Bioscene

Bioscene
Volume- 21 Number- 02
ISSN: 1539-2422 (P) 2055-1583 (O)
www.explorebioscene.com

An Unusual Presentation of a Bleeding Vallecular Cyst in a Newborn

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Abstract: Congenital vallecularcystsarevery rarely occurring cysts in newborns .It usuallypresents with feeding difficultiesin the newborns and an associated failure to thrive is common.Early suspicion and prompt direct laryngoscopy usually clinch the diagnosis of this rare type of cyst. The standardrecommendedtreatmentis surgical excision or marsupialization. We present a case report of a twenty-day old newbornwhopresentedat the casualty with life threatening respiratory distress and bleeding from mouth. The newborn was resuscitated and stabilized.Furtherinvestigationsfor coagulation and bleeding disorders, surfactant deficiency, sepsiswere normal. The newborn then had stridor after 12hrs during the course of treatment and again developed severe respiratory distress with desaturation, seizures and central cyanosis.The bleeding from oropharynx persisted and initial during difficult reintubationthe bleeding vallecular cyst in oro pharynx was noticed.Thiscystwas excised by the paediatricsurgeon.Thepost operative period was uneventful and the newborn survived.

Keywords : Laryngeal cyst,Vallecular cyst, Neonate , OropharyngealBleeding, Airway obstruction

Case Report :

A 20-day old male term baby born out of non consanguineous marriage to a primi mother withknowncomorbidities presented at the casualty of our medical college hospital. The newborn was observed to have respiratory distress and blood stained

secretions from mouth.A careful examination and the clinical history revealed the following.

The mother's antenatal history was uneventful with no associated decreased fetal movements, leaking PV, bleeding PV or any maternalillnesses.Antenatal scans were normal.The baby was delivered at 40weeks of gestation by emergency LSCS in view of failure of progressionoflabour.

The newborn had abirth weight of 2.580kg,APGARscore was normal. The initial postnatal days were uneventful and the baby passed urine and meconium in 24hrs.There was no exaggerated physiological jaundice,centralcyanosis,feeding difficulty and breathing difficulty.The newborn was discharged on day 5 and was exclusively breastfed.On 20th day of life mother noticed her newbornwas holding breath and then became unresponsive for 10 seconds. The baby was brought back from home to local hospital immediately.The newborn was found to have oxygen desaturation which did notimprovewithadministration of oxygenby nasal prongs/mask. The baby was thus referred to our medical college hospital with a level 3 NICU.On arrival at the casualty there was blood-stained frothy secretions from mouth of the newborn,the newborn was tachypneic, sub costal retractions bilaterally present and frequent episodes ofdesaturations.Noassociated seizure like movements, abnormal cry,umbilical stump bleeding or evidence of any other site bleeding was noticed. The newborn was shifted to NICU.The newborn was stabilized and support of and respiratory support by high frequency humidified nasal cannula support with empirical antibiotics was initiated.The newborn had persistent blood streaking from the oropharynx.Vitamin K injection was administered suspecting hemorrhagic disease of newborn followed by fresh frozen plasma infusion.

The routine Hemoglobinwas 14.7g/dl, platelet 4.3 lacs /mm3,Total count 11,000/mm^3,CRP negative,PT/INR normal, sepsis screen negative,renal ,liver function tests normal and serum electrolytes were normal.ChestXraydoneshowedonly minimal bilateral heterogenous opacities**(fig. 1**)probably due to aspiration of blood-stained secretions with no evidence of cardiomegaly or any signs of lung anomalies.Echodone was normal with no evidence of pulmonary artery hypertension . Neurosonogram showed no bleeding or abnormalities in brain, ultrasonogramofabdomen and thorax were also normal.

A trial of surfactant was given.12hours post admission baby was noted to have worsening of respiratory distress and SpO2 levels were dropping.The baby was immediately intubated, and mechanical ventilation initiated,but the bleeding from oropharynx persisted.Baby was noted to have stridor while on mechanical ventilator by next day and in view of suspected choanal atresia ENT opinion was sought and they opined that it was normal. On day 3 of admission baby was extubated and was stable for 12hrs and again developed respiratory distress

andincreased intensity ofstridor. Baby also developed one episode of seizure, probably hypoxic episode with a saturation drop to 60% O2.Intubation was reattempted and this time it was a difficult intubation with a cystic mass found in the root of tongue which probably caused these symptoms**(fig. 2).**The newborn was intubated,andmechanicalventilator support given and general condition improved.Hemogram repeated revealed a sharp dip of Hemoglobin to 5 g/dland blood transfusion was given.The results were as follows**(Table 1).**FlexibleBronchoscopy done showed cystic lesion above epiglottis making laryngeal view difficult and edematous supraglottic structures. Due to unavailability of pediatric surgeon,thenewbornwas referred to higher center where the presence ofVallecularcystat base of tongue was confirmed and microscopic laryngeal surgery was done. The cyst was coablated and the post operative period was uneventful. There was no further bleeding from the oropharynx and the newborn wasdischarged on day 7 post surgery.

Discussion:

Oropharngeal and laryngeal cysts are a great challenge to a paediatrician,neonatologist and the pediatric surgeon as they are rare.Thetimely diagnosis and skillful management can reduce the morbidity and mortality associated with these cysts[1]. Laryngeal Vallecular cyst is a type of benign cyst appearing in the throat and early diagnosis and treatment is challenging. They may usually present with dysphagia, hoarseness and stridor or very rarely with bleeding. The individualized approach for each type for diagnosis is affirmed and surgical cure is mandatory [1,2].It is mentioned under different nomenclaturewhich includes epiglottic cyst, mucous retention cyst, ductal cyst and base of tongue cyst. These are described under De santo's classification in 1970 as saccular, ductal, throidforaminal types [2].

In the newborn period a vallecular cyst is a very rare condition.Literture review has mentioned thatandit usually presents with hoarseness of voice, foreign body sensation and pain in oropharynx in adults[3,4].A few infants in the pediatric age groups presented with feeding difficulties,failure to thrive and dysphagia [5]. Congenital laryngeal cysts can also present as an apparent life-threatening episode with central cyanosis and respiratory distress [3,4,5].

Early suspicion and direct laryngoscopy usually clinch the diagnosis. This can be treated with surgical excision or marsupialization [6]. If symptomatic, surgical and electrocautery options are also available for excision ofvallecular cyst[7].In few reported cases airway management of children with vallecular cyst was done under general anesthesia due to difficulty in intubation[8].Another study showed the role ofMRIoropharynx,flexiblelaryngoscopyand thorough clinical examination whichcoulddiagnose these rarely occurring cysts[9].We could find a

casedescription of 2020 where a neonatal vallecular cyst presented with a similar catastrophic event with life threatening respiratory distress but without bleeding[10]. Lateral radiography,flexible laryngoscopy/bronchoscopy,CT,MRI,USG of neck are the various diagnostic tools available and cyst aspiration, marsupialization,resection and excisionusingmicroinstruments, lasers, electrocautery are the definitive treatment options available.[1].Literature review has revealed that vallecular cyst has been reported by Collin et al [11]in a 3-day old baby presented with stridor and underwent flexible laryngoscopy which revealed airway mass obstructing laryngeal inlet underwent bronchoscopy with excision of cyst, a similar scenario.

Conclusion:

Vallecular cyst usually gets unrecognized in pediatric and neonatal population. Themyriad of different clinical presentations can be challenging to the paediatrician and neonatologist.Bleeding from oral cavity is very a rare presentation of valleculcyst.Clinical diagnostic skills also play a vital role in identifying such rare cases.So a prompt clinico-diagnostic examination withemergency surgical managementcan help the newborns to survive without morbidities.

Author Declaration:

- Financial or Other Competing Interests: None
- Was informed consent obtained from the subjects involved in the study? No
- For any images presented appropriate consent has been obtained from the subjects. NA

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Annexures-Table 1 and 2 Figures

s.no	Day 1	Day 3	Day 5
Hemoglobin	14.7g/dl	14.6g/dl	8.9 g/dl
PCV	46%	46.9%	27.8%
RBC	4.37million/mm^3	4.45million/mm^3	2.57million/mm^3
Platelet	5.46lakhs/mm^3	6.1lakhs/mm^3	4.36lakhs/mm^3
TC	11900/mm^3	5800/mm^3	10,170/mm^3
CRP	negative	19.2mg/dl	7mg/dl
Prothrombin time	11secs	12.8secs	12 secs
INR	0.92	1.07	1.07
APTT	Normal	Normal	normal
Blood urea	14mg/dl		
S.Creatinine	0.3mg/dl		

SGOT	72mg/dl		
SGPT	28mg/dl		
ALP	169 IU/L		
Total protein	5.8g/dl		
S.Albumin	3.5g/dl		
S.Globulin	2.3g/dl		
A/G ratio	1.5		
S.sodium	132meq/l		
S.potassium	5.10meq/l		
S.bicarbonate	20 meq/l		
S.chloride	100meq/l		
S.Calcium	9.7mg/dl		
Stool occult blood	positive		
USG	Normal		
Echo	Normal study		

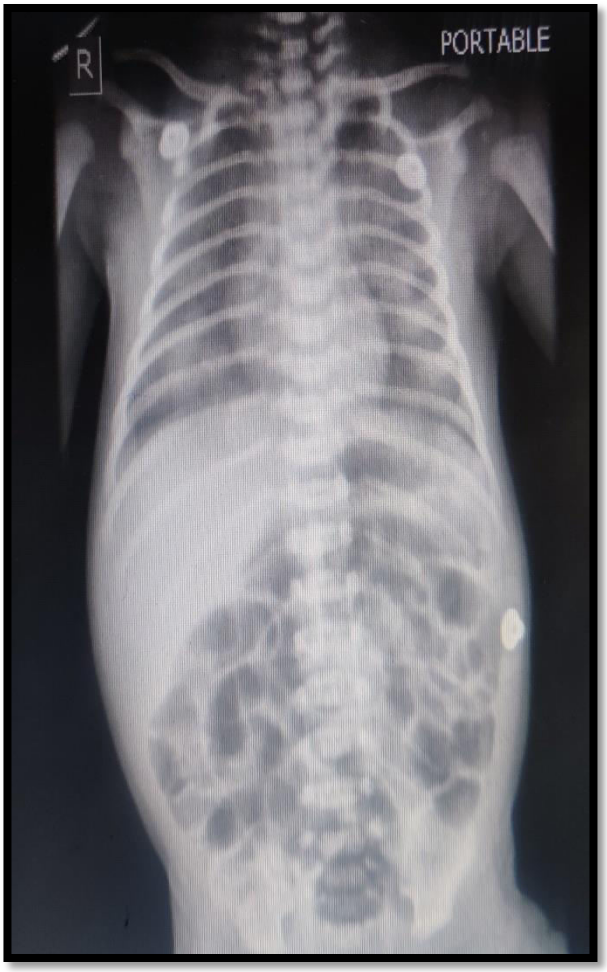

Figure 1: Chest Xray with abdomen of the newborn showing mild infiltrates in the bilateral lung fields probably due to aspiration of secretions with blood from the cyst.

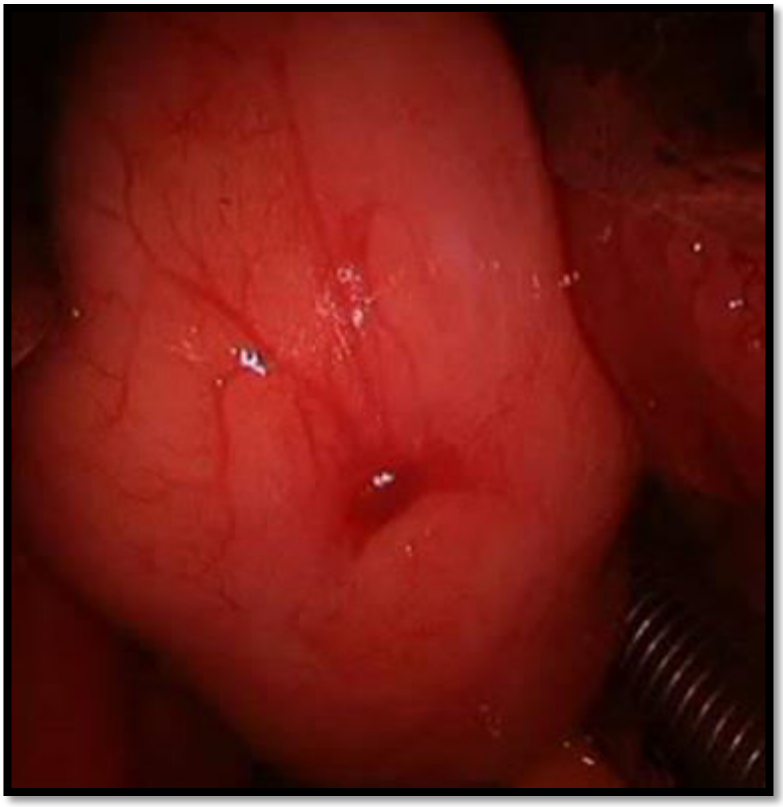

Figure 2 Thevallecular cyst visible at the root of the tongue