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CASE REPORT

Bilateral maxillary brown tumours as the first presentation of primary hyperparathyroidism

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Abstract
Introduction Symptomatic skeletal disease in primary hyperparathyroidism is over 30 times more common in India compared to the west. The classical "brown tumour" is commonly seen with the major sites being ends of long bones, the pelvis and ribs. Facial involvement is rare and, when present, usually involves the mandible.
Case report We report a 68-year-old gentleman with a rare initial presentation of primary hyperparathyroidism with bilateral maxillary brown tumours.
Discussion and conclusion Successful parathyroid surgery resulted in a regression in the tumours. The report highlights the need to consider primary hyperparathyroidism in the initial differential diagnosis of bony lesions of the jaw.

Keywords Maxillary brown tumours · Primary hyperparathyroidism

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Introduction

Routine screening for serum calcium has lead to the pattern of changing clinical presentation of primary hyperparathyroidism in western countries. Symptomatic primary hyperparathyroidism (PHPT) accounted for only 2% of patients in a series from Minnesota (United States) [8]. In India, PHPT is rarely recognised but remains a predominately symptomatic disease. Advanced skeletal involvement with formation of brown tumours or pathological fractures was seen in 60% of our patients [3]. The common sites of brown tumours are the long bones, pelvic girdle, clavicle, ribs and the mandible. Tumours involving the maxillae are rare.

We report the case of a 68-year-old gentleman who presented with brown tumours of both maxillae with PHPT. The successful excision of a right inferior parathyroid adenoma resulted in the gradual decrease in the maxillary tumours. The report highlights the need to consider PHPT in the differential diagnosis of tumourous lesions of the maxillae.

Case report

A 68-year-old gentleman was referred to the Department of Otolaryngology with progressive tumourous growth in both maxillae over the last 2 years. (Fig. 1). As the paranasal sinus X-ray was suggestive of a malignancy (Fig. 2), a computed tomographic (CT) scan was ordered. The CT scan of the maxillae revealed (Fig. 3a,b) large expansile lucent lesions at the alveolar process of the right and left maxilla. A bone scan of the maxillary region with technetium-99, which showed increased uptake in both maxillae, was reported as to be suggestive of an inflammatory lesion (Fig. 4). The patient underwent a biopsy of

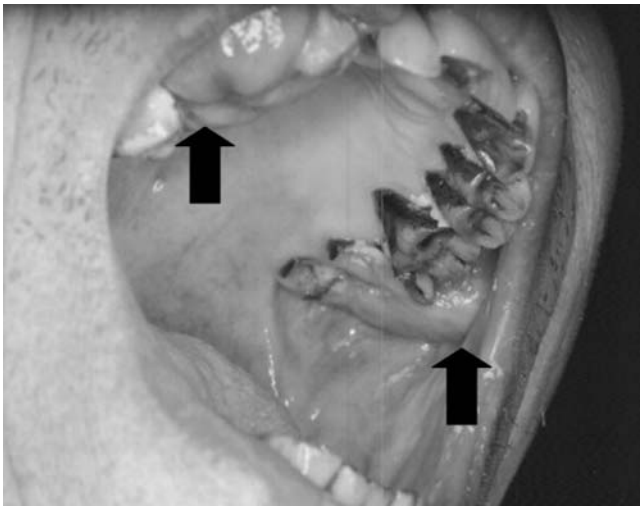

Fig. 1 Photograph of the patient's oral cavity showing bilateral soft tissue masses in relation to the maxillae

the right-sided tumour. The biopsy revealed multiple giant cells, fibroblasts with areas of haemorrhage, which was consistent with a giant cell reparative granuloma (Fig. 5). At this point, biochemical evaluation revealed the presence of hypercalcemia with elevated serum parathyroid hormone levels.

The patient was referred to the endocrine department. Further questioning revealed the history of symptomatic renal stones with bilateral renal lithiasis and mild renal impairment. The patient gave prior history of a Colles' fracture sustained 5 years back. Review of the CT scan revealed the presence of a nodule below the right inferior pole of the right lobe of the thyroid. The location of the parathyroid adenoma was confirmed with a sestamibi scan. The patient underwent right inferior parathyroid adenoma removal along with percutaneous nephrostomy for his

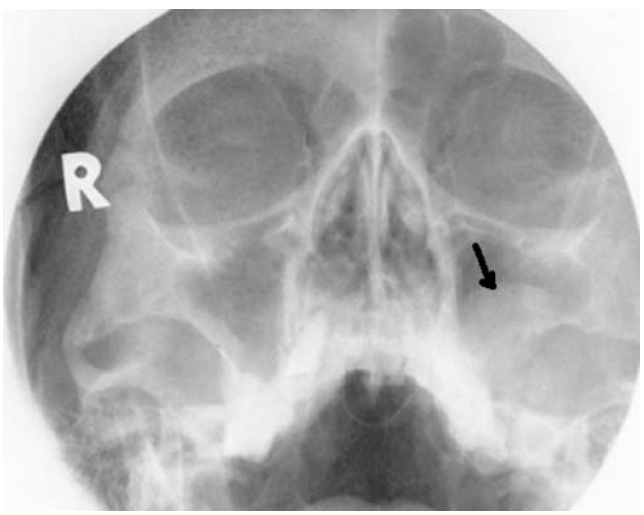

Fig. 2 The X-ray PNS revealed a homogenous opacity with a convex upper margin in the region of the left maxillary sinus with loss of definition of the upper alveolar margin and lucency of the left zygoma

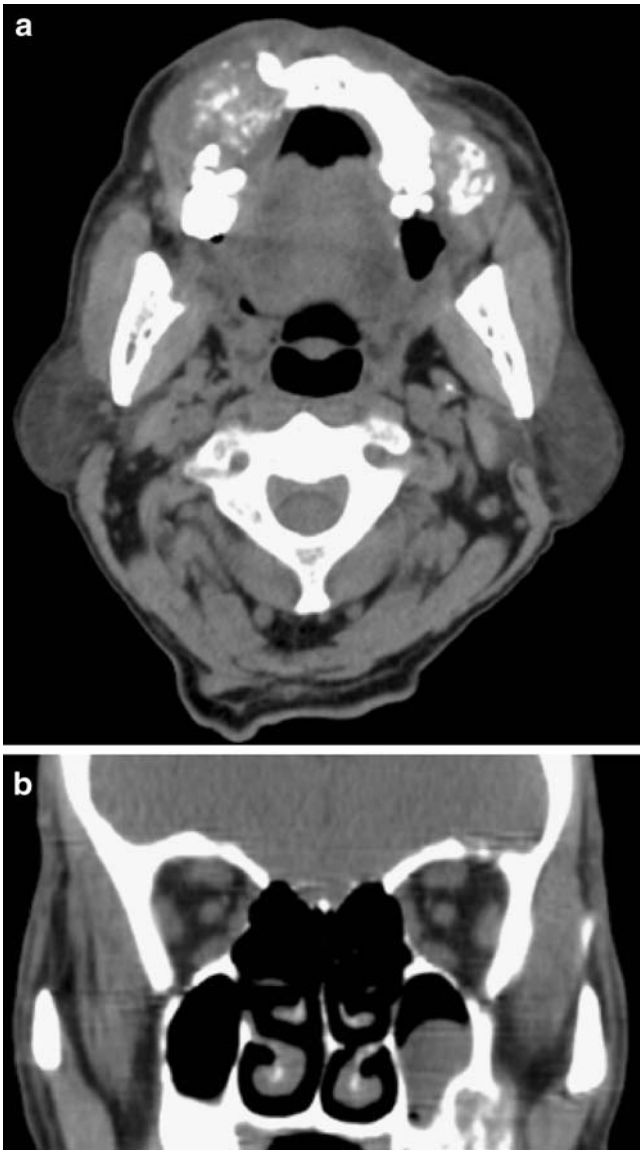

Fig. 3 **a** and **b** The CT scan shows two large expansile lucent lesions at the alveolar process of the right and left maxilla with multiple hyperdense foci s/o calcifications cortex is expanded outwards with mild expansion of lingual surface on right side. Cortex is extremely thinned out and not visualised at places. The lesions are extending superiorly within the floor of the ipsilateral maxillary sinuses

obstructive renal stones. He had an uneventful recovery with normalisation of serum calcium levels. There was noticeable shrinkage of the maxillary brown tumours within a fortnight of parathyroid surgery.

Discussion

Skeletal involvement in "classical" primary hyperparathyroidism reflects a striking increase in osteoclastic bone resorption and is accompanied by replacement of the marrow spaces with fibrovascular tissue and increase in

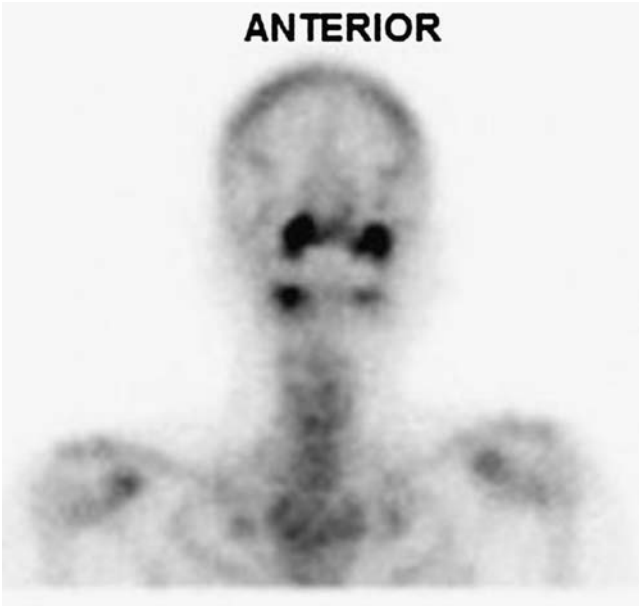

Fig. 4 Technetium-99 bone scan revealed increased uptake in both maxillae suggestive of an inflammatory pathology

new bone formation by osteoblasts. The initial manifestation of generalised demineralisation is later followed by subperiosteal resorption, formation of cysts and osteoclastomas. Osteoclastomas or "brown tumours" are composed of multiple giant osteoclasts (giant cells) mixed with stromal cells and matrix. Cysts develop as a result of intraosseous bleeding and tissue degeneration. The cystic spaces are filled up by clusters of giant cells, haemosiderin-laden macrophages and plump fibroblasts. The presence of haemorrhage, haemosiderin and hypervascularity lead to the brownish colour, which is responsible for the name. These lesions are essentially cellular reparative processes and are non-neoplastic. The term "tumour" is a misnomer. Although alternative designations have been offered they are not widely accepted. These bony tumours are most commonly found in the trabecular portion of the long bones, pelvis, ribs and the jaw.

Although the mandible is a common site for the detection of brown tumours, rare cases of the involvement of the maxillary bone are reported [2, 9]. The present case represents the second report of the bilateral maxillary brown tumours in a patient with primary hyperparathyroidism [4]. Most patients with maxillary lesions present with hard, clearly visible and palpable masses of the upper jaw. Occasionally, the lesion is painful. Radiologically, mandibular lesions present as well-circumscribed osteolytic lesions. In the maxillae, they commonly present as space-occupying lesion in the sinus, as was seen in our patient (Fig. 2).

Histologically, the presence of the giant osteoclasts in the lesions along with plump fibroblasts lead to confusion of the tumour with other jaw lesions that contain giant cells like giant cell granuloma, aneurismal bone cysts or cherubism [6]. Histological features alone cannot establish a certain diagnosis of a brown tumour. However, a clinical history of more widespread skeletal involvement, pathological fractures and renal stones may suggest the presence of PHPT. The diagnosis is readily confirmed by establishing elevated serum calcium and parathyroid hormone levels. There is a familial form of hyperparathyroidism associated with jaw tumours in which the histology of the jaw tumour shows an ossifying fibroma. This can be readily distinguished from brown tumours on histological grounds [7].

The clinical value of recognising primary hyperparathyroidism with maxillary giant cell lesions is in the fact that many of the tumours may spontaneously regress once the primary parathyroid lesion is removed. The parathyroid lesion is most commonly an adenoma. A single case report of an atypical parathyroid adenoma with a mandibular brown tumour has been reported [1]. After the successful removal of the offending parathyroid pathology, tumour regression and healing are expected in a majority of cases. Recurrence of the brown tumour can occur because of persistent or recurrent PHPT. There have been case reports of patients where the brown tumour continued to grow despite the successful removal of the tumour [5]. An occasional large brown tumour or persistent deformity may require operative intervention.

Conclusion

Brown tumours of the jaw may commonly involve the mandible and rarely involve the maxilla in association with

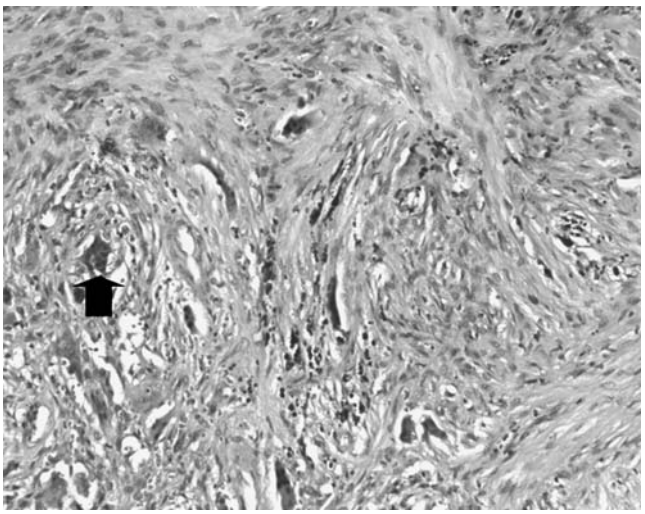

Fig. 5 ×40 magnification of haematoxylin–eosin stained section revealing multiple giant cells (*bold arrow*) with stoma-containing fibroblasts, blood vessels and areas of haemorrhage. A diagnosis of a giant cell reparative granuloma was made based on the basis of this combination

primary hyperparathyroidism. The presence of underlying primary hyperparathyroidism should be sought in all unexplained mandibular and maxillary lesions. A majority of these lesions may disappear with the removal of the parathyroid pathology.

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Conflicting interest None.

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