University Journal of Pre and Para Clinical Sciences

ISSN 2455–2879 2018, Vol.4(4)

SPHENOID SINUS BROWN TUMOR IN PRIMARY HYPERPARATHYROIDISM- A CASE REPORT

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Abstract : Brown tumors are expansile osteolytic lesions of bone, occurring in hyperparathyroidism. Skeletal involvement in primary hyperparathyroidism secondary to parathyroid adenoma is very uncommon and brown tumor has become extremely a rare clinical entity. Brown tumors occur most commonly in ribs, mandible, clavicle, and pelvis, and are uncommon in the facial bones. We report a rare case of primary hyperparathyroidism presenting with brown tumor in the sphenoid sinus and the relevant literature is reviewed.

Keyword : Brown tumor, primary hyperparathyroidism, paranasal sinuses

INTRODUCTION

Hyperparathyroidism is a clinical disorder characterized by increase in circulating concentration of parathyroid hormone (PTH). It is generally classified into primary, secondary, and tertiary depending on its etiology. Primary HPT is usually due to a single parathyroid adenoma .The most common presentation of primary HPT is hypercalcemia which is asymptomatic. Brown tumor is a benign reactive destructive process of bone that occurs in patients with either primary or secondary hyperparathyroidism (HPT). The skull base is composed of membranous bone and has little trabecular bone. Brown tumor usually occurs in the medullary shaft of long bones. Common locations for brown tumor include the mandible, maxilla, clavicle, metacarpal bones, ribs and pelvic bones and rarely the cranium and face. We report a case of primary hyperparathyroidism presenting with brown tumor in the sphenoid sinus and the relevant literature is reviewed.

CASE PRESENTATION

Our patient was a 23 years old male with complaints of head-ache- on and off for 3 years duration. A computed tomography (CT) scan of the brain revealed an expansile soft tissue density lesion with an attenuation value of 44.8 HU within the sphenoid sinus (figure 1). For further characterisation of the lesion MRI scan of the brain (plain & contrast) was done. MRI scan showed the lesion to be T1 isointense, T2 heterointense with blooming foci on susceptibility weighted images (SWI) and showing homogenous enhancement on contrast administration (figures 2, 3, 4 & 5). The lesion was seen causing expansion and remodelling of the sphenoid sinus. Clinical investigations revealed the following parameters · Serum calcium- 11.6 (N: 8.2 -10.6 mg/dL) · Serum phosphorus- 2.9 (N: 2.5-4.8 mg/dL) · ALP (alkaline phosphatise)- 1852 (N: 80-360 U/L)
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PTH(parathyroid hormone)- 1578 (N: 12-70 pg/mL) Ultrasonographic examination of the neck showed a well defined 2x 1.5 cm hyperechoic lesion in the posterior aspect of the left lobe of thyroid gland characteristic of a parathyroid adenoma (figures 6 & 7). Surgical excision and removal of the adenoma was done and histopathologically proven to be an adenoma.

DISCUSSION

Brown tumor represents localized accumulation of fibrous tis-sue and giant cells, which can replace bone and may even produce osseous expansion and subsequently undergo necrosis and liquefaction, producing cysts as a result of intraosseous bleeding and tissue degeneration (1,5,6). Brown tumors typically cause bone expansion rather than bone destruction, as they are the products of a benign, slow growing process. Brown tumors are due to the direct effect of parathyroid hormone on bone (2). In primary hyperparathyroidism, a parathyroid adenoma is the cause in 81 %, while other causes include hyperplasia (15%) and carcinoma (4%).These tumors are considered as the end-stage complication of hyperparathyroidism and are rarely the first presentation of the disease which is seen in our case. Involvement of the orbito-frontal region and the skull base is considered rare(4). In the skull base, they usually involve one or more of the paranasal sinuses.The most commonly involved sinus is maxillary sinus and the frontal sinus is the least commonly involved, with the sphenoidal and ethmoidal sinuses lying somewhere in between (3,5). The differential diagnoses of bone-expanding or lytic lesions in the skull base and paranasal sinuses include plasmacytoma, metastatic lesions, fibrous dysplasia, primary bone tumors, inflammatory lesions and giant cell lesions. Bone expanding giant cell lesions that can arise in the skull base include true giant-cell tumor, aneurysmal bone cyst, giant cell reparative granuloma and brown tumor (1). Radiographically, brown tumors are usually well-defined purely lytic unilocular or multilocular lesions that stimulate little reactive bone formation (8). The pathologically similar giant cell tumors often have indistinct borders and blend into the normal bone, a finding that may serve to distinguish them from brown tumors (6,8). However, occasionally a solitary brown tumor is the only skeletal manifestation of HPT. On CT scan, brown tumors appear as a lytic lesion with associated soft tissue mass.

The appearance on CT is not specific, and metastasis or myeloma may have a similar appearance (7). On MR imaging, brown tumors usually have hypointense signal on T1 weighted images, variable signal on T2 weighted images, and loss of signal on in-phase images because of magnetic susceptibility related to hemosiderin, osteoid matrix or air within the tissue (8). Brown tumors enhance intensely after contrast injection (9).The early and intense enhancement is due to rich vascularity When an osteolytic lesion of the skull base is diagnosed, especially when it is a giant cell tumor, even with a normal serum calcium level, PTH assay is necessary to exclude HPT. Treatment of brown tumor should start with treatment of HPT -removal of offending adenoma and if persisted after this primary treatment, enucleation and curettage should be added.

There is limited evidence about natural history of brown tumor in the skull base. When parathyroid adenoma is removed and consequently serum level of parathormone and calcium decrease, brown tumors rarely progress.

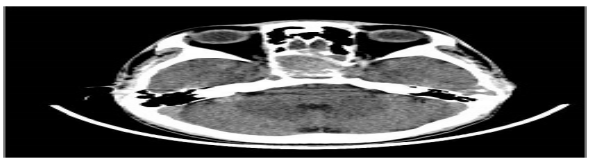


Fig 1

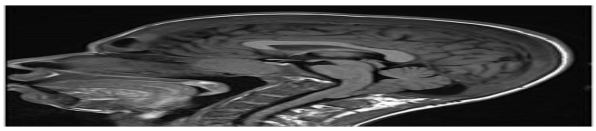


Fig 2

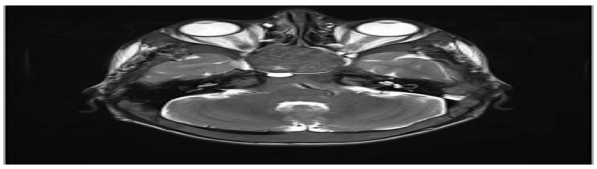


Fig 3

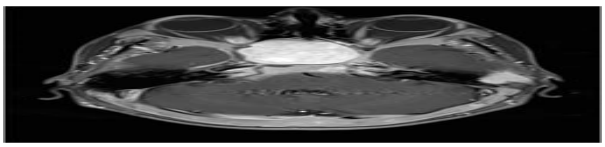


Fig 4

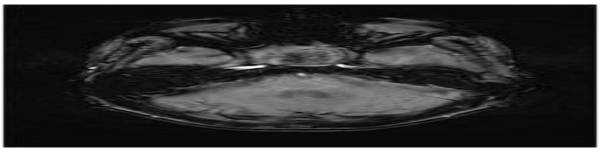


Fig 5

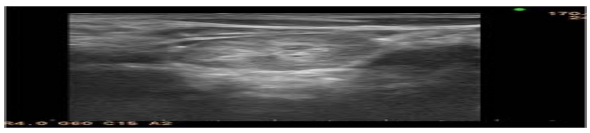
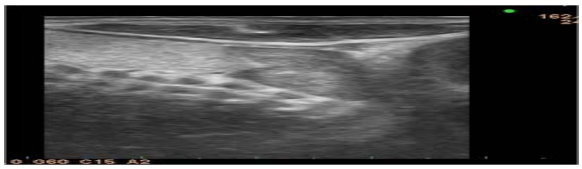


Fig 6

Fig 7



CT brain- expansile soft tissue lesion in sphenoid sinus
MRI TI weighted image- hypointense lesion in sphenoid sinus
MRI T2 weighted image- heterointense lesion in sphenoid sinus
Post contrast T1 weighted image- intense contrast enhancement of the lesion SWI – blooming foci within the lesion
USG neck- well defined hyperechoic lesion in the left lobe of thyroid posteriorly

CONCLUSION

In conclusion, brown tumors of the anterior skull base region are rare. They are extremely rare in primary HPT. The point in reporting this case is that they may be the first clinical sign of HPT. To prevent unnecessary surgical procedures, PTH assay is necessary in any patient with expansive-lytic lesion or giant cell tumor of the skull base.

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