Painless osteoid osteoma over anterior hard palate: A case report and review of literature

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Abstract

Introduction: Osteoid osteoma (OO) is a benign osteoblastic tumor which constitutes about 3% of all the primary bone tumors, about 10% of all the osseous benign tumors. Only 1% of the cases occur in the jaws. The present case reports a painless osteoid osteoma, which is a rarity.

Case presentation: A 35 year old male patient reported with a bony hard swelling on anterior part of hard palate. Radiograph showed a small radiopacity along the root of maxillary right central incisor. A provisional diagnosis of peripheral ossifying fibroma was made. An excisional biopsy under local anesthesia was performed. Platelet rich fibrin obtained from patient's blood was used as a surgical dressing. Healing was uneventful at the biopsy site. Histopathological investigation revealed it to be a benign bone forming tumor- a rare variant of osteoid osteoma with the absence of characteristic pain.

Conclusion: Osteoid osteoma is a benign tumor that occurs rarely in jaw bones. The present case reports a painless variant of osteoid osteoma of anterior part of hard palate in a young adult. This will add a rare variant of an otherwise rare lesion to the present sparse literature of osteoid osteoma.

Keywords: Diagnosis. Periodontal healing. Regeneration. Gingiva. Osteoid osteoma.

Introduction

Osteoid osteoma (OO) is a benign bone tumor which rarely involves the craniofacial bones. It accounts for 3% of all the primary bone tumors and approximately 10-12% of all benign bone tumors. About 80% of OO occurs in long bone while less than 1% occurs in jaws. It is most frequently reported in second and third decade of life, more common in males than females (2:1) (Ida *et al*., 2002). The hallmark symptom includes severe nocturnal pain which gets relieved by salicylates. It was first described as a distinct clinical entity by Jaffe (1935), who set the following diagnostic criteria: (1) It is a benign neoplasm, (2) mostly seen in young adults, (3) is unlikely an inflammatory lesion, (4) It has characteristic radiographic features (focal rarefaction and reactive bone formation), (5) forms large amount of osteoid tissue which gets calcified, (6) the most outstanding feature is the nocturnal pain which characteristically gets relieved by salicylates. The true nature of OO is unknown. Some considered it to be a variant of osteoblastoma because of its similar clinical and histopathological features, but the radiographic presence of radiopaque nidus surrounded by sclerotic bone formation favors the diagnosis of OO (Singh & Solomon, 2017).

The present case is reported because of its occurrence in anterior maxillary palatal region with no associated pain.

Case Presentation

A 35 year old healthy male reported with a swelling over gums on the palatal aspect of maxillary incisor teeth since 6 months. The patient incidentally noticed a small swelling which gradually increased in size. It was painless with no blood/ pus discharge. On examination there was a single, isolated, discrete hard swelling on the palatal aspect of maxillary right canine, lateral incisor and central incisor (Figure 1a). The swelling was 1.5x1.0 cm in size, sessile, non-tender, the overlying palatal mucosa was of same color as the adjoining normal gingiva with areas of slight erythema and ulceration, most likely due to interference in occlusion with the

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mandibular anterior teeth. The associated teeth were non-mobile, non-tender and vital. The maxillary right lateral incisor was slightly labially displaced. There was no reported drug intake. The patient was otherwise systemically healthy with no relevant medical or family history.

The differential diagnosis included: ossifying fibroma, cementoblastoma, idiopathic osteosclerosis, complex odontoma, osteoblastoma, and osteoid osteoma. Radiographs were advised. The intraoral periapical and maxillary occlusal radiograph showed a well-defined radiopacity in relation to the root of maxillary right lateral incisor in the middle third region (Figure 1b and 1c). The radiopacity was uniform and the margins were irregular but well-defined. There was no root resorption and root displacement. Based on the clinical and radiographic findings, a provisional diagnosis of peripheral ossifying fibroma was made.

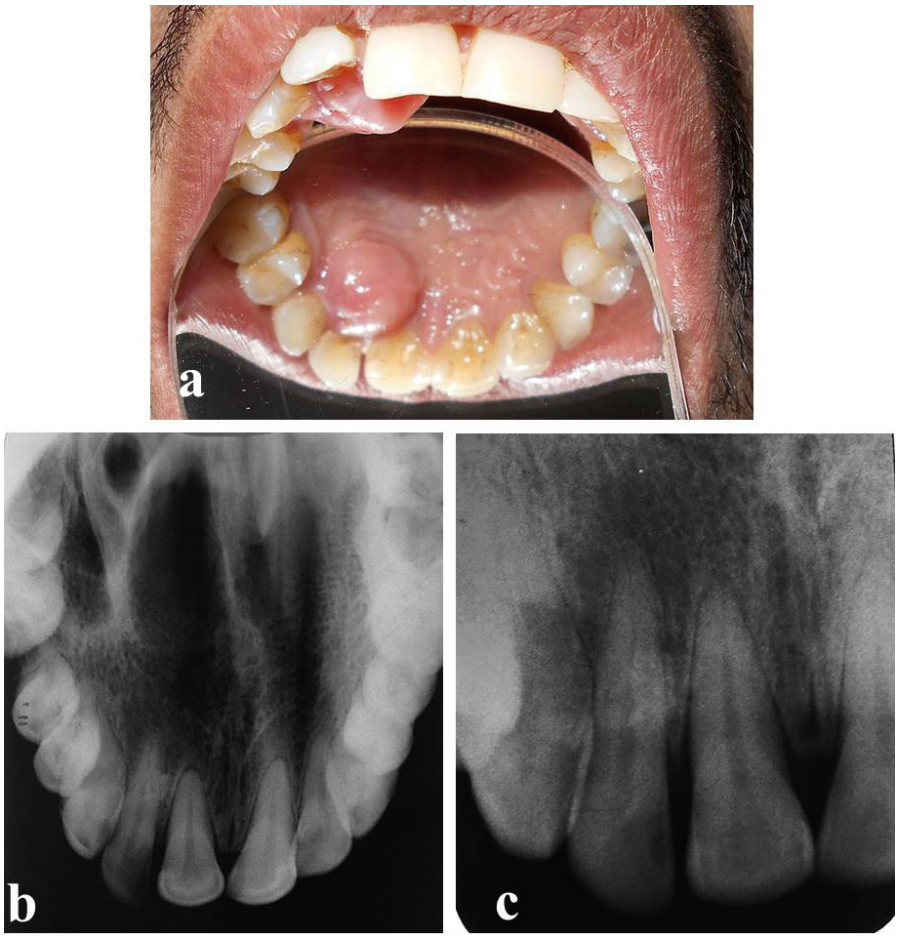

Figure 1. a) Pre-operative clinical picture of the sessile swelling over the palatal aspect of #11-13. b) Pre-operative Maxillary occlusal radiograph showing well defined radiopacity with irregular margins, in relation to the middle third region of #12 with distinct root outline and periodontal ligament space. c) Pre-operative intraoral periapical radiograph showing the same radiopacity in relation to root of #12.

Case Management

Oral prophylaxis was done. Routine blood investigations were advised and excisional biopsy was done under local anesthesia (2% lidocaine with 1:1,00,000 epinephrine) using scalpel and blade, keeping clear margins of 2 mm (Figure 2a). The surgical site was thoroughly curetted and inspected for any bony prominences. The center of the lesion offered bony hard resistance to the use of scalpel, so chisel and mallet were used. Platelet rich fibrin (PRF) was prepared from patient's blood to be used as a surgical dressing and very gently sutured with the adjoining palatal gingiva at the biopsy site (Figure 2b), using resorbable suture. To avoid any trauma during the healing phase, non eugenol periodontal dressing was placed under an acrylic removable maxillary stent. Patient was prescribed antibiotics and anti-inflammatory medications (Amoxicillin 500mg TDS and Ibuprofen 400 mg TDS for 5 days). He was also advised 0.12% chlorhexidine rinses twice a day for one week.

The patient was reviewed five days postoperatively when the acrylic stent was removed. The biopsy site showed very good healing and the patient was encouraged to continue using the mouthwash for another week. The patient was seen regularly during the healing phase. There were no signs of any recurrences over a follow-up period of five years (Figure 3).

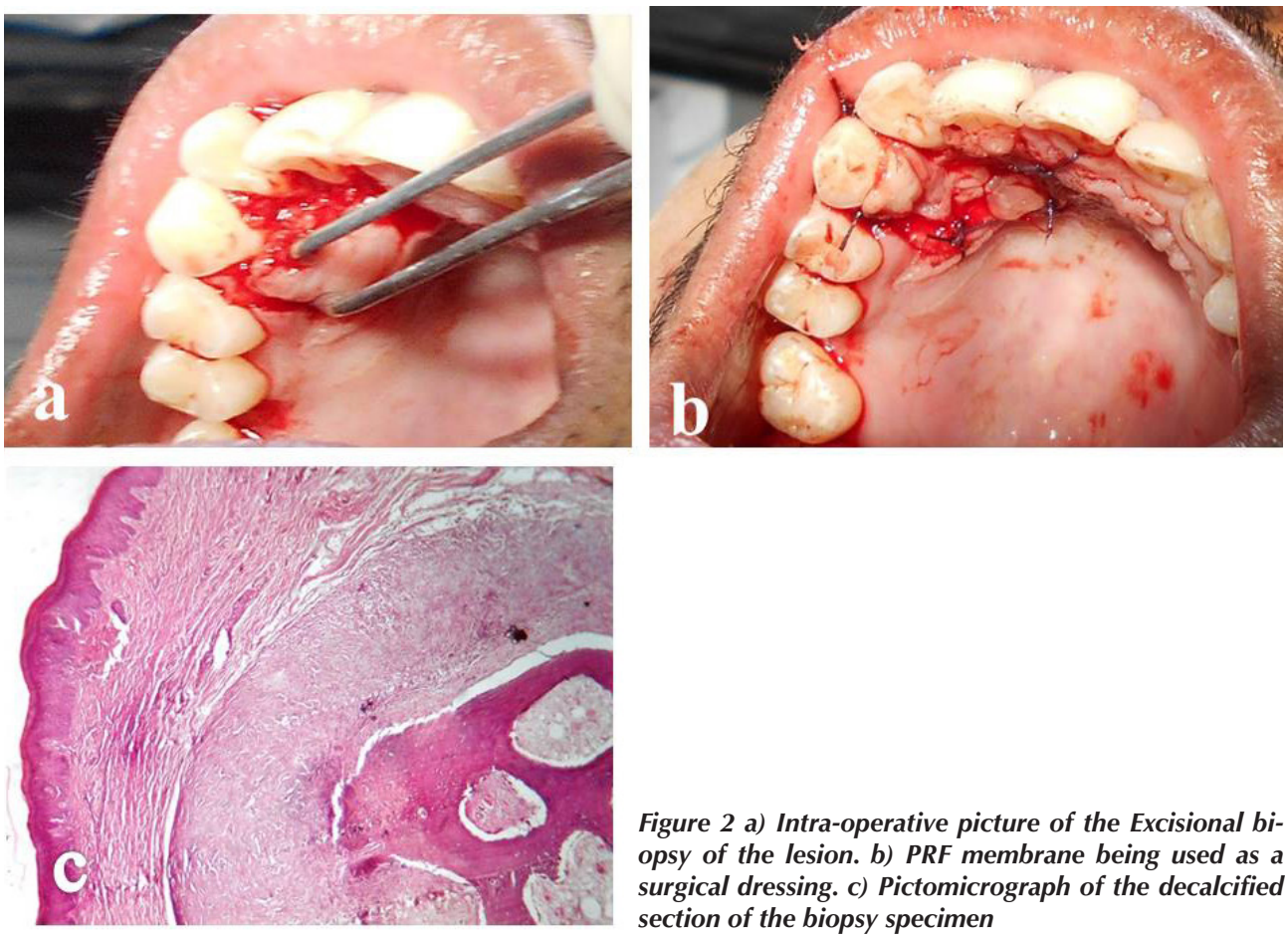

Figure 2 a) Intra-operative picture of the Excisional biopsy of the lesion. b) PRF membrane being used as a surgical dressing. c) Pictomicrograph of the decalcified section of the biopsy specimen

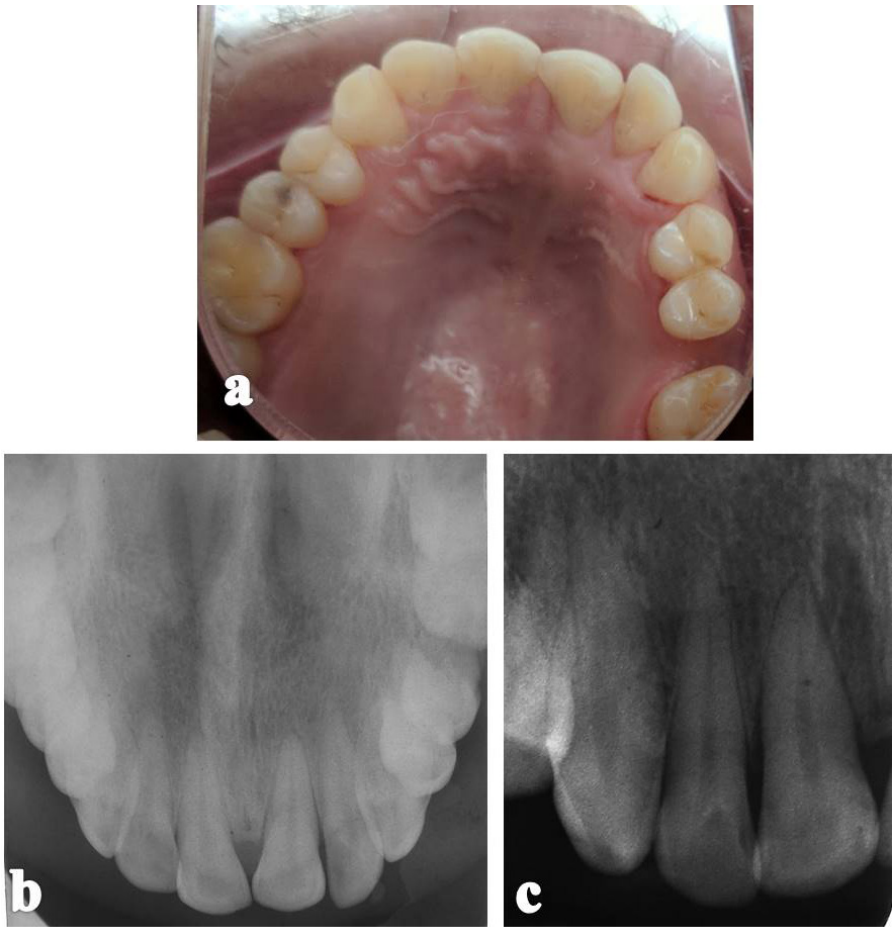

Figure 3. a) Clinical appearance of the biopsy site after one year of biopsy showing clinically indistinguishable appearance of the biopsy site as compared to the adjacent area. b) Maxillary occlusal radiograph showing complete healing with no signs of any recurrence after one year of biopsy. c) Intraoral periapical radiograph after one year of biopsy with good bone healing and no signs of recurrence.

The Hematoxylin & Eosin (H & E) stained decalcified section showed orthokeratinized stratified squamous epithelium overlying connective tissue. There was mature bone in the center along with areas of new immature bone and large areas of osteoid tissue at the periphery. There were abundant plump osteocytes entrapped in the osteoid tissue. Dense bundles of collagen fibers and few fibroblasts were seen at the periphery of the lesion. Several small blood vessels were evident throughout the lesion. There were no prominent nerve bundles seen. None of the cells showed any atypia (Figure 2c). The histological impression was of some bone forming benign lesion.

Discussion

Osteoid osteoma (OO) has been described as a small but characteristically painful bony lesion which seldom manifests in the jaws. Lichenstein (1965) described it as 'a small round tumor like nidus composed of osteoid and trabeculae of newly formed bone deposited within a highly vascularized osteogenic connective tissue'. According to Singh and Solomon (2017), who reviewed about 20 cases of OO from the English literature, reported that females were more commonly affected than males (1.2:1) and the predominant site was mandibular molar region during the second and third decade of life. In the present case, the tumor was present in a 35 year old male in anterior palatal region which is a rare site of its occurrence.

The clinical hallmark of OO is the pain which is continuous or intermittent, is usually dull and boring but worsens during the night, and characteristically gets relieved by salicylates. Many theories have been given to explain the curious pain seen in OO (Chaudhary and Kulkarni, 2007). Golding (1954) attributed the pain to the presence of the vascular components of the lesions, whereas, Jaffe (1958) postulated that the pain was because of the arteriolar blood supply of the lesion. Sherman and McFarland (1965) attributed the pain to the presence of non-myelinated nerve fibers accompanying the blood vessels in the reactive fibrous zone surrounding the nidus. Schulman and Dorfman (1970) suggested that the pain was generated and transmitted by the autonomic nerves sensitive to pressure. McDermott *et al*. (1996) reported an incidental case of painless OO of rib detected during preoperative scintigraphic study of a 59 year old male with prostatic carcinoma. They also reviewed the English literature for painless OO and found only 18 such cases, out of which only 3 cases (17%) were seen in the cranio-facial bones (1 in frontal bone, 1 in mandible and 1 in maxilla); others were reported in the phalanges (44%) and long bones (39%). They suggested that the site of origin may influence whether pain is present or not. The intramedullary or subperiosteal location of OO may permit painless growth as there is usually no surrounding zone of sclerotic bone which can cause nerve fiber compression and may incite pain. An absence of nerve fibers within the nidus may also cause painless occurrence of OO. An *et al*. (2013) reported a rare multifocal case of OO affecting the right mandibular body in a 10 year old boy and also reviewed the literature for OO of jaw bones.The most commonly reported complaint in cases of OO was pain (66.7%), followed by swelling (52.4%) and tenderness (14.3%). Of the osteoid osteomas of the jaws reviewed, 34.3% of the cases were not related to pain history. Out of 21 cases, 5 cases involved maxilla, mandible was involved in 16 cases and only one case of painless OO was reviewed in left anterior maxilla in a 77 year old male as incidental finding with a radiographic finding of a dense nidus with surrounding sclerosis (Brynolf, 1969). So it may be postulated that the site of occurrence, i.e. anterior maxilla may present painless variant of OO only. Stoopack (1958) reported a case of painless OO in a 25 year old male as an incidental finding in a patient having a sialolith of the submandibular gland with concurrent OO of the mandible. Absence of pain in our case could be explained by the subperiosteal location of the lesion and the absence of any prominent nerve bundles in the histologic section.

The characteristic radiographic features of OO is the presence of central radiolucent nidus of <2cms in diameter with a sclerotic margin. An *et al*. (2013) in their review on OO of the jaw bones concluded that the nidus of OO may have dense (38.1%) or mixed (38.1%) radiographic appearance rather than radiolucent appearance. They also cited that the subperiosteal osteoid osteoma arises as a soft tissue mass adjacent to the affected bone with almost no reactive sclerosis. In our case, the radiographic picture was that of a well-defined, uniform radiopacity with irregular margin and the subperiosteal location explains the absence of sclerotic margin.

The treatment of choice for OO is complete surgical excision. In our case, Platelet rich fibrin (PRF) was used as an adjunct to the biopsy site healing. PRF is a second generation platelet concentrate obtained from autologous blood without the need of any complicated biochemical processing. Aravindaksha *et al*. (2014) coined the term "Palatal Bandage" for the PRF membrane to be used as a dressing at the free gingival graft (FGG) donor sites. Reports on using PRF as an adjunct for wound healing in oral cavity are scarce. To the authors' knowledge, ours is the first case where PRF has been used as a "physiologic dressing" at the "biopsy site" to promote healing and reduce post-operative pain. While osteoid osteoma generally does not recur after complete removal of the nidus; in recurrent lesions, it may be inferred that may be the lesion was not removed completely in the first instance or may have

multiple nidi (An *et al.,* 2013). Seeding of OO has not been reported. Few cases of malignant transformation of osteoblastoma have been reported but none of OO (Chaurasia and Balan, 2008).

Conclusions

Osteoid osteoma is a rare benign jaw tumor. This reported case is a rarity in terms of its clinical presentation of being painless lesion in anterior palatal region. Clinicopathologic and radiographic correlation is important to differentiate it from other benign bone forming tumors. Clinical presentation and symptoms play a critical role in the pathologic diagnosis of osteoid osteoma with other similar lesions.The small number of reported cases of OO necessitates an increased awareness among the dentists to report additional cases in literature.It will provide a medium for better understanding and show a clear picture of such rare bony lesions so that they could be diagnosed at a much earlier stage.

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