Brown tumor of the facial bones: Case report and literature review

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Abstract
Brown tumor, an uncommon focal giant-cell lesion, arises as a direct result of the effect of parathyroid hormone on bone tissue in patients with hyperparathyroidism. The initial treatment involves the correction of hyperparathyroidism, which usually leads to tumor regression. We report a case of brown tumor of the right nasal fossa in a 71-year-old woman. The tumor had caused nasal obstruction and epistaxis. Laboratory evaluation revealed that the patient had primary hyperparathyroidism. Anatomicopathologic investigation revealed the presence of a giant-cell tumor. We performed a partial parathyroidectomy, but the tumor in the right nasal fossa failed to regress. One year later, we performed surgical resection of the lesion. The patient recovered uneventfully, and she remained asymptomatic and recurrence-free at the 1-year follow-up. Facial lesions with histologic features of a giant-cell tumor should be evaluated from a systemic standpoint. Hyperparathyroidism should always be investigated by laboratory tests because most affected patients are asymptomatic. Surgical resection of a brown tumor should be considered if the mass does not regress after correction of the inciting hyperparathyroidism or if the patient is highly symptomatic.

Introduction
Brown tumor, an uncommon focal giant-cell lesion, arises as a direct result of the effect of parathyroid hormone on bone tissue in some patients with hyperparathyroidism. Brown tumors can affect the mandible, maxilla, clavicle, ribs, and pelvic bones. Radiographic and histologic changes associated with the presence of this lesion may be very similar to those of other bone lesions of the face. Therefore, the diagnosis requires a systemic investigation for lesion differentiation.

We report a case of brown tumor in an elderly woman, and we discuss the clinical history, differential diagnosis, diagnosis, and treatment of this type of lesion.

Case report
A 71-year-old woman was referred to our otorhinolaryngology service for evaluation of a painful mass in the right nasal fossa that had developed 1 month earlier. The size of the mass had progressively increased and had eventually caused nasal obstruction and epistaxis. Six years earlier, the patient had presented with a nodule in the right mandibular region. At that time, she underwent an excisional biopsy and hemimandibulectomy, and the mass was diagnosed histologically as a giant-cell tumor. Until the development of the new mass, she had remained asymptomatic. She had not been evaluated for hyperparathyroidism during her previous episode.

Otorhinolaryngologic examination revealed that the mass had caused a septal deviation to the left. Oroscopy identified a bulge in the right upper gingival-labial sulcus. Computed tomography (CT) of the paranasal sinuses revealed a dense area in the right nasal fossa (figure 1). No bone erosion was evident. Biopsy of the mass revealed that it was a giant-cell tumor.

Laboratory evaluation revealed that the patient had primary hyperparathyroidism, as her parathyroid hormone level was 988 pg/ml (normal: 12 to 72). Other laboratory measurements were total serum calcium, 11.2 mg/dl (normal: 8.8 to 11.0); ionized calcium, 5.6 mg/dl (normal: 4.0 to 5.4); phosphorus, 2.0 mg/dl (normal: 2.5 to 4.8); and alkaline phosphatase, 145 U/L (normal: 32 to 104). Ultrasonography of the neck revealed an enlargement of the lower left parathyroid gland (2.1 × 1.4 × 0.8 cm). The finding of hyperparathyroidism confirmed the diagnosis of brown tumor.

To correct the hormonal imbalance, we performed a partial parathyroidectomy with a self-implantation of the right lower parathyroid into the prethyroid muscles. Histopathologic examination of the lower left parathyroid revealed the presence of an adenoma.

During the first year of postoperative follow-up, the mass...
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in the right nasal fossa failed to regress and the patient’s nasal symptoms persisted. Therefore, she underwent exeresis of the mass through a sublabial incision. Intraoperatively, the mass appeared as a hard, brownish-white, well-delimited, 2.5 × 1.2 × 1.1-cm lesion that extended into the upper portion of the hard palate. Curettage of the palatine bone was also performed.

Anatomicopathologic examination revealed that the fibrous lesion contained numerous giant cells, again confirming our diagnosis of brown tumor. The patient recovered uneventfully, and she remained asymptomatic and recurrence-free at the 1-year follow-up (figure 2).

Discussion
Brown tumor is an uncommon sequela of hyperparathyroidism. The lesion localizes in areas of intense bone resorption, and the bone defect becomes filled with fibroblastic tissue that can deform the bone and simulate a neoplastic process. These tumors have a brown or yellow hue.

Brown tumors arise secondary to both primary and secondary hyperparathyroidism. They have been reported to occur in 4.5% of patients with primary hyperparathyroidism and 1.5 to 1.7% of those with secondary disease. Hyperparathyroidism is frequently caused by the development of a parathyroid adenoma and less often by hyperplasia or a carcinoma. Some parathyroid adenomas and hyperplasias are familial (5% of cases), and others are part of multiple type I, IIa, and IIb endocrine neoplasias. The size of a parathyroid adenoma is correlated with the level of parathyroid hormone in blood. Anatomicopathologic study of the parathyroid in our patient revealed the presence of an adenoma.

Albright et al, in their classic description of primary hyperparathyroidism, reported that 2 of 17 patients (11.8%) had tumors involving the jaws. In a review of 220 patients, Rosenberg and Guralnick found that 10 patients (4.5%) had a mass involving one or both jaws as their presenting complaint. Brown tumors arise more frequently in the mandible than in the maxilla. Our patient originally had a brown tumor in the mandible and later in the right nasal fossa.

The reported prevalence of brown tumor is 0.1%. The disease can manifest at any age, but it is more common among persons older than 50 years. It is three times more common in women than in men. Our patient was 65 years old the first time she was diagnosed with a giant-cell tumor.

Most patients with hyperparathyroidism are asymptomatic, as was our patient. Hypercalcemia is often discovered incidentally during routine laboratory testing; hypophosphatemia and increased alkaline phosphatase levels in blood may also be seen. These metabolic changes were detected in our patient.

Parathyroid adenomas are usually small and almost never palpable in the neck. When clinical manifestations are present, the signs and symptoms can be divided into two types: urologic changes secondary to hypercalcemia and skeletal changes. Urologic changes include polyuria,
polydipsia, and the development of kidney stones. Again, hypercalcemia is usually asymptomatic, but in severe cases it may cause anorexia, vomiting, constipation, fatigue, weight loss, muscle weakness, psychiatric symptoms, and pancreatitis. Skeletal changes may represent the first manifestations of the disease, with a loss of cortical bone and an increase in trabecular bone. Bone pain and arthralgia are the most common symptoms. Bone demineralization is uncommon in mild hyperparathyroidism, but cystic fibrous osteitis is detected in some cases along with pathologic fractures and brown tumors. Radiologically, parathyroid adenomas may or may not exhibit well-defined margins; they may also cause cortical expansions associated with lytic lesions. Finally, patients may exhibit hyperparathyroidism-induced bone changes, such as medullary bone demineralization of the mandible. Brown tumors exhibit no pathognomonic histologic changes. Examination will reveal a dense fibroblastic stroma, areas of cystic degeneration, osteoid, microfractures, hemorrhage, macrophages with hemosiderin, and multinucleated osteoclastic giant cells. Hemorrhage and hemosiderin confer the lesion’s characteristic color. Similar changes may occur in fibrous dysplasia, true giant-cell tumors, and reparative granulomas. Differentiating between a brown tumor and other giant-cell tumors may be very difficult, even with histology.

Fibrous dysplasia affects the bones of the face, and it is most common among young women. Histology reveals trabecular bone with a stroma rich in fibrous tissue and multinucleated giant cells that are visible in areas of hemorrhage secondary to focal degeneration. True giant-cell tumors are more infiltrative than brown tumors. Their cause is unknown. They are rare, but when they do occur, they usually develop in older patients and they primarily involve the long bones. They also have a tendency to recur. Because of their infiltrative nature, true giant-cell tumors are difficult to remove surgically. Histologic analysis reveals giant cells around a fibrous stroma and some degree of cellular atypia. Reparative granulomas are localized tumors detected in young patients. They primarily involve the mandible. Their cause is still unknown, but some investigators believe that they are a result of trauma. They can be easily removed by curettage or local excision. A reparative granuloma can be differentiated from a brown tumor by the absence of hyperparathyroidism. Histologically, they contain giant cells, but their stroma is less dense and more vascularized. Therefore, patients with giant-cell tumors should be investigated for the presence of hyperparathyroidism and hypercalcemia in order to differentiate these granulomas from brown tumors. Our patient was tested for hyperparathyroidism and hypercalcemia only during her second episode.

The initial treatment of brown tumor involves control of hyperparathyroidism, regardless of whether it is primary or secondary. Treatment of primary hyperparathyroidism requires a parathyroidectomy. Once hyperparathyroidism is controlled, the tumor tends to regress, although in some cases surgical removal is necessary, especially for patients who have large, symptomatic tumors. Our patient required surgical removal. Brown tumor can recur if hyperparathyroidism persists or recurs.

References