

## Case Report

# Osteoid Osteoma of the Oral and Maxillofacial Region: Report of Two Cases

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## ABSTRACT

**Background:** Osteoid osteoma frequently affect long bones such as femur and tibia but seldomly affect jaw bones. It is neither locally aggressive nor does it have the potential for malignant transformation. Reports suggest that it occurs in young adults below 30 years of age. **Case presentations:** We document and discuss 2 cases of osteoid osteoma, one, a 22-year old with a giant lesion on the right maxillary region, partially occluding the vision. The patient complained of associated pain. The second patient, a 25 year old female who presented with a mandibular swelling of about 1 cm which was also associated with pain. These two patients had temporary relieve of their pain by ingestion of analgesic. They both had surgeries that were uneventful. **Conclusion:** We documented and discussed 2 cases of Osteoid osteoma on the maxillofacial region in young patients with both of the cases almost occluding the patient's vision.

**Keywords:** Osteoid Osteoma, Maxillary tumors, Mandibular tumors, Oral and Maxillofacial Region

## INTRODUCTION

Osteoid osteoma(OO) represents about 10 % of all benign bone tumours that was first described in 1935 by Jaffe<sup>[1]</sup>. It is neither locally aggressive nor does it have the potential for malignant transformation and frequently affect long bones of the femur and tibia. It seldomly occurs in the jaw bones.<sup>[1]</sup> Reports suggest that it occurs in young adults below 30 years.<sup>[1,2]</sup> As at 2015, the largest documented OO was 7.0x6.5x6.4cm.<sup>[3]</sup> Three types of osteoid osteoma have been described viz., cortical (classic type), medullary and subperiosteal types.<sup>[4]</sup> The etiology of OO remains unclear, it is believed that it is either a benign neoplasm or it may be due to

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trauma or could result from an inflammatory process.<sup>[2,3]</sup> Pain is a characteristic feature of this lesion and is accompanied by vasomotor disturbances.<sup>[1]</sup> OO is said to be very rare in the head and neck region<sup>[5]</sup> Hence, we present 2 cases of OO. a 22-year old with a maxillary lesion of 3 years and a 25-year old female with a mandibular mass of a year duration We also reviewed the relevant literatures.

#### Case Report

Case 1. A 22 -year old male presented at our facility with a 3-year history of Right (RT) maxillary lesion, slow growing, painful, and obstructing his vision(Fig1).



**Fig 1: The male patient with massive osteoid osteoma occluding the eyelids.**

The pain was intermittent, of short duration and relieved by analgesics. There was no history of trauma and no discharge from the swelling. Examination showed a well-defined non-tender right maxillary swelling that measured 10x15 cm in dimension. It was bony hard on palpation and attached to the underlying structures. There was obstruction of the right eye by the lesion. The mouth opening was adequate, with full complement of the teeth and no deranged occlusion. The swelling was bony hard and did not extend into the oral cavity. A clinical diagnosis of osteoma was made. Patient could not afford a CT Scan and so we ordered for plain radiographs which showed an area of opacity over the antral region.

Excision of the lesion was carried out under General Anesthesia(Fig2)



**Fig 2: Same patient after the lesion has been exposed.**

for histopathological analysis of the surgical specimen carried out. Post operatively, the patient was placed on antibiotics and analgesics and was discharged home after 10 days(Fig3).

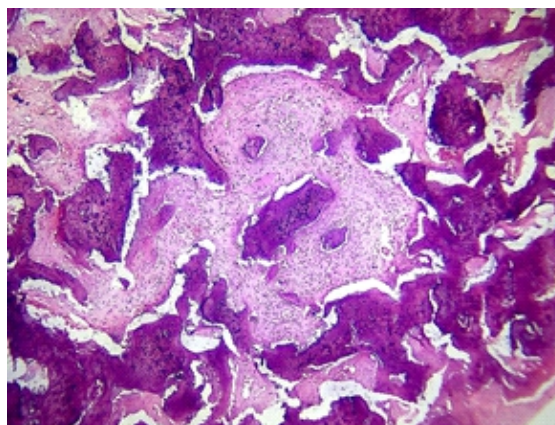
The gross specimen consisted of bony hard,



**Fig 3: Post-operative**

irregularly shaped, grey-brown tissue, grey brown tissue measuring 6.5x4.5x3cm and weighing 55g. Cut surface of the specimen showed white hard areas. Microscopic sections showed a lesion composed predominantly of cortical type bone and mature lamellar bone. Nidus of interlacing network of osteoid trabeculae with variable mineralization which demonstrated prominent osteoclastic rimming and the inter-trabecular space which contained dense fibro-vascular stroma within which are bland looking spindle cells and multinucleated giant cells (osteoclasts) with mild infiltration by inflammatory cells(Fig4).

It was diagnosed as Osteoid osteoma. The histopathological diagnosis of OO was consistent with the preoperative diagnosis



**Fig 4: Photomicrograph at x40: Haphazardly arranged bony trabeculae rimmed by osteoblasts within vascularized fibroblastic stroma**

Case 2: A 25 years old female presented with a 1 year history of painful swelling which was not increasing in size since she noticed it, there was no associated history of discharge nor previous history of trauma. Although there was associated Pain, however, it was relieved by analgesics. Examination revealed a well-defined mandibular swelling measuring 1cmx1cm, the swelling was not tender or hard but was attached to the underlying bone. The overlying mucosa was also relatively intact. It was located around the alveolus and basal bone. There was no derangement of occlusion. Patient was unable to take computerized tomographic scans as she could only afford a plain radiograph which showed an area of radiopacity surrounding a radiolucent point.

Surgical excision was done under general anaesthesia. She had antibiotics and analgesics and was discharged home after 4 days.

The gross specimen was made of bony hard, irregularly shaped, grey-brown tissue measuring 1.0x1.0x0.9cm and weighing 15g

Histology showed interlacing network of osteoid trabeculae surrounded by mature lamellar bone and a cellular fibrovascular stroma within which were bland spindle cells and occasional osteoclast type giant cell.

## DISCUSSION

Less than 1% of OOs occur in the jaw bones

compared to about 80% in long bones.<sup>[1]</sup> As at 2015, only about 26 cases of OOs of the jaws were reported in the literature.<sup>[3]</sup> It occurs in the second and third decade of life with more female in the ratio of 1.08:1 to 1.2:1.<sup>[1,3]</sup> The present cases were 22 years old male with a large maxillary lesion and 25 years old female with mandibular OO. Sunil et al<sup>[3]</sup>, reported a lesion measuring 7.0x6.8x6.4 cm on the mandible while our largest case was 6.5x4.5x3cm. Al Sadhan et al<sup>[6]</sup>, reported a case of 3.5mm in diameter. The lingual aspect and the lower border of the body of the mandible have been found to be the most common sites of involvement.<sup>[7]</sup> Our mandibular case was on the lingual aspect of the mandible.

The commonest symptom of OO is pain, which might be either continuous or intermittent.<sup>[1]</sup> This pain is frequently dull and boring, and often becomes worse at night.<sup>[1]</sup> Previous authors pointed out that vascular elements were responsible for the pain and the osseous reaction. Others believed it was attributable to the arteriolar blood supply of the OOs.<sup>[1]</sup> In the present cases, there was history of intermittent and radiating pain. The duration of the lesions did not arouse any suspicion of malignancy nor was the lesion intraoral for our first case for any odontogenic pathology to be suspected. The second case was however an intraoral lesion and pain was the main reason for presentation. These findings were in agreement with the literatures.<sup>[1]</sup> As at now, the exact nature of OO has a diverse pathogenesis suggesting a range of presentation from neoplasm to inflammatory reaction to unusual healing.<sup>[3,8]</sup> Karandikar et al<sup>[9]</sup>, reported a case with previous history of trauma, but non of our patients gave history of trauma.

The usefulness of the radiographic appearance of osteoid osteoma in its diagnosis cannot be over emphasized. According to Jaffe, cited by Singh and Solomon<sup>[1]</sup>, the nidus is more lucent than opaque and is surrounded by a reactive opacity of variable distance from the nidus. The less matured lesion has a radiopaque presentation while the fully matured one has a radiolucent nidus. When osteoid calcification occurs at the later end of

development, there is resultant central opacity of the body which varies in density as the calcification progresses.<sup>[1,6]</sup> In the case reported by Singh and Solomon<sup>[1]</sup>, the radiographic picture was that of a well-defined radiopacity with radiolucent rim showing a central radiopaque nidus surrounded by a radiolucent border. This was also noted in the first case presented here. The second case however did not present with a clear radiographic finding. The radiographic differential diagnosis includes ossifying fibroma, idiopathic osteosclerosis, cementoblastoma, complex odontoma, osteoblastoma, and osteoid osteoma. Though Ossifying fibroma causes root resorption, OO does not. Idiopathic osteoclerosis and complex odontoma are asymptomatic and do not cause bone resorption which OO does. Cementoblastoma is asymptomatic and characterized by the propagation of cementum in close association with the root of the tooth.<sup>[1]</sup>

The histopathology as described by Huvo has three distinct evolutionary phases of ossification. The initial stage characterized by the presence of actively proliferating, densely packed prominent osteoblasts in a highly vascularized stroma. The intermediate phase, in which the osteoid is deposited between the osteoblasts. The mature stage of the lesion, the osteoid is transformed into a well-calcified, compact trabecula of an atypical bone, which are histopathologically peculiar because they are neither typically woven nor typically lamellar.<sup>[6]</sup> The case reported by Singh and Solomon<sup>[1]</sup>, was a mature type of the lesion which is similar to the histopathologic findings in both of our cases.

Treatment of OO is excision and does not recur.<sup>1</sup> Malignant transformation is very rare though a case has been reported in the literature which transformed into osteosarcoma.<sup>[10]</sup> The present cases were excised and there has not been recurrence after 1 year and 9 years respectively.

## CONCLUSION

We reported 2 cases of OO which is a benign bone lesion in a 22 years old male and a 25 years old

female. The lesion in the first patient was massive and occurred in the maxilla which is an uncommon site, with partial obstruction of vision. With respect to the search literature, it was found to be the second biggest in term of size. The second occurred on the mandible around the alveolus and basal bone. They both presented with pain that was temporarily relieved by analgesics.

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