

Bronchogenic cyst of the tongue in a Sudanese adult lined by respiratory and gastric epithelium: A very rare Localization

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Abstract-*Bronchogenic cyst in the tongue is a rare heterotopic cysts, considered as a choristoma. It is lined by respiratory epithelium. Thoracic bronchogenic cysts originate from the ventral foregut that forms the respiratory system. The authors report a case of bronchogenic cyst of the tongue in adult Sudanese patient presented with macroglossia. Ultrasound and MRI Imaging revealed a median cystic lesion, suggesting an embryonic tumor. Histopathologic examination after surgical excision confirmed the diagnosis of lingual choristoma of the bronchogenic cyst type. Considered that enucleation is the best treatment option. There were no post-operative complications or recurrence.*

Keywords-*Bronchogenic cyst, lingual cyst, choristoma, heterotopic cyst, macroglossia*

I. INTRODUCTION

Choristoma are non-malignant embryonic tumors, characterized by the presence of normal tissue in abnormal site. The bronchogenic cyst is a choristoma affecting the upper aero digestive tract. It is lined with respiratory or oral epithelium. When this lesion affects the tongue, it is considered as an extremely rare congenital malformation especially in adult. The purpose of this article is to describe a new case of lingual bronchogenic cyst with respiratory and oral epithelium in Sudanese adult.

II. CASE REPORT

Twenty years Sudanese patient presented in maxillofacial surgery clinic in Khartoum Teaching Dental Hospital with a painless swelling in the tongue appeared 10 years ago that increased in size gradually. There was a disturbance in speaking and eating. There is no history of trauma or previous operation. He denied any medical problems or medical treatment.

On general examination, the patient looks well, not pale, not jaundiced, and not cyanosed. There is no facial dysmorphism or congenital malformation in the body. Examination of the tongue showed huge well circumscribed swelling occupied the anterior two third, 4*3 cm in diameter, with normal colour. The lesion is not tender, not pulsatile and fluctuant in consistency. The

surface texture is smooth and there is no ulceration. Primarily hyperechoid cystic lesion appeared in ultrasound. MRI demonstrated cystic lesion with high intensity signal on T2 and low intensity signal on T1.

Needle aspiration was performed for preoperative assessment and revealed a gray fluid. Surgical exploration was performed under nasotracheal general anesthesia via a midline sagittal glossotomy incision. The lesion was completely dissected and excised, the histopathological examination of the surgical specimen revealed a benign cyst lined by respiratory epithelium and focally lined by stratified epithelium. The cyst wall shows smooth muscle and seromucinous glands along with chronic inflammatory cell infiltrates. After surgery eating and drinking returned to normal. The immediate postoperative and long term course was uneventful with no sign of recurrence. Healing was normal.

III. DISCUSSION

Bronchogenic cyst is considered as congenital oral cysts of foregut origin or lingual alimentary cyst. Other congenital oral cysts of foregut origin include enteric, and oesophageal cysts. They are much rarer than the well described dermoid, epidermoid and thyroglossal cysts. There are several synonyms for lingual cyst of foregut origin and for lingual alimentary tract cyst, these included: lingual cyst, intralingual cyst of foregut origin, gastric cyst, heterotopic gastric cyst, cyst with intestinal epithelium, enteric duplication cyst, congenital duplication cyst, choristomatic cyst, gastric choristoma and enterocytoma [1].

Bronchogenic cyst is aberration secondary to abnormal detachment of accessory lung buds from the primitive foregut [2]. The exact aetiology is poorly understood, but they are thought to arise from misplaced embryonic rests of the primitive foregut. During differentiation in the 3rd week of embryonic development, the foregut undergoes a dichotomy; a ventral part that forms the respiratory tract and a dorsal part that creates the esophagus, stomach, and duodenum.

Due to close proximity of the primitive foregut to the pharyngeal arches, embryonic rests may be misplaced and entrapped between the parts of the tongue and the tuberculum impar, during the 4th week. These entrapped rests are exposed to different inductive influences and can differentiate to respiratory or well differentiated gastrointestinal epithelium. These cysts are located close to the trachea or main stem bronchi. Rarely there is communication of the cyst with the tracheobronchial tree [1].

Bronchogenic cysts are widely reported in the literature. In adult, they are found in mediastinum and lung parenchyma. However they may be located in the thoracic wall, abdomen or cervical region. Rarely affects the tongue. Presentation may range from life-threatening to completely asymptomatic in all age groups of patients. Most of these lesions are discovered at birth and may appear later in adult life [3].

Manor Y et al [1] reviewed 53 Lingual bronchogenic cyst which 29 could be grouped into lingual alimentary tract cysts and 24 into lingual cyst of foregut origin. Kun-Darbois et al [4] reported two cases of bronchogenic cyst in tip of the tongue in a nine day old boy and in a three months old girl. Other cases of lingual bronchogenic cysts have been reported in the literature most of them affect neonates or young children [5,6,7,8] or rarely affects adults [9] as in our case.

Very rare cases of a congenital dermoid cyst together with a bronchogenic cyst of the tongue (teratoid cyst of the tongue) have been accounted [10,11]

Bronchogenic cysts can be located in the neck. Perez JA et al [12] reviewed 70 published cases of cervical bronchogenic cysts, 75% were located in the midline of the neck, in the upper third or in the lateral lines mostly in the lower third.

Investigation of such lesions may be varying from simple needle aspiration to assess fluid contents, to ultrasound which reveals cystic cavity, but MRI is the best imaging modality to evaluate bronchogenic cyst in the tongue. [4]

The main differential diagnosis for congenital cyst in the tongue is lymphangioma [13], followed by dermoid cyst, haemangioma or even ranula involving the ventral surface of the tongue. The lesion must therefore be surgically enucleated for diagnosis, treatment and prevention.

The epithelial lining of the lingual cyst of foregut origin is composed mainly of respiratory tract epithelium-pseudostratified ciliated columnar and cuboidal, and the epithelial lining of the lingual alimentary tract cyst is composed mainly of gastric or intestinal mucosa [1]. However, there are several reports in the literature in which the epithelial lining of the lingual cyst was

composed of both types of epithelium, respiratory and gastric or intestinal epithelium as in our case.

Malignant transformation to adenocarcinoma is a very rare complication of chronic untreated lingual bronchogenic cyst, cases were reported in literature [14].

IV. CONCLUSION

Although lingual bronchogenic cysts are infrequent especially in adult, they should be considered as part of the differential diagnosis for cystic lesions in the tongue. Most of the cases affects the anterior two third, either the dorsal or ventral surface.

V. CONFLICTS OF INTERESTS

The authors have no conflicts of interests to declare

VI. COMPLIANCE WITH ETHICAL STANDARD

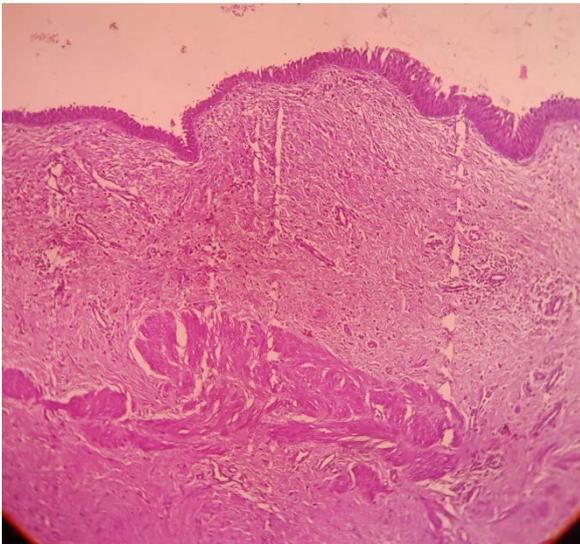
Ethical approval was obtained from Khartoum Teaching Dental Hospital medical director, and informed consent was obtained from the patient.

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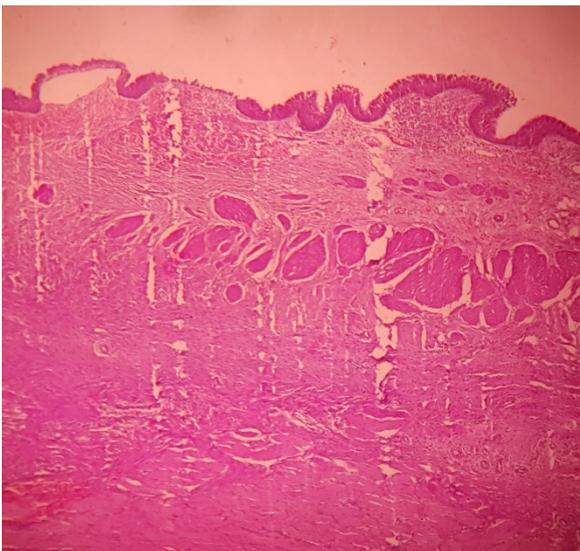
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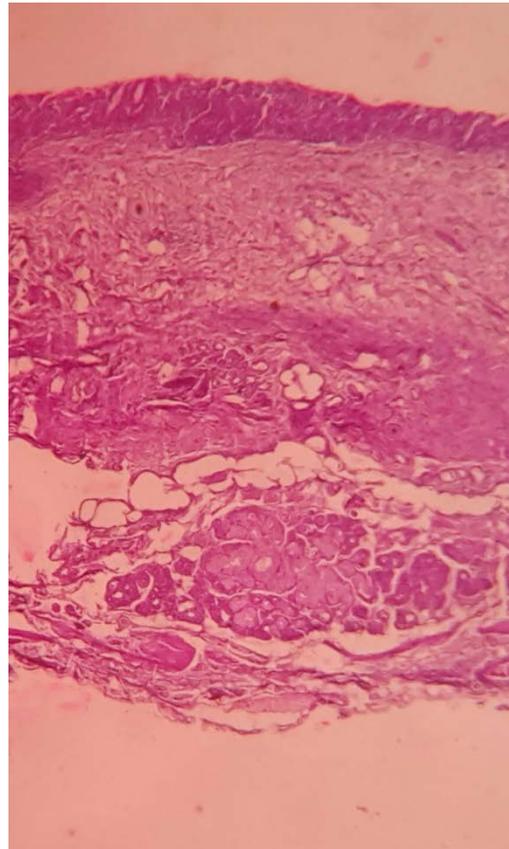
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(A) : Cystic lining composed of ciliated columnar epithelium with goblet cells (H&E original magnification 10X).



(B) : Clusters of smooth muscle fibers under the lining (arrowheads, H&E original magnification 10X).



(C) : Seromucous gland (arrowhead, H&E original magnification 10X).