operation day 2. We managed patient with observation of vital signs, air passage, emphysema subcutis, infusion of D5 NS 21 ml/hour, suction and humidification, Cefotaxime 4 x 150 mg IV, Metronidazole 3 x 30 mg IV, Methylprednisolone 3 x 6 mg IV, Paracetamol 3 x 50 mg IV drip, gauze stoma change daily, and we plan to perform marsupialization on general anesthesia with microlaryngoscopy approach but it was delayed because of the restrictions on elective surgery, due to Covid-19 pandemic, at Hasan Sadikin Hospital.

The patient was treated for 5 days with intravenous antibiotics, steroids, analgesics and was evaluated with flexible laryngoscope. The examination on epiglottis, shaped like omega shape, and found cystic mass reduction attached to the left side of the vallecula and epiglottis measuring 1x1x1 cm. The bronchopneumonia was improved. Because the limitations of elective surgery during the pandemic at Hasan Sadikin Hospital the patient was allowed to outpatient, with take home medication cefixime 2x20 mg sir per

OGT, methylprednisolone 3x2 mg per OGT, and N-acetyl cysteine syrup 2x50 mg and plan for control 1 week later.



Picture 5. Clinical Picture cyst size 1x1x1cm was found in oropharynx after 5 days observation and antibiotic.

After the patient is observed at home, the cyst is not visible at oropharynx. On neck, at region colli anterior there was an attached tracheotomy cannula. The follow up treatment planning was cyst marsupialization, but marsupialization has not been performed because operating services at Hasan Sadikin Hospital are still limited due to the increasing number of COVID-19 cases and the availability of operating rooms and inpatient rooms. As a result of this, the patient was temporarily given tapering-off azithromycin 1x30mg.

After 2 months of home observation, our patient was scheduled for flexible laryngoscope and we found the cyst disappeared. we decannulate the tracheostomy tube and re-observe again. Currently, the patient is still in control at the ENT division Laryngopharynx clinic.

We discuss the diagnosis and treatment of vallecular cyst in neonates and share our experience with this condition. In order to provide comprehensive understanding of how this illness is managed, we also review the literature

DISCUSSION

Congenital valvular cysts are uncommon but can be a threatening cause of wheezing in newborns and young children. The resulting airway obstruction can contribute to severe morbidity and mortality.⁷ Although rare, vallecular cyst can be life-threatening because the localization can lead to sudden airway obstruction.⁸ This patient has an upper airway obstruction due to a vallecular cyst. Terms used include mucus-retaining cysts, epiglottic cysts, base of the tongue cysts, congenital cysts, and most recently ductal cysts. DeSanto et al. classified laryngeal cysts according to their location and surface mucosa, hence the term ductal cyst. Newman divide laryngeal cysts into epithelial cysts, tonsillar cysts, and oncolytic cysts.⁹

Vallecular cysts are rare in children but most common in newborns and infants. This case report presents a neonatal case. Most affected infants develop symptoms within the first week of life. It is important for doctors to recognize symptoms and comprehend the proper way to evaluate these patients.

Respiratory disease, particularly life-threatening airway obstruction, is the most prevalent manifestation observed in cases of congenital vallecular cysts. The severity of symptoms can vary depending on factors such as the size of the cyst, its extension into the airway, and the age of the patient. Shortly after birth, infants with vallecular cysts commonly exhibit symptoms such as

inspiratory stridor and dyspnea. As children grow older, additional issues related to development and feeding may arise and become apparent. Other notable symptoms associated with vallecular cysts may include a hoarse cry, choking, coughing, episodes of apnea or cyanosis, chest retraction, and postprandial vomiting. A case report detailing a baby with a vallecular cyst presenting with chest retraction exemplifies the potential impact on airway dynamics. Progressive enlargement of the cyst can lead to alterations in airway dynamics, resulting in increased negative inspiratory pressure and contributing to the development of supraglottic prolapse and laryngomalacia.¹⁰⁻¹¹

The diagnosis of vallecular cysts can be challenging due to several potential obstacles. One of these obstacles is that flexible fiberoptic laryngoscopy may not always clearly show the involvement of the trough region. This limitation can be attributed to the coexistence of laryngomalacia, a condition that commonly occurs alongside vallecular cysts. In such cases, flexible endoscopy may be employed to differentiate between trough cysts and laryngomalacia. However, it is important to note that flexible endoscopy does not necessarily provide a definitive means of narrowing down the diagnosis.

To establish a definitive diagnosis of laryngeal cysts, direct laryngoscopy remains the gold standard. This procedure allows the surgeon to not only confirm the presence of vallecular cysts but also assess any additional airway lesions and facilitate surgical treatment when necessary. The use of direct laryngoscopy is essential in securing the airway and ensuring the appropriate management of vallecular cysts.

Vallecular cysts pose a unique clinical challenge, and a comprehensive approach involving a thorough medical history, radiological techniques, clinical evaluations, and endoscopic investigations is crucial to confirm the diagnosis accurately. By gathering information from various sources and conducting a comprehensive evaluation, healthcare professionals can obtain the necessary data to confidently diagnose vallecular cysts and plan appropriate treatment strategies.¹²⁻¹³

Endoscopic examinations and imaging studies can provide valuable insights; however, a conclusive diagnosis of vallecular cysts requires pathologic evaluation. The formation of these cysts may result from blockages in the ducts of the submucosal glands within the vallecula. It is important to note that pseudocysts do not possess a significant internal mucosal lining. To differentiate vallecular cysts from cases of cystic lymphangioma vallecular tumors, immunoreactivity testing for D2-40, a monoclonal antibody specific to lymphatic endothelium, can be employed. This immunoreactivity testing can be particularly helpful in distinguishing cases of vallecular cysts from those featuring a single large cyst filled with fluid, which are characteristic of cystic lymphangioma vallecular tumors. By conducting pathologic evaluations and utilizing immunoreactivity testing, healthcare professionals can achieve a more definitive diagnosis of vallecular cysts, enabling appropriate treatment planning and management.¹⁴

In patients with vallecular cysts, it is essential for an anesthesiologist to conduct routine pre-anesthetic evaluations and implement appropriate airway management measures. If a patient is found to have airway issues associated with vallecular cysts, it is crucial to promptly inform the anesthesiologist to ensure necessary preparations are made. It may be beneficial to avoid placing the endotracheal tube in close proximity to the base of the tongue or the area of vallecular cyst formation.

Vallecular cysts can be treated through various surgical procedures. Standard treatment options include conservative medical approaches, cyst aspiration, endoscopic excision, microlaryngoscopy and bronchoscopy with endoscopic marsupialization, as well as using CO₂ laser or microdebrider techniques for deroofting the cyst. Some authors recommend complete removal of the cyst using a CO₂ laser following aspiration

to decompress the cyst. Additionally, new transoral resection techniques have been developed. During surgical interventions, it is crucial to preserve the lingual surface of the epiglottis to prevent vallecular scarring.

In cases where vallecular cysts are recurrent or large, expanding beyond the boundaries of the larynx, surgical excision through an external route may be necessary. This approach involves removing the cyst via an external incision. This option is particularly suitable when conservative measures or less invasive procedures have not been effective.

By employing appropriate anesthetic evaluations, implementing tailored airway management strategies, and considering the range of surgical treatment options available, healthcare professionals can optimize the management and outcomes for patients with vallecular cysts.¹⁴⁻¹⁵

Endoscopic excision of vallecular cysts can sometimes be ineffective, leading to multiple recurrences. This fibrosis creates an environment that allows the cyst to enlarge along the path of least resistance, away from the area of fibrosis. Due to the rarity of vallecular cysts, it is challenging to directly compare treatment techniques and determine the most effective approach. Therefore, additional multi-institutional randomized prospective trials are necessary to gain a better understanding of the optimal diagnostic and therapeutic strategies for these patients. These trials would help identify the most effective methods for diagnosing vallecular cysts and selecting appropriate treatment options.

By conducting further research and implementing rigorous clinical trials involving multiple institutions, healthcare professionals can gather more comprehensive data on vallecular cysts. This will enable them to establish evidence-based guidelines and protocols for diagnosing and treating these complex conditions. Improved understanding and standardized approaches will contribute to better outcomes for patients affected by vallecular cysts.¹⁶

The diagnosis of vallecular cyst was confirmed through the use of fiberoptic bronchoscopy, a diagnostic procedure. In a separate study, it was found that 88% of patients experienced symptomatic improvement after undergoing either complete removal or marsupialization of the cyst using a CO2 laser. For the remaining patients, marsupialization was performed using cold instruments. In the case of this patient, if the cyst does not shrink, marsupialization will be planned as the next course of action.

The recurrence rate of vallecular cysts can vary, but according to the majority of authors, marsupialization techniques, whether utilizing CO2 lasers or not, have shown to be successful. A minimal recurrence rate was reported following marsupialization when compared to complete excision of the cyst. In the reported case, the cyst was initially aspirated, and then a partial excision of the cyst was performed using a micro-laryngeal instrument.

By utilizing fiberoptic bronchoscopy for diagnosis and tailoring the treatment approach based on the individual patient's condition, healthcare professionals can effectively manage vallecular cysts. The success rates of different techniques, such as complete removal, marsupialization with CO2 lasers, or cold instrument marsupialization, highlight the importance of personalized treatment plans. Continual research and clinical observations will further contribute to the refinement of treatment strategies and the improvement of patient outcomes in cases of vallecular cysts.¹⁷

Treatment options for patients with infected vallecular cysts may vary depending on the severity of the airway obstruction. For patients with a low risk of airway obstruction, a combination of intravenous antibiotics and steroids may be administered to reduce inflammation and alleviate symptoms. This conservative approach aims to manage the infection and inflammation without invasive procedures. However, in cases where the airway obstruction is more significant, surgical excision is commonly performed as the primary treatment for vallecular cysts. Various surgical techniques

can be employed, including cyst aspiration, marsupialization, surgical debulking, and excision using methods such as CO2 laser or electrocautery. It is important to note that cyst aspiration alone is not recommended as a definitive treatment due to its high recurrence rate. The remaining cyst wall after marsupialization or surgical debulking poses a risk of recurrence, as it may reaccumulate over time. In a specific study, the patients were treated with a combination of antibiotics, steroids, and analgesics. Following two months of homecare monitoring, a subsequent flexible laryngoscope examination revealed the disappearance of the cyst. This highlights the potential effectiveness of a comprehensive medical approach in managing vallecular cysts.¹⁸

It is crucial for healthcare professionals to carefully assess each patient's condition and tailor the treatment plan accordingly. The choice between conservative management or surgical intervention depends on factors such as the severity of symptoms, risk of airway obstruction, and recurrence rates associated with different procedures. Continued monitoring and follow-up examinations are necessary to ensure the successful resolution of vallecular cysts and the prevention of complications.¹⁹

Vallecular cysts can sometimes be challenging to identify, particularly after the resolution of supraglottic inflammation. Physicians may need to perform flexible nasoendoscopy to visualize these cysts, as they can be easily missed during routine examinations. A retrospective cohort study conducted by Burger et al. revealed a consistent association between vallecular cysts and the development of suppurative epiglottitis infections. It is worth noting that supraglottic infections are commonly caused by *Haemophilus influenzae* or group A streptococci, with a higher incidence among children compared to adults. Other pathogens, such as *Bacteroides* and *Pneumococcus* species, as well as *Candida* species and Herpes Simplex Virus, can also contribute to supraglottic infections. Additionally, physical irritants like heat, caustic substances, and direct trauma can play a role. In cases where elective surgeries are limited due to hospital conditions, such as during the COVID-19 pandemic, alternative management approaches need to be considered. For instance, in this particular patient, antibiotics (e.g., azithromycin) were administered while closely monitoring the progress of the patient, considering the relatively high recurrence rate associated with conservative treatment alone. As the patient responded well to this therapy, the decision was made to postpone the marsupialization surgery while maintaining vigilant monitoring. During challenging times or when surgical interventions may be restricted, a multidisciplinary approach involving medical management, close observation, and regular assessments can be employed to ensure appropriate care for patients with vallecular cysts. It is essential to balance the need for immediate surgical intervention with the risks and limitations posed by external factors, while still prioritizing the well-being and safety of the patient. Regular reassessment of the patient's condition and ongoing communication between the medical team and the patient are vital in managing vallecular cysts effectively during these circumstances.¹⁹

2. CONCLUSION

Vallecular cysts are rare upper aerodigestive tract congenital diseases. A complete medical history, ENT examination, imaging, flexible laryngoscope, and clinical investigations are all recommended for diagnosis. Management therapy for vallecular cyst usually with aspiration and marsupialization. Antibiotics can be used for therapy but do not guarantee recurrence.

ACKNOWLEDGMENT (IF ANY)

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