Case Report

Intraoral osseous choriostoma---Case report and review on its biological occurrence

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Abstract

The terminology choriostoma describes a tumor-like lesion belonging to hamartomatous group of pathology, that is, normal tissue found in abnormal location. Osseous choriostomas in oral cavity are rare and mostly the cases have been reported in tongue but rarely arising from the interdental gingiva. There are various pathological entities arising from gingiva that give a thought of wider differential diagnosis starting from inflammatory to traumatic origin. Hamartomas in oral cavity are very rare which can definitely be attributed to the sparse literature which leads to misdiagnosis as well failure to understand its biology. In this report, we present a case of osseous choriostoma arising from interdental gingiva in a young female patient in the lower arch.

Keywords: Hamartoma, interdental gingiva, osseous choriostoma, osteoma

Introduction

"Choristoma" is a tumor-like growth of common tissue in an unusual anatomical location. It was initially described by Krolls (1971). But earlier described by Monserrat (1913) as a tumor-like growth of lamellar bone in an unusual location where the bone is not normally present. It is different from a hamartoma, which is a tumor-like mass of mature normal cells located where they are normally found. Choristoma can be comprised of different tissue types. We can find bone, cartilage, gastric mucosa, glial tissue and sebaceous glands. Osseous choristoma is a well circumscribed growth and benign in nature. It is described as mature osseous tissue in ectopic sites.[1-3]

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The osseous choristoma is caused due to the following theories:

1. The developmental malformation theory described there is entrapment and ossification of mesenchymal cell originated from embryonic branchial arches. This theory is widely accepted.
2. The other theory proposes reactive or posttraumatic metaplasia and calcification of the affected area.

Conventionally, osseous choristoma is considered a developmental anomaly. Although it normally occurs in younger individuals, cases involving older patients and a wide range of age groups have been reported. Osseous choristomas can be easily misdiagnosed as osteomas and osteolipoma. Osteomas are attached to the cortex of the jaw by pedicle or wide base. Osseous choristoma appears as a well-circumscribed mass. Lamellar bone with haversian system is present here. Dense fibrous connective tissue covered by stratified squamous epithelium is observed.[4,5]

Mainly the affected individuals belong to third or fourth decade of lives. Usually, patients are asymptomatic but in case of large

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lesions airway obstruction, dysphagia, gag reflex may occur.[6] The lingual lesions were initially termed as lingual osteoma, later renamed as osseous choriostoma.[7] As a primary care physician, its early diagnosis and management is important as most patients are asymptomatic, but larger lesions may lead to feeding and breathing difficulties. Pathologic evaluation and surgical excision remain the mainstay of diagnosis and treatment, respectively.

Case Report

A 21-year-old female presented with a chief complaint of a bony, long-standing pedunculated growth in the lower right segment of the mandible with respect to first molar region. The mass was firm, pink, non-pulsatile. The growth was slow growing in size and was first noticed by the patient three months back. The growth was painless, non-ulcerative and asymptomatic. There was no history of trauma, difficulty in speech, mastication, or other daily activities.

Clinical examination revealed the presence of a firm pedunculated mass adjacent to the right first molar occupying buccal vestibule [Figure 1]. The overlying mucosa appeared normal pink. The growth seemed free from the periosteum. There were no palpable lymph nodes in the submandibular region. The radiographic findings were insignificant. The lesion was surgically excised from the interdental region site under local anesthesia. The specimen measured 1.0 cm × 1.5 cm × 0.6 cm. It was stored in 10% buffered formalin and sent for histopathological examination.

Histological findings showed that the entity was composed of well-organized and compact bone with lamellar structures beneath the epithelium [Figure 2], and lined by parakeratinized stratified squamous epithelium. [Figure 3]. The patient's postoperative recovery was uneventful and now on regular follow-up. No trace of recurrence seen.

Discussion

Normal epithelial cell have a relation with basement membrane with the help of basal surface. Epithelial mesenchymal transition is often defined as a biological process which influences epithelial cells to undergo several changes. Cells achieve mesenchymal phenotype. Cells obtain increased migratory capacity. They become more invasive, resistant to apoptosis. Mesenchymal cell is produced via this transition. They can migrate now and degradation of basement membrane will be evident. "Epithelial mesenchymal transformation" was first described by Elizabeth Hay. The term "transformation" was replaced with "transition" due to reversibility nature and because being distinct from neoplastic transformation.[8,9]

Various cells of a body initially generated from a single cell that is fertilized ovum. During development, a cell can achieve different phenotype with a process known as differentiation. As per recent study during development certain cells in epithelium becomes plastic and can go back and forth between epithelial and mesenchymal state with the

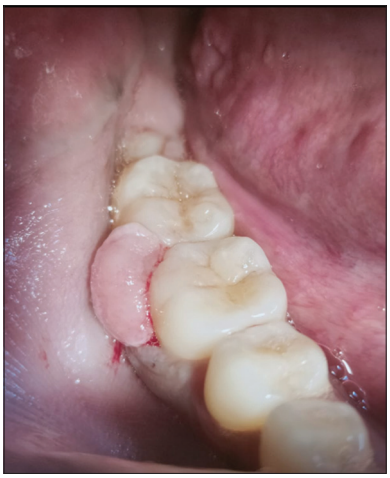

Figure 1: Firm pedunculated growth from the interdental region in the right mandibular arch

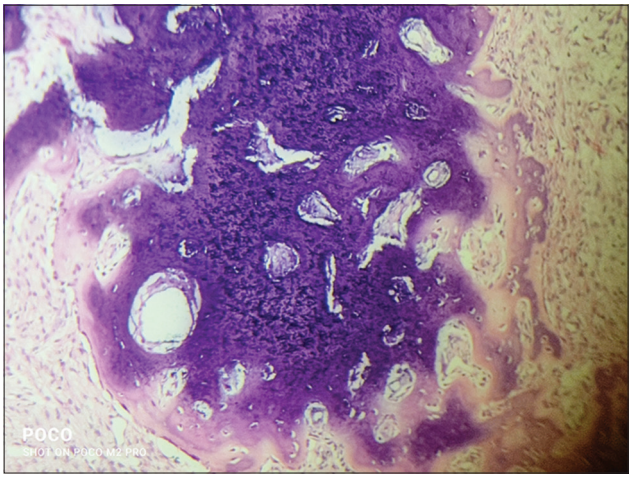

Figure 2: Stroma depicting well-organized compact bone with lamellar structures

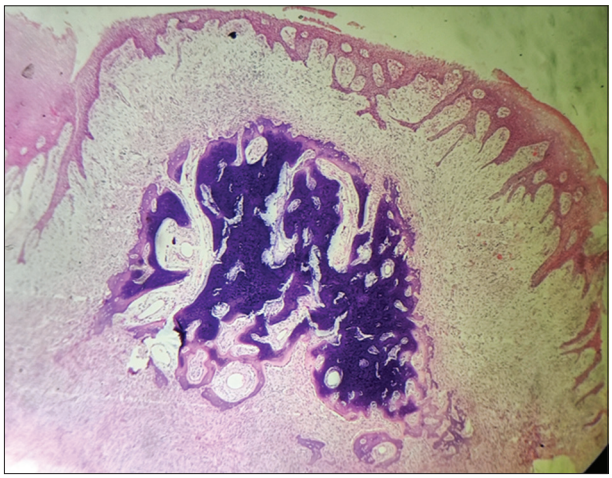

Figure 3: The scanner view of the specimen. Overlying epithelium is parakeratinised. The inderlyingstroma is fibro-collgenous with loose collagen around the bony entity. A area of hyalinization seen around the woven structure

process of EMT (Epithelial mesenchymal transition). In two consecutive meetings, one organized in Poland (2007), other one at Cold Spring Harbor Laboratories (2008) it was decided to classify EMT into three types based on the biological context.[10] EMT type 1 includes EMTs which are related to implantation, formation and development of embryo. This neither was related to fibrosis nor got invasive property. They can produce mesenchymal cells which can undergo an MET to produce secondary epithelia.[11] Type 2 of EMT is associated to wound healing, regeneration of tissue and fibrosis. This type is basically associated with repair.[11] Type 3 EMT is associated with neoplasm. Genetic changes and thus formation and growth of tumor is evident here. Changes seen in this type can affect oncogene, tumor suppressor gene, etc.; cancer cells achieve the ability to metastasize. These cells can invade other tissue. Among these cancer cells, some retain their epithelial characteristics and some acquire mesenchymal. But what exactly induce type 3 EMTs in cancer cells is still not clear. EMT uses various cell markers (epithelial and mesenchymal). As per example we can list E-cadherin, cytokeratin, ZO-1, laminin-1, entactin.syndecan, MUC1, desmoplakin, miR200 family (EPITHELIALPHENOTYPE) and FTS binding protein FAP, N-cadherin, vimentin, fibronectin, snail, slug, ETS, twist, goosecoid, miR21 (MESENCHYMAL PHENOTYPE). If there is a colocalization is seen of both the sets it indicates it is passing partly through EMT. Once transition is completed, there will be mesenchymal phenotype markers only. According to the studies, there are two different school of thoughts on development of choriostoma. One explained it to be a developmental anomaly, other explained it to arise due to trauma. Whatever may be the reason of development a trace to EMT can be thought.

Primary care physicians need to provide special attention in oral care as osseous choriostoma is not quite common, these lesion are often diagnosed as other lesions with similar clinical features. Diagnosis like peripheral giant cell granuloma and fibrous hyperplasia are made when present on alveolar ridge. If lesion is pedunculated it might be misdiagnosed to be papilloma too.[12] Osseous choristoma of oral cavity is rare. The range of age of patients can be from 5 to 73 years with a mean age of 28.7 years. Female predilection observed. The size of the lesion can go upto 5 cm and the duration ranges from 3 days to 50 years. The lesions present as a hard pedunculated or sessile mass. Hamartoma is a similar morphological entity which is often confused with choristoma. Choristoma is a histologically normal tissue proliferation of a type that is normally not found in the anatomic site. On the other hand, hamartoma is a disorganized proliferation of mature tissues, composed of elements that are normally found in the specific location in which it develops, often with one predominating element.[13] Primary care physicians' aim is to achieve improvement in these patients' overall health condition with main focus on oral health as it plays a main role diet, mastication that acts to maintain along quality of life.

Conclusion

This is a reported rare case of osseous choristoma of the mandibular buccal vestibule in a 21-year-old female. Though osseous choristoma is very rare in the oral cavity, it should be considered when a patient presents with a peripheral lesion not attached to underlying bone that has a hard consistency on palpation. Our diagnosis is based on clinical, radiographic examinations and histopathological analysis. Surgical excision is the recommended treatment. Recurrences are rare.

Declaration of patient consent

The patient has given consent for publication of her data and images.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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