Case Report

Choristoma: A rare congenital tumor of the tongue

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ABSTRACT

Most congenital masses of the mouth floor are cystic, irrespective of their origin. They may obstruct airway or digestive tract or may present other complications. Recognizing and treating these masses expeditiously is imperative. Choristoma is a mass of normal tissue in an abnormal location; they are classified according to the predominant epithelial lining. Few cases have been reported lined with gastric and respiratory epithelia within the same cyst. This case report presents a 5-month-old boy with an infected choristoma involving the floor of the mouth and its successful management.

Keywords: Floor of the mouth, heterotopic, lingual cyst

INTRODUCTION

Congenital masses of the floor of the mouth are not rare, most of them being cystic. The most common are lymphatic malformations and teratomas, followed by dermoid, epidermoid, and thyroglossal cysts. Salivary glands' obstruction could be explained by anatomical variations of the ducts or an inflammatory process, and they are not uncommon in children; sialolithiasis occurs frequently in adults and only 3% of them affect children. Tumors of the surrounding tissues should be ruled out.[1]

Cysts lined by respiratory and gastrointestinal epithelia in the oral cavity are unusual.[2]

We present a case of a cyst on the ventral surface of the tongue lined with mucosal ectopia, which was diagnosed only after histological examination.

CASE REPORT

A 5-month-old boy was admitted with a swelling under the tongue and a history of fever in the past 72 h; the bulging had increased in size in the past 2 days. He had no swallowing or feeding problems. His parents had noticed soon after birth that he had an elevated tongue. An ultrasound study had been scheduled.

Clinical examination revealed an erythematous, tender, fluctuant sublingual mass (5 cm × 3 cm) reaching from the submental area to the mouth floor.

The patient was started on intravenous amoxicillin and clavulanic acid. He underwent submental drainage of the abscess. After this procedure, the patient remained intubated in the pediatric Intensive Care Unit to prevent complications secondary to local swelling. The bacteriological culture examination was negative.

The mass did not disappear after drainage. Magnetic resonance imaging (MRI) demonstrated a multicystic mass showing inflammatory signs, reaching from the floor of the mouth to the base of the tongue [Figure 1].

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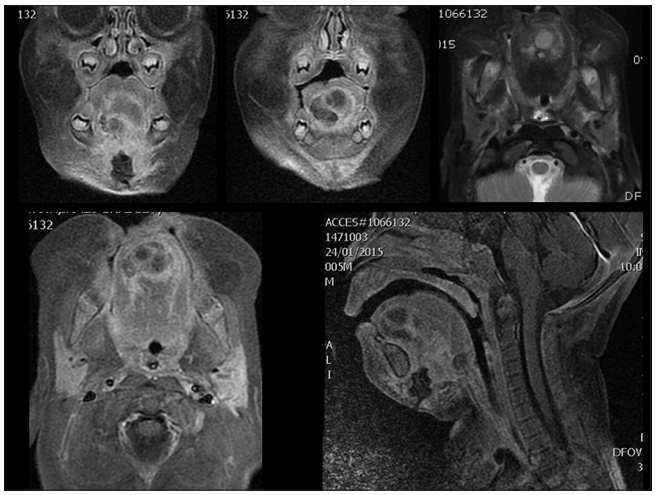

Figure 1: Magnetic resonance imaging shows multicystic mass with inflammatory signs, reaching from the floor of the mouth to the base of the tongue

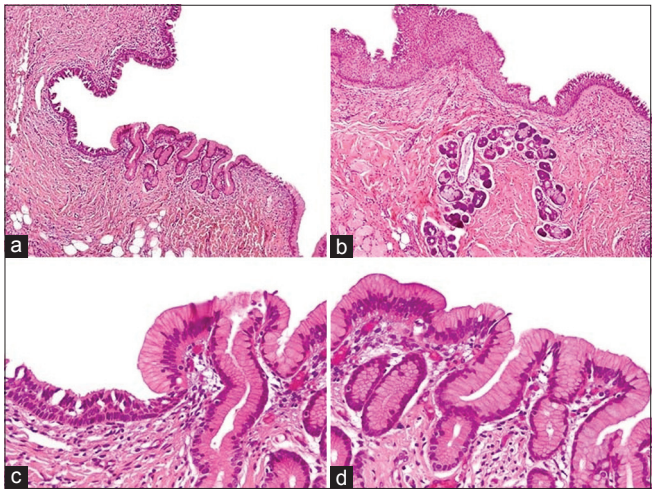

Figure 3: Heterotopic gastrointestinal cyst (H and E). (a) 40x. The cyst wall is lined by respiratory and gastric epithelia. (b) 100x. Transition between stratified squamous and respiratory epithelia. Seromucous glands are also seen in the subepithelial tissue. (c) 200x. Ciliated columnar epithelium next to gastric epithelium. (d) 400x. Note that features in gastric epithelium are typical mucous cells of gastric foveolar glands

Mass excision was performed through a midline incision along the lingual frenulum, under general anesthesia with nasoendotracheal intubation. Dissection of the mass was carefully accomplished identifying and preserving both Wharton's ducts and lingual nerves.

On gross examination, the specimen consisted of a cystic mass measuring 2 cm in diameter containing colloid material. Figure 2 shows details of the procedure and specimen.

The histological examination showed a cystic lesion lined with respiratory epithelium, stratified squamous epithelium, and tall columnar mucus-secreting epithelium. The latter type is characteristic of gastric surface epithelium. Seromucous glands were also identified in subepithelial tissue [Figure 3]. A fistulous connection to the epidermal surface was observed, related to the

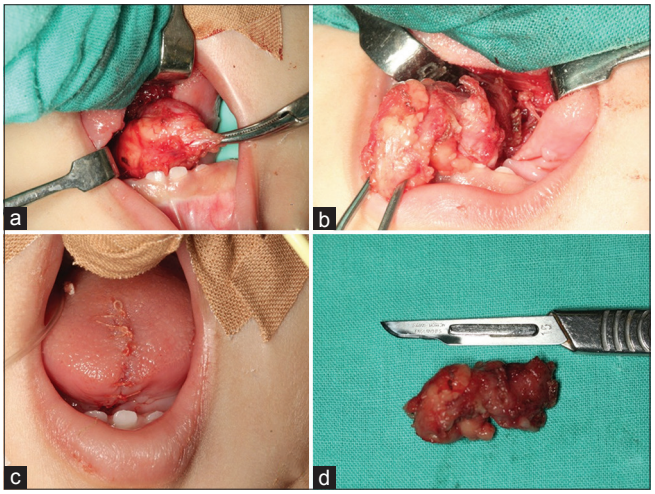

Figure 2: Surgical (a) Intraoral approach through a midline incision along the lingual frenulum. (b) Surgical excision of the mass. (c) 5/0 vicryl suture. d) Surgical specimen

previous drainage. In addition, the lesion showed rupture signs as granulomatous reaction, chronic inflammatory infiltrate, and interstitial fibrosis.

It has not been identified as thyroid parenchyma after inclusion of all materials submitted.

The floor of the mouth and tongue returned to their normal position immediately after surgery. Postoperative follow-up has been uneventful. Two months after surgery, a 2 mm whitish, subcutaneous, noninflammatory node was noted near the submental drain scar. An ultrasound detected a small retention cyst in the subcutaneous tissue, not extending beyond the submental fat. A follow-up ultrasound was performed, and an epidermoid inclusion cyst was removed 3 months later. The patient remains uneventful until now.

DISCUSSION

Choristoma is a mass consisting of normal tissue in an abnormal location.[3] The pathogenesis of lingual choristoma is still uncertain; they are probably derivatives of primitive foregut endoderm and can contain different epithelia: glial, cartilaginous, osseous, thyroid, gastric, and respiratory.[2] The foregut contains components of the endoderm and mesoderm that lead to the development of the trachea, bronchi, esophagus, liver, stomach, and intestine. Abnormal separation of the embryonic tracheoesophageal septum can lead to the formation of this kind of cyst.[4-6]

The mass/cyst usually presents as asymptomatic swelling in the anterior two-thirds of the tongue, but one-third of the cases can cause increased salivation, altered speech, and difficulties with feeding and respiration. Infection, hemorrhage, asphyxia, or ulceration are rarely seen findings.[1,6,7]

There is a slight predilection for males (1.6:1).[2]

An imaging including MRI, computed tomography scan, and/or ultrasound assessment may be used to confirm location of the

mass and its relation to the anatomic structures to correctly plan the surgical approach. Congenital lingual cystic masses can be detected prenatally using ultrasound.[8]

Surgery is the treatment of choice; the approach to resection is based on accessibility to the mass, size, and airway control. Recurrence is uncommon.[7,9,10]

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Conflicts of interest

There are no conflicts of interest.

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