Sphenoid sinus brown tumor, a mass lesion of occipital bone and hypercalcemia: An unusual presentation of primary hyperparathyroidism

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ABSTRACT. Brown tumor is a focal lesion of the bone caused by primary or, less commonly, secondary or tertiary hyperparathyroidism (HPT). While the mandible is the most frequently involved bone in the head and neck region, atypical involvement of the cranium in the area of the sphenoid sinus is exceedingly rare. In the literature, a unique case of brown tumor of the sphenoid sinus was reported in a patient with primary HPT. We present a case of sphenoid sinus and occipital bone brown tumor associated with primary HPT. A 47-year-old woman presented a 2-yr history of headaches, dizziness, diffuse body and articular pain, fatigue, and a 6-month history of intermittent nausea and vomiting, polydipsia, and polyuria. Magnetic resonance imaging (MRI) demonstrated an expansive mass lesion in the sphenoid sinus with erosion of the sellar floor and medial wall of the right orbit, and expansion in the medulla of bone. Examination of biopsy specimens obtained from sphenoid sinus mass confirmed the diagnosis of brown tumor. The biochemical laboratory studies showed elevation of parathyroid hormone and confirmed the diagnosis of primary HPT. Excision of a parathyroid adenoma affected the metabolic status into normalizing. At the follow-up of 12 months postoperatively, the size of sphenoid sinus brown tumor decreased and the mass of occipital bone disappeared. In conclusion, this is a first report of primary HPT masquerading as a destructive fibrous sphenoid sinus brown tumor associated with a mass lesion of occipital bone and hypercalcemia in the literature. (J. Endocrinol. Invest. 27: 366-369, 2004)
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INTRODUCTION

Brown tumor is a focal lesion representing the terminal stage of the bone remodeling process during primary and secondary hyperparathyroidism (HPT) (1, 2). Despite cases of brown tumors with HPT secondary to renal failure being increasingly reported in the literature, those associated with primary HPT have nearly disappeared in the last three decades because of early diagnosis and successful treatment of this form of HPT. Classical skeletal lesions which are bone resorption, bone cysts, brown tumors and generalized osteopenia, now occur in fewer than 5% of cases (3). The ribs, clavicles, pelvic girdle, and the mandible are the most often involved bones (4, 5). There are only a few publications that describe the unusual involvement of the facial bone, the orbits, maxillae and sphenoid sinus (2, 6-8). A unique case of brown tumor of the sphenoid sinus was reported in a patient with primary HPT in 1986 (8). This paper describes a clinically and histologically diagnosed case of brown tumor that was located in the sphenoid sinus and occipital bone.

CASE REPORT

A 47-yr-old woman came to the department of Neurosurgery with a 2-yr history of headaches, dizziness, diffuse body and articular pain, fatigue, and...
a 6-month history of intermittent nausea and vomiting, polydipsia, and polyuria. She had a history of arterial hypertension over the last 5 yr and had been treated with antihypertensive medication. The patient was hospitalized in the Neurosurgery clinic and transferred to Internal Medicine due to high serum calcium levels. On physical examination, blood pressure: 160/110 mmHg, arterial pulse: 88/min, rhythm. Laboratory values were as follows: Hb: 9.9 g/dl, Hct: 28%, white blood cell (WBC): 9700/mm³, blood urea nitrogen (BUN): 22 mg/dl, Cr: 2 mg/dl, total calcium: 16.5 mg/dl, p: 2.1 mg/dl, serum alkaline phosphatase (ALP): 766 U/l (N: 135-258), Cl/p: 52, serum albumin: 3.2 g/dl, intact parathyroid hormone (iPTH): 2415 pg/ml (N: 10-65). Urinary calcium: 486 mg/d, urinary p: 873 mg/d, tubular reabsorption of phosphate (TRP): 0.40 (N: 0.80-0.97). The levels of serum immunoglobulins, calcitonin, 25-OH-vitamin D (21 ng/ml), insulin, and the examination of bone marrow aspiration biopsy, hypophysial functions including GH and prolactin and 24-h urinary catecholamines and their metabolites were within the normal ranges. Plain radiographs demonstrated diffuse osteopenia, salt-and-pepper appearance (in the skull) (Fig. 1) and subperiosteal resorption of cortical bone. Total bone scan with 20 mCi technetium-99m-methylene diphosphonate (99m-Tc-MDP) showed prominent increased activity areas in cranium, lumbar vertebrae and femur (Fig. 2). Parathyroid ultrasonography (US) demonstrated a heterogenous solid mass measuring 26x18 mm, located in inferior-posterior of right thyroid lobe. A cervical magnetic resonance imaging (MRI) demonstrated a hyperintense mass in the same localization. Cranial MRI revealed an expansive lytic mass measuring 5x3.5x3.5 cm in the sphenoid sinus with erosion of the sella floor and the medial wall of the right orbit, as well as the posterior ethmoid sinuses, and an expansive mass lesion measuring 3x2x2 cm on the right occipital bone with expansion in medulla of bone (Fig. 3A).

The history and the physical, laboratory and radiological findings are suggestive of primary HPT presenting brown tumors in sphenoid sinus and on occipital bone. The patient was operated on and the right inferior parathyroid gland with adenoma measuring 3x2x1 cm was totally removed. Histopathological examination of the excised parathyroid gland revealed an adenoma. Microscopical examination of parathyroid adenoma revealed chief cells with round-oval uniform nucleus and large cytoplasm forming solid nests in a thin fibrovascular stroma (Fig. 4). Since the patient refused the operation, the mass in sphenoid sinus could not be removed. Biopsy specimens were obtained from this mass. Histopathologically the sphenoid sinus lesion consisted of groups of osteoclast type multinucleated giant cells in a vascularized and cellular fibrous stroma. There were hemorrhage areas and clusters of hemosiderin-laden macrophages. Reactive woven bone, which displayed osteoclastic activity, was seen in same areas (Fig. 5).

At the follow-up of 12 months postoperatively, the patients did not have any complaints; the iPTH was 70 pg/ml, serum calcium: 9.4 mg/dl, phosphorus: 3.0 mg/dl, and ALP: 241 U/l. The size of sphenoid

![Fig. 1 - Lateral skull radiography reveals “salt and pepper” appearance.](image1)

![Fig. 2 - Technetium-99m-methylene diphosphonate of skeleton. Increased activity in cranial bones, lumbar vertebrae and femur.](image2)