Rare disease

A bilaterally recurring exophytic mass on the lower jaw

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Summary

Brown tumour is a rare focal giant-cell lesion that arises as a direct result of the effect of parathyroid hormone on bone tissue in patients with hyperparathyroidism (HPT). The lesions localise in areas of intense bone resorption, and the bone defect becomes filled with fibroblastic tissue. It can affect the mandible, maxilla, clavicle, ribs and pelvic bones. Therefore, diagnosis requires a systemic investigation for lesion differentiation. The authors present a case of a 27-year-old woman, with a rare case of brown tumour of mandible due to HPT secondary to osteomalacia. The lesion showed a tendency to recur even after excision till her parathyroid levels became normal.

BACKGROUND

Secondary hyperparathyroidism (SPT) can manifest with no symptoms except brown tumour as in our case. Differential diagnosis of exophytic growths especially in the posterior mandible should also include brown tumour.

CASE PRESENTATION

A 27-year-old female patient reported with recurrent swellings on both sides of the lower jaw since 2 years. She noticed a small swelling on the right side of the lower jaw in the molar region which gradually increased in size over a period of 8 months and was painless. She gave no history of trauma from opposing teeth in the region of the swellings during the early stages of the lesion. There was displacement of the associated teeth. Examination revealed

a similar lesion on the contralateral side of the same jaw (figure 1). Her oral hygiene status was found to be good.

Both the lesions were excised but they recurred within 1 month. They were excised again with removal of the molar teeth on the right side. A histological diagnosis of reparative giant cell granuloma was obtained.

Two months later, a similar swelling reappeared on the left side of the lower jaw. This time, it was a rapidly growing lesion associated with pain when occluding. Extraoral examination revealed a diffuse, non-tender and firm swelling (figure 2) on the left lower side of her face with an associated tender, mobile submandibular lymph node. Intraoral examination revealed an irregular, firm and non-tender exophytic mass (figure 3) measuring 40×60 mm on the alveolar ridge, displacing the lower first and second molars. The

Figure 1 Panoramic radiograph taken earlier (when the lesions first appeared on either side).

Figure 2 Diffuse non-tender firm swelling on the left lower side of face.

surface of the mass was roughened due to trauma from the upper teeth. Bleeding was present on palpation.

INVESTIGATIONS

Intraoral periapical radiograph of 36, 37 revealed a diffuse radiolucency, surrounding the entire root with loss of lamina dura depictive of massive bone destruction. Panoramic radiograph (figure 4) showed extensive bone loss in the lower left molar area which gave a floating appearance of the first molar and the lesion had displaced the teeth.

Serum parathormone level was 264 pg/ml and serum calcium levels were 9.6 mg/dl. Her serum phosphorous levels, alkaline phosphatase levels, renal function tests, liver function tests were within normal limits.

Her vitamin D level was 3.2 pg/ml.

Ultrasonography (USG) of parathyroid glands revealed that a 15×15 mm hypoechoic lesion on the left side near the posteroinferior surface of the thyroid gland with few specks of calcification seen within it. USG guided fine needle aspiration cytology of the lesion revealed colloid cells with no presence of oxiphyl or chief cells suggestive of solitary thyroid nodule.

DIFFERENTIAL DIAGNOSIS

- ▶ Pyogenic granuloma
- ▶ Peripheral giant cell granuloma
- ▶ Central giant cell granuloma
- ▶ Brown tumour of hyperparathyroidism (HPT).

Figure 3 Firm and non-tender exophytic mass present on the left alveolar ridge.

Figure 4 Panoramic radiograph showing extensive bone loss in the molar area giving a floating appearance of the first molar and the lesion displacing the teeth out of occlusion.

TREATMENT

She was administered 500 mg calcium with 250 IU vitamin D₃ daily and cholecalciferol 60 000 IU weekly for 6 months.

OUTCOME AND FOLLOW-UP

After 3 months her parathormone level was reassessed and found to be 154 pg/ml. She was advised to continue her medications, and after 6 months her parathormone levels were found to be normal. This confirmed the diagnosis of HPT secondary to osteomalacia. The lesion was surgically excised and had not recurred after 9 months of follow-up (figures 5 and 6).

DISCUSSION

Brown tumour is a non-neoplastic lesion that develops in patients with HPT, occasionally affecting the jawbones.¹ This lesion results from microfractures and secondary haemorrhages that lead to an influx of multinucleated macrophages and the growth of reactive fibrous tissue.²⁻⁴ Prevalence of brown tumour is about 0.1%, more common in persons over 50 years and more frequent in mandible than in maxilla.⁵ Although Neville *et al* implied that these lesions are most common in the jaws – other literatures do not agree with them.^{6 7}

Primary HPT is characterised by high circulating levels of parathyroid hormone, possibly due to hyperplasia or

Figure 5 Postoperative panoramic radiograph.

Figure 6 Postoperative intraoral photograph.

benign/malignant neoplasia of the parathyroid glands.^{1 7 8} Secondary HPT involves an increase in parathyroid hormone (PTH) levels to meet some bodily requirement. It is a compensatory mechanism that may be due to rickets, osteomalacia, pregnancy, renal insufficiency, calcium deprivation or maternal hypoparathyroidism.⁹ Tertiary HPT is due to the development of autonomous parathyroid hyperplasia after long-standing secondary HPT, most often in renal failure.¹⁰ There is increase in bone breakdown (osteoclastic resorption) with subsequent fibrous replacement and reactive osteoblastic activity.¹¹ There may be microfractures, with subsequent haemorrhage and growth of fibrous tissue and an influx of macrophages. The resulting mass is called a brown tumour because of the brown colour of the vascular elements and blood in the mass. This results in the characteristic radiological features of generalised bone demineralisation, resorption, cysts, brown tumours, erosion of the dental lamina dura and pathologic fractures.¹⁻³

Kalathas *et al* stated that osteitis fibrosa cystica/brown tumour as a manifestation of HPT was first described by Von Recklinghausen (1891).¹² The first case in the literature

reporting a brown tumour was published in 1953 that described a fronto-ethmoidal brown tumour.¹³ It is linked to abnormalities that appear in HPT such as excess PTH production in primary hyperparathyroidism, or phosphate retention, skeletal resistance to PTH, impaired degradation of PTH and altered feedback regulation of PTH by calcium in SPT.¹³⁻¹⁶

Brown tumours of mandible and maxilla are uncommon but can be the earliest manifestation of undiagnosed HPT (6%).^{17 18} They usually present clinically as slowly enlarging, locally destructive swellings.^{9 19} They may occasionally behave aggressively and grow to a huge size. On rare occasions, lesions indistinguishable from peripheral giant cell granulomas have been seen in patients with HPT.² Occurrence as an exophytic growth as in our case is very rare.^{20 21}

Radiographically, brown tumours present with reduced bone density, generalised osteoporosis, loss of lamina dura and 'salt and pepper' appearance of the skull,^{22 23} but localised brown tumours in jaws pose diagnostic difficulties as they simulate common lesions like ameloblastoma, central giant cell granuloma, osteomyelitis, odontogenic cysts,

etc.^{23 24} Additionally, they are histologically indistinguishable from the other giant cell lesions like aneurysmal bone cyst, cherubism and giant cell granuloma. Consequently, all patients with giant cell lesion in the jaws should be screened for both HPT and hypercalcaemia to rule out brown tumour. Definitive diagnosis validates a meticulous clinical, radiological and biochemical assessment.^{4 15 25}

In a review of 220 patients, Rosenberg and Guralnick found that 10 patients (4.5%) had a mass involving one or both jaws as their presenting complaint.²⁶ Brown tumours are more commonly seen in the mandible than in the maxilla.¹⁻³ The reported prevalence of brown tumours is 0.1%. The disease can manifest at any age, but it is more common among persons over 50 years, and is three times more common in women than in men.^{4 26 27} Brown tumours are histologically similar to giant-cell tumours, giant-cell regenerative granulomas, cherubism and aneurysmal osseous cysts.^{2 3}

On imaging, brown tumours appear as lytic lesions with thinned cortical bone that may be fractured. Concurrent changes that suggest brown tumours such as osteopaenia, a 'salt-and-pepper' bone appearance, subperiosteal bone resorption and disappearance of the lamina dura around the roots of the teeth, may help differentiate it from other entities.²⁸⁻³⁰ Tomographic imaging shows an osseous mass, with no cortical disruption, no periosteal reaction or inflammatory signs, a heterogeneous centre and areas that suggest cysts. MRI shows variable intensities on T2-weighted images and intense enhancement on T1-weighted contrast MRI. MRI may be better for determining the presence of cysts or fluid filled levels; a finding that is very suggestive of a brown tumour. Osseous gammagraphy is not indicated for the diagnosis of brown tumours; however, isolated hypermetabolic lesions or simultaneous hypercapitation of bone lesions and parathyroid adenomas, when done with Tc 99m, have been described.²³

Arabi *et al* stated that healing of rickets and osteitis fibrosa cystica with vitamin D therapy in a 14-year-old girl with non-azotemic rickets, while some reports have shown that intravenous alfacalcidol or calcitriol is effective treatment for secondary HPT in subjects undergoing dialysis. These agents reduce PTH secretion and parathyroid cell hyperplasia, and in some cases they resulted in regression of brown tumours without surgical treatment.²⁵

Learning points

- ▶ Reactive hyperplasia is common in the oral mucosa.
- ▶ Brown tumour of HPT should be included in the differential diagnosis of gingival hyperplasia adjacent to areas of bony involvement, particularly in the posterior mandible.
- ▶ Investigate such lesions for serum calcium, serum alkaline phosphatase, serum parathormone and serum vitamin D.
- ▶ Surgery is to be deferred till the serum PTH has returned to normal otherwise the lesion is likely to recur.
- ▶ The lesion may undergo spontaneous regression once PTH has returned to normal.

Competing interests None.

Patient consent Obtained.

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