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Case report

Unusually rapid growth of brown tumour in the mandible after parathyroidectomy associated with the presence of a supernumerary parathyroid gland

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ABSTRACT

The aim of this study is to report the case of a quick growing brown tumour in the jaw after a parathyroidectomy due to the presence of a rare fifth parathyroid gland. The patient had chronic renal disease and the diagnosis was tertiary hyperparathyroidism. Thirty days after the parathyroidectomy, the patient returned with a significant increase in the tumour size. The suspicion of a supernumerary gland was confirmed by parathyroid scintigraphy. The treatment of brown tumour is dependent on the treatment of the hyperparathyroidism. However, curettage should be considered if a large lesion is disturbing mastication. In conclusion, this case should attract the attention of general practitioner dentists, since they may be the first professionals who have contact with the patient with a brown tumour in the jaws. Likewise, this case emphasises the importance of knowing the type of hyperparathyroidism involved to allow for effective treatment planning.

1. Introduction

The brown tumour is an unusual giant cell tumour of the maxillofacial skeleton that appears as late bony manifestations of severe hyperparathyroidism (HPT), as a consequence of undiagnosed or untreated HPT where the bone metabolism is abnormal (Smith and Ward, 1978). The increased circulating levels of parathyroid hormone increase osteoclastic bone resorption, primarily in cortical bone (Triantafillidou et al., 2006).

The HTP may be classified into three categories: primary, secondary and tertiary. Primary hyperparathyroidism is not a rare disease and the primary form is due to excessive production of parathyroid hormone (PTH) secretion by an autonomous gland resulting in hypercalcaemia. Secondary hyperparathyroidism as a result of hypocalcaemia, vitamin D deficiency or chronic renal insufficiency, which acts as a stimulus for PTH production. Tertiary HPT, first described in 1963, is the rarest form of hyperparathyroidism, affecting patients with long-standing secondary HPT who, in the setting of renal failure, develop hypersecretion of PTH (Davies et al., 1968).

Histologically there is no difference between a brown tumour and central giant cell granuloma (CGCG). Thus, the diagnosis of brown tumour can be made through the use of biochemical tests such as serum calcium, alkaline phosphatase, phosphorus, sodium, potassium and intact PTH levels, and also through the findings of PTH (Pinto et al., 2006).

Its treatment is initially based on treating the underlying endocrine abnormality. Surgical excision of the diseased parathyroid gland to control PTH is the first choice of treatment for a brown tumour because the normalisation of parathyroid function should lead to a reduction in size or disappearance of the tumour (Pinto et al., 2006). However, brown tumour management depends on the severity of the lesions present.

The aim of this paper is to report a rare case of a rapidly growing brown tumour in the jaw associated with tertiary hyperparathyroidism and present the importance of a thorough early diagnosis for successful treatment. This should allow a dentist to understand the disease and to refer the patient for the correct treatment.

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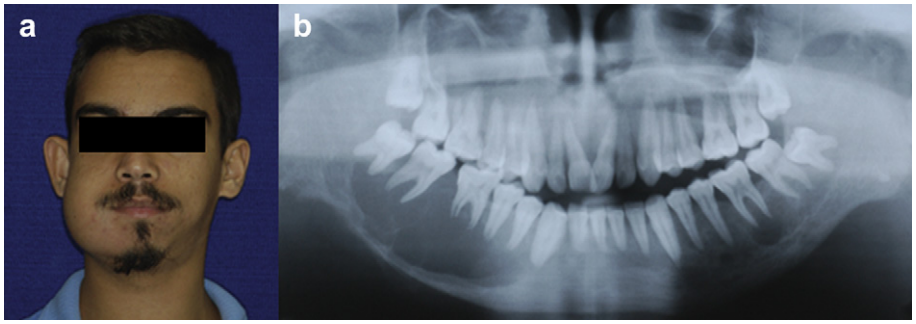

Fig. 1. (a) Patient with 21 years old presenting swelling of the posterior right mandible. (b) Orthopantogram showing a large, well-defined, unilocular, radiolucent lesion right mandible.

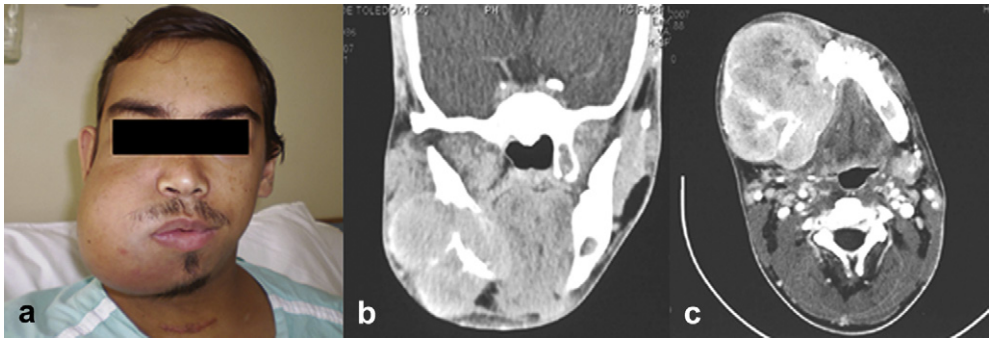

Fig. 2. (a) Dramatic enlargement of the lesion in the anterior and posterior mandible 30 days after first surgery. Computed tomography scan of the related areas showed the expansive nature of the brown tumour in mandible, 30 days after first surgery. (b) Coronal CT and (c) axial CT.

2. Report of a case

A 21-year-old male patient was referred to the Department of Diagnosis and Surgery at São Paulo State University in May 2007 for the evaluation of asymptomatic facial asymmetry which was noticed 30 days previously. Swelling of the posterior region of the right side of the mandible was evident (Fig. 1a). Medical history was significant, with chronic renal insufficiency and anaemia. The patient had been undergoing dialysis three times a week which he began two years before the clinical onset of the lesion.

The swelling was firm and painless to the touch. The dentition was initially unaffected. A panoramic radiograph showed a large, well-defined, unilocular, radiolucent lesion extending from the roots of the canine to the third molar on the right side, and a small lesion between the first pre-molars and the first molar on the left side of the mandible (Fig. 1b).

A biopsy taken from multiple areas was performed for histopathologic evaluation. The results were consistent with a giant cell lesion. The differential diagnosis included CGCG, brown tumour of HPT and aneurismal bone cyst.

Serum chemistry was performed and revealed: an elevated parathyroid hormone (PTH) level at 1088 pg/dL (15–65 pg/dL); serum calcium, 8.9 mg/dL (8.8–11); increased phosphorus, 6.7 mg/dL (2.5–5.0); alkaline phosphatase 2393 IU/L (65–300). Based on the medical history, clinical manifestations and laboratory tests, the final diagnosis was brown tumour of HPT.

A whole body scan showed skeletal involvement, focal areas of increased concentration of radiotracer in the ribs, iliac bone, skull and mandible in the regions. After correction of his anaemia, the patient underwent a total parathyroidectomy identified in the usual location by scintigraphy, with auto-transplantation of parathyroid tissue in the right brachioradialis muscle. At the same time, he underwent partial removal of the thyroid due to its proximity to the parathyroid glands. The histological findings were compatible with parathyroid hyperplasia.

Thirty days after surgery, the patient returned with an enlargement of the lesion in the anterior and posterior mandible and a slight increase of the palate causing difficulty in eating and speaking (Fig. 2). The second whole body scan showed increased osteogenesis and the presence of several other new osteogenic lesions compared to the previous examination performed before surgery (Fig. 3a). The new laboratory findings were: parathyroid hormone (PTH) level at 413 pg/dL (15–65 pg/dL); alkaline phosphatase 1627 IU/L (65–300); the serum calcium levels remained within the reference values.

This form of persistent disease usually results from an ectopic or supernumerary parathyroid gland so parathyroid scintigraphy was performed. This scan identified a focal area of increased radiation posterior to the left sternoclavicular joint compatible with a diagnosis of hyperfunctioning parathyroid tissue (Fig. 3b).

The patient underwent further surgery with re-exploration of the neck and upper mediastinum, at which time a thymectomy and resection of a supernumerary parathyroid gland were performed (Fig. 3c). At the same time curettage of the mass in the mandible via an extra oral approach was performed under general anaesthesia (Fig. 3d).

Post-operatively, the patient developed symptomatic hypocalcemia and was managed with oral calcium supplementation in addition to vitamin D3 and Renagel to reduce serum concentrations of phosphorus.

A CT scan five months after the second procedure showed reduction of the lesion (Fig. 4a and b). Two years later, there was no recurrence of the brown tumour (Fig. 4c). Serum chemistry was performed and revealed: serum calcium, 10.7 mg/dL (8.8–11.0 mg/dL);