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Case Report

Osseous choristoma: Report of two cases in oral cavity

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ABSTRACT

Osseous choristoma is the formation of normal mature bone at an abnormal position. These are rare benign tumor-like growth usually asymptomatic except growth or swelling as the clinical presentation. In oral cavity, osseous choristoma is rare and commonly found in posterior third of the tongue. Clinically it is misdiagnosed as malignancy because of its hardness. Clinically, it is also mistaken for other oral cavity lesions, especially those of minor salivary gland lesions of oral cavity. Hence, awareness amongst clinicians, general practitioners and oral pathologists about oral osseous choristomas is necessary. Here, we report two cases of osseous choristoma in the oral cavity located at sites other than tongue.

1. Introduction

Choristoma is the development of normal tissue at the ectopic site.[1] The tissue with this kind of growth might include bone, cartilage, glial tissue, gastric musoca or sebaceous glands, etc. Osseous choristoma is a heterotropic lesion to describe the benign tumor like growth of a mature bone mass at an abnormal position where osseous tissue is normally not a part of the morphology.[1,2] In oral cavity, choristomas are covered by normal mucosa with osseous or cartilaginous mass in the subepithelial connective tissue. Most common site for oral osseous choristoma is posterior third of the tongue. Localization of these lesions in oral cavity sites other than tongue is relatively uncommon.[1–8] Here, we report two cases of osseous choritstoma of the oral cavity in soft palate and alveolar mucosa.

2. Case Report

2.1. Case 1

A thirty-year-old female reported with a chief complaint of swelling on soft palate since three months with a history of trauma to soft palate three months back. We received an excisional biopsy of greyish white tissue measuring 0.7 x 0.5 x 0.3cm. Microscopic examination showed a small nodular elevation lined by stratified squamous epithelium with acanthosis and hyperkeratosis. The subepithelial tissue in the nodular region showed presence of normal mature compact bone without osteoblastic rimming.

2.2. Case 2

A 52-year-old male reported with a chief complaint of swelling over the lower front jaw region. Clinical examination revealed a firm hard swelling over lower anterior mandibular gingiva. Clinically, the case was diagnosed as traumatic fibroma based on the presentation. Excision of the mass was carried out and sent for histopathological evaluation. Grossly, a globular, smooth

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whitish mass with mucosal covering and core of hard connective tissue was received. The tissue measured 1.5 x 1 x 0.6 cm in dimensions. Microscopically, the H & E stained section of the lesion revealed bits of tissues with covering of stratified squamous epithelium and subepithelial well organized circumscribed lesion composed of dense mature bone with compact and unremarkable osteocytes and no prominent osteoblastic rimming. The osseous trabeculae were separated by fibrous tissue.

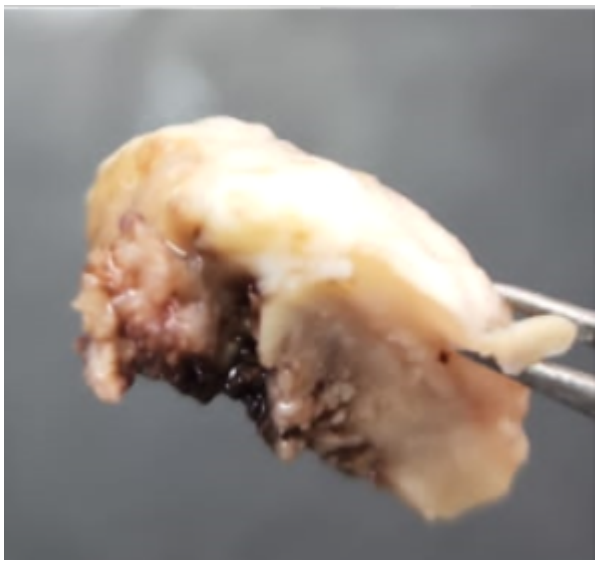

Fig. 1: Macroscopic image of the case 2 showing excised mass with overlying mucosa and underlying tissue showing firm hard areas

3. Discussion

The term osseous choristoma was coined by Krolls et al. in 1971.[4] Osseous choristoma is a term used to describe the growth of a tumor-like mass of normal bone in an abnormal position. A recent systematic review carried out by Zaid Shareef et al in 2020 included 35 articles from 1917 to February 2020 about osseous choristoma cases of oral cavity. They studied the reports of a total of 69 cases in all 35 articles and evaluated for all the specifics of the cases. Osseous choristoma was stated to be found in age ranging from 5 to 73 year with the highest incidence in the third decade of life. Female predilection was observed in overall database.[3] Weitzner suggested that 80% of these lesions occur in women and in patients <40 years old when he reported three new cases and reviewed 38 previously reported cases.[9] In the present article, one case was 30 years female and the other cases 52 years male.

Previous reviews have concluded that, the lesion is commonly found in posterior tongue adjacent to circumvallate papillae. However, the two cases reported in the present study are found in the soft palate and the

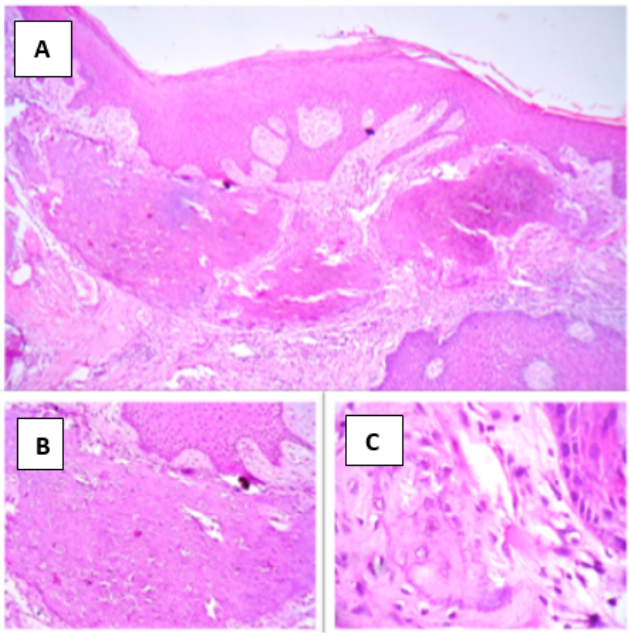

Fig. 2: Microscopic images of Case 1 showing: **A**): Small nodular elevation lined by acanthotic and hyperkeratotic stratified squamous epithelium. Subepithelial tissue shows osseous trabeculae showing mature compact bone (H & E ×40); **B:** Osseous trabculae showing mature compact bone in the subepithelial tissue (H & E ×100); **C:** Dense, mature, compact bone with osteoblastic rimming (H & E ×400)

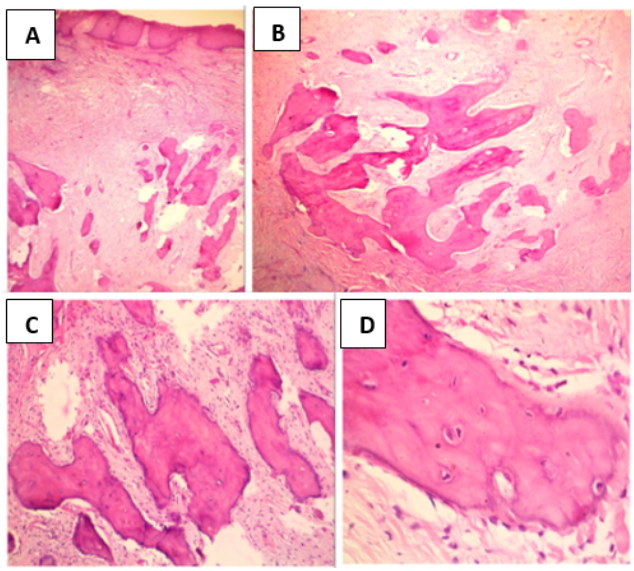

Fig. 3: Microscopic images of Case 2 showing: **A**): Intact squamous epithelium with osseous trabeculae in the subepithelial tissue (H & E ×40) **B & C**): Osseous trabeculae showing dense, mature compact bone (H& E ×100) **D**): Compact bone without prominent osteoblastic rimming (H & E ×400)

lower anterior mandibular gingiva.[3] An extensive review of literature revealed that total 85 cases have been reported in the oral cavity; of which 69 cases have been described in tongue, 14 cases in buccal mucosa and 2 cases in soft palate.[7,8] The present case represents third case of osseous choriostoma in soft palate and eighth in gingiva.

Clinical presentation of the lesion most commonly include dysphasia, gagging, foreign body sensation, mass effect, hoarseness, swelling and pain. Both the cases in this report presented with swellings. The diagnosis of osseous choristoma is histological examination of the excised mass which reveals the presence of normal lining epithelium over the subepithelial tissue containing mature bony trabeculae without osteoblastic rimming and osteolytic activity. Both the cases in the present study have histopathological features as described earlier. Surgical excision is the treatment of choice and has a very low recurrence rate as observed in most of the studies. The present cases were also treated by surgical excision.[3,4,8]

The etiopathology of osseous choristoma is still unknown. Two different hypotheses have been proposed; whereas one suggests that this is a type of malformation, the other concludes that it is formed as a result of trauma or chronic irritation. According to some investigators, choristoma refers to a tumor like growth that has developed from groups of primordial cells located at a site remote from the original tissue or organ.[10]

Monserrat was the first to propose the developmental malformation theory and he attributed the origin of the lesion to the ossification of the branchial arch remnants as a result the lesions are found in the foramen caecum area.[11] The possibility of entrapment of mesenchymal pluripotential cells originate from these embryonic branchial arches, and subsequent development of an osseous lesion in the tongue seems an attractive theory of its origin. This theory is strongly supported by Begel et al,[12] Engel and Cherick.[13] Cataldo et al[14] and Jahnke and Daly.[15] Other theories include epignathous formation and that of degenerating fibroma undergoing the ossification.[16] The latter theory suggests osseous lesions to represent a reactive or posttraumatic center of ossification.[17] In our case, the later theory probably explains the occurrence of the lesions.

This article is presented for its rarity and its likelihood to be clinically misdiagnosed as malignancy because of the hard consistency of the lesions. Clinically, it can also be mistaken for other oral cavity lesions including salivary gland lesions, mucocele, fibroma, lipoma and neural tumors to mention a few. Hence, awareness amongst clinicians, general practitioners and oral pathologists about the oral osseous choristomas is necessary. They usually appear as a tumorous mass of normal bony structure with mature cells in an ectopic position. The etiology being development or reactive still remains debatable.

4. Source of Funding

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5. Conflict of Interest

None.

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