



CHONDRO-OSSEOUS CHORISTOMA OF TONGUE: A RARE CASE REPORT AND REVIEW OF LITERATURE

Pathology

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ABSTRACT

Choristoma is a benign tumor where new bone formation occurs. It occurs exclusively in flat bones of the skull and face but rarely present in oral cavity. However when it occurs in the oral cavity, mucosa of tongue is most commonly affected. The various tissue types that can present in the oral cavity as choristoma are cartilage, bone, glial tissue, salivary gland and thyroid tissue. Cases presenting both bone and cartilage simultaneously are named as chondro-osseous or osteo cartilaginous choristoma are even rarer. We report 11th case of chondro-osseous choristoma of tongue in the world of a 47 year old male presenting with nodular swelling over left lateral border of tongue where histopathological examination of the excised specimen showed nodule composed of cartilaginous and bony trabeculae covered with intact squamous mucosa and hence the diagnosis of chondro-osseous choristoma was made.

KEYWORDS

choristoma, nodular swelling, chondro-osseous, histopathologic examination

INTRODUCTION

Choristoma is a term used to describe a non neoplastic growth of a histologically normal tissue in an abnormal site^{1,2}. Oral cavity can develop choristomas from different tissues, such as salivary, sweat, sebaceous and thyroid glands, cartilage, bone, glia, adipocytes and gastric mucosa^{1,3}.

Chondro-osseous choristoma is a well-defined nodular mass, containing both osseous and cartilaginous tissues⁴. In the oral cavity, these lesions especially involve the dorsum of the tongue in women from fifth decade of life⁴. The lesion mostly presents as an asymptomatic mass, however, globus sensation, dysphagia and even airway obstruction have been reported⁵. Etiology remains uncertain⁶ and histopathology is characterized by a mass of osseous and cartilaginous tissues⁴. Management is based on complete surgical resection⁷.

Herein, considering its rarity, the purpose of this article is to report 11th case of chondro-osseous choristoma of tongue in the world after thorough review of literature.

Case Report

A 47 year male presented to the OPD of a government hospital with a swelling over left lateral border of tongue since 3 years (Figure 1). There was preceding history of trauma due to tongue bite. The swelling was painless, progressively increasing in size but not associated with difficulty in mastication, deglutition, speech or airway obstruction. Patient was fully dentate with good oral health status. There was also no history of similar swelling over head and neck region.

An intraoral examination revealed a firm, non tender, mobile, nodular mass over left lateral border of tongue measuring 3.5x3x2.5 cm. The overlying mucosa was normal and there were no associated inflammatory changes or ulcerations. Excision of mass was done under local anaesthesia and histopathological examination was done at our centre.

Grossly, the specimen was firm and nodular with a diameter of 3.5 cm. Cut surface of the mass was solid, greyish white and gritty. Representative sections were taken, processed and stained with hematoxylin and eosin. Sections revealed a well circumscribed nodule composed of cartilage and bony trabeculae (Figure 2 & 3). Marrow elements were seen in the entire lesion and the nodule was covered with intact squamous mucosa. Hence the histological features were consistent with diagnosis of chondro-osseous choristoma of tongue.

DISCUSSION

Proliferation of bone and cartilage in soft tissues are uncommon findings. Proliferation of both bone and cartilage in the same lesion is

extremely rare. On review of medical literature we found only ten reported cases of osteo-cartilaginous choristomas of tongue⁸; present case being the 11th one in the world medical literature (Table 1).

It is important to note that choristoma pathogenesis is still uncertain. Overall considering tongue as the main oral osteo-cartilaginous choristoma location, there are five theories to explain histogenesis including: metaplasia; proliferation of cartilaginous embryonic rests; pluripotential cell derivation; mixed tumor with preponderance of cartilage; neoplasms or teratomas with preponderance of cartilage⁶.

It is believed that the main theory for osteo-cartilaginous choristomas histogenesis is metaplasia, since, in fact, some reported cases are preceded by a history of trauma⁶ (as in our case). Presence of trauma would lead to a chronic inflammation that stimulate the metaplastic transformation based on the transformation of mesenchymal cells into chondrocytes and, as a consequence, in the oral cavity, the lesion would be more frequently located on the dorsum and anterior region of the tongue, or on the lateral border of the tongue³.

Clinically, oral choristomas are observed as firm, nodular pedunculated or sessile swelling, ranging from 0.5 to 2 cm in diameter. Most patients are unaware of the lesion, but symptoms of pain, dysphagia, gagging, choking, and nausea have been reported⁸.

Main differential diagnosis for intraoral choristomas are fibrous hyperplasia, pyogenic granuloma, a cyst, a salivary gland tumor and an ectopic thyroid tissue^{6,10,11}.

On histopathology, oral choristomas are composed by a well-circumscribed mass that can present variable lamellar bone with well-developed haversian system (osseous choristoma), a well defined viable mature cartilage mass (cartilaginous choristoma), or a combination of bone and cartilage surrounded by a fibrous connective tissue (osteo-cartilaginous choristoma)^{4,9}. Furthermore, it is important to emphasize that there is no presence of atypia, infiltration to adjacent tissues, and the presence of salivary gland tissue, therefore discarding the possibility of malignant neoplastic lesions or glandular origin⁸.

Choristoma treatment is based on complete surgical excision of the lesion and recurrence has not been reported^{7,9}.

Table.1 Cases Of Chondro-osseous Choristomas Of Tongue Reported In Medical Literature

N	Authors	Age	Sex	Site
1	Roy et al (1970) ⁸	20	F	Near to foramen
2	Gabriele Kaufman (1978) ⁸	22	F	Anterolateral to foramen magnum

3	Wesley and Zielinsk (1978)8	57	F	Ventral surface of the tongue
4	Shimono et al.(1994)8	47	M	Dorsal surface of the tongue base
5	Landini et al.(1989)8	35	F	Left lateral border of tongue
6	Watson et al.(1990)8	67	F	Junction of anterior two thirds with the posterior third (midline)
7	Piattelli et al.(2000)8	64	F	Right ventral surface of the tongue
8	Dermiseren et al.(2007)8	28	M	Dorsum of the tongue, in front of the foramen cecum
9	Majeed and farah (2011)12	57	M	Anterior right dorsal aspect of tongue
10	Camara et al.(2017)8	53	M	Dorsum of tongue
11	Present case	47	M	Left lateral border of tongue



Figure1. Nodular Mass In Lateral Border Of Tongue.

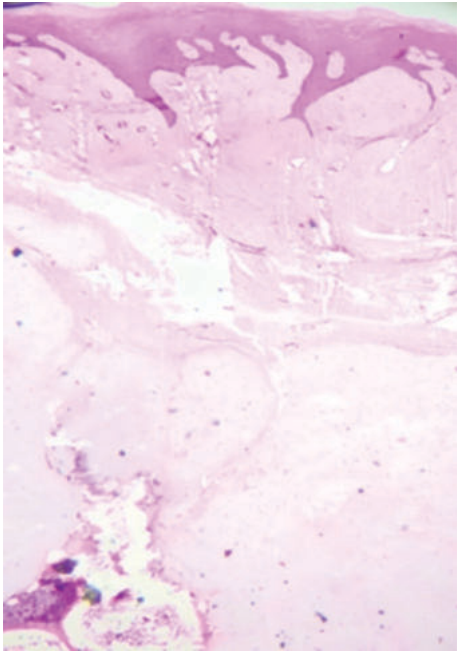


Figure 2. H&E Stained Section Shows The Lesion Separated From The Overlying Epithelium By Fibrous And Collagenous Zone(100x).

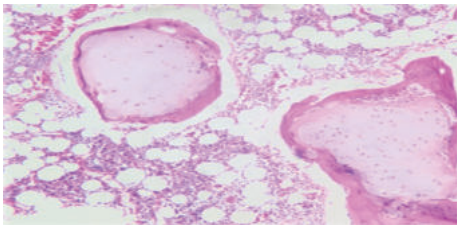


Figure 3. H&E Stained Section Shows Mature Viable Cartilage

And Bone Marrow Elements(400x).

CONCLUSION

In conclusion, osteo-cartilaginous choristoma should be included in the differential diagnosis of any firm swelling on any region of the tongue. Awareness of its benign behaviour is important to avoid aggressive surgical management. Nevertheless definitive diagnosis is obtained only after histopathologic examination.

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