Rare disease

Brown tumour of the jaw

Preeti P Nair,¹ Harshkant P Gharote,¹ Shaji Thomas,² Guruprasad R,¹ Neha Singh¹¹Department of Oral Medicine and Radiology, People's College of Dental Sciences and Research Centre, Bhopal, Madhya Pradesh, India;²Department of Oral and Maxillofacial surgery, People's College of Dental Sciences and Research Centre, Bhopal, Madhya Pradesh, India**Correspondence to** Professor Preeti P Nair, shajihoss@gmail.com**Summary**

Brown tumours are classic bony lesions that arise as a result of the effect of parathyroid hormone on bone tissue in some patients with hyperparathyroidism. They are erosive bony lesions caused by rapid osteolysis and peritrabecular fibrosis, resulting in a local destructive phenomenon. Facial skeleton is involved in about 2% of all cases of which the mandible is frequently affected. A 35-year-old female who was diagnosed with osteomalacia and brown tumour in posterior mandible as the sign of secondary hyperparathyroidism secondary to vitamin D deficiency is presented.

BACKGROUND

The reported prevalence of brown tumour is 0.1% and its incidence in secondary hyperparathyroidism is 1.5 to 1.7%. This report describes a case of lytic bony lesion in the mandible that raised the suspicion of giant cell lesion, compelling further investigation. Positive results in serum chemistry propelled referral to an endocrinologist revealing vitamin D deficiency (osteomalacia). This signifies the role of the oral physician in early diagnosis of systemic conditions in individuals who present it with the oral manifestations. This paper is an attempt to throw light that nutrition deficiency such as of vitamin D can cause bony lesion which can be an early indication of systemic illness.

Figure 1 Extraoral swelling present on the lower one third of face.

CASE PRESENTATION

A 35-year-old female reported with a swelling in lower left jaw since 1 year. She had undergone extraction of lower left carious second and third molars 1 year back. One month later, she noticed a swelling in the same region, which progressed slowly to attain the present size. Over the last 3 months, she experienced pain that was dull and intermittent initially but later became severe. Her medical and family history was not significant. On extraoral examination, a single diffuse swelling was present on the lower one third of face extending 2 cm from the left corner of the mouth up to angle of the mandible and superoinferiorly 4 cm from ala of the nose to 2 cm below lower border of the mandible (figure 1). The temperature of the swelling was slightly raised and it was firm and tender on palpation. Intraorally a well-defined swelling was present over the alveolar ridge extending from distal aspect of 36 upto the retromolar area and from buccal vestibule to the floor of the mouth on the left side, measuring 2.5×2.7cm in size, with well-defined margins and ulcerated surface (figure 2). It was tender on palpation and firm to hard in consistency.

Figure 2 Intraoral swelling over the left lower posterior alveolar ridge.

Figure 3 Periapical radiograph of 36, 37 region.

There was bicortical expansion of the mandible. The hard tissue findings included root stump with 46, deep proximal caries with 48 and supra-eruption with 27, 28 impinging over the swelling of the lower jaw. The periodontal status of the patient was poor with generalised periodontal pockets and gingival recession with 31, 41.

Based on history and clinical examination, a provisional diagnosis of benign tumour of mandible with secondary infection was arrived at.

INVESTIGATIONS

Periapical radiograph revealed a mixed radiopaque-radiolucent lesion in 37, 38 region (figure 3). An occlusal view showed expansion of lingual cortex from 36 extending posteriorly. The overlying bony covering is of poor density (figure 4). A panoramic view exhibited a lesion of mixed density with ill defined margins extending from distal of 36 to middle of ramus of mandible (figure 5). CT showed evidence of expansile lytic lesions of size $2.7 \times 2.5 \times 1.9$ cm noted in the left mandibular body with thick sclerosis in outer cortex and thinning in the inner cortex. Multiple irregular bony septae with hyperdense foci were also noted in the lesion (figure 6).

The serum chemistry showed that parathyroid hormone (PTH) level increased to 77.10 pg/ml (normal range 15–68 pg/ml) and 25-hydroxy cholecalciferol level decreased to 3.47 ng/ml (normal range 30–100 ng/ml). However, serum alkaline phosphatase, calcium and phosphorus levels were within normal limits. Renal function test was within normal limits.

Incisional biopsy was performed. Histopathological examination showed highly cellular connective tissue stroma composed of plump spindle shaped cells interspersed throughout with numerous multinucleated giant cells. The giant cells varied in size and shape and showed upto 25 nuclei. The stroma also showed many small blood capillaries with extravasated red blood cells. Periphery of the tissue showed bony trabeculae (figures 7 and 8). The histopathological features were suggestive of giant cell granuloma.

Thus, a final diagnosis of brown tumour of jaw with secondary hyperparathyroidism and osteomalacia were arrived at.

Figure 4 Occlusal radiograph showing poorly defined expanded lingual cortical plate.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis of ameloblastoma, central giant cell granuloma, odontogenic keratocyst, dentigerous cyst, odontogenic myxoma and calcifying epithelial odontogenic tumour were put forth.

TREATMENT

The treatment was directed towards improvement of the systemic condition and the patient was treated with 60000 IU of vitamin D3 once a week for 6 weeks and calcium (500 mg) twice daily.

OUTCOME AND FOLLOW-UP

The patient was kept under observation for a period of 3 months and it was noticed that the size of the lesion showed mild reduction. The patient is currently under regular follow-up.

DISCUSSION

Von Recklinghausen was the first to describe hyperparathyroidism (HPT) as a bone disease in 1891. Askanazy, in 1904, was the first to describe a patient with a parathyroid tumour and osteitis fibrosa cystica. However, he did not correlate these two entities. HPT may occur in one of three clinical forms: primary, secondary or tertiary. In all instances the disease is characterised by increase in PTH level and mobilisation of calcium from bone. Primary parathyroid hyperplasia, parathyroid adenoma or parathyroid carcinoma can result in primary HPT. Secondary HPT results from a compensatory mechanism due to the primary condition producing hypocalcemia such as rickets, osteomalacia, pregnancy or chronic renal insufficiency.

Figure 5 Orthopantomograph showing mixed density lesion with ill defined margins.

Figure 6 CT image of the mandible.

Tertiary HPT may occur after long-standing secondary HPT.¹ Increased level of PTH causes bone resorption that result in hypercalcaemia.² A fourth type of HPT, (ectopic HPT) as described by Guimarães *et al*⁶ is thought to arise from increased PTH levels synthesised in patients with malignant diseases.

The name 'brown tumour' for bony lesions seen in HPT was first coined by Jaffe. The term is used because of reddish brown colour caused by its vascularity, haemorrhage and deposits of haemosiderin.⁴ It is considered as a kind of giant cell lesion that presents as an osteolytic lesion of the bone. It is reported to occur in 4.5% of patients with primary hyperparathyroidism and 1.5 to 1.7% of those with secondary disease.⁵ It may be the first clinical manifestation of hyperparathyroidism as in the present case. Mandible is more often affected as compared to maxilla. It is three times more frequently seen in women aged over 50 years⁶ unlike the present case who was 35 years old. The lesion may present as a soft to firm mass with varying degrees of tenderness,⁴ altered masticatory function, facial deformities⁶ or may be asymptomatic as in our case report.

Brown tumour appears as a well-defined unilocular or multilocular radiolucency.^{2 7 8} Ashrafi described their radiographic appearance as well-defined lytic lesions with little reactive bone formation, cortical thinning or expansion.⁴ Langland *et al* described them to be monocystic without marginal scalloping, showing internal septae.¹ They described that the borders may be poorly defined resembling the present case. Jebasingh *et al* described the mandibular lesions as well-defined osteolytic lesions.⁹ In the present report, CT revealed expansile lytic lesion in the left

Figure 7 Photomicrograph of the lesion (low power).

Figure 8 High power resolution of the lesion.

posterior mandible with thick sclerotic outer cortex and thinning of inner cortex. Multiple irregular bony septae with hyperdense foci were also noted in the lesion that gave it a multilocular appearance.

Histologically, brown tumour or osteoclastoma depicts multiple giant cells mixed with stromal cells and matrix. Intraosseous bleeding and tissue degeneration results in cyst formation. The cystic spaces are filled with clusters of giant cells, haemosiderin laden macrophages and plump fibroblasts.⁹ All giant cell lesions consist of two main components: mononuclear stromal cells and multinucleated giant cells.³ However, no pathognomonic histologic changes are found in brown tumour and similar changes may be seen in conditions like fibrous dysplasia, giant cell tumour and reparative granuloma. Reparative granuloma is detected in young patients as localised lesion. It primarily affects mandible. The cause is still unknown but some investigators believe that they are results from trauma, and can be differentiated from brown tumour by absence of hyperparathyroidism.⁵

Secondary hyperparathyroidism may be caused by chronic renal failure or vitamin D deficiency (osteomalacia). Secondary hyperparathyroidism results in high levels of PTH as a compensation for hypocalcaemia rather than a primary abnormality of parathyroid gland. This high level

of PTH then causes resorption of bone.¹⁰ Brown tumours are mainly the result of secondary hyperparathyroidism in patients with renal insufficiency, but they have also been described as a rare manifestation of calcium malabsorption and some forms of osteomalacia.¹¹

Histopathological presentation of brown tumour cannot be distinguished from that of giant cell lesion; hence diagnosis is based on its association with HPT. There is no single test that establishes the diagnosis of secondary HPT and is based on the high level of PTH associated with low or normal serum calcium. In the present case, PTH level were higher than normal but did not show any alteration in calcium level.³

The management of the secondary hyperparathyroidism depends on the successful treatment of the primary causative condition. According to Pellegrino¹² surgical excision of the brown tumour is indicated under following circumstances: if the lesion is large and disfiguring or if the affected bone is weakened. This author stated that healing and remodelling is accelerated if the brown tumour is removed.¹

Learning points

- ▶ Brown tumour can appear as an early manifestation of osteomalacia with secondary hyperparathyroidism.
- ▶ Role of oral physician is important in identifying systemic conditions.
- ▶ In developing nations nutritional deficiency should rank high in the differential diagnosis of disorders.
- ▶ There is a significant incidence of osteomalacia which remains asymptomatic, resulting from poor dietary intake in addition to lack of exposure to solar radiation.
- ▶ Osteomalacia leading to secondary hyperparathyroidism must be addressed before surgical removal of brown tumour.
- ▶ A multi-disciplinary approach to this entity can result in successful treatment.

Competing interests None.

Patient consent Obtained.

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