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Osteocartilaginous choristoma of buccal mucosa: A rare entity

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INTRODUCTION

Choristomas or heterotopias are characterized by presence of tumor-like growth of histologically normal appearing mature tissues in an abnormal anatomical location.^[1-3] Commonly observed choristomas of oral cavity are those of bone, cartilage or both and are called as soft tissue osteomas or soft tissue chondromas.^[3] In the oral cavity they are commonly seen on posterior tongue (85%) near foramen cecum and about 70% of them have been reported in women.^[3] The osseous choristoma of buccal mucosa is most frequently seen in the 5th decade with an age range of 5–75 years.^[4] The histological variants include lipocartilagenous or osteocartilagenous choristoma.^[5] This article reports an osteocartilagenous soft tissue choristoma in the buccal mucosa that presented as a swelling in a 48-year-old female patient.

CASE REPORT

A 48-year-old female patient reported to the Dental OPD College with a chief complaint of swelling on the left buccal mucosa, which was more evident since past 4 months. Intraoral examination revealed a sessile oval-shaped swelling with well-defined margins, measuring 1.5 × 1 cm on the left buccal mucosa along the line of occlusion in relation to left mandibular second molar. The lesion was provisionally diagnosed as irritational fibroma, excised under local anesthesia and sent for histopathological examination.

The gross specimen received was oval shaped, well-circumscribed, creamish white in color measuring 0.7 × 1 cm in diameter. A radiograph taken of the cut surface of the specimen revealed a well circumscribed lesion with central radiopacity, representing a calcified area in the center.

Microscopic examination of the hematoxylin and eosin stained section showed predominantly parakeratotic stratified squamous surface epithelium. The lesional area was separated from the overlying epithelium by fibrocollagenous zone [Figure 1]. The lesion was well-circumscribed compressing the surrounding fibrocollagenous connective tissue creating a pseudocapsule. The central core of the lesion showed woven bone with interconnecting bony trabeculae lined by active osteoblasts and enclosing plump osteocytes. Marrow tissue consisting of blood vessels and connective tissue fibers was found between the trabeculae of bone [Figures 2 and 3]. The area of the bony trabeculae close to the epithelium showed a lining of osteoclasts encased in Howship's lacunae [Figures 4 and 5]. It can be suggested that bone formed was under the process of remodeling as there were areas of both bone formation and resorption. The area surrounding the core of woven bone consisted of plump fibroblasts, fibrocytes and loosely arranged collagen fibers resembling immature connective tissue [Figure 6]. Myxoid areas and endothelial lined blood capillaries were also seen. Serial sections of the tissue showed circumscribed chondroid areas enclosing chondrocytes separated by fibrous stroma [Figure 7]. With these features a final diagnosis of soft tissue osteocartilagenous or chondrosseous choristoma was given.

DISCUSSION

Chou *et al.*, recognized the following categories of oral choristomas:^[6]

- Salivary gland choristoma
- Central
- Gingival
- Cartilaginous choristoma
- Lingual thyroid choristoma
- Glial choristoma
- Gastric mucosal choristoma.

The other choristomas that occur in oral cavity include tumor-like masses of sebaceous glands (Fordyce spots),^[3] hairy polyp that arises from remnants of ectodermal and mesodermal germ layers in the palatine tonsil and nasopharynx consisting of fibrovascular core, skin appendages with hair, adipose tissue and muscle tissue.^[2] Lymph node may contain salivary

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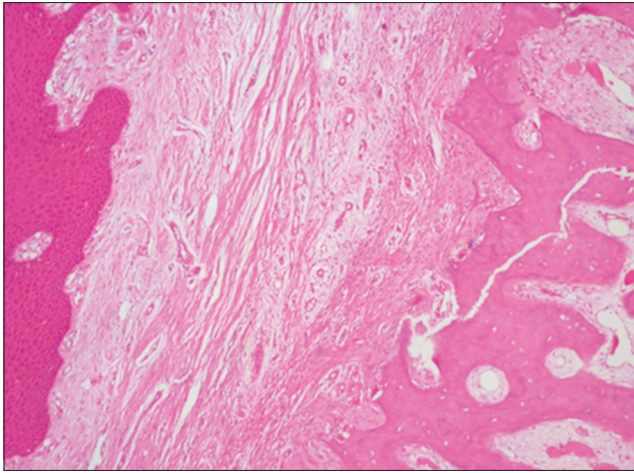


Figure 1: Photomicrograph showing the lesional tissue separated from the overlying surface epithelium by fibrocollagenous tissue (H&E stain, x40)

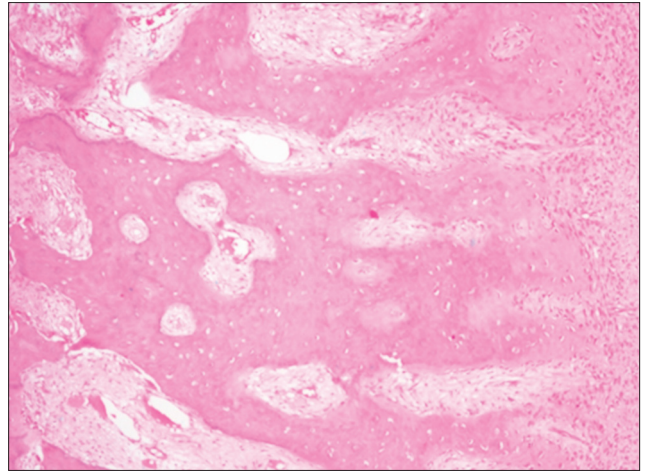


Figure 2: Photomicrograph showing core of the lesion containing woven bone with interconnecting bony trabeculae lined by active osteoblasts and enclosing plump osteocytes. Marrow tissue could be appreciated between the trabecular bone (H&E stain, x40)

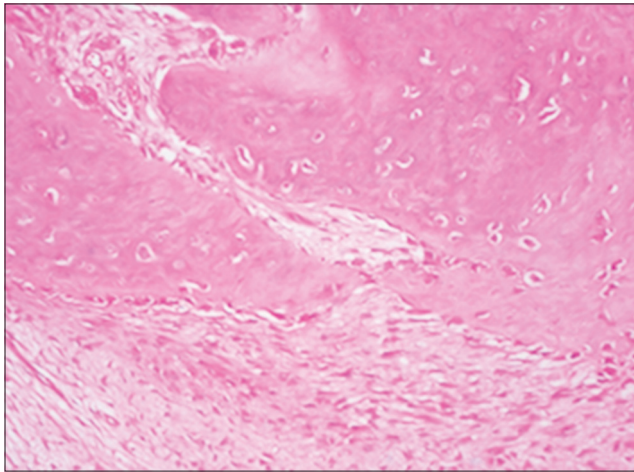


Figure 3: High power view of the trabecular bone showing osteoblastic rimming and osteocytes enclosed within the trabecular bone (H&E stain, x200)

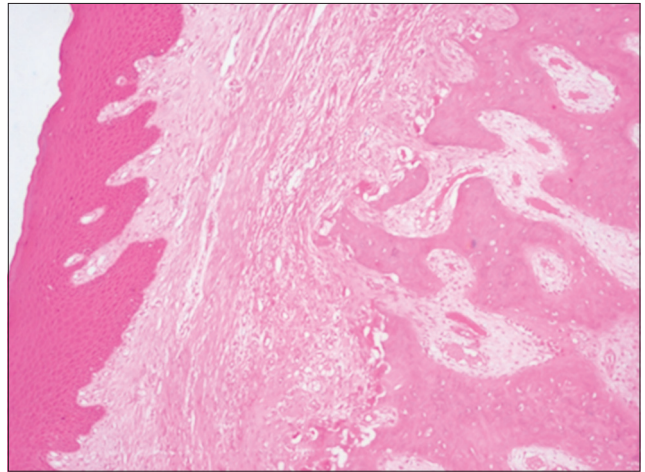


Figure 4: Photomicrograph showing osteoclasts encased in Howship's lacunae lining the bony trabeculae close to the epithelium (H&E stain, x40)

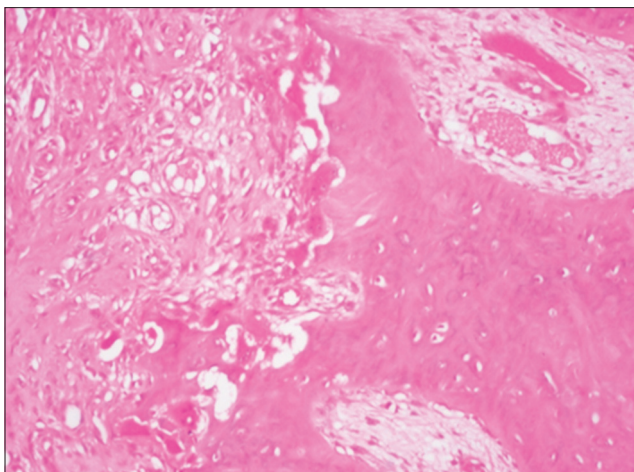


Figure 5: Photomicrograph showing osteoclasts encased in Howship's lacunae lining the bony trabeculae (H&E stain, x200)

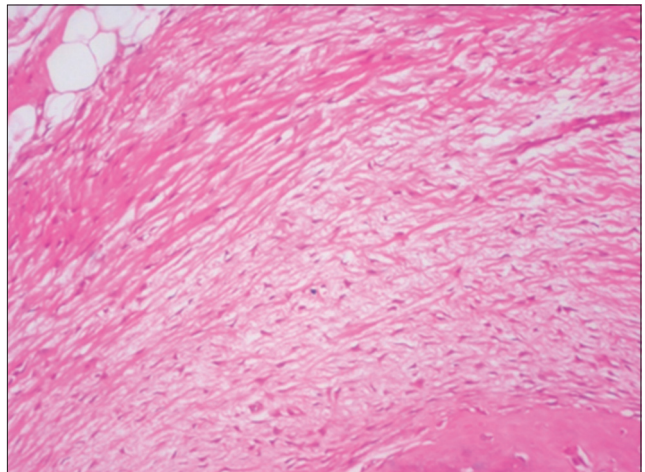


Figure 6: Photomicrograph showing the ossifying area surrounded by loosely arranged collagen fibers interspersed with fibroblasts and fibrocytes. Adipose lobules were seen interspersed (H&E stain, x100)

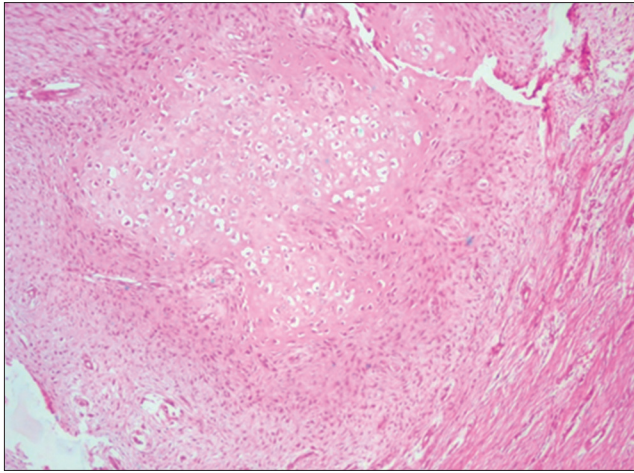


Figure 7: Photomicrograph showing areas of normal chondroid tissue (H&E stain, x100)

gland, thyroid follicles, squamous or Mullerian epithelium and nevus cell rests.^[7]

According to Chen *et al.*, about 13 cases of buccal osseous choristoma have been reported.^[4] Chou *et al.*, have reported 20 cases of cartilaginous choristoma of which two occurred in buccal mucosa.^[6]

Differential diagnoses

- Peripheral ossifying fibroma (POF): Histologically, POF is a reactive lesion and presents with inflammatory cells, which was not seen in our case. POF does not present with marrow tissue enclosed within the trabeculae of bone. These features were observed in our case pointing more towards a choristoma^[8]
- Heterotopic ossification/Myositis ossificans: Heterotopic ossification is a reactive bone-producing soft-tissue proliferation of muscle or other connective tissues associated with trauma. Ossification occurring in subcutaneous or submucosal fat is called as panniculitis ossificans or fasciitis ossificans. The lesion shows poorly organized fibroblastic proliferation with considerable mitotic activity, but cytologic atypia is not seen. The fibroblasts also form fascicles towards the periphery with an admixture of osteoblasts and reactive bone formation. Myositis ossificans is typically associated with muscle bundles. Alternating areas of calcification and muscle tissue is seen histologically. These features could not be appreciated even though the lesion was close to buccinator muscle^[8]
- Dystrophic calcification found in old thrombi or hematoma or keratin-filled soft tissue cysts: Dystrophic calcifications are not organized and present as irregular calcifications. The present case showed well-organized trabeculae of woven bone and hence could be easily excluded^[8]

- Extraskeletal osteochondroma: These show a characteristic pattern of arrangement. They present with peripheral capsule surrounding lobules of mature cartilage which show central areas of mature ossification. The pattern of arrangement of cartilage and bone were not in favor of a diagnosis of extraskeletal osteochondroma in the present case^[9]
- Cutright tumor: Presence of discrete cartilaginous islands within the soft tissue of flabby anterior maxillary ridge is known as Cutright tumor. This represents hyperplasia of existing embryonic cartilaginous rests due to inflammatory factors induced by ill-fitting dentures. They differ from soft tissue choristomas in that they arise directly from the bone beneath the lightly bound mandibular alveolar ridge.^[10]

CONCLUSION

Extraskeletal proliferation of bone and cartilage in oral and maxillofacial soft tissues probably reflects the multipotential nature of primitive mesenchymal cells throughout the region. Osseous and chondroid choristomas are commonly found in the tongue. Here we present a rare case of chondrososseous choristoma in the buccal mucosa.

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