

## Case Report

### Osseous choristoma of submental region: A rare occurrence

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#### ABSTRACT

Osseous choristoma is a rare, benign lesion of the oral cavity. This report presents a case of osseous choristoma in the submental region of a 30-year-old female subject. Her chief complaint was a painless swelling in the submental region. Panoramic radiography showed a well-defined, round, radiopaque lesion near the inferior border of the left mental region. The lesion was diagnosed as an osseous choristoma based on the histopathological examination of the surgical specimen. This paper is an attempt to bring forward a unique occurrence of osseous choristoma, which would further help the medical fraternity in improvising their knowledge, diagnosis, and treatment of this entity.

**Key Words:** Biopsy, calcification, osseous choristoma, osteocytes, submental

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## INTRODUCTION

Choristomas can be defined as developmental malformations that have developed from a group of primordial cells representing a tumor-like growth of microscopically normal cells or tissues in an ectopic location.<sup>[1]</sup> Several different tissue types may occur in the oral cavity as choristomas. These include gastric mucosa, glial tissue and tumor-like masses of sebaceous glands. However, the most frequently observed choristomas of the oral cavity are those that consist of bone, cartilage or both.<sup>[2-4]</sup> These lesions sometimes have been called osteoma mucosae, soft-tissue osteomas, but choristoma is a better term because they do not appear to be true neoplasms.<sup>[5]</sup> They occur at any age and present as a firm nodule ranging up to 2 cm in diameter. More than 70% of osseous choristomas have been reported in women. Oral osseous choristomas show a striking predilection for the tongue, which accounts for 85% of cases. The

most common location is the posterior tongue near the foramen cecum.<sup>[6]</sup> Less commonly, it can occur on other sites such as buccal mucosa and alveolar mucosa.<sup>[7,8]</sup>

## CASE REPORT

A 30-year-old female was referred from the local dentist to the Department of Oral and Maxillofacial Surgery, Sudha Rustagi College of Dental Sciences and Research, Faridabad, with a chief complaint of a painless, hard swelling in the lower left front jaw region of 3-year duration.

The patient's history revealed that she was apparently normal 3 years ago when she noticed a hard, painless swelling in the submental region, which was pea-sized initially and gradually increased to attain the present size, which was consistent for the past 6 months. On taking the past medical history, it was found that she

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had tuberculosis 2 years back, which was successfully treated. Dental history revealed no history of trauma or pus discharge.

Extraoral examination revealed a nodular swelling in the submental region, facial asymmetry and normal-appearing overlying skin along with no history of localized raised temperature, extraction or any other dental treatment [Figure 1]. On palpation, the swelling was hard, painless, single and nodular, extending 2–4 cm lateral to the midline of the lower jaw, immobile, nonfluctuant, non-compressible and nontender. There were no signs of suppuration, sinus tract formation and lymph nodes, or temporomandibular joint involvement.

Intraoral examination exhibited no associated swelling or sinus formation in the affected region or any associated occlusal disturbance. All the teeth were present except the third molars. Orthopantomogram revealed a well-defined, oval-to-round, homogeneous, radiopaque area approximately 2 × 1.5 cm in size, extending from #33 to the mesial aspect of #35 near the submental region [Figure 2a]. There was neither any evidence of radiolucent band around the radiopaque area nor

any radiolucency within the radiopacity. Mandibular occlusal radiograph showed a diffuse radiopaque area of approximately 1.8 × 1.2 cm in size, extending from #33 to the mesial aspect of #35 near the submental region [Figure 2b]. The patient underwent an excisional biopsy procedure which was performed under local anesthesia [Figure 3].

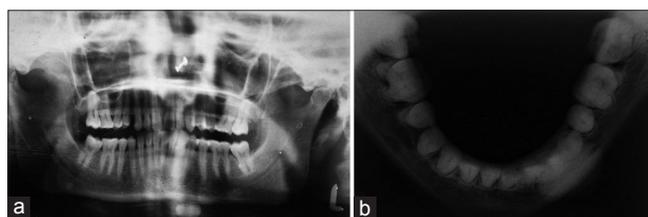
The gross examination of the excised specimen revealed a well-circumscribed, hard tissue without any associated connective tissue. The excised mass was approximately 1.3 × 1.2 × 0.7 cm in size, cream in color, hard in consistency and smooth in texture [Figure 4]. On histopathological examination of the surgical specimen, the lesion was diagnosed as an osseous choristoma. Microscopic examination showed a mass of woven bone arranged in an interconnecting trabecular pattern, containing intact osteocytes within the lacunae and fibrous marrow tissue with no osteoblastic rimming [Figure 5].

## DISCUSSION

The osseous choristoma by definition is a normal bony tissue, but in an abnormal location; this can be in the skin (previously known as osteoma cutis)



**Figure 1:** Preoperative clinical appearance of the patient showing nodular swelling in submental region.



**Figure 2:** Orthopantomogram and occlusal radiograph showing an oval-to-round-radiopaque mass near the submental region, extending from #33 to #35.

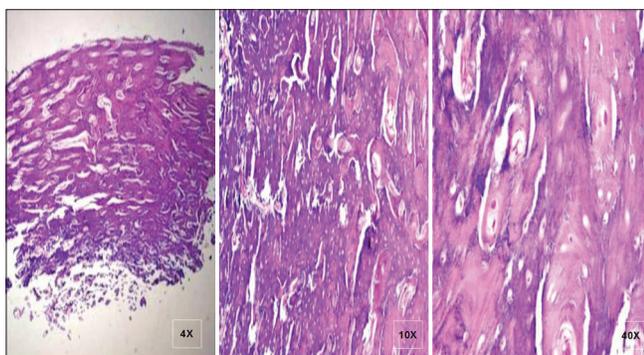


**Figure 3:** Excisional biopsy performed under local anesthesia.

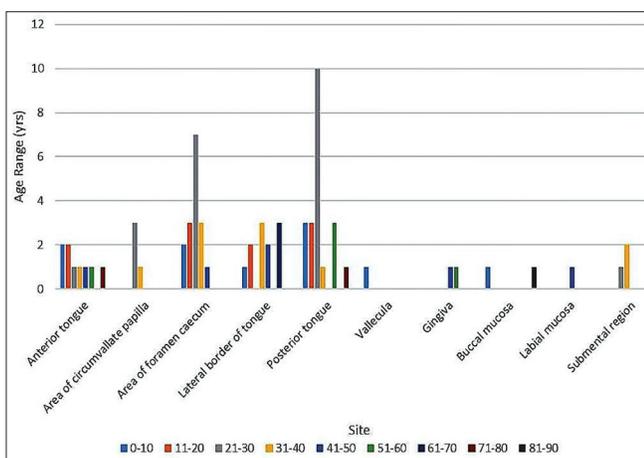


**Figure 4:** The excised specimen which was hard, cream-colored and smooth-textured, measuring 1.3 × 1.2 × 0.7 cm.

or in the oral cavity mucosa (previously known as osteoma mucosae).<sup>[9]</sup> Osseous choristomas, which have also been referred to as “soft tissue osteomas,” are rare benign lesions of the oral cavity. Krolls *et al.* were the first to use the term osseous choristoma to identify normal, lamellar bony structures that developed in soft tissue.<sup>[5]</sup> The present case was unique in itself because choristomas rarely occur in the maxillofacial region, and only 79 cases have been reported so far.<sup>[1,10-13]</sup> In the maxillofacial region, they are most frequently located on the dorsum of the tongue near the circumvallate papillae or foramen caecum. On rare occasions, they can be seen in the buccal mucosa, labial mucosa, gingiva, submental region, masseter muscle and submandibular region. The occurrence of a number of cases of osseous choristomas, in terms of location and age, have been well described with the help of bar charts [Figure 6]. Classification of choristoma by Chou *et al.* is presented in Table 1.<sup>[2]</sup>



**Figure 5:** Photomicrograph of hard tissue section showing woven osseous trabeculae containing intact osteocytes within the lacunae (H and E, x4, x10, x40).



**Figure 6:** Bar chart representing the number of reported cases of osseous choristomas in terms of location and age.

Clinically, the lesion is usually a firm, smooth-surfaced, sessile, or pedunculated nodule, measuring between 0.5 and 2.0 cm in diameter. Many patients are unaware of the lesion, although some complain of gagging or dysphagia.<sup>[6]</sup>

Several theories have tried to explain the pathogenesis of these lesions. In general, these theories are subgrouped in two categories: Developmental theory and reactive (posttraumatic) theory. However, the exact etiopathogenesis remains unknown.<sup>[14]</sup> The developmental theory suggests that these lesions are derived from the mesenchymal primordial cells or branchial arch remnants.<sup>[15]</sup>

The posttraumatic theory suggests that osseous choristoma can result in metaplastic osseous differentiation after mechanical injury and that it acts in a manner similar to that of posttraumatic myositis ossificans and dystrophic calcification.<sup>[16]</sup>

Microscopic examination of osseous choristomas shows a well-circumscribed lamellated mass of dense, vital bone with haversian canals surrounded by dense, fibrous connective tissue and covered with stratified squamous epithelium in the exophytic part of the swelling. Osteocytes may be seen in the lacunae within the small bony sphere.<sup>[12]</sup> Unilateral submental masses may be attributed to a number of pathological conditions of various origins.

In the differential diagnosis of the present case, lymph node calcification, sialolith and calcinosis cutis were also considered. Calcified lymph node is associated with a history of chronic inflammation (e.g., sinusitis and tonsillitis), tuberculosis, metastasis of thyroid cancer and treated lymphoma. However, they were ruled out as the patient presented the history of successfully treated tuberculosis, and there were no other associated

**Table 1: Chou *et al.* classified choristoma as**

Salivary gland choristoma
Central
Gingival
Cartilaginous choristoma
Osseous choristoma
Lingual thyroid choristoma
Lingual sebaceous choristoma
Glial choristoma
Gastric mucosal choristoma
Cystic
Solid

diseases. In addition, there was no distribution along the course of the cervical, submandibular and digastric node chain. Radiographically, it did not exhibit cauliflower-like, multiple radiopacities, and histologically there was no presentation of necrosis, Langerhans giant cells, histiocytes and fibrosis along with dystrophic calcification. There was no relevant fibrous capsule or attachment of lymph nodes nor was there any residual structure of lymph node or the histopathological presence of tubercular granuloma. Sialolith was ruled out as there was no history of pain and swelling in the salivary gland, which would intensify at meal time when salivary flow is stimulated, and histopathologically there was no associated salivary gland tissue. Calcinosis cutis is an accumulation of calcium salts which are often attached to the dermis and usually associated with calcinosis, Raynaud phenomenon, esophageal dysmotility, sclerodactyly, and telangiectasia syndrome, which made it easy to rule out the diagnosis.

Choristomas are best treated by local surgical excision and recurrence has not been reported.<sup>[6]</sup> The present case was also treated by surgical excision, and there has been no report of recurrence to date.

## CONCLUSION

To the best of our knowledge, the present case is the third to be reported in the submental region and is of interest due to the rare occurrence of the osseous choristoma in the submental region. The number of reported cases is insufficient to describe it completely. To increase the available knowledge regarding it, new case reports should be added to the literature, which should include clinical, radiographic and histological findings, as well as follow-up reports and treatment protocols.

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## Conflicts of interest

The authors of this manuscript declare that they have no conflicts of interest, real or perceived, financial or nonfinancial in this article.

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