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Rare disease

Osteoid osteoma of mandible

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Summary

Osteomas are benign, slow-growing osteogenic tumours rarely occurring in the craniofacial bones. Osteomas are characterised by the proliferation of compact and/or cancellous bone. It can be of a central, peripheral, or extra skeletal type. The peripheral type arises from the periosteum and is rarely seen in the mandible. The lingual surface and lower border of the body are the most common locations of these lesions. They are usually asymptomatic and can be discovered in routine clinical and radiographic examination. In this paper, the authors present a large solitary peripheral osteoma located in the inferior surface of the left angle of mandible and causing facial deformity in a 14-year-old boy. Radiographic examination by diagnostic radiographs revealed mixed appearance (radiopaque- radiolucent), well-circumscribed, pedunculated mass approximately 3.5 cm in size. The osteoma was removed surgically, and no recurrence has been observed.

BACKGROUND

Osteoid osteoma (OO) is considered as a low growth neoplasm or even more, like an inactive neoplasm. The lack of knowledge concerning the genesis of OO and its confusion with similar lesions in the bone make accurate compilation of data concerning the lesion a difficult task. It is obvious that the small number of reported cases of OO in maxilla and mandible does not allow drawing conclusions about this lesion's behaviour in the jaws.

CASE PRESENTATION

A 14-year-old male reported to the oral diagnosis and radiology department with a complaint of facial and extraoral swelling on the inferior aspect of the left angle of mandible. He had been aware of the slow but steady increase in the size of the lesion over the past 4 years. The lesion was associated with occasional pain at night, and there was no difficulty in opening of mouth or chewing. He had facial trauma about 5 years back, and his medical history was not contributory. Clinical examination revealed extraoral swelling on the left side (figure 1). The regional lymph nodes were non-palpable. Extraoral examination revealed a well-defined, round, immobile mass. The lesion was bony-hard on palpation. The overlying skin was normal. A solitary, round, 3.5×3.5 cm well-defined radio-opaque radiolucent lesion at inferior border of mandible was noticed with panoramic radiography and postero-anterior view of mandible (figures 2 and 3). No similar lesions were found in maxilla, mandible, ribs and long bones.

These clinical and radiographic features were supportive of the working diagnosis of peripheral osteoma.

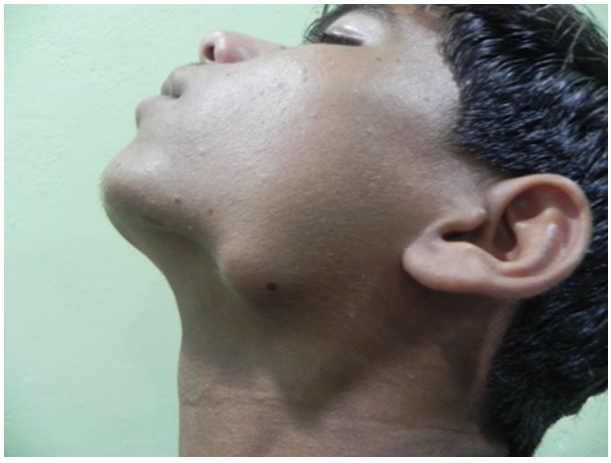

Figure 1 Clinical presentation.

INVESTIGATIONS

- Complete haemogram and routine urine
- Serum calcium
- Serum alkanine phosphatase
- Serum prostaglandin estimation
- Liver function test
- Kidney function test
- Orthopantomograph
- Postero-anterior view of mandible.

DIFFERENTIAL DIAGNOSIS

- Garre's osteomyelitis
- Eosinophillic granuloma
- Osteosarcoma (early detection)
- Osteoblastoma
- Complex odontoma.

TREATMENT

Under local anesthesia with controlled intravenous sedation, the lesion was completely excised using oscillating saw and chisels via an extraoral approach. Curettage and fulgaration of the inferior border of mandible (near the point of origin of tumour) was accomplished (figure 4). Standard protocol of perioperative antibiotics

and anti-inflammatory regimen was followed. The excised specimen was submitted for histopathological examination. The histopathology confirmed the clinical diagnosis of OO (figure 5).

OUTCOME AND FOLLOW-UP

The patient is undergoing regular follow-up after uneventful recovery and removal of sutures. Patient shows good healing with cosmetically acceptable scar. Yet so far there is no recurrence since 6 month.

DISCUSSION

OO is a benign lesion consisting of a small round nidus. It accounts for 3% of all primary bone tumours, and about 10% of benign bone tumours. About 80% of OO occur in long bones, while less than 1% occurs in jaws. OO is most frequently observed in the second and third decades of life, more commonly in males than females, at a ratio approximately 2: 1. Mild pain is the principal symptom as in the present case.[1–4]

OO was described as a specific entity by Jaffe in 1935. Jaffe described a type of nidus which appeared as a hard osseous core composed of densely set trabeculae of newly formed bone which was atypical. Jaffe perceived the initial notable changes in this lesion as an increased vascularisation and destruction with replacement by new atypical bone following resorption of the destroyed tissue. The stroma consisted of osteogenic connective tissue containing numerous blood channels. A cortical lesion which produced this bony replacement stimulated the overlying periosteum to lay down new bone of fairly normal architecture.[5]

Real prevalence of osteoma is unknown. Sondergaard *et al*[6] in their study demonstrated that the prevalence of osteoma in 50 patients with ulcerative colitis is 4% and 2% in the control group. It has been reported that osteomas have no sex predilection.[7 8]

Osteoma of the jaw bones is quite rare. These lesions are more frequent in the mandible than the maxilla. Sayan *et al*[9] reported finding 22.85% of the lesions in the mandible and 14.28% in the maxilla in their study. Kaplan *et al*[7] reported 81.3%, Chaurasia and Balan[8] reported 83% and Woldenberg *et al*[10] reported 64% of cases occurred in the mandible.

The lingual surface and lower border of the body are the most common locations of mandibular lesions.[11–13] The exact aetiology and pathogenesis of peripheral osteoma is unknown. Neoplastic and reactive causes have been suggested as possible aetiologic factors. Kaplan *et al*[7 14] and Woldenberg *et al*[10] suggested that some peripheral osteomas may be reactive rather than neoplasms, probably associated with trauma. Also, some authors have reported that as many of the peripheral osteomas are located on the lower border of the mandible, it is possible that muscle traction plays a role in the development of peripheral osteomas.[10 11] However, in the case described in this paper, we have information as to the possible cause, there being history of previous trauma to the same region 5 years back.

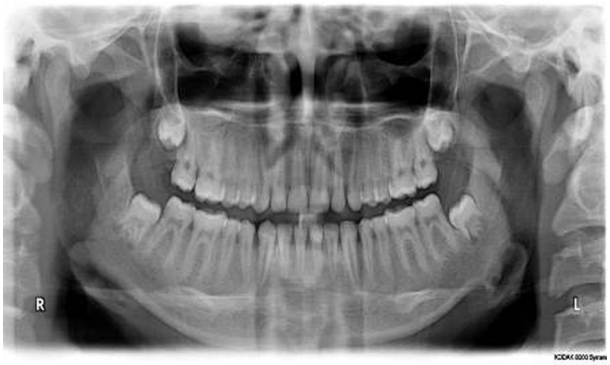

Figure 2 Orthopantomograph.

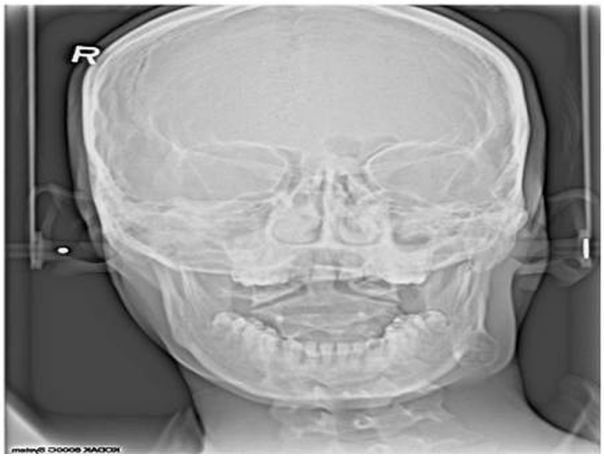

Figure 3 Postero-anterior view mandible.

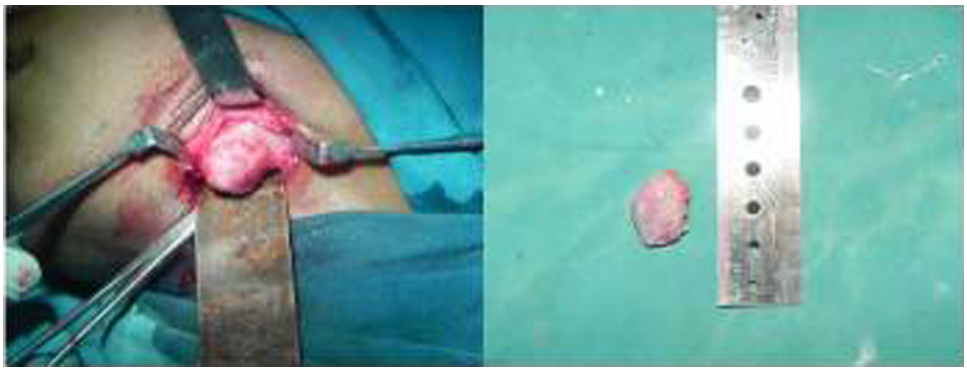

Figure 4 Surgical exploration and excised specimen.

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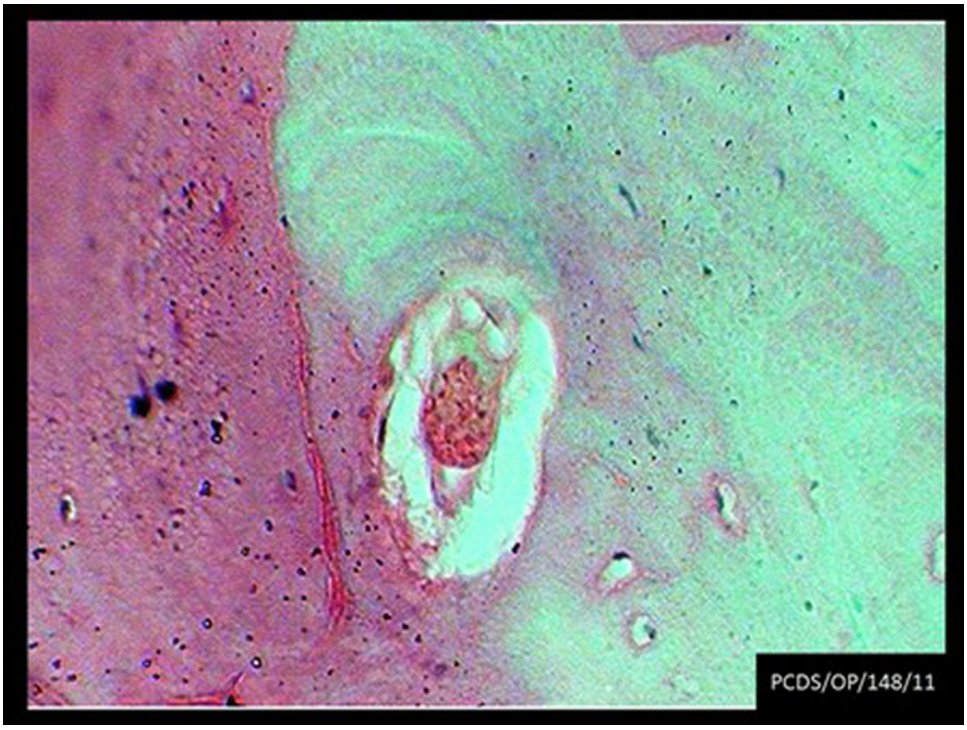

Figure 5 Histopathological picture.

Histologically osteomas consists of mature, lamellar bone or cancellous bone with abundant fibrofatty marrow between bony trabeculae,[15 16] as reported in our case. Histologically there is narrow margin of differentiation between osteoma, osteochandroma and tori, it can only be differentiated clinically and radiographically. Radiographically osteoma shows as well-circumscribed, densely sclerotic and radiopaque mass. Endosteal osteomas are generally identified on routine radiographic examination.[16] Osteomas are diagnosed and treated by local excision.[15]

Recurrence of peripheral osteoma after surgical excision is extremely rare; the goal of follow-up is to look for new osteomas or other signs indicative of Gardners syndrome, as it is ruled out in our case as our patient is young male adult.

OO can occur anywhere. It can involve a single bone or several bones. It is reported to occur in the cortex of the shafts of long bones in 80–90% of cases, epiphysial and metaphyseal regions of both small and large bones of the axial and appendicular skeletons, especially the femur, tibia and humerus.

The lower extremities are the most common sites of OOs. Barei *et al* reported that in 50–60% of cases, OO occurs in the femur and tibia.[17 18]

Pettine *et al* noted that 50% of lesions occur in the cervical spine, and up to 78% of OOs in the lumbar spine are associated with scoliosis.[19–22] The tumour has a predilection for the posterior elements, most commonly affecting the cancellous lamina, spinous process and pedicle but sparing the vertebral bodies.

OOs of the hand and wrist are rare, most commonly involving the phalanges, and often result in atypical clinical and radiologic characteristics. The findings are similar to those of tumours involving the foot and ankle. Intra-articular OO occurs in 10% of cases and can involve the hip, elbow and ankle.[23–25]

Malignant transformation of peripheral osteoma has not been reported in the literature. Patients with osteomas should be evaluated for Gardner's syndrome. This syndrome is an autosomal dominant disease characterised by gastrointestinal polyps, multiple osteomas, skin and soft tissue tumours and multiple impacted or supernumerary teeth. Intestinal polyps are predominantly adenomas and may progress to malignancy in almost 100% of patients.[26 27]

Learning points

- The rate of growth is slow with common complaint of pain.
- Can cause facial asymmetry.
- Obtaining proper history is important.
- Thorough physical examination is necessary to rule out polyosteotic origin.
- Histopathological confirmation aids in diagnosis.

Competing interests None.

Patient consent Obtained.

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