

# Osseous choristoma of the tongue: A rare disease

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## ABSTRACT

Choristoma is described as the growth of normal tissue in an unusual location. Intraoral choristoma may be comprised of various tissue types such as cartilage, bone, gastric mucosa and, even glial tissue. Osseous choristoma is a rare, benign normal bony tissue in an abnormal location, commonly over the dorsum of the tongue though cases at

maxillofacial region have been reported. This entity should be kept in mind because it can be confused with other tumor and tumor-like lesions of the tongue. We report a case of lingual osseous choristoma along with discussion regarding its epidemiology, pathogenesis, clinical features, and treatment.

**Keywords:** Osseous choristoma, Ectopic, Lingual, Rare

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## INTRODUCTION

The term osseous choristoma was first described by Krolls et al. in 1971. He had collected 25 cases for analysis and proposed the term osseous choristoma based on the nature and pathogenesis of the lesion.<sup>1</sup> Choristoma is defined as lesion composed of normal histological tissue, but at a location which is remote from the tissue origin site. Hence osseous choristoma refers to bony lesion away from the normal bony structures.<sup>2</sup> With just under a hundred cases reported,<sup>2</sup> knowledge development of such particular uncommon case remains devious, especially concerning its etiopathogenesis. Lingual involvement with osseous histotype, as reported here, was the most common variant.<sup>3</sup> Surgical management is usually mandatory and favors good outcome.<sup>4</sup>

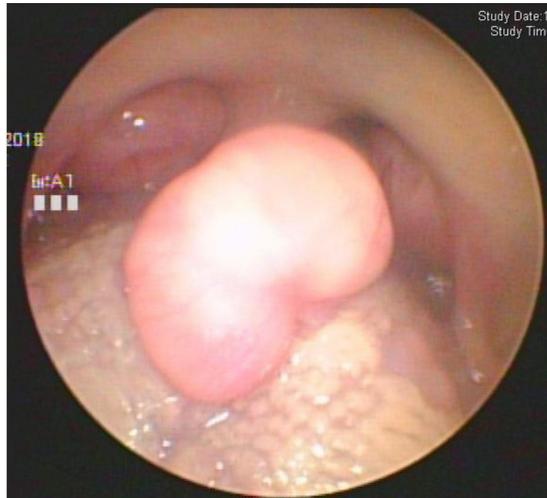
## CASE REPORT

An 18-year-old Malay female was admitted with a painless swelling over her tongue since her childhood. She was referred to our centre by a general practitioner with the impression of malignancy of the base of tongue. The concerning increase in size urged her to have an ENT consultation. She denied any history of trauma to the swelling site. Despite the discomfort it caused, there was no notable dysphagia, odynophagia, difficulty in breathing. During her childhood, the lesion was small and had not caused her any feeding problem. Examination showed a pedunculated swelling at the center of her tongue over the circumvallate papillae, just anterior to the foramen caecum of the tongue (Figure 1). The mass with overlying pink and healthy mucosa sized approximately 2 × 3 cm

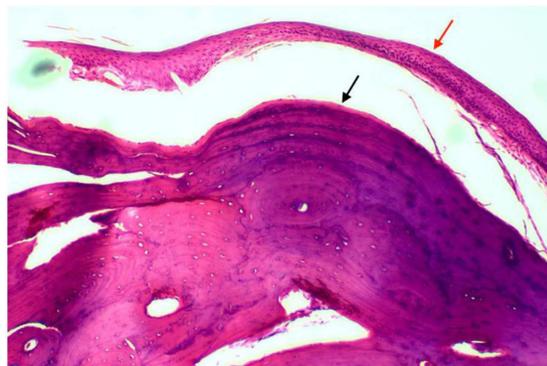
in dimension. It was firm to hard in consistency and non-tender. No ulceration and palpable neck swelling was detected. Endoscopic examination revealed no significant abnormality over the base of tongue, vallecula, bilateral piriform fossa, arytenoid cartilage, and epiglottis. Bilateral vocal folds were normal and mobile. The patient's thyroid function test was within normal limit. Initial clinical diagnosis of osseous choristoma of the tongue was made, and the differential diagnosis included a hamartoma, lingual thyroid, thyroglossal duct cyst, epidermoid carcinoma, salivary gland tumors, peripheral ossifying fibroma and fibrous hyperplasia. Ultrasonography of her neck showed that both lobes of the thyroid gland and the isthmus were at their normal position with normal echo pattern.

She underwent surgical excision of the tongue lesion under general anesthesia. A solid mass (2 cm × 3 cm × 1 cm) was excised from the midline of circumvallate papillae region with sharp dissection and hemostasis accomplished with electrocauterization. The specimen was fixed with 10% formalin and sent for histopathological examination. The surgical excision site was closed primarily using vicryl 4/0 suture. Histopathology of the specimen processed with a haematoxylin and eosin staining revealed a well-circumscribed lesion partly covered by benign stratified squamous epithelium and composed of irregular mature bony trabeculae (Figure 2, Figure 3, Figure 4). There was no osteoblastic activity or nuclear atypia seen. The diagnosis of osseous choristoma was made. The surgical wound healing was good, with no visible scar seen after six weeks post-surgery. One year of regular follow-up was uneventful, with no recurrence of the mass detected.

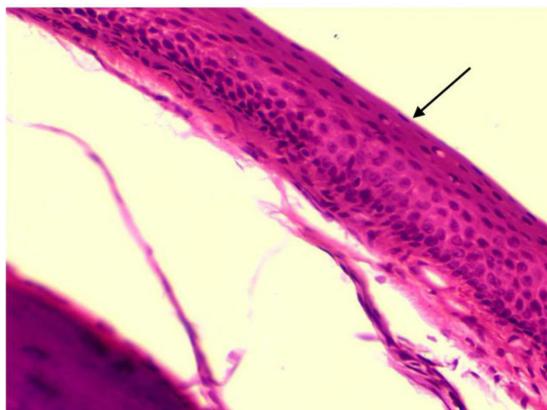
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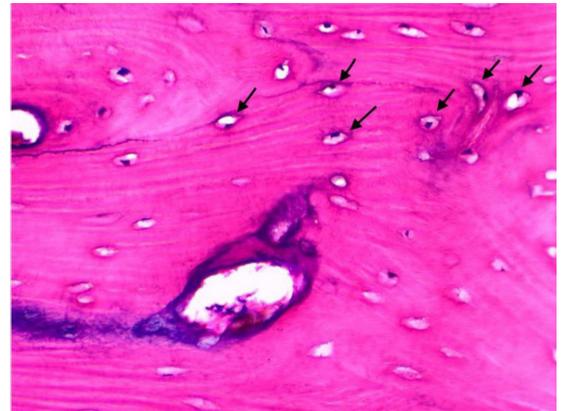
**Figure 1** Smooth-surfaced mass over the tongue dorsum, anterior to the foramen caecum



**Figure 2** Histopathological appearance of the lesion (haematoxylin and eosin staining, 100x magnification). A well-circumscribed lesion, partly covered by benign stratified squamous epithelium (red arrow) and composed of mature compact (black arrow)



**Figure 3** Histopathological examination of the lesion (haematoxylin and eosin staining, 400x magnification) showing mature stratified squamous epithelium (black arrow)



**Figure 4** Histopathological examination of the lesion (haematoxylin and eosin staining, 400x magnification) showing mature compact bone with osteocytes (black arrows)

## DISCUSSION

A recent literature review by Yoshimura et al<sup>2</sup> reported a total of 97 osseous choristoma case reports published in English. Its locations with respective frequencies in decreasing order are as follows; tongue (76 [78%]), buccal mucosa (14 [15%]), alveolar mucosa (2 [2%]), submandibular region (2 [2%]), submental region (1 [1%]), masseter muscle (1 [1%]), and hard plate (1 [1%]). The most frequently affected region in lingual involvement is the posterior third of the tongue dorsum in proximity to foramen caecum and circumvallate papillae, and sometimes it arises at the lateral border or middle third of the tongue.<sup>2,3</sup> Female gender predilection was also noted with up to 4 times higher prevalence.<sup>5</sup> Both characteristics in location and gender were consistent in our case.

Histologically, the osseous choristoma is normal bony tissue consisting of well-circumscribed, lamellated mass of bone with well-developed Haversian canal system. The lesion is surrounded by dense fibrous connective tissue, with stratified squamous epithelium covering exophytic part of the swelling.<sup>6</sup> Osteoblastic activity usually presents in the developing or young osseous choristoma.<sup>1</sup> The fact that the lesion is slow-growing and composed of very mature bone may be the reason that osteoblastic activity is hard to visualize in this case, although it was observed in previous studies.<sup>2,7</sup>

The exact etiopathology underlying osseous choristoma has yet to be fully elucidated. Proposed hypotheses are mainly divided into embryological and trauma or inflammation.<sup>4</sup> Embryological malformation mechanisms postulated were ossification of fusing first and third branchial arches, primitive foregut endoderm derivative from abnormal tracheoesophageal septum separation,

osseous proliferation in undescended intraglossal thyroid tissue, and metabolic alterations in pluripotent mesenchymal cells.<sup>8-11</sup> While most of these developmental-related theories remain speculative, the latter served feasibility for further research. Immunohistochemical study by Yoshimura et al.<sup>12</sup> revealed the potential role of bone morphogenetic protein-2 and -4 as ectopic ossification trigger in the soft tissue. Trauma with subsequent chronic inflammation, on the other hand, could be accounted for the reactive ossification center emerging in susceptible area at posterior third of the tongue.<sup>4</sup> Despite the appealing explanation for the childhood-onset osseous choristoma by embryological hypotheses, possible role for other hypotheses to constitute multifactorial pathogenesis could not be ruled out. In the light of these diverse hypotheses, it is crucial to put age of onset and variable inflammation-inducing factors into consideration in future reviews.

Osseous choristoma was diagnosed at wide ranges of age (5-73 years) with a mean age of 28.7 years. Most cases (31/60 [51.7%]) were in their second and third decades of life.<sup>3</sup> Lesion size varies from 3 mm to 5 cm with sessile or pedunculated appearance and outlining normal mucosa.<sup>4,13</sup> Symptoms rely on the lesion size, tumour localization, and surrounding tissue phlogosis. Most cases are asymptomatic, but one-third of the cases might experience swelling, pain, dysphagia, gagging, nausea, increased salivation, altered speech, and feeding or breathing difficulties.<sup>2,10</sup> Vast array of differential diagnosis including tumor-like lesions, benign tumors, and malignant tumors requires preliminary imaging workup with histopathological examination to obtain a final definitive diagnosis.<sup>3</sup> Therefore, surgical management was preferred in spite of paucisymptomatic nature of this disease. Surgical excision is the mainstay treatment for osseous choristoma. The postoperative course was mostly uneventful with a slight chance of recurrence.<sup>4</sup>

## AUTHOR CONTRIBUTIONS

All authors contributed to the concept, design, definition of intellectual content, literature research, clinical studies, data analysis, along with manuscript preparation, editing, and review; Te BC and Noorasmaliza MP worked on data acquisition; Ahmad MA, Noorasmaliza MP and Yunus MRM served as guarantors for this study.

## CONFLICT OF INTEREST

The authors have nothing to disclose.

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## ETHICAL CONSIDERATIONS

Written informed consent was obtained from the patient and its copy was available to be reviewed by the Editor-in-Chief of this journal.

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