

Lingual choristoma

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ABSTRACT

Osseous choristoma is a rare, benign tumour of the oral cavity. It is composed of normal lamellar bone occurring at an abnormal site. The aetiology and pathogenesis of this lesion remains debatable. Most are asymptomatic and usually present with mass effect. We report a case of a young lady who presented with globus pharyngeus secondary to a lingual choristoma.

Keywords: Lingual, choristoma, osseous, osteoma

INTRODUCTION

Osseous choristoma is commonly found on the dorsum of tongue. ¹ Choristoma in the oral cavity may be made of different types of tissues including bone, cartilage, gastric mucosa, glial tissue and tumour like masses of sebaceous gland. ² Still, bone tissue remains the predominant tissue of choristoma of oral cavity. ³ Choristoma has a female predilection of approximately 2.3:1. ⁴ It is commonly seen in the third and fourth decades of life. ⁵ The lesions mostly presents as an asymptomatic mass however, globus sensation, dysphagia and even airway obstruction have been reported. ⁶

CASE REPORT

A 25-year-old lady with no underlying medical

illness was referred to our clinic for an incidental finding of a painless lump on the tongue which was noted during a dental procedure. She was having occasional globus sensation for the past six months. However, there were no symptoms of pain, bleeding from the lump, dysphagia, odynophagia or noisy breathing. There was also no history of local trauma or denture application. She denied any signs and symptoms of hypothyroidism or hyperthyroidism. There was also no complain of similar swelling over the head and neck region.

An intraoral examination revealed a whitish, ovoid, firm mass measuring 0.8 x 0.8 cm over dorsum of tongue at the foramen cecum which is at the junction between anterior two-third and posterior one-third. The overlying mucosa was intact and normal looking. Upon palpation, the mass was non-tender, firm and adherent to the underlying

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tissue. There was no lymph node palpable over the neck and thyroid gland was normal. Laryngoscopy performed revealed no abnormality. Other systemic examination was normal. Thyroid function test performed was within normal range.

Excision of mass was performed under local anaesthesia. A bony hard mass measuring 1 x 1cm was removed and was sent for histopathological examination. No complication was encountered during or post procedure. It was reported as a polypoidal bony lesion covered by non-keratinising stratified squamous epithelium, containing mature lamellar bone trabeculae at the centre resembling an osseous choristoma (Figure 1). Upon follow up after one year, she was well and asymptomatic.

DISCUSSION

Osseous choristoma was originally described in 1913 by Monserrat as lingual osteoma which implies a neoplasm.⁷ Later it was renamed by Krolls *et al.* in 1971 as osseous choristoma.⁸ Although several theories have been proposed to explain the aetiology and pathogenesis of lingual choristoma, it can be grouped into two main categories: reactive or post-traumatic theory³ and developmental malformation theory.⁴ The former theory suggest that the local tissue reaction caused by recurrent trauma to the alleged area leads to metaplasia and calcification hence, choristoma formation. The latter developmental malformation theory suggests that the remnant of branchial arch undergoes endochondral ossification leading to the choristoma formation. The developmental theory is most widely accepted as it explains why the lesion usually occurs in the foramen caecum

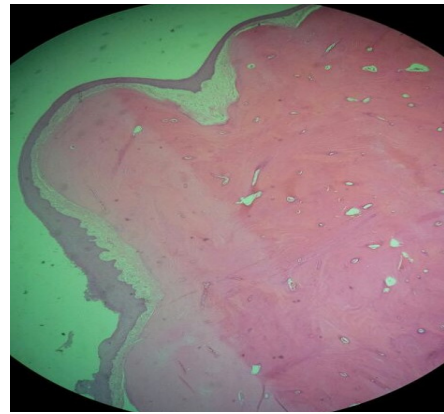


Fig. 1: Mature lamella bony trabeculae covered by mucous membrane of tongue (H&E stain; magnification x4)

as the case reported here. Lingual choristoma are slow growing tumours, hence symptoms only appear when normal lingual function is disrupted as the lesion enlarges⁹ which may explain why patients are not symptomatic at birth.

Differential diagnosis of lesion in the foramen caecum includes: benign lesions including hemangioma, lymphangioma, fibrous tumours, thyroglossal duct cyst, lingual thyroid, teratoma, hamartoma, mucocoele as well as malignant tumours such as sarcoma, rhabdomyosarcoma and epidermoid carcinoma.¹⁰ Nevertheless, the definitive diagnosis is only obtained after histopathological examination as in our case.

Treatment of choice proposed is surgical excision and no recurrence or malignant transformation has been reported.¹¹ As in our patient, no recurrence was noted after one year of follow up.

In conclusion lingual choristoma is rare and mostly presents on the dorsum of the tongue. It is usually an incidental finding

following a procedure. Surgical excision remains the main modality of treatment and definitive diagnosis is achieved via a histopathological examination. Awareness of this entity may avoid unnecessary burden to the patient.

REFERENCES

1: Sheridan SM. Osseous choristoma: a report of two cases. *J Oral Maxillofac Surg.* 1984; 22:99-102.
2: Chou LS, Hansen LS, Daniels TE. Choristomas of the oral cavity: a review. *Oral Surg Oral Med Oral Pathol.* 1991; 72:584-93.
3: Tohill MJ, Green JG, Cohen DM. Intraoral osseous and cartilaginous choristomas: report of three cases and review of the literature. *Oral Surg, Oral Med, Oral Pathol.* 1987; 63:506-10.
4: Vered M, Lustig JP, Buchner A. Lingual osteoma: A debatable entity. *J Oral Maxillofac Surg.* 1998; 56:9-13.

5: Liu JY, Tan KK. Lingual osteoma: case report and literature review. *Singapore Med J.* 2011; 52:198-200.

6: Horn C, Thaker HM, Tampakopoulo DA, De Serres LM, Keller JL, Haddad J Jr. Tongue lesions in the paediatric population. *Otolaryngol Head Neck Surg.* 2001; 124:164-9.

7: Monserrat M. Osteome de la langue. *Bulletin de la Societe D'anatomie.* 1913; 88:282-3.

8: Kroll SO, Jacoway JR, Alexander WN. Osseous Choristomas (Osteomas) of intraoral soft tissue. *Oral Surg Oral Med Oral Pathol.* 1971; 32:588-95.

9: Naik VR, Wan Faiziah W, Musa MY. Choristoma of the base of tongue. *Indian J Pathol Microbiol.* 2009; 52:86-7.

10: Lutcavage GJ, Fulbright DK. Osteoma of the tongue. *J Oral Maxillofac Surg.* 1993; 1:697-9.

11: Andressakis DD, Pavlakis AG, Chrysomali E, Rapidis AD. Infected lingual osseous choristoma. Report of a case and review of literature. *Med Oral Patol Oral Cir Bucal.* 2008; 13:627-32.