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Lingual Cyct

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Abstract

Epidermoid cysts encountered throughout the body, only 7% occurs in the head and neck area, with the oral cavity accounting for only 1.6%. Intraoral this benign slow growing and painless entity is usually located in the submandibular, sublingual and sub mental region. They can cause symptoms of dysphagia and dyspnea and have a malignant transformation potential. Surgical excision is the treatment of choice. Described here is a case of gigantic sublingual cyst.

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Introduction

Epidermoid cysts are benign pathologies that can occur anywhere in the body, predominantly seen in areas where embryonic elements fuse together[1]. Most cases have been reported in the ovaries and the testicles (80%), with head and neck accounting for 7% [1,2]. Dermoid and epidermoid cysts in the mouth are uncommon and comprise less than 0.01% of all the oral cysts [2-4]. Majority of them occur in sublingual region, but there are rare case reports of occurrence in other sites

The tongue is a complex of muscle groups with a fibrous scaffold consisting of the hyoglossal membrane and midline lingual septum.

The root of the tongue is an important sub region of the oral cavity, associated with very specific differential diagnoses. It is relatively resistant to primary neoplastic and infectious processes due to its high percentage of skeletal muscle and lack of significant lymphatic tissue .

The majority of lesions found in the root of the tongue are congenital and benign, representing ectopic tissues of thyroidal, epidermal, dermal, foregut, venous, and lymphatic origin.

The most common midline mass at the base of the tongue is a lingual thyroid which should be confirmed or excluded by radionuclide scanning.

Case Presentation

9 month old boy admitted through ER complain of cough, shortness of breath, fever for 3 days, and Vomiting for 7 months

This boy was product of full term, c/s because of breech presentation, admitted to NICU for 3 days because of jaundice.

This boy condition started since he was 37 days old and admitted as case of bronchiolitis for 7 days and discharge in a good condition.

The second admission at age of 2 months as case of bronchopneumonia for 4 days.

The third admission at age of 4 months again as bronchopneumonia stayed in a hospital for 2 weeks.

Child was fully vaccinated with normal developmental skills except mild gross motor delay. Was on bottle feeding with good preparation and feeding technique.

Parents are not relative. Mother is teacher. Father is jobless. They have 4 other siblings, the eldest one got bronchial asthma, and the youngest one got cystic fibrosis.

On Examination

Patient was active, alert, but very marasmus,vital Sign was stable Temp= 37.2 c pulse= 146/min

B.P= 99/78 oxygen saturation =99% in room air and respiratory rate= 40/min.

His weight = 4.8 kg markedly below 3rd centile

Has mild subcostal retractions but no dysmorphic features, cyanosis, jaundice or pallor.

Chest revealed bilateral crepitation's but no wheeze.

Other systemic examination unremarkable.

So, differential diagnosis for repeated chest infection were gastro esophageal reflux (GERD), Cystic fibrosis, immunodeficient disorder, and a remote possibilities of HIV and TB .

and to roll out structural abnormalities ,such as H-fistula

The baby was investigated accordingly

His CBC revealed WBC = 11, neutrophils= 43.5, lymphocyte= 46, Hb= 11.1, MCV=81.4 MCH =30.6 platelet =530

CBG PH= 7.3 pco2 =24.3 Hco3= 23.5

All his electrolytes, liver, renal, bone profile, immunological test and sweat chloride test were normal.

His HIV, PPD test, PCR all were -ve

CT chest showed multiple patchy consolidations.

So, patient treated for his bronchopneumonia with cefuroxime and Salbutamol inhaler and improved.

Meanwhile he was observed for complete feeding process and he is noticed to have milk coming from his both nostrils with frequent aspirations and desaturations. Video 1 So scopic swallowing test done by ENT consultant to rule out any structural obstruction

or anomalies which Revealed a mass protruding at the back of the tongue obstructing the passage of milk ingestion and part of the milk refluxed back to the nostril .

CT scan with contrast studies revealed a cystic lesion at the posterior third of the tongue Thyroid function test were normal TSH 2.2 suggesting lingual cyst vs thyroglossal duct cyst. T4 1.38.

Thyroid scan showed normal uptake in the thyroid gland and no uptake in the cystic lesion at the posterior third of the tongue So, the mass was excited by ENT consultant. histo-pathological study revealed oral mucosa, no specific changes and no thyroid tissue detected.

Microscopic Description

Section reveals fragments of tissue lined by stratified squamous epithelium showing mild acanthosis,spongiosis and Para keratosis. the sub epithelial tissue shows scattered mononuclear inflammatory cells. Minor salivary glands are also seen. there is cyst like space surrounded by mononuclear inflammatory cells. There is no evidence of dysplasia or malignancy.

Final Diagnosis was Lingual Cyst

Follow Up

Patient was followed in the clinic regularly where he was growing smoothly, increasing in weight steadily and no more vomiting ,aspiration or admission were recorded for 8 months Video 2

Discussion

Lingual cysts are extremely rare in the neonate and young infant Epidermoid and dermoid cysts are rare, benign lesions found throughout the body, with 7% occurring in the head and neck area, 1.6% of which occurs in the oral cavity. Of all the oral cysts dermoid cysts account for only 0.01% [2-4] Roser was the first to designate dermoid cysts in the floor of the mouth as epidermoid tumors [5] Even though the expression "dermoid cyst" characterizes a distinct entity, the word "dermoid" has been used to designate true dermoid cysts, epidermoid cysts, and teratoid cysts [2,3,6,10,11] Based on the histopathological picture Meyer divided the floor of the mouth cysts into following types: [1,2,4]

Â Epidermoid cysts - where in the cystic cavity is lined with epithelium without skin appendages (like our case)

.Â Dermoid cysts - here the epithelial lined cystic cavity encloses skin appendages such as hair, hair follicles, sebaceous, and sweat glands.

Â Teratoid cysts - in this entity, the cystic cavity in addition to skin appendages also encloses mesodermal derivatives such as bone, muscle, gastrointestinal and respiratory tissue.

All these three cysts owing to their squamous epithelium lining may enclose cheesy keratinaceous material within their lumen. Hence the fundamental difference between the dermoid and the epidermoid is the presence of skin appendages within the wall of the former and the lack of the same in the latter [3,6].

Epidermoid cysts may be categorized as congenital or acquired based on their origin although there is no disparity between the two either clinically or histologically [2,4,7]

In medical literature, a variety of terms have been used for tongue base cysts, such as epiglottic cyst, lingual cyst, vallecular cyst,or laryngeal cyst[1–8].Two major hypotheses to explain the pathogenesis are that this cyst is a consequence of either ductal obstruction of mucus glands or an embryological malformation [2]. Histologically, the cyst contains respiratory epithelium with mucous glands, with an external lining of squamous epithelium [2–4]. Most affected infants have symptoms during the first week of life [5]. Clinical manifestations consist of various degrees of upper airway obstruction such as inspiratory stridor, chest retraction, apnea, cyanosis, and feeding difficulty. Stridor is the most common symptom [1–8]. About 60% of children with stridor have laryngeal obstruction such as laryngomalacia, vocal cord paralysis, subglottic stenosis, hemangioma, or laryngeal cysts; 25% have lesions in the upper airway, including choanal atresia, macroglossia follow-up is necessary in case of recurrence [1]. Primary diagnostic approach to laryngeal or vallecular cysts should be a flexible nasopharyngeal laryngoscopy or bronchoscopy. CT and MR imaging often help narrow the differential diagnosis such as

lingual thyroid, proximal cystic dilatation of the thyroglossal duct, lymphangioma or hemangioma, dermoid cyst, lipoma, fibroma, or carcinoma [6]. Although surgical removal may be the treatment of choice, other modalities such as endoscopic marsupialization, excision, and deroofing of the cyst have been recently developed. Marsupialization under general anesthesia is a safe and definitive procedure, especially when performed by CO2 laser. Sometimes, preincubation aspiration becomes necessary before the insertion of the endotracheal tube. Simple aspiration of the cyst is not advised because of its high recurrence rate [1–6]. Spontaneous disappearance of a tongue base cyst after oropharyngeal suctioning has not been previously reported [1–8]. In conclusion, tongue base cysts should be considered in differential diagnosis in new born with stridor, respiratory difficulties, or swallowing problems. An endoscopic laryngobronchoscopy has to be performed d before making the diagnosis of laryngomalacia. Definitive therapy requires large marsupialization under general anesthesia,].

Traumatic or iatrogenic inclusion of epithelial cells or the blockage of a sebaceous gland duct have been postulated as the pathogenesis of acquired cyst [4,7]. However, some authors have also stated that midline cysts may represent a diverse form of thyroglossal duct cyst [4,6,7].

Although floor of the mouth in the midline is most favored site, occasional occurrence involving the buccal mucosa, tongue, lips, uvula, temporomandibular joint dermal graft, intradiploic, intracranial, and intraosseous location within the mandible and maxilla also have been cited in literature [4,8,9]. These lesions show variation in size and weight from few millimeters to centimeters and a gram to several hundred grams respectively [3,5]. Symptoms of dysphagia, dyspnea and dysphonia may occur due to upward displacement of tongue by these sublingual swellings [4,5]. Furthermore growth in an inferior direction may give rise to appearance of characteristic "double chin" [1,4,8]. These well encapsulated lesions typically feel "dough like" on palpation, although they may be fluctuant and cyst like based on consistency of the luminal contents, that may range from a cheesy, sebaceous to liquefied substance [1,5,6].

Fine needle aspiration cytology, ultrasound, CT and MR imaging provide essential information on the cyst location that allows optimal preoperative planning. Ultrasonographic findings comprise solid and cystic structures within a heterogeneous mass [3]. On CT scans, the dermoid appear as moderately thin walled, unilocular masses filled with a homogeneous, hypoattenuating fluid substance with numerous hypo attenuating fat nodules giving the pathognomonic "sack-of-marbles" appearance [3]. On MR imaging dermoid cysts give variable signal intensity on T1-weighted images and are usually hyper intense on T2-weighted images [3,9]. Fine needle aspiration cytology has been advocated as an essential investigation. Although not equivalent to CT and MRI, it is safe, economical and dependable technique and is therefore useful for analysis of sublingual lesions [1,6].

Treatment comprises total surgical excision [1,2,4]-[6]. Caution should be taken not to rupture the cyst, as cystic contents may act as irritants to fibro vascular tissues, causing postoperative inflammation [3]. Recurrences are unusual after absolute surgical excision [1,3]. Reports of malignant transformation of sublingual dermoid and epidermoid to squamous carcinoma and basal cell carcinoma are present [1,2,12]. A 5% rate of malignant transformation of the teratoid variety of oral dermoid cysts has also been quoted in literature [3]-[5].

Conclusion

we present a very rare case of lingual cyst located at the posterior third of the tongue. Lingual cyst has a verities of presentations ranging from asymptomatic child down to severe obstruction as in our patient with florid manifestation such as frequent vomiting, aspiration, dysphagia, failure to thrive, repeated aspiration, pneumonia ,frequent wheeze and respiratory distress. High index of suspicion together with watching a baby during his complete process of feeding is very beneficial to reach a final diagnoses.

Consent

The authors had obtained Oral informed consent from the patient's legal guardian for publication of this case report and accompanying image.[13,14]

Conflict of Interest

The authors declare that they have no conflict of interests

Authors Contribution

Author was saw the case and advised be seen ENT Consultant who did bronchoscopy and excited the mass and sent to histopathology, endocrine consultant saw the case and asked for thyroid scan, family medicine consultant brought the review and NICU Consultant wrote the case

References

1. University of Rochester Medical Center. (2015). Thyroglossal duct cyst. Retrieved from http:// www.urmc.rochester.edu
2. Deaver M. J.; Silman E. F.; Lotfipour S. (2009). "Infected thyroglossal duct cyst". Western Journal of Emergency Medicine. 10 (3): 205.
3. SRB's Manual of Surgery 3rd edition 2009;405;406.
4. Stahl W.M.; Lyall D. (1954). "Cervical cysts and fistulae of thyroglossal Tract Origin". Annals of Surgery. 139 (1): 123–128. doi:10.1097/00000658-195401000-00018. PMC 1609283.
5. The State of Queensland. (2011) Thyroglossal cysts/ fistuka. Retrieved from http:// www.health.qld.gov.au
6. Koca H, Seckin T, Sipahi A, Kaznac A. Epidermoid cyst in the floor of the mouth: report of a case. Quintessence Int. 2007;38:473–477. [PubMed]
7. Ozan F, Polat HB, Ay S, Goze F. Epidermoid cyst of the buccal mucosa: A case report. J Contemp, Dent Pract. 2007;3:90–96. [PubMed]
8. Pancholi A, Raniga S, Vohra PA, Vaidya V. Midline submental epidermoid cyst: A Rare Case. Internet J Otorhinolaryngol. 2006;2
9. Kandogan T, Koc M, Vardar E, Selek E, Sezgin O. Sublingual epidermoid cyst: a case report. J Med Case Reports. 2007;1:87. doi: 10.1186/1752-1947-1-87. [PMC free article] [PubMed] [CrossRef]
10. Damle MV, Irani DK, Hiranandani NL. Epidermoid cyst of the floor of the mouth. case report. Bombay Hosp J. 2002;44
11. Seah TE, Sufyan W, Singh B. Case report of a dermoid cyst of the floor of the mouth. Anna Acad Med Singapore. 2004;33:77S–79S. [PubMed]
12. Hemaraju N, Nanda SK, Mediker SB. Sublingual dermoid cyst. Indian Journal of Otolaryngology and Head and Neck Surgery. 2004;3:218–220. [PMC free article] [PubMed]
13. Shear M, Speight P. Cysts of Oral and Maxillofacial Region. 4. Blackwell Munksgaard; 2007. pp. 181–183. [CrossRef]
14. Rutherford SA, Leach PA, King AT. Skull Base: An Interdisciplinary Approach. 2006. pp. 109–115. [PMC free article] [PubMed]