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WisdomGale

CASE REPORT

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Multifocal maxillomandibular brown tumors associated with primary hyperparathyroidism: Imaging findings and diagnostic challenges in a young woman

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SUMMARY

Brown tumors are rare benign osteolytic lesions resulting from excessive osteoclastic activity in the setting of hyperparathyroidism. They occur in approximately 1.5%–4.5% of patients with primary or secondary hyperparathyroidism and may occasionally represent the initial manifestation of the disease. We report the case of a 31-year-old woman who presented with progressive left facial swelling and mandibular deformity. Laboratory investigations revealed severe primary hyperparathyroidism with marked hypercalcemia and significantly elevated parathyroid hormone levels. Computed tomography demonstrated multifocal osteolytic lesions involving both the maxilla and mandible, associated with diffuse craniofacial demineralization displaying a characteristic "salt-and-pepper" appearance. Additional lesions were identified in the ribs and humerus, together with a right inferior parathyroid adenoma. The diagnosis of multifocal brown tumors secondary to primary hyperparathyroidism was established based on the combination of clinical, biochemical, and imaging findings. Surgical excision of the adenoma resulted in normalization of biochemical parameters and progressive regression of the mandibular lesion. This case is noteworthy because of the unusual coexistence of multiple maxillomandibular brown tumors with multifocal skeletal involvement in a young patient. It highlights the pivotal role of imaging in suggesting the diagnosis and preventing misinterpretation as metastatic bone disease or other giant-cell lesions.

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Introduction

Brown tumors are benign osteoclastic lesions that can affect various parts of the skeletal system. They are rare and occur in 1.5% to 4.5% of patients with primary or secondary hyperparathyroidism. Maxillomandibular involvement is relatively frequent in this rare condition. It is important to recognize them, as multiple lesions may be misdiagnosed as metastatic disease. They predominantly involve the facial bones, particularly the mandible, but may also affect the pelvis, ribs, and femur. We report an unusual case of multifocal brown tumors involving both the maxilla and mandible, associated with additional skeletal lesions and severe primary hyperparathyroidism in a young woman. This case underscores the importance of recognizing the characteristic imaging findings that may guide diagnosis and avoid unnecessary invasive procedures.

Case Presentation

A 31-year-old woman with no significant past medical history presented with a progressively enlarging left mandibular swelling, without associated inflammatory signs. Clinical examination revealed a mandibular mass with left facial deformity.

Laboratory investigations showed hypercalcemia (156 mg/l; normal: 88–104), hypophosphatemia (23 mg/l; normal: 26–45), corrected calcium of 151 mg/l, and markedly elevated parathyroid hormone (PTH 1–84) at 1,376.5 pg/ml (normal: 16.3–72.3).

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Facial computed tomography (CT) scan, performed before and after contrast administration, revealed a left mandibular osteolytic, multiloculated mass without periosteal reaction, displacing adjacent structures without evidence of invasion, measuring 73 × 60 × 54 mm (Fig. 1). Additional lesions with similar characteristics were noted in the right maxilla (Fig. 2), as well as in the ribs and humerus on lower sections (Fig. 3).

Diffuse bone involvement of the facial bones and cranial vault was also observed, characterized by bone demineralization with multiple small osteolytic lacunae, giving a "salt-and-pepper" appearance (Fig. 4).

CT imaging also revealed an oval, well-defined lesion adjacent to the inferior pole of the right thyroid lobe, with partially irregular margins, measuring 26 × 22 × 20 mm, consistent with a parathyroid adenoma (Fig. 5).

The diagnosis of brown tumors secondary to a right parathyroid adenoma was established based on clinical, biochemical, and radiological findings. Surgical excision of the adenoma was performed, resulting in subsequent normalization of PTH 1–84 levels and phosphocalcic parameters. The patient's clinical follow-up was marked by a reduction in the size of the mandibular swelling.

Discussion

Brown tumors result from hyperparathyroidism-induced bone remodeling mediated by PTH, in which normal bone tissue is replaced by fibrous tissue, hemorrhagic components, and hemosiderin deposits [1]. These histopathological features are relatively similar to those observed in other giant-cell tumors and fibrous dysplasia. Brown tumors are most often asymptomatic; however, depending on their size and location, they may present as swelling associated with facial deformity. Lesions may be solitary or multiple and can involve any bone [2]. They most commonly affect the pelvis, ribs, and femurs, although atypical locations such as the hand have also been reported [3]. Maxillomandibular involvement is less common, accounting for approximately 4.5% of cases [4].

The present case exhibits several uncommon features. First, the patient was relatively young at presentation. Second, simultaneous involvement of both the maxilla and mandible associated with additional lesions in the ribs and humerus is

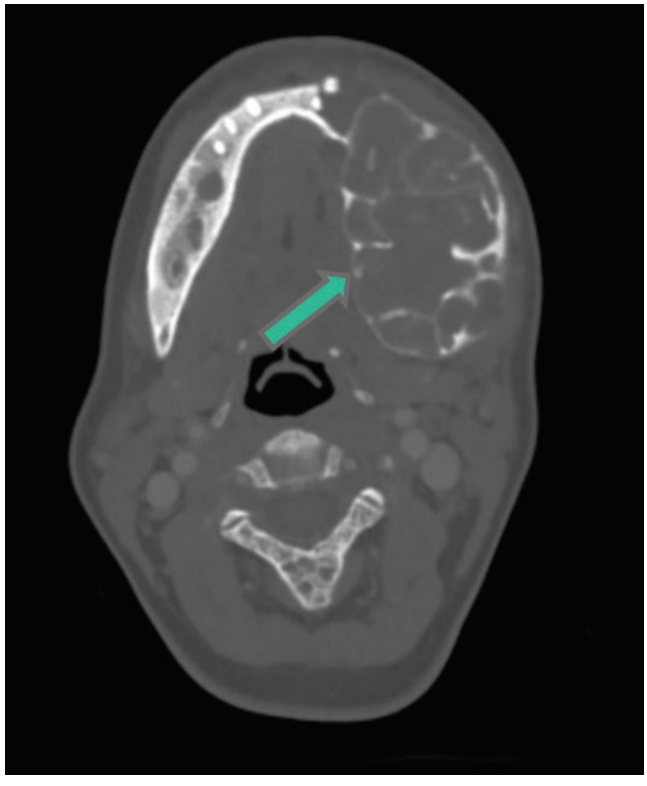


Figure 1. Axial facial CT scan demonstrating a left mandibular brown tumor.

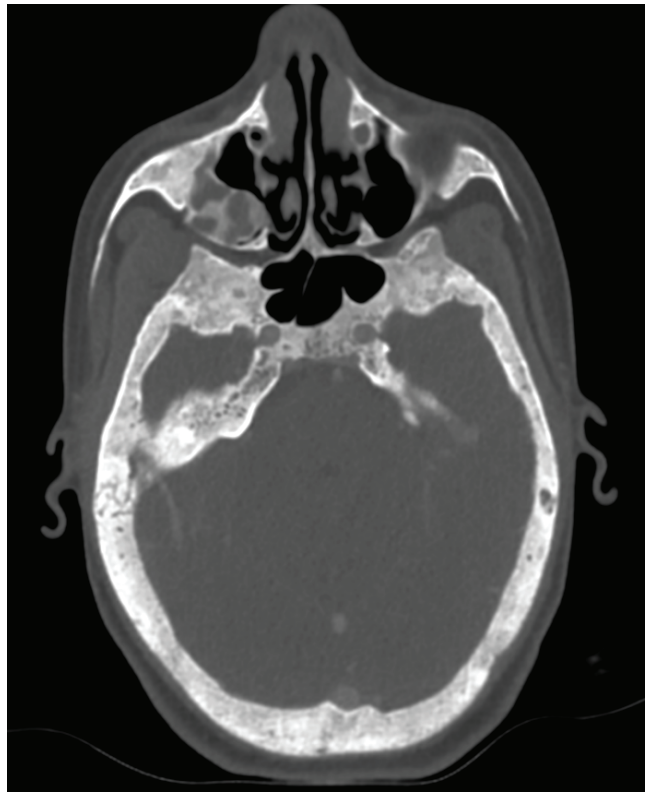


Figure 2. Axial facial CT scan demonstrating a right maxilla brown tumor.

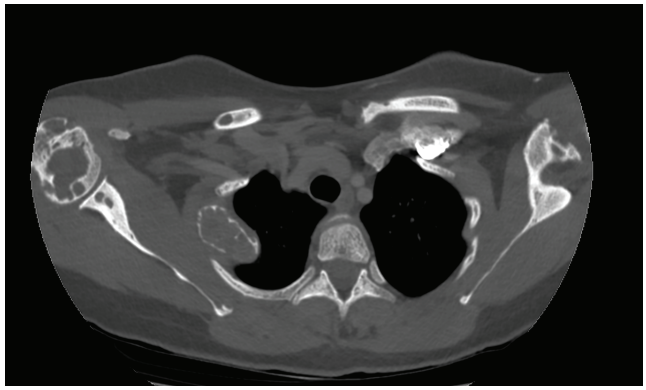


Figure 3. Axial CT scan demonstrating a brown tumor in the right rib and humerus.

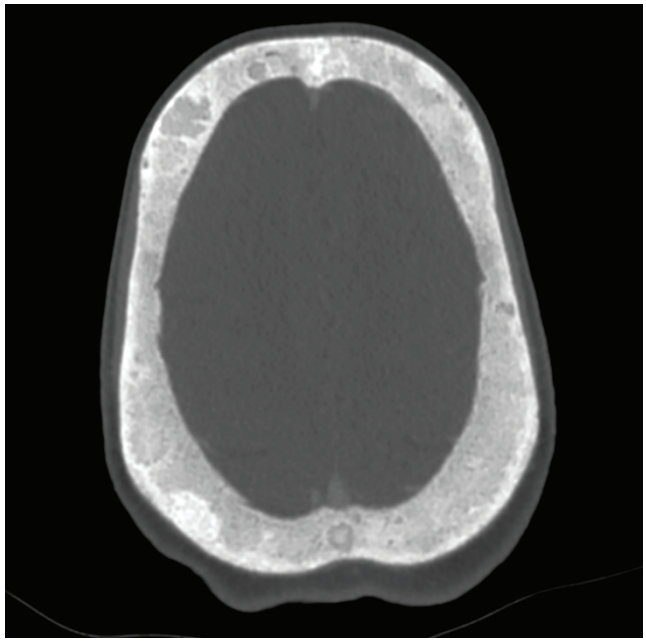


Figure 4. Axial CT scan demonstrating a small osteolytic lacuna, giving a "salt-and-pepper" appearance in the cranial vault.

uncommon. Third, the coexistence of multifocal brown tumors and diffuse skeletal manifestations of hyperparathyroidism, including the characteristic "salt-and-pepper" appearance of the skull, provided a comprehensive radiological depiction of advanced primary hyperparathyroidism. Such extensive skeletal involvement may closely mimic metastatic disease, multiple myeloma, or multifocal giant-cell lesions, making radiological recognition crucial for accurate diagnosis and appropriate management.

Biologically, patients typically present with disturbances in calcium–phosphate metabolism associated with elevated PTH levels. Radiologically, brown tumors appear as well-circumscribed osteolytic lesions, either unilocular or multilocular,

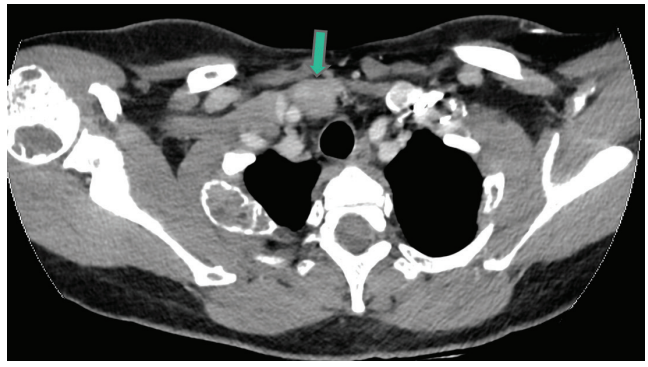


Figure 5. Axial CT scan demonstrating a right parathyroid adenoma.

with heterogeneous internal content. Within the craniofacial region, mandibular involvement is more frequent than maxillary localization [5]. Diffuse bone demineralization with a characteristic "ground-glass" appearance may also be observed [6]. Additional imaging findings related to primary hyperparathyroidism may include subperiosteal bone resorption, loss of the lamina dura surrounding the tooth roots, and generalized osteopenia [7]. CT is useful for evaluating extension into adjacent structures and for detecting concomitant brown tumors at other skeletal sites. CT also helps differentiate brown tumors from bone cysts, as the latter typically does not enhance after contrast administration, unlike brown tumors [4]. Magnetic resonance imaging (MRI) findings are non-specific and depend on lesion composition, which may be solid, cystic, or mixed solid-cystic [8]. Fluid–fluid levels may also be observed on MRI due to intratumoral hemorrhage. Bone scintigraphy is non-specific but may detect osseous brown tumors through focal increased uptake of technetium-99m methylene diphosphonate within the lesions [9]. It is also useful for assessing the extent of multifocal skeletal involvement [4]. In some cases, diffuse heterogeneous uptake throughout the skeleton may be observed secondary to metabolic bone disease associated with hyperparathyroidism. Technetium-99m sestamibi (99mTc-sestamibi) scintigraphy allows localization of hyperfunctioning parathyroid glands [10]. The differential diagnoses include bone metastases, multiple myeloma, chondromas, giant-cell tumors, and aneurysmal bone cysts. Elevated serum PTH levels and uptake of 99mTc-sestamibi within the lesions may suggest the diagnosis of brown tumors even before histopathological confirmation. This distinction is important because brown tumors are non-neoplastic lesions, unlike true giant-cell tumors, which carry a risk of malignant transformation and pulmonary metastases

[11]. Treatment depends on several factors, including etiology, extent, location, and symptomatology. In 80%–85% of cases, a solitary parathyroid adenoma is identified, and surgical excision generally leads to regression of the bone lesions [12]. In brown tumors secondary to secondary hyperparathyroidism, treatment is primarily medical and aims to reduce PTH levels and restore calcium–phosphate balance. Management is based on calcium carbonate supplementation, vitamin D therapy, and calcimimetics [13].

Conclusion

Brown tumors remain an uncommon manifestation of hyperparathyroidism and may occasionally represent the presenting feature of the disease. This case illustrates an unusual combination of multifocal maxillomandibular and extraskeletal involvement associated with a parathyroid adenoma in a young patient. Recognition of the characteristic imaging findings, together with biochemical abnormalities, is essential for establishing the diagnosis, avoiding confusion with malignant bone lesions, and guiding appropriate treatment. Early management of the underlying hyperparathyroidism can result in substantial regression of these lesions and favorable clinical outcomes.

Conflicts of interest

The authors declare no conflicts of interest.

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