CASE REPORT

Mandibular brown tumor as the first manifestation of primary hyperparathyroidism: A case report

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Abstract Patients with primary hyperparathyroidism (brown tumor) are usually asymptomatic and clinical presentation of the tumor in the jaws is rarely the first sign of the disease. We report a 45-year-old female patient who presented with a mandibular swelling as the first sign of the disease.

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1. Introduction

Primary hyperparathyroidism (Primary HPT) is defined as hypercalcemia resulting from the overproduction of parathyroid hormone (PTH) by one or more hyperfunctioning parathyroid glands. Most cases, 80% to 85%, are caused by a single parathyroid adenoma. The rest are caused by parathyroid hyperplasia with parathyroid cancer accounting for 1% to 2% of cases (Owens, 2009). Patients with primary HPT are usually diagnosed incidentally as hypercalcemia, but less frequently the clinical presentation includes renal calculi, osteoporosis, neuropsychiatric symptoms and, rarely, peptic ulcer disease or pancreatitis (Rodgers et al., 2008; Fraser, 2009). A variety of imaging techniques, including ultrasonography, computed tomography, magnetic resonance imaging and Technetium-99 (Tc-99m) can be used in investigating the disease.

In this case report, a large brown tumor in the mandible revealed Primary HPT, an unusual first manifestation of the disease.

2. Case report

A 45-year-old female patient presented to the dental department in a general district hospital in a rural part of Jordan, complaining of a large swelling in the anterior mandible of three months duration. Pain and paresthesia were not noted, but she had grade 3 mobility in the lower anterior teeth. Her medical history was unremarkable. A Panoramic radiograph (Fig. 1) revealed a radiolucent lesion of the mandible extending from right premolar region to left incisors; it had irregular margins and evidence of root resorption.

On surgical exploration of the lesion, both the buccal and lingual cortical plates were found to be expanded. The surgeon performed an excisional biopsy followed by curettage of the
bony walls with a large round bur. Histopathology of the mandibular lesion showed a proliferation of spindle cells with extravasated blood and haphazardly arranged, variably sized multinucleated giant cells (Fig. 2). At this time the lesion was identified as giant cell lesion and the patient was referred to the University of Jordan Hospital for further treatment.

Blood biochemistry revealed elevated serum calcium level 11.41 mg/dl (normal range, 8.5–10.5), alkaline phosphatase 183 IU/L (normal range up to 100), parathormone 94 pg/ml (normal range, 15–65) and normal phosphorus 3.9 mg/dl (normal range, 2.4–4.1). Sestamibi scan of the parathyroid glands reported parathyroid adenoma in the lower pole of the left lobe of the thyroid gland (Fig. 3). A second surgical procedure was undertaken to excise the left inferior parathyroid gland. The excised tissue measured 2.2 cm × 2 cm × 1 cm and weighed 2.4 g. The patient was prescribed calcium carbonate and alfa-calcidol orally postoperatively to combat transient hypocalcemia.

A follow up at 18 months revealed satisfactory clinical recovery with adequate bone filling radiographically and normal calcium, phosphorus, alkaline phosphatase and parathyroid hormone levels. Calcium supplement treatment was discontinued after 6 months.

3. Discussion

Brown tumors represent the terminal stage of hyperparathyroidism-dependent bone pathology. They have been described as resulting from an imbalance of osteoclastic and osteoblastic activity that causes resorption with fibrous replacement of the bone (Suarez-Cunqueiro et al., 2004). Although mandibular involvement has been reported in 4.5% of subjects in a 220-case hyperparathyroidism study (Rosenberg and Guralnick, 1962), it is rare to find a brown tumor as the first clinical manifestation of primary HPT before the onset of general manifestations (Suarez-Cunqueiro et al., 2004). Clinically, brown tumors present as painful, hard, clearly visible and palpable swelling. Radiographically, they appear as well-demarcated

Figure 1 Panoramic radiograph revealing a well-defined radiolucent lesion in the anterior mandible with evidence of root resorption.

Figure 2 A photomicrograph of the surgical section (hematoxylin–eosin staining; magnification ×20) showing proliferation of spindle cells with extravasated blood and variably sized multinucleated giant cells, arrow pointing to a large multinucleated giant cell.

Figure 3 Technetium-99 (Tc-99m) Sestamibi scanning. The patient had one “hot spot” in her neck corresponding to a parathyroid tumor in the lower left parathyroid gland.
monolocular or multilocular osteolytic lesions, with root resorption and loss of the lamina dura seen occasionally.

Diagnosis is readily confirmed by establishing elevated serum calcium and parathyroid hormone levels, because histological features alone are insufficient. Sestamibi scanning is one of the most common ways to localize diseased parathyroid glands prior to surgery (Rodgers et al., 2008). Tc-99m, a mild and safe radioactive agent, is injected intravenously and absorbed by the hyperactive parathyroid gland and detected by scanning.

Treatment involves control of hyperparathyroidism and for primary HPT, partial parathyroidectomy is required. Small osteolytic jaw lesions may regress spontaneously, however, with large disfiguring and symptomatic lesions, excision may be indicated as was done in this case (Suarez-Cunqueiro et al., 2004; Daniels, 2004). Surgical excision of the jaw lesion, when required, is usually done after parathyroid surgery. However, in this case the surgeon’s decision to excise the lesion rather than to perform initially incisional biopsy was based on the assumption that patients in remote regions of Jordan do not attend follow-up appointments regularly.

Postoperative hypocalcemia may occur in patients who undergo partial parathyroidectomy, therefore, calcium supplements could be required as was done with this patient.

4. Conclusion

Radiolucent lesions of the jaws showing giant cells on histopathology should raise suspicion of hyperparathyroidism. This case highlights the importance of a thorough diagnostic work-up for all lesions in the mandible.

References