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Case Report

Osteocartilaginous choristoma of bilateral palatine tonsil

Preeti Jain*, Arsala Mulla, Imtisangla Jamir, Pradeep Rajendran

Department of Pathology, Bharatratna Dr. Babasaheb Ambedkar Municipal General Hospital (BDBA), Kandivali(W), Mumbai, Maharashtra, India

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***Correspondence:**
Dr. Preeti Jain,
E-mail: hodpathologybdba@gmail.com

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ABSTRACT

Choristoma is defined as the presence of normal tissues in abnormal anatomical locations. This entity is extremely rare, with only few cases reported in the literature. We hereby report a case of tonsillar osteocartilagenous choristoma in a 37-year-old male who presented at our hospital ENT outpatient department (OPD) with bilateral tonsillitis. He underwent bilateral tonsillectomy surgery and we received the specimen at Pathology department. Grossly both the tonsils were enlarged with cut surface showing chalky white firm to hard areas. The histopathological examination revealed unexpected presence of mature cartilage and bone surrounded by lymphoid follicles along with areas of calcification in both tonsils. Osseous and cartilaginous choristoma are the most frequently observed choristoma in oral cavity. However cartilaginous choristoma of oral cavity is commonly seen in the tongue, buccal mucosa and soft palate. This case report shall create awareness about occurrence of choristoma at rare sites like the palatine tonsil; among pathologists and clinicians.

Keywords: Palatine tonsil, Osteocartilaginous choristoma, Tonsillitis, Histopathology

INTRODUCTION

The palatine tonsils are paired nodular masses of lymphoid tissue situated on either side of the oropharynx having an extremely remarkable role in the antimicrobial defense of the body.[1,2] The tonsils are prone to infection with tonsillitis being one of the commonest infectious diseases seen commonly in the young age group. Choristoma is tumor like mass consisting histologically of normal tissue that is foreign to the site at which they are located. Various embryological anomalies occur in the neck region due to its complex development. It is one of the developmental anomaly of the second pharyngeal arch.[3] It defers from heterotopia which may not necessarily present as a mass or tumor, thus choristoma is a specific type of heterotopia.[4] The different types of tissues that can occur as choristoma includes cartilage, bone, thyroid tissue, gastric mucosa, respiratory, glial tissue, salivary glands and they are named after the tissue of origin.[5] Cartilaginous choristoma is a rare entity, whereas the presence of cartilage and osseous metaplasia is even rarer with very few reported cases in medical literature.[6,7] Choristoma of the head and neck region is reported in the pharynx, hypopharynx, oral cavity and middle ear. Osseous and cartilaginous choristomas are commonly found in the mouth, with predilection in dorsum of tongue.[6,7] These are incidentally diagnosed during routine tonsillectomies and presents as recurrent tonsillitis with enlarged tonsil. Management is based on complete surgical excision.[8] Here we report a case of chronic tonsillitis with osteocartilaginous choristoma of bilateral tonsils.

CASE REPORT

A 37-year-old male presented to ENT Department with chief complaints of recurrent episodes of throat pain, difficulty in swallowing and burning sensations in the throat. On local examination, tonsils were enlarged and inflamed with exudates.

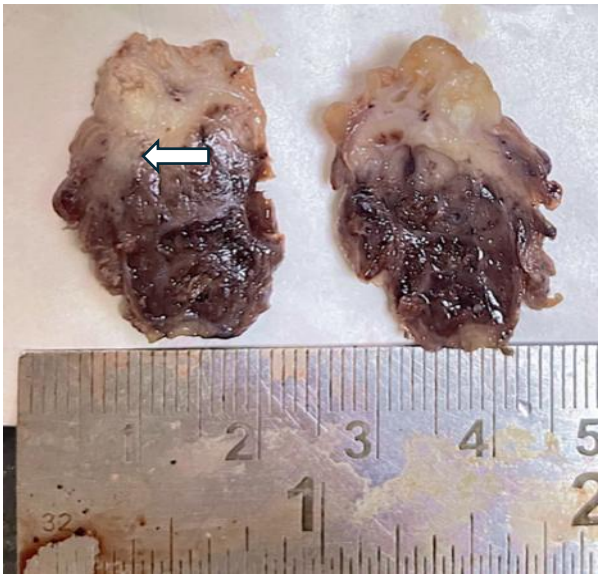

Figure 1: F cut section of bilateral tonsils revealing grey brown with chalky white glistening area.

A clinical diagnosis of bilateral tonsillar hypertrophy with tonsillitis was made. Patient underwent bilateral tonsillectomy, and the specimen was received in the Pathology Department for histopathological examination. Grossly, excised right and left tonsils were globular, firm and grey-brown in color. Tonsils measured 2.5x1.7x0.5cm (right tonsil) and 2x1.5x0.5 cm (left tonsil) respectively.

Cut surface of both the tonsils appeared gray-white with few hard chalky white areas (Figure 1). Histopathological examination revealed tonsils lined by stratified squamous epithelium with lymphoid follicular hyperplasia (Figure 2A).

Both tonsils showed islands of mature hyaline cartilage and focus of osseous metaplasia surrounded by lymphoid follicles were seen embedded in fibro collagenous tissue. (Figure 2B and 2C). Based on these microscopic features, the diagnosis of osteocartilaginous choristomas was rendered.

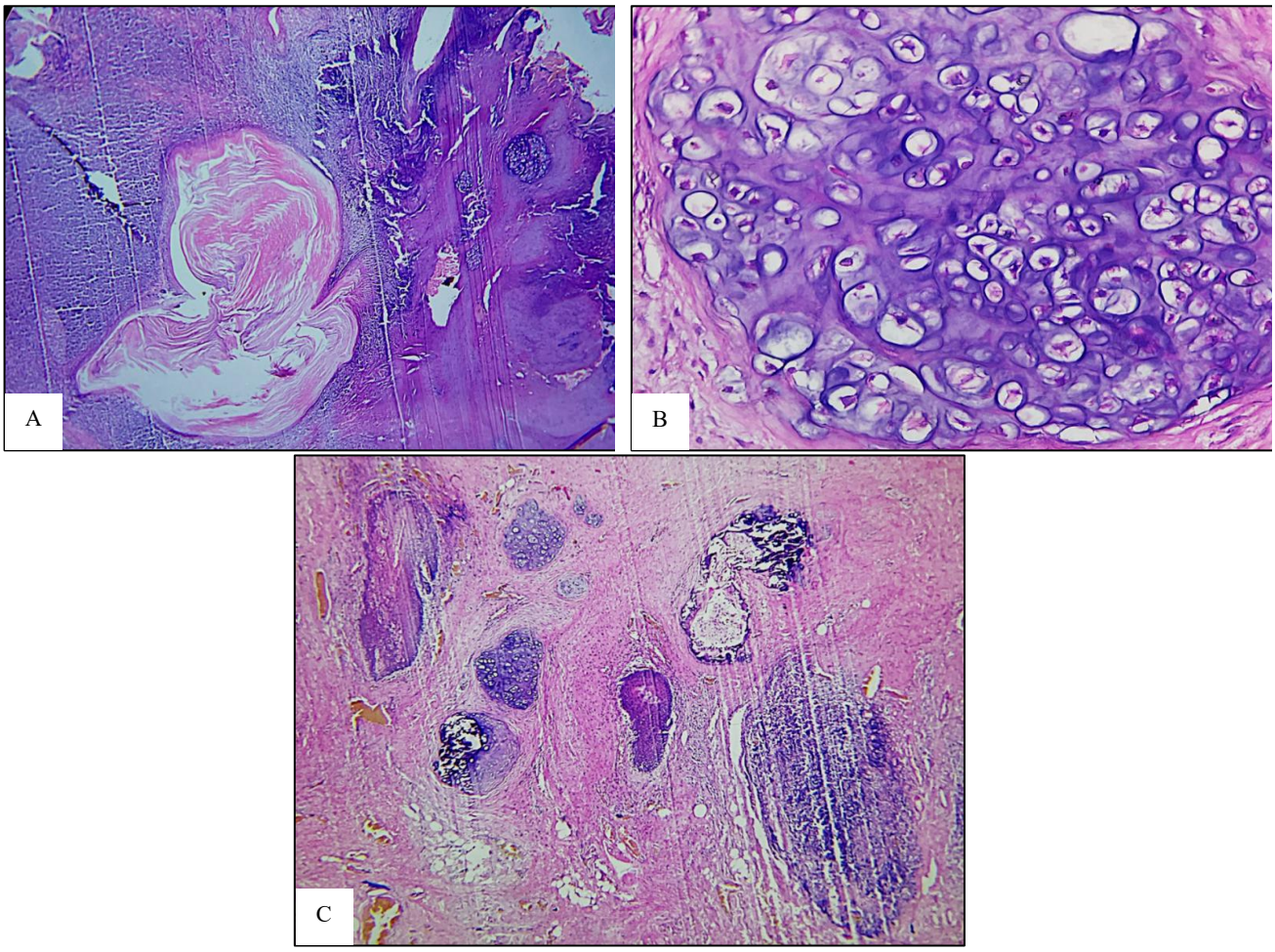

Figure 2: (A) microscopic picture showing mature hyaline cartilage with follicular hyperplasia along and intervening crypts showing keratinous flakes (H and E, 40x), (B) section shows chondrocytes of formed mature cartilage arranged surrounded by amorphous cartilaginous matrix (H and E, 400x) and (C) islands of mature hyaline cartilage along with lymphoid hyperplasia and osseous metaplasia. (H and E, 40x).

DISCUSSION

Cartilaginous choristomas was first described by Berry in 1890.[8] The age group ranges widely from 10 to 80 years with a reported incidence of 3% on histopathological examination on tonsillectomy species, showing no gender predilection.[9]

In the oral cavity, osseous and cartilaginous choristomas are commonly seen involving the tongue. Hence, osseocartilaginous choristomas involving palatine tonsil is a rare entity. These lesions usually manifest as chronic tonsillitis accompanied by tonsillar enlargement.[6,7]

Natural history of this lesion is unclear but several proposed hypotheses explaining the cause and pathogenesis of these lesions include osseocartilaginous development from heterotopic fetal remnants and due to the pluripotency of primitive mesenchymal cells. It can be developmental anomaly of the second pharyngeal arch.[3] Choristomas differentiate from multi-lineage mesenchymal progenitor cells as suggested by Haemel et al.[3]

Lindhalm et al had proposed that chronic inflammation leading to liberation of osteogenic substances may lead to heterotopic cartilage proliferation and heterotopic bone formation.[3] Some postulate that the multipotent nature of primitive mesenchymal cells stimulated to grow by trauma, irritation or inflammation may cause extra skeletal proliferation of cartilage in oral cavity and maxillofacial soft tissue.

Sulhyan K.R. et al in a large study reported distribution of cases of tonsillectomy specimens at their institute with majority cases being acute tonsillitis, chronic tonsillitis, epidermal inclusion cyst, reactive lymphoid hyperplasia; few cases of inflammatory Myo fibroblastic tumors and squamous cell carcinoma were present with cartilaginous choristoma accounting only for 2.84% cases.[11]

Differential diagnosis includes the cartilaginous metaplasia. In the oral cavity metaplasia is usually seen in soft tissue beneath ill-fitting dentures. It is histopathological characterized by single or clustered chondrocytes at various stages of maturation, whereas only mature tissue is present in choristoma.[12] In our case, mature bone and cartilage that is not a normal constituent of the nasopharyngeal epithelium was present; thus, ruling out the possibility of cartilaginous metaplasia and therefore was diagnosed as choristomas.

Choristoma is a benign tumor and treatment of choice is simple excision and no further treatment is required. Although recurrence has not been documented in the head and neck, some extraoral cases have been reported to be recurrent, so all perichondrium should be removed during surgical excision as it may have potential to develop new cartilage.[12] Our patient was followed for 1 year with no signs of recurrence.

CONCLUSION

Osteocartilaginous choristoma is a rare entity and of academic interest. It comprises a small minority of all nasopharyngeal masses and expected to follow a benign course. Complete surgical excision remains the preferred mode of treatment. Routine histopathological examination is required to detect these cases, and the pathologist must be aware of this entity as larger choristomas can be clinically mistaken for true neoplasm. Thus, although tonsillitis is the commonest lesion diagnosed clinically; histopathology plays an important role in diagnosis of various other non-neoplastic and neoplastic lesions of tonsils.

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