Review Article

Foregut duplication cyst presenting as lingual swelling: Case report and review of literature

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ABSTRACT

Congenital lingual cystic masses are challenging entities that can be detected prenatally or are discovered in various forms after birth. Foregut duplication cyst of tongue is an extremely rare condition. Here we present the eleventh case in literature on foregut duplication cyst in tongue lined with gastric mucosa. Complete excision was curative with no recurrence on followup.

Key words: Foregut, duplication cyst, lingual swelling

Introduction

A lingual cystic mass in a newborn is an interesting challenge for the pediatric surgeons. Although most cases are brought to attention following birth, some are detected prenatally. Some of these masses present with respiratory distress requiring emergency intervention. [1,2] Among the different lesions that can arise in the tongue, the foregut duplication cyst is of special interest because of its rarity, variable characteristics, and its puzzling embryogenesis. Until now, only ten cases of intralingual foregut duplication cysts lined with gastric mucosa have been reported in the literature [Table 1].[3-9]

Review Report

This neonate presented with mass in tongue on tenth day of life [Figure 1], which was diagnosed as tongue lymphangioma in anomaly scan at the 8th month of gestation. Fullterm child with 3kg birth weight was not able to take breast feed and was on spoon feeding. On examination, his right side of the tongue and floor of mouth was diffusely enlarged, and a bulge was noted at the right submandibular region. On palpation, cystic, nonpulsatile mass was felt in the body of the tongue. Ultrasound revealed a 4 × 4 × 2.2 cm mixed cystic mass, probably lymphangioma. Computed tomography (CT) scan detected a 3.6 × 3.4 × 4.2 cm solid mass with cystic areas confined to the tongue with involvement of floor of mouth. There was diagnostic dilemma between lymphangioma and cystic variety of teratoma. Malignancy workup was negative. Marspulisation and wedge biopsy was taken as complete excision was not possible. Histopathology was suggestive of mucous retention cyst and ruled out malignant lesion.

Postoperatively the mass gradually increased in size and by the age of 2.5 months he began exhibiting difficulty in feeding. Magnetic resonance imaging (MRI) revealed multicystic purely tongue-based lesion with involvement of floor of mouth and with no involvement of other surrounding structures [Figure 2]. Patient underwent surgery under general anesthesia with nasotracheal intubation and throat pack [Figure 3]. The cyst was excised completely. The cyst wall was thick and adherent to the lingual muscles. The lesion extended to the base of the tongue [Figure 4]. The excised mass measured 3 × 2.4 × 1.2 cm. Postoperative course was uneventful [Figure 5].

Histopathology revealed oral cystic lesion lined

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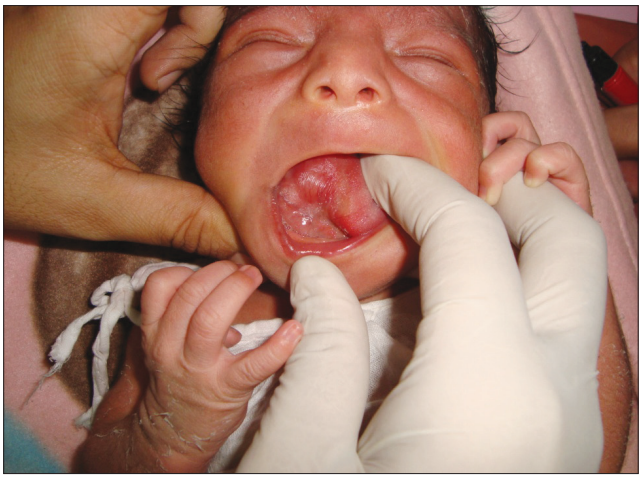

Figure 1: Tongue mass

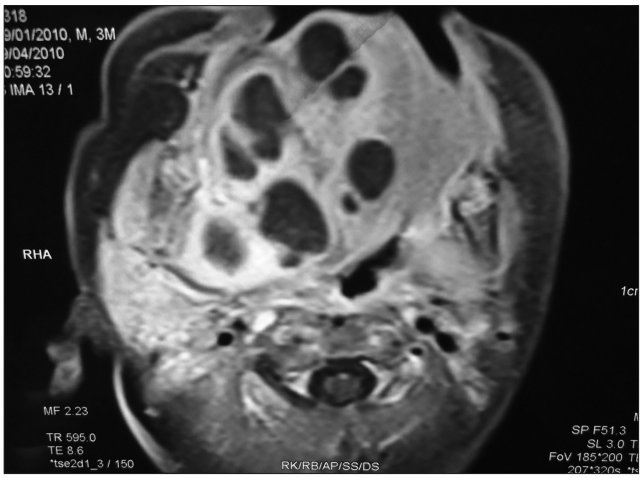

Figure 2: Magnetic resonance imaging

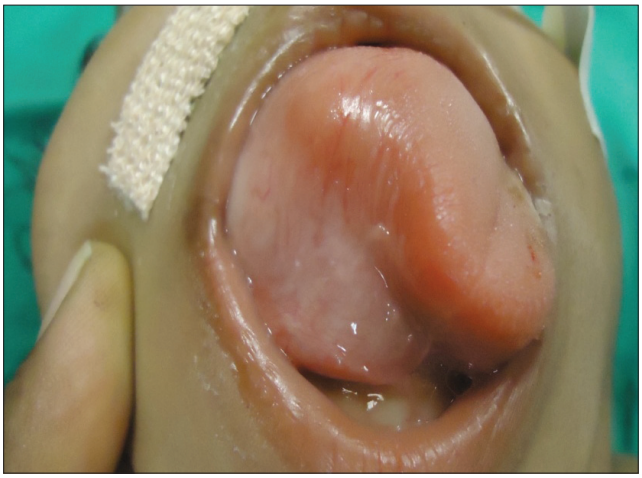

Figure 3: Pre op

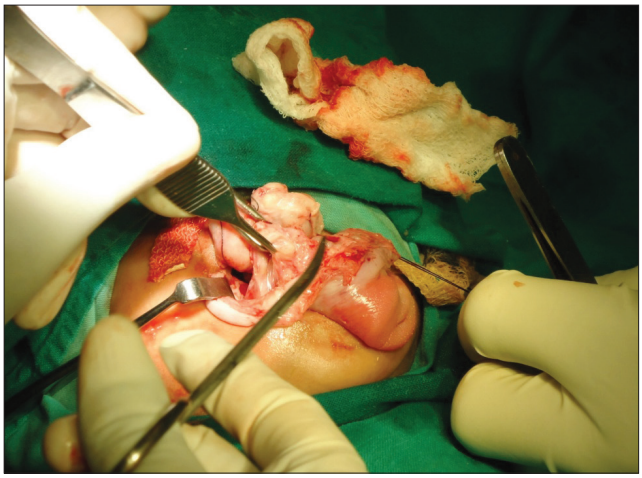

Figure 4: Intra op

Table 1: Reported cases of gastric variety of intralingual foregut duplication cyst

Author	Age and sex of patient	Presentation
Duncan and Daniel[3]	2 mo/M	Cyst had enlarged
Lister and Zachary[4]	11 mo/M	Emerging teeth damaged cyst
Lister and Zachary[4]	Newborn/F	Cyst caused tongue protrusion
Willis[5]	4 mo/M	Not specified
Harris and Courtemanche[6]	6 mo/M	Cyst caused tongue protrusion
Brown and Kerr-Wilson[7]	11 mo/M	Cyst had enlarged
Willner, *et al*,[8]	8 mo/F	Asymptomatic and stable
Said Al Naief[9]	2 yr/F	Cyst affected speech and feeding
El- Bitar, *et al*,2003	2 mo/M	Cyst affected feeding
Kumar S, *et al*,[20]	New born/M	Cyst asymptomatic
Present case	Day 10/M	Cyst caused tongue protrusion

by gastric type of epithelium. It featured an outer smooth-muscle layer (muscularis propria, with circular and longitudinal layers), submucosa, and areas of mucosal lining that contained pits and glands and were consistent with gastric mucosa. Other areas of the mucosa were of the simple columnar type [Figure 6].

Discussion

A cystic mass in the anterior two-thirds of the tongue in a newborn can represent ranula, lymphangima, haemangioma, and dermoid cyst.[4,10,11] Since first reported by Duncan and Daniel[9] in 1942, cysts lined with gastric and/or intestinal mucosa have rarely been seen in the tongue or the floor of the mouth. These cysts have usually been reported along the alimentary tract, from the esophagus to the colon,[10,12] and in the gallbladder, pancreas, lungs, larynx, and urinary bladder.[13]

Congenital gastric and intestinal cysts of the oral cavity are more common in boys than in girls.[2,8,9,14] These masses involve the anterior aspect of the tongue in 60% of cases.[9,10] Some are asymptomatic, and some cause various degrees of feeding and breathing difficulties or manifest in unexpected ways, such as recurrent bleeding[13] or a brownish discharge from a lingual sinus.[10] Most of these cysts are solitary, but in some cases more than one cyst has been present.[14,15]

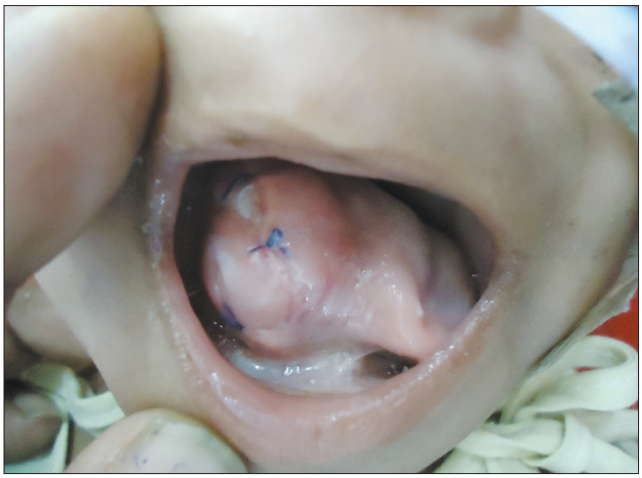

Figure 5: Postop appearance

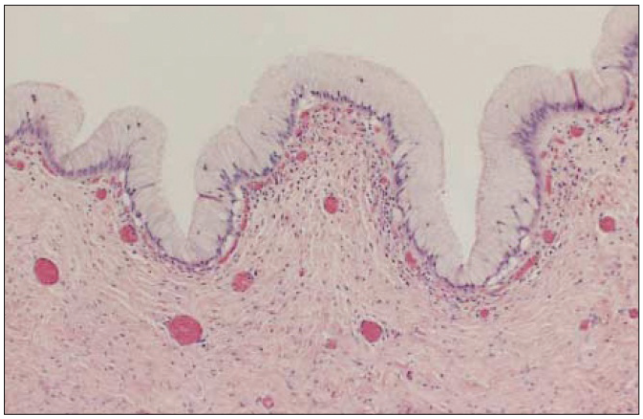

Figure 6: Histopathological picture

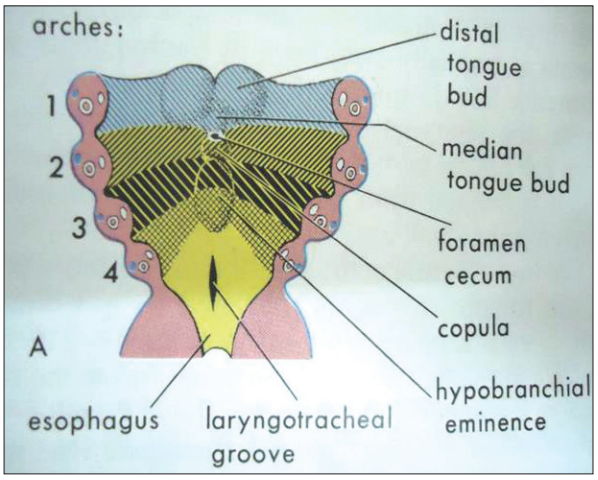

Figure 7: Developmental picture

These lesions have been given several names, including cystic choristoma, heterotopic gastrointestinal cyst, enterocystoma, and duplication cyst.[9] Our case fulfilled Rickham's diagnostic criteria for duplication cysts including the presence of a smooth-muscle coat, an attachment of the cyst to a part of the alimentary tract, and the presence of a mucosal lining from the alimentary tract.[4,7,8,11,16]

Extensive research revealed only ten cases of which only two were multilocular.[3-9,17] Lingual foregut duplication cysts are believed to arise from endodermal cells that become trapped during the fusion of the lateral lingual swelling (distal tongue bud) and the tuberculum impar (median tongue bud) in the 3- to 4-mm embryo.[2,10,14] These cells are believed to derive from the stomodeum.[18] These pluripotential cells can differentiate into various types of epithelium, including gastric, intestinal, colonic, and even respiratory [Figure 7].

In addition to a clinical examination,[10,13] the initial evaluation of a newborn with a lingual mass can include ultrasonography, Computed tomography(CT), and/or magnetic resonance imaging(MRI).[8,9] Clinical examination alone cannot differentiate among the wide variety of possible lesions. Anomaly scan is also a useful tool for antenatal diagnosis of the lesion.

Excising an intralingual cyst whose wall adheres to the surrounding muscles is a tedious exercise, especially in an infant with a large cyst. Total, if not possible then subtotal excision is curative.[15,17,19,20]

Conclusion

Intralingual foregut duplication cyst should be considered in the differential diagnosis of a lingual cystic mass in an infant. Early surgical excision in a symptomatic patient is desirable in order to avoid failure-to-thrive, respiratory distress, or more extensive surgery later.

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