CASE REPORT

MULTIPLE MAXILLO-MANDIBULAR BROWN TUMORS OF TERTIARY HYPERPARATHYROIDISM

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ABSTRACT

Brown tumor, also known as fibrous-cystic osteitis, is rare and terminal stage bony complication of hyperparathyroidism (HPT). It represents a reparative cellular process rather than a real neoplastic lesion. Involvement of the maxillofacial region is rare and if occured, the mandible is the predominantly affected region. The maxillary involvement remains very rare. Radiological evaluation of the lesion should be made in cooperation with clinical history and the laboratory findings. Here, we present an extremelly rare case of multiple brown tumors localized in bilateral mandible and maxillary sinuses and associated with tertiary HPT in our 30 year old female patient with long standing end stage renal disease (ESRD).

Key words: Brown tumor, tertiary hyperparathyroidism, chronic renal failure

Introduction

Brown tumor, also known as fibrous-cystic osteitis, is a uni-or multifocal, late metabolic bony manifestation of hyperparathyroidism (HPT). It represents a reparative cellular process as a result of a local destructive phenomenon rather than a real neoplastic lesion. The increased level of parathyroid hormone (PTH) leads to imbalance between osteoblasts and osteoclasts, resulting in bone resorption predominating over the formation. These lesions are rarely seen in the skull and if occured, the mandible is the preferentially affected region. The involvement of maxillary bone and paranasal sinuses is very rare.[1,2] Here, we present an extremelly rare case of bilateral mandibular and maxillary sinus brown tumors associated with tertiary HPT in a 30 year old patient with long standing end stage renal disease (ESRD).

Case Presentation

A 30 year old female patient presented with slowly growing, painless swellings on both cheeks. She had been on hemodialysis for 10 years and her transplanted kidney was rejected 2 years ago. She had no malignancy history. On physical examination, large mass lesions were identified on both cheeks, more prominent on the left side. Her laboratory results revealed the following: increased levels of serum calcium (12 mg/dL), serum phosphate (8 mg/dL), alkaline phosphatase (360 IU/L) and PTH (1665 pg/mL). On ultrasonograpy (US), heterogeneous, hypoechoic solid lesions with multiple foci of calcifications on both sides of the buccal region were revealed. In addition, a hypervascular hypoechoic lesion measuring 10 mm were detected posterior and extracapsular to the lower pole of the left thyroid lobe, suggesting a parathyroid gland adenoma which was confirmed by parathyroid scintigraphy. Maxillofacial magnetic resonance imaging (MRI) revealed that the buccal lesions were located within bilateral mandibular bones and maxillary sinuses with intermediate to high signal intensity on T2w images and low signal intensity on T1w images and marked enhancement following

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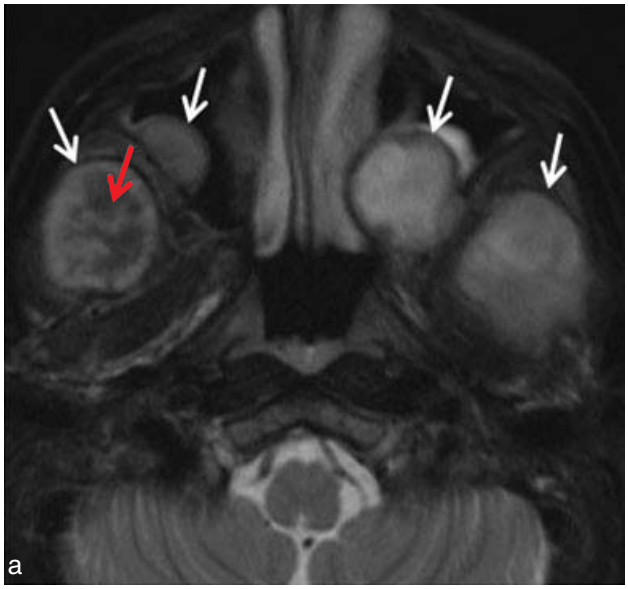
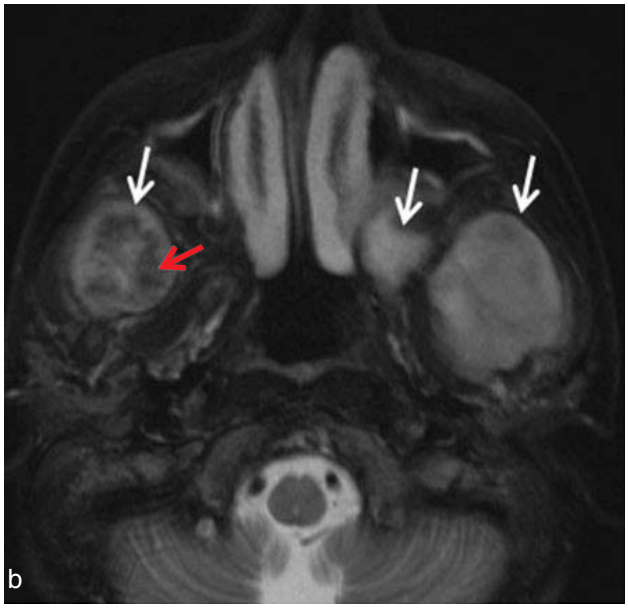
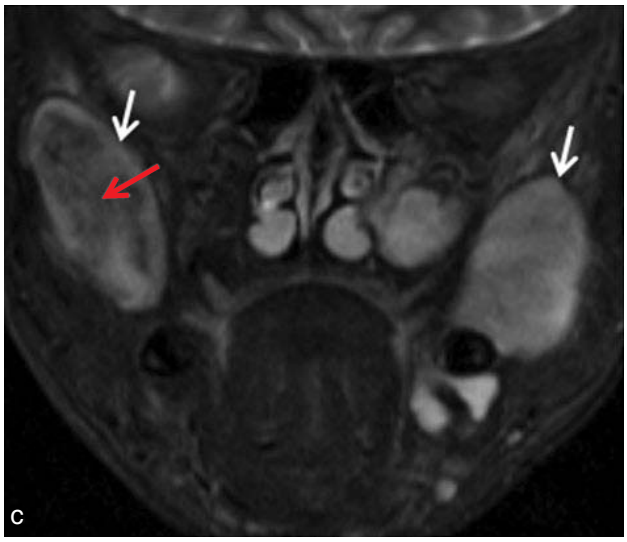
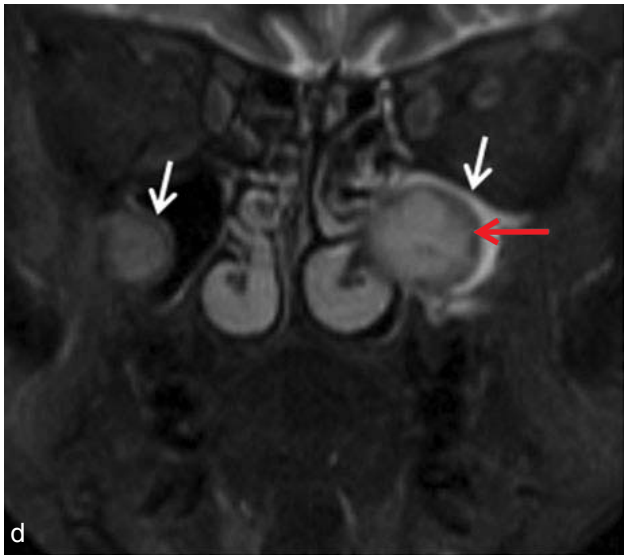
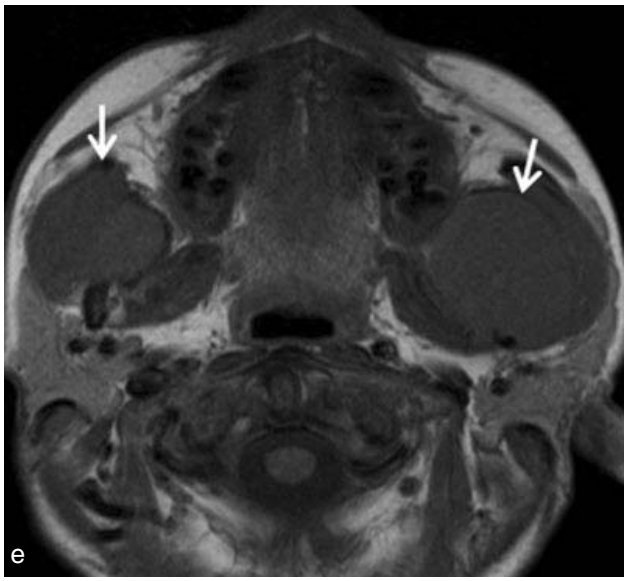
contrast administration. The largest lesion was the left mandibular lesion with the size of 4.5 x 4 x 3 cm (Fig.1). The lesions were well defined and hypodense on computed tomography (CT) scan. The left maxillary sinus lesion and bilateral mandibular lesions were expansile, lobulated with sclerotic peripheral rims and central components without bony destruction. The left maxillary lesion was extending through the nasal cavity and the pterygopalatine fossa. The right maxillary lesion was sitting on the lateral wall of the maxillary sinus and had lamellerappearence within the lesion. The mixed lytic and sclerotic nature of the











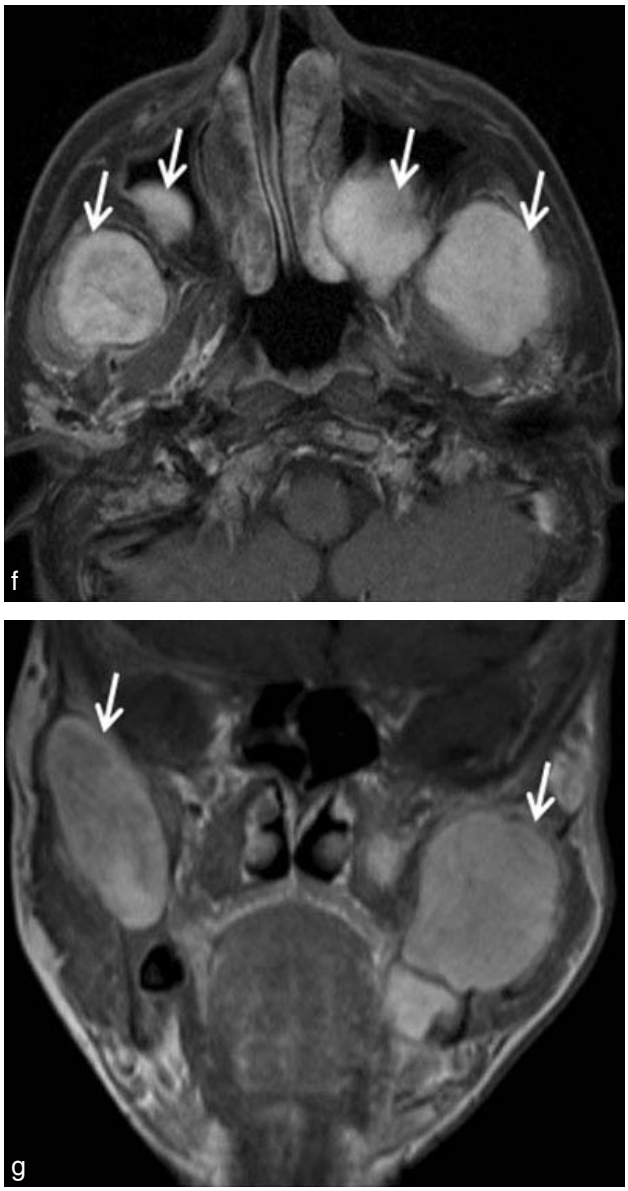
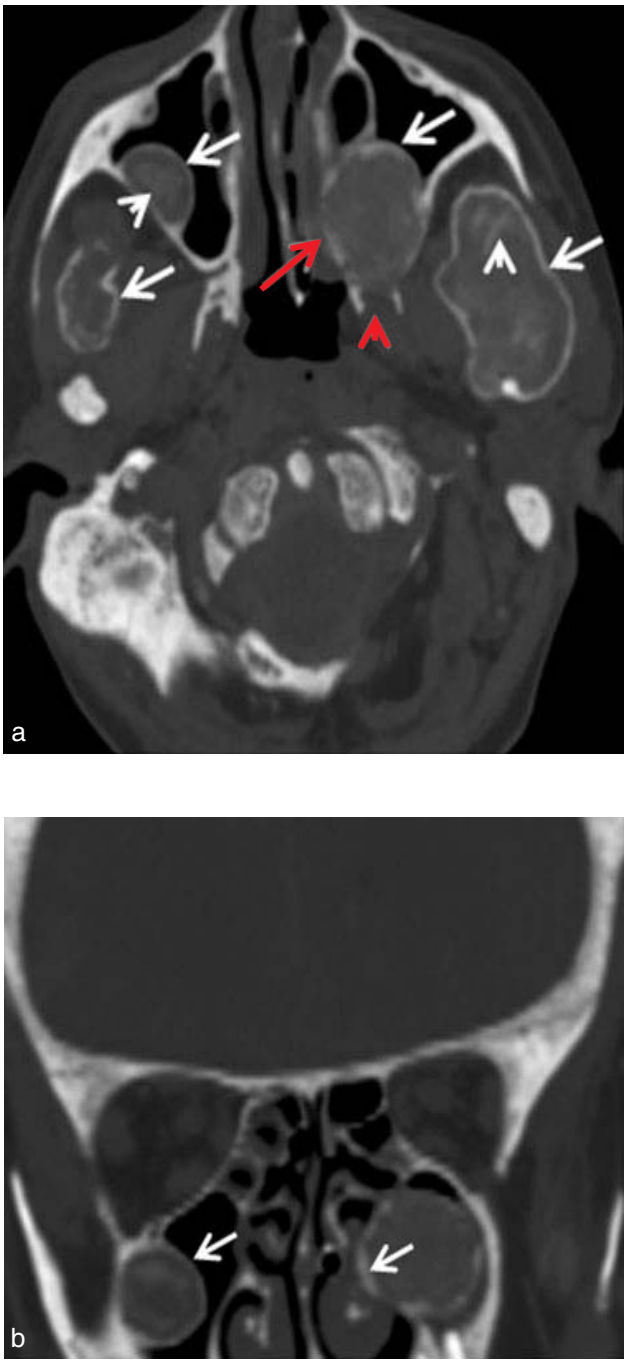


Figure 1: Axial **(a,b,e,f)** and coronal **(c,d,g)** IV contrast enhanced maxillofacial MRI shows bilateral maxillary and mandibular lesions which have heterogenous intermediate to high signal intensity on T2 w images (**a,b,c,d;** white arrows) and low signal intensity on T1 w image (**e,** white arrows). Note the low signal sclerotic components within (**a,b,c,** red arrows) and at the periphery of the lesions (**d,** red arrow). Following contrast administration marked enhancement of the lesions were demonstrated (**f,g,** white arrows)

lesions suggested the stages of bone resorption and formation occuring at the same time. There were no bony destruction or adjacent soft tissue involve-ment. The calvarium bones had a mottled appearance, the characteristic salt and pepper pattern of hyperpara-thyroidism (Fig. 2).

Together with these typical radiological findings and multifocality of the lesions, clinical history, laboratory results and scintigraphic verification of accompanying parathyroid adenoma, they were radiologically dignosed as brown tumors associated with tertiary HPT. Unfortunatelly, the patient died due to subarach-noid hemorrhage prior to planned operations for the maxillo-mandibular lesions and the parathyroid adenoma.

*informed consent has been taken from the patient.



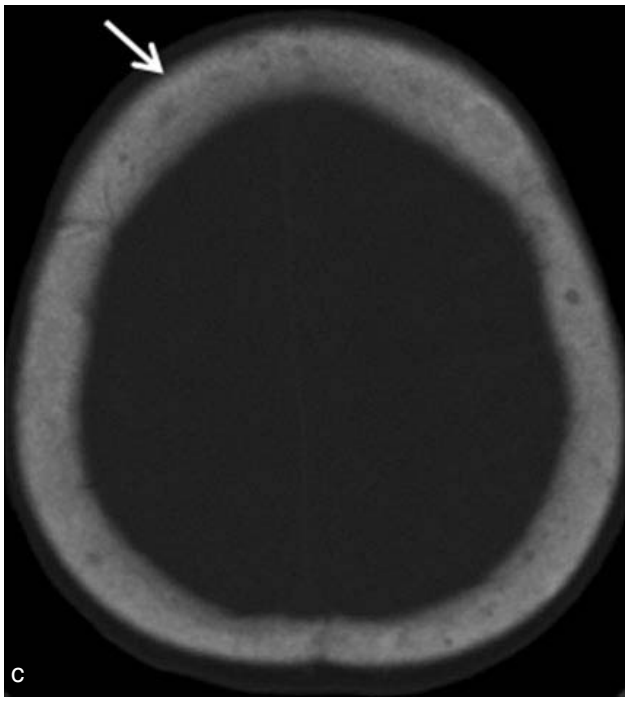


Figure 2: Axial **(a,c)** and coronal **(b)** maxillofacial CT images shows bilateral mandibular and maxillary lesions with radiolucent cystic appearance. Note the sclerotic periheral rims (**a,b,** white arrows) and central sclerotic lineer components (**a,** white arrow heads). The left maxillary sinus lesion extending through the nasal cavity (red arrow) and pterygopalatine fossa (red arrow head). The expansion of the left maxillary sinus and the left masticator space are visible. The mottled appearance of the calvarium is visible (**c,** white arrow).

Discussion

HP Trefers to excess PTH and its effects in the body. It can be primary, secondary, or tertiary. In primary HPT, the PTH secretion is increased due to the presence of adenoma or hyperplasia of one or more of the glands and rarely due to carcinoma. The secondary HPT occurs mostly as a complication of chronic renal failure (CRF) in which decrease in the serum calcium level due to continual and excessive urinary calcium excretion leads to increased PTH secretion which in turn results in rapid osteoclastic turnover and mobilization of skeletal calcium to maintain serum calcium level.[1] The other causes may be vitamin D deficiency or malnutrition.[3] Rarely, hyperplasia or adenoma can develop in one or more parathyroid glands as a result of chronic overstimulation in patients with longstanding renal insufficiency and secondary HPT, resulting in tertiary HPT. With the increased level of PTH, the increased bone catabolism results in characteristic imaging features, predominantly involving the skeletal system, ranging from cortical thinning (subperiosteal resorption), generalized osteopenia to bone resorption and brown tumors.[2,4] In localized regions where bone loss is particularly rapid, intraosseous bleeding, reparative granulation tissue and vascular, proliferating fibrous tissue replace the normal marrow contents with a characteristically expansile behaviour, resulting in a brown tumor.[5,6] Hemosiderin imparts the brown color. Incidence of brown tumour is about 3-4% and 1.5-1.7% in primary and secondary HPT, respectively with a slightly greater frequency in primary HPT.[7] However, since secondary HPT is much more common than primary HPT, most brown tumors are seen in associatiation with secondary HPT. The radiological features depend on the relative proportion of its components and the similar radiological features can also be present in many other lesions having cyst like appearance. They are usually well defined lesions with expansile behaviour which may be lytic or mixed lytic/sclerotic in appearance. There may be a little provoked reactive bone or the cortex may be thinned, but will not be penetrated. On CT scan, the lesions have cystic radiolucent appearance with attenuation values in the range of blood and fibrous tissue. On MRI, the lesions have low signal intensity on T1-w images and intermediated to high signal intensity on T2-w images with possible fluid-fluid levels. Following contrast administration they can show enhancement. Histopathological examination can not guarantee the exact tissue diagnosis and they can be easily misdiagnosed as giant cell tumor since both share similar macroscopical and microscopical features of osteoclastoma. In addition, aneurysmal bone cyst and cherubism also have similar features.[8] Multifocalty of the lesions which strongly suggests a systemic pathology is very guiding in the diagnosis. However, although very rare, brown tumors may be ill defined with associated bony destruction or adjacent soft tissue involvement.[9] In these situations, a malignant lesion should also be considered in the differential diagnosis. However, since bone scans can show intense uptake and may cause false positive results.[10] Therefore, for definitive diagnosis evaluation of the lesion should be made in cooperation with clinical history and the laboratory findings.

Conclusion

Brown tumors are rare and terminal stage bony complications of HPT and should be considered in the differential diagnosis of expansive cystic lesions of mandible and also the paranasal sinuses. Radiological imaging features along with the clinical and biochemical findings are very guiding in the diagnosis.

Conflict of Interest: Declared none by authors.

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