HIDDEN FOR YEARS: A CASE OF AN OSSEOUS CHORISTOMA ARISING FROM BASE OF THE TONGUE

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ABSTRACT

Osseous choristoma is a soft tissue present in an ectopic position. The choristomas, are benign tumors having bony tissues. In the oral cavity it is more commonly located on the dorsal aspect of the tongue. Such type of lesions are not true neoplasms but presenting as a mass at an abnormal location. Disease is diagnosed only after histopathologic examination. The only treatment option is surgical excision. Lingual osseous choristomas are rarely occurring entities and till now only 72 cases have been reported in the literature. In this paper we present a case of a lingual osseous choristoma presenting as a stalked soft tissue structure at the tongue base in a 36-year-old male.
Key Words: Choristoma, Base of tongue, Stalked lesion.

This case report can be cited as: Akbar FA, Sawal HA, Mushahid U, Ahmad S. Hidden for years: A case of an osseous choristoma arising from base of the tongue. Pak J Pathol. 2020; 31(1): 23-25.

INTRODUCTION

Choristoma is a tumorous mass of normal bony structure with mature cells in an ectopic position [1]. Different choristomas may present in the mouth with tissue components consisting of bone, cartilage, gastric mucosa, glial tissue, or tumor-like masses of sebaceous glands [2]. These types of lesions are also called soft tissue osteomas, but lesions are not actually neoplasms so osseous choristoma is a more specific term [3]. Choristomas mostly occur in the tongue and less commonly in buccal mucosa and alveolar mucosa [4].

PATIENT INFORMATION

A 36-year-old male presented to the ENT department Combined Military Hospital (CMH) Rawalpindi with complaints of a mass protruding and hanging from the mouth. He first noticed the mass when he was 25 years old. He also complained of slight neck discomfort, but no dysphagia, odynophagia, hoarseness or respiratory difficulty was present. He was a non-smoker and reported no comorbids. Past medical and surgical history was not significant. On examination, the oral cavity appeared normal and no obvious mass was noted. Patient forced himself to gag after which a stalked mass suddenly appeared and hung out from mouth. The mass was soft, smooth and cylindrical in shape, measuring 6x3x2 cm, with a stalk (measuring 6x2 cm) that seemed to attach to the base of tongue as shown in Figure-1.

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Received: ***22 Jul 2019****; Revised:* ***04 Oct 2019****; Accepted:* ***18 Nov 2019***

CLINICAL FINDINGS

The mass itself was painless. Fiber optic direct laryngoscopy (FODL) showed the stalk attaching to left base of tongue, and the mass appeared to be hiding within the esophagus once patient took a sip of water. Rest of FODL examination was unremarkable. Imaging showed a mass in the esophagus, with no other masses or lesions in the neck as shown in Figure-3. FNAC of the mass was inconclusive. Initially the mass was thought to be a malformation of thyroglossal duct remnant. After informed consent, patient was planned for surgical excision of the mass under G/A, both for alleviation of the patient's symptoms and for histologic diagnosis. Direct laryngoscopy (D/L) was done; the stalk of the mass was separated from its attachment to the left base of tongue and the resultant wound cauterized. The mass was sent for histopathological analysis. Patient made an uneventful post op recovery and his throat and neck discomfort resolved.

MACROSCOPIC APPEARANCE

The tissue specimen consists of a flask like piece of soft tissue collectively measuring 9.5x2x1.5cm. Thicker part measuring 5x2x1.5cm and cylindrical part measuring 4.5x0.8x0.6cm. Cut surface shows hemorrhagic white solid structure. Representative sections are taken as blocks D. Cylindrical part, full thickness RST2 as shown in Figure-2.

MICROSCOPIC APPEARANCE

The sections examined revealed a tissue covered by stratified squamous epithelium. The subepithelial loss connective tissue comprised predominantly of adipose tissue and seromucinous

salivary gland acini with increased vascular proliferation. Focal areas of cartilaginous metaplasia were also seen. No evidence of malignancy was seen in the sections examined.

At the time of writing, the patient was three months post-surgical treatment. At the clinical follow-up visit post-surgery, the patient was symptom free and seemed to be recovering well. He is planned to be reevaluated again at 3 months later on.

Figure-1: Patient showing mass appeared to hang out of his mouth.

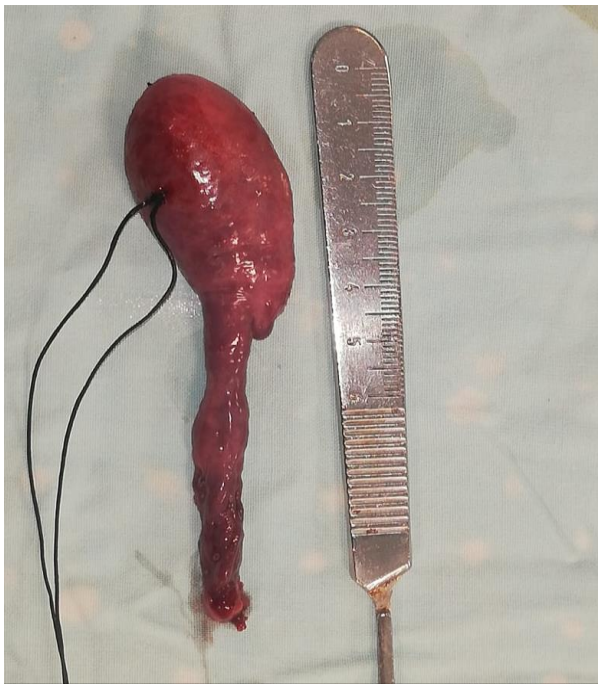

Figure-2: Macroscopic appearance.

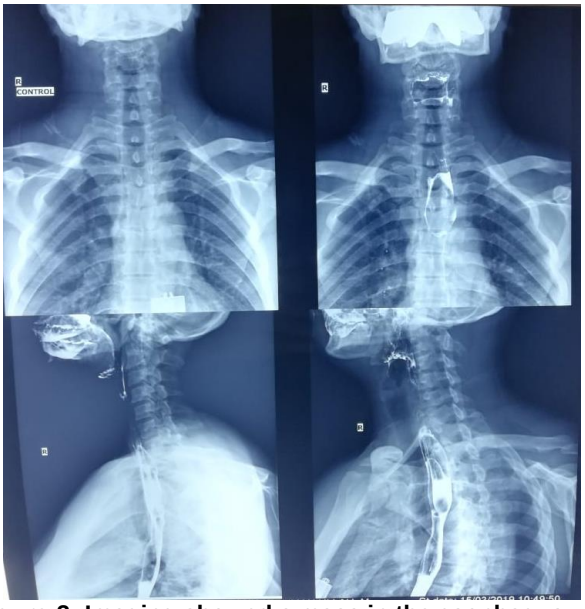

Figure-3: Imaging showed a mass in the esophagus.

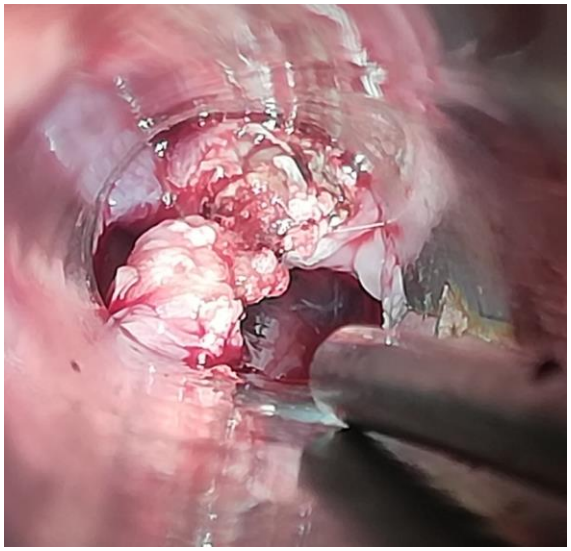

Figure-4: Direct laryngoscope view, showing stalk of the tumor partially cauterized during separation from base of tongue.

DISCUSSION

A choristoma is usually formed by heterotopic tissue, which is rarely present in the throat [5]. If they are formed parallel to respiratory epithelium they are called as bronchogenic cysts [6]. According to the reported literature of Choristomas, 72 cases have been reported till 2017 [7]. The part which is affected most frequently is the posterior part of the tongue, dorsal surface near the foramen caecum and circumvallate papillae. Lingual osseous choristomas vary in size from 3mm to 5 cm and present clinically as hard masses that can be either pedunculated or sessile. Mostly the outer mucosa shows a normal clinical appearance [8, 11]. The pathogenic cause of the choristoma is still unknown and under debate. There are numerous theories

which attempt to explain the pathogenesis of this disease. According to few authors the remnants of the undescended thyroid tissue might produce an osseous lesion but in rare cases choristoma is not localised in midline but on the border of the tongue [8]. Another hypothesis suggests recurrent irritation and trauma of the tongue as an etiology [8, 9]. A third hypothesis suggests that pluripotential cells of the branchial arches play a role in pathogenesis [13]. Cervical osseous choristomas are infrequent, but they are an important differential diagnosis to be considered in cases of cervical tumor [12]. The treatment of osseous choristomas is surgical excision by using sharp dissection, KTO laser, or electronic scalpel. Malignancy has not been reported [10]. Recurrence has been reported in 2 cases [14, 15].

AUTHORS CONTRIBUTION

Farhan Akbar: Principal author, literature review, paper writing

Humaira Aziz Sawal: Literature review, paper writing and results compilation

Uzair Mushahid: Paper writing, Literature review and final proof reading

Shoaib Ahmad: Overall supervision

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