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RESEARCH ARTICLE

MAXILLARY BROWN TUMOUR: THE ONLY MANIFESTATION OF NORMOCALCEMIC PRIMARY HYPERPARATHYROIDISM (A CASE REPORT).

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ABSTRACT

Brown tumour is a rare complication of hyperparathyroidism, usually associated with high serum calcium levels. As the only and first symptom of normocalcaemic primary hyperparathyroidism (NPHP), it is an extremely rare occurrence. In the facial region, mandibular involvement is most common but maxillary involvement is exceptional. Their treatment is generally conservative and is based on the normalisation of calcium, phosphorus and parathormone levels. NPHP is becoming an increasingly emerging entity due to the frequent testing of different parameters of the phosphocalcic balance. Before making the diagnosis of NPHP, other etiologies of secondary hyperparathyroidism should be ruled out. According to international guidelines, NPHP will be managed in the same way as asymptomatic hyperparathyroidism. We report a rare case of a patient with a maxillary tumour whose investigations concluded to be a brown tumour secondary to NPHP.

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INTRODUCTION

The brown tumour is a pathology that has become rare, often associated with hypercalcaemia, rarely with normocalcaemia (1). only about ten observations of primary hyperparathyroidism revealed by a maxillofacial tumour have been published (2). Objective of the work: to highlight the value of parathyroid hormone testing in the presence of any osteolytic lesions and review of the literature on NPHP.

OBSERVATION

A 52 year old female patient, with no previous history, consulted for a painful left jugal swelling that had been evolving for a year and was progressively increasing in volume, making mastication difficult.

A CT scan showed a benign-looking tissue lesion centred on the left geno-labial soft tissues and lysing the alveolar ridge opposite teeth 23, 24 and the lower wall of the homolateral maxillary sinus, which could suggest an ondotogenic tumour or a cemento-ossifying fibroma (Figure 1,2). The tumour was removed and the anatomopathological study with immunohistochemical complement was in favour of a brown tumour (Figure 3,4). The phosphocalcic assessment showed normal blood calcium levels on several occasions, phosphorus levels at the lower limit of 25 mg/l, and parathyroid hormone levels of 345 pg/ml (5xN). The diagnosis of NPHP was made after exclusion of all causes of secondary hyperparathyroidism. The localization workup did not find a parathyroid adenoma and the patient was scheduled for exploratory surgery.

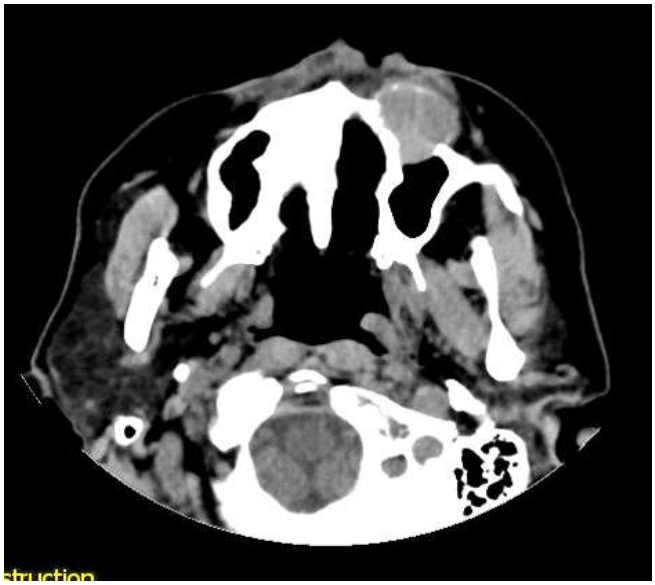

Figure N1: CT scan show a benign-looking tissue lesion centred on the left geno-labial soft tissues and lysing the alveolar ridge.

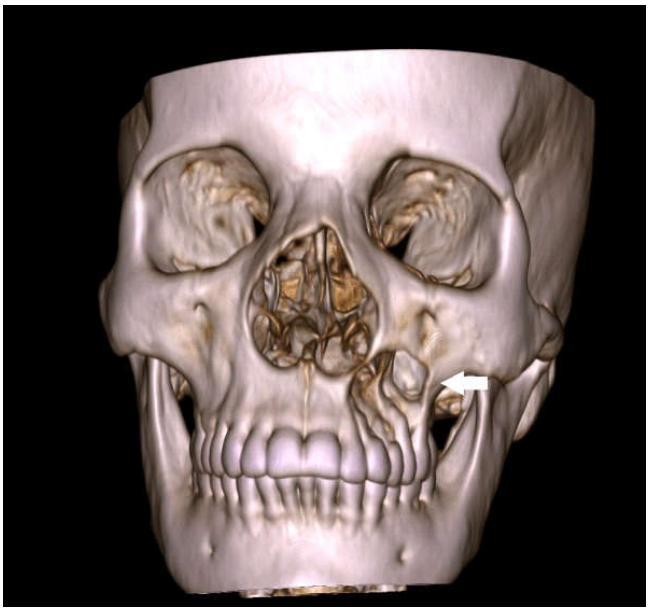

Figure N2: 3D CT image of the lesion.

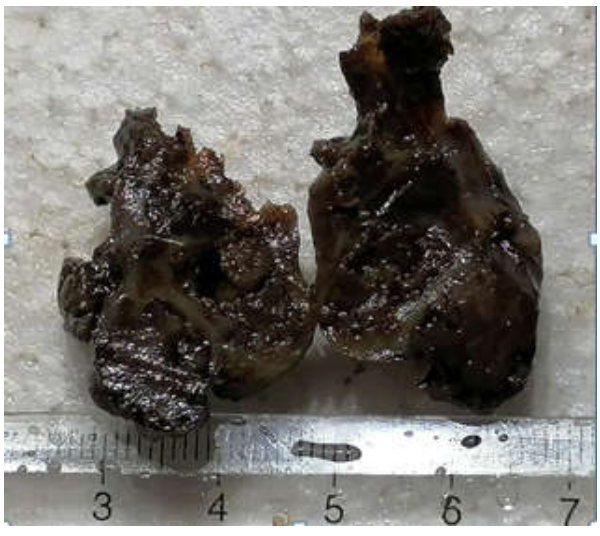

Figure N3: macroscopic image of the operative specimen.

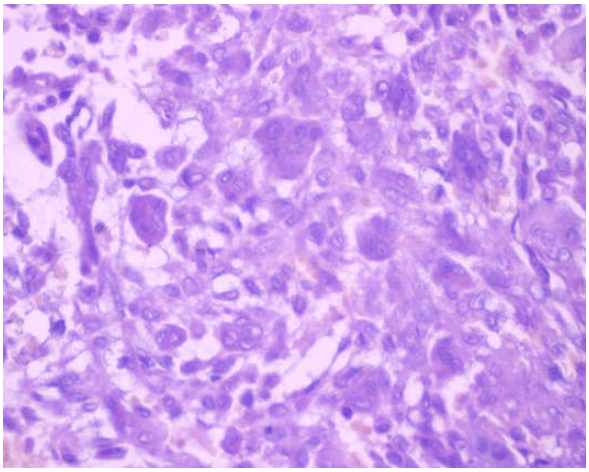

Figure N4. Morphological appearance showing Gx40 giant cell tumour proliferation

DISCUSSION

Brown tumour is a rare clinical entity exceptionally complicating NPHP. The term brown tumour is related to the macroscopic appearance of the tumour, which results from deposits of haemosiderin-laden blood pigments (3). It usually presents as a slowly enlarging, painful mass that may be locally aggressive but without metastatic potential. In the facial region, mandibular involvement is most common, maxillary involvement is exceptional, only about ten observations of primary hyperparathyroidism revealed by a maxillofacial tumour have been published (2). The treatment of these tumours depends on the extent of the tumour mass and the associated functional problems. Close control of blood levels of: calcium, phosphorus, 1,25-dihydroxy Vitamin D and parathyroid hormone is often sufficient to control and mitigate the growth of the tumour mass. Removal of the hyperfunctioning parathyroid(s) leads to regression of the brown tumours in many cases. Surgical removal of tumour masses may be necessary for patients who do not respond satisfactorily to conservative treatment, and who present aesthetic and functional complaints (4).

NPHP is an increasingly emerging entity, due to frequent testing of calcium and phosphorus parameters. The pathophysiology of this entity is not yet elucidated. To date, several hypotheses have been proposed: that NPHP is an early form of classical primary hyperparathyroidism and that in some patients the hypercalcaemia becomes frank with time. The second hypothesis is a possible partial tissue resistance of the bone and kidney to the action of PTH. The third hypothesis is probably related to the normal range of serum calcium considered by some authors to be very narrow (5). Diagnostic criteria for NPHP: should include a consistently normal total serum calcium (adjusted for albumin) and ionised calcium level. In addition, causes of secondary hyperparathyroidism should be excluded: vitamin D deficiency, decreased creatinine clearance, certain drugs (Hydrochlorothiazide, Lithium), hypercalciuria as a primary renal abnormality and gastrointestinal disorders associated with calcium malabsorption (6). There are no data regarding the management strategy for normocalcaemic patients. The Fourth International Workshop guidelines consider NPHP an entity of asymptomatic primary hyperparathyroidism (7). Parathyroidectomy will be offered to patients with bone or

renal complications. Available data suggest that patients with NPHP develop renal and bone complications to a similar or greater extent than hypercalcaemic patients. Therefore, clinical, biological (total and/or ionised calcium, parathyroid hormone, phosphorus, 24-hour calciuria) and radiological (renal ultrasound, bone densitometry) monitoring is recommended every one to two years in patients without complications at the time of diagnosis. If the disease worsens or a complication develops, parathyroidectomy of one or more hyperfunctioning glands may be considered (8). Patients who develop frank hypercalcaemia during follow-up should be managed according to the guidelines for asymptomatic hyperparathyroidism (7,8). Preoperative localisation is an important element of successful parathyroid surgery. Although data are limited for patients with NPHP, it appears that localisation tests are less likely to locate a parathyroid lesion. A study by Cunha-Bezerra showed that the sensitivity of imaging studies is lower for normocalcaemic patients compared to hypercalcaemic patients (9).

CONCLUSION

Normocalcaemic primary hyperparathyroidism is an entity of interest. The multiplicity of presumed pathophysiological mechanisms underlines the need for further research in this field in order to standardise both diagnostic criteria and therapeutic management. Our observation underlines the rarity of the localization of the brown tumor and the possibility of occurrence of these even in normocalcemia, hence the necessity of parathyroid hormone assay in front of the discovery of a brown tumor whatever the serum calcium value.

Conflict of interest statement: The authors do not have any financial, real or perceived conflict of interest in the publication of this manuscript.

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