Case Report

Bronchogenic Cyst of the Tongue in an Infant: A Case Report and Review of Literature

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ABSTRACT

Bronchogenic cysts are rare congenital lesions and represent the most common primary cysts of the mediastinum. However, extra thoracic bronchogenic cysts are uncommon occurrences and very few bronchogenic cysts in intraoral sites have been reported. Lingual bronchogenic cysts are rare and are considered by many authors to be choristomas. Although the aethiopathogenesis is not known with certainty, it has been suggested that the cysts may arise from undifferentiated embryonic rests of primitive foregut that are misplaced or entrapped between parts of the developing tongue. This case report discussed the case of a large lingual bronchogenic cyst of the tongue in 31 day old infant that enucleation of cyst done without recurrence 12 months after treatment.

INTRODUCTION

Bronchogenic cysts are rare congenital lesions and represent the most common primary cysts of the mediastinum.[1] However, extra thoracic bronchogenic cysts are uncommon occurrences and very few bronchogenic cysts in intraoral sites have been reported.[2]

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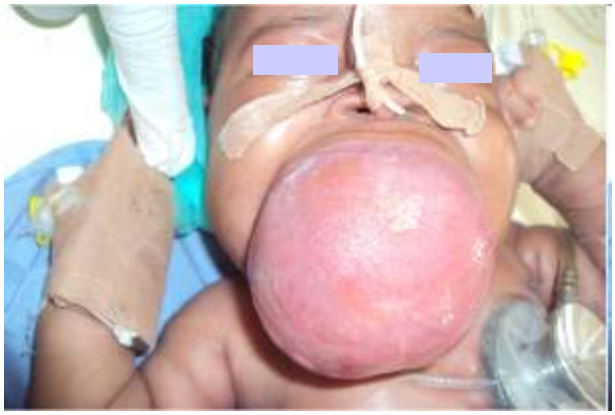
Bronchogenic cysts are lined by respiratory epithelium, that is ciliated, pseudo stratified squamous epithelium, comprising mucus-secreting cells, smooth muscle cells and cartilaginous tissue and this suggests that they are choriostomas.[2] However, authors have reported that choristomas of the oral cavity may comprise of bronchogenic cysts (lined by respiratory epithelium) and gastrointestinal duplications (lined by gastro-intestinal epithelium) while lesions comprising both types epithelium have rarely been reported.[3,4]

We present this case report of bronchogenic cyst in a newborn African infant to add to existing literature.

Case report

A 31 day old baby was brought to the children emergency unit of the Federal Medical Centre Abeokuta (a tertiary referral centre) on account of a progressively increasing massive tongue mass present from birth which resulted in inability to breastfeed. Examination revealed a small for age baby (weighed 2.7 kilogram) that was not pale, anicteric. There was a massive swelling within the substance of the tongue, extending from the tip of the tongue to beyond the junction of the anterior two thirds and posterior one third with obliteration of the oral cavity (Fig 1). There were areas of ulcerations on the ventral aspect of the tongue and increased vascular markings on the dorsal aspect. The swelling was soft to palpation and trans-illuminated light. A clinical diagnosis of dermoid cyst was made.

Fig 1: shows large tongue swelling filling almost the entirety of the tongue

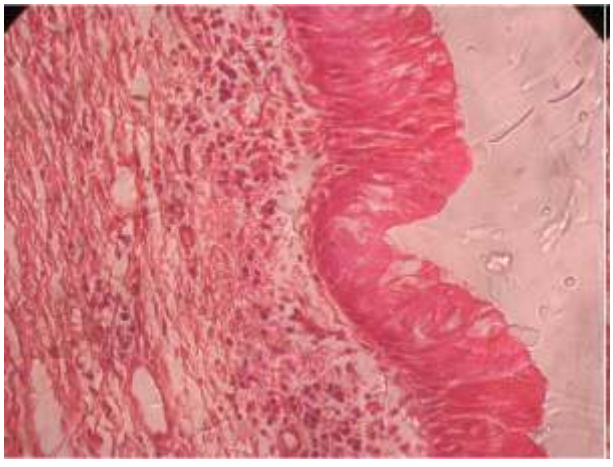


Patient was admitted and commenced on nasogastric tube (NG) feeding, antibiotics were also administered. Ultrasound scan revealed a well marginated cystic tongue mass with regular outline measuring 46.5x47.9x49.3mm (L X AP X T) with a volume of 57.8 millilitres. It had fine internal echoes with associated single layering. The lesion was non-vascularised on Doppler interrogation. While on admission, serial aspiration of the cystic mass was done on three different occasions and a total of 350 millilitres of dirty creamy aspirate was evacuated. Patient had cystic enucleation done under general anaesthesia at 64 days of life. Fig (2).

Fig 2: Shows patient 4 weeksafter cystic enucleation

Histology revealed a large unicystic lesion lined by pseudo-stratified epithelium with isolated ciliated areas. The features were descriptive of a bronchogenic cyst.

Fig 3: H&E (x400) shows cystic lesion with a pseudo-stratified epithelium with sparse cilia and mild inflammatory cells infiltrate in connective tissue.



DISCUSSION

Lingual bronchogenic cysts are rare occurrences and have been variously considered to be choristomas (or heterotopic cysts).[2] Manor et al.[5] had previously described 52 lingual cysts considered to be bronchogenic or gastrointestinal choristomas:12 of which were characterized by a respiratory epithelium (bronchogenic cysts), 25 were characterized by gastrointestinal epithelium and 15 were characterized by a combination of the two types of epithelium. Many authors have previously used the same terms to qualify bronchogeniccysts, gastrointestinal duplications or a combination of the two.[6,7,8] Previous reports have noted a predilection of these cysts for the anterior two-third of the tongue and the floor the mouth mostly in infant boys.[8,9,10]; these were in concordance with the present case, although, the lesion extended beyond the anterior two-thirds of the tongue.

Although, several hypothesis have been suggested, the aetiopathogenesis of bronchogenic cyst has not

been described with certainty.[4] Bronchogenic cyst may result from an early aberrant nodule that becomes detached from the primitive tracheobronchial tree or maybe derived from cells of the oesophagotracheal ridge, forming an isolated bronchial structure.[4] Another hypothesis of the aetiopathogenesis of bronchogenic cyst suggested thatthe cysts may arise from undifferentiated embryonic rests of primitive foregut that are misplaced or entrapped between parts of the developing tongue.[4,11] Similarly, some authors have hypothesised that bronchogenic cyst may arise from retention cysts of salivary gland origin of the tongue, or may even be due to the metaplasia of oral epithelium but these last two hypothesis seem unlikely.[4]

The differential diagnosis of lingual bronchogenic cysts include choristomas such as foregut duplication and bone choristomas. Other differentials of bronchogenic cyst will include cystic lymphangioma, haemangioma, thyroglossal duct cyst, dermoid cyst or sialocele.[12]

Lymphangiomas are benign congenital or hamartomatous malformation of lymphatic vessels and similar to this case, they typically appear at birth and 90% will appear by 2 years of age[13]. In the same vein, oral lymphangiomas have a predilection for anterior two-thirds of the tongue. However, unlike in this case, histologically, they present as multiple intertwining lymph vessels lined by thin endothelium[13].

Dermoid cyst are rare congenital cysts that are believed to arise as a result of entrapped pluripotent cells during embryogenesis[14]. Dermoid cyst are mainly seen in the midline of the body and in the testis and ovaries but the floor the mouth is the commonest intraoral site[15]. Dermoid cyst are extremely rare in the tongue, more so, histologically they arelined by epidermis and characteristically contain skin adnexa[15].

On the other hand, thyroglossal ductal cysts are congenital neck masses and result from the embryologic remnants of the descending thyroglossal duct and are found along its path from the foramen caecum to the pyramidal lobe of the thyroid gland[16]. They typically present in anterior neck and tongue lesions are rare and when they occur in the tongue, they usually present at the base of the tongue[16]. Thyroglossal duct cysts may however be histologically similar to Bronchogenic cyst as it may be lined by pseudo stratified epithelium, by stratified squamous epithelium or by both pseudo stratified epithelium and stratified squamous epithelium[16].

Bronchogenic cysts are often reported at birth as in this present case, however, asymptomatic cases may be diagnosed later in life.[4]This case presented with difficulty in feeding which resulted in stunted growth. This is a common complication of Bronchogenic cyst. Other reported complications include upper airway obstruction, respiratory difficulties and infection and possibly abscess formation.[4,5] These other complications were not observed in the case being reported. Malignant transformation of untreated chronic lingual cyst to adenocarcinoma has been previously reported and early treatment, weighing the balance of anaesthetic complications and the complications of untreated cases should be considered in the management of cases.[17] In our case, though difficult intubation was anticipated, patient successfully had a nasotracheal intubation which was uneventful.

CONCLUSION

This was a review of a rare case of lingual bronchogenic cyst. The presentation in the tongue, in a male infant were similar to several reports. Also, like previous authors, we recommend early surgical enucleation of the cyst and regular follow up. There were no post-surgical complications in this patient and the lesion has not recurred after 12 months of follow up.

REFERENCES

1. Sarper A, Ayten A, Golbasi I, Demircan A, Isin E. Bronchogenic cyst. Tex Heart Inst J. 2003; 30(2):105-8.
2. Kun Darbois JD, Breheret R, Bizon A, Pare A. Laccourreye L. Bronchogenic cyst of the tip of the tongue: report of two cases. Eur Ann Otorhinolaryngol Head Neck Dis. 2015; 132 (1):49-51.
3. Blanchard M, Kadlub N, Boudjemaa S, et al. [Tongue cyst in children:foregut duplication, a possible diagnosis]. Rev StomatolChirMaxillofac2012; 113:442–7.
4. Joshi R, Cobb AR, Wilson P, et al. Lingual cyst lined by respiratory and gastric epithelium in a neonate. Br J Oral MaxillofacSurg 2013; 51:173–5.
5. Manor Y, Buchner A, Peleg M, et al. Lingual cyst with respiratory epithelium: an entity of debatable histogenesis. J Oral MaxillofacSurg 1999; 57:124–7.
6. Wiersma R, Hadley GP, Bosenberg AT, et al: Intralingual cysts of foregut origin. J Pediatr Surg. 1992; 27 (11):1404-6.
7. Mirchadani Y, Sciubba J, Gloster ES: Congenital oral cyst with heterotopic gastrointestinal and respiratory mucosa. Arch Pathol Lab Med. 1989; 113:1301.
8. Kinoshita Y, Honma Y, Otuka T, et al: Gastrointestinal mucosal cyst of the oral cavity: Report of a case and a review of literature. J Oral Maxillofac Surg. 1994; 52 (11):1203-5.
9. Bailey BM. A detached bronchogenic cyst occurring in the tongue of a neonate. *Br J Oral Surg*. 1982; 20:288–93.
10. Eaton D, Billings K, Timmons C, Booth T, Biavati JM. Congenital foregut duplication cysts of the anterior tongue. *Arch Otolaryngol Head Neck Surg*2001; 127:1484- 7.
11. Gorlin RJ, Jirasek JE. Oral cysts containing gastric or intestinal mucosa. An unusual embryological accident or heterotopia. *Arch of Otolaryngol*. 1970; 91:594–7.
12. Akyol MU, Orhan D. Lingual tumors in infants: a case report and review of theliterature. Int J PediatrOtorhinolaryngol 2004; 68:111–5.
13. Pammar C, Kotrashetti VS, Nayak R, Hosmani J. Lymphangioma of the buccal mucosa: A case report with a literature review. J OrofacSci 2015; 7:129-31.
14. Bonet-Coloma C, Mínguez-Martínez I, Palma-Carrió C, Ortega-Sánchez B, Peñarrocha-Diago M, Mínguez-Sanz JM. Orofacialdermoidcysts in pediatricpatients: a review of 8cases. Med Oral Patol Oral Cir Bucal. 2011: 1; 16(2):e200-3.
15. Fomete B, Saheeb B D, Onyebuchi E P, Ogbeifun J O. Dermoid cysts of the oral cavity as seen in a Nigerian Tertiary Institution. Niger J Surg Res 2013; 15:3-6.
16. Thompson LD, Herrera HB, Lau SK. A Clinico-pathologicSeries of 685 Thyroglossal DuctRemnantCysts.Head Neck Pathol. 2016; 10(4):465-474.
17. Volchok J, Jaffer A, Cooper T, et al. Adenocarcinoma arising in a lingual foregut duplication cyst. Arch Otolaryngol Head Neck Surg. 2007; 133:717–9.