**Case Report**

**Primary hyperparathyroidism with Brown’s Tumour-A case report**

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**Abstract**

Parathyroid adenoma is the commonest cause of primary hypercalcemia and usually presents with symptoms/signs of hypercalcemia. Brown tumours are rare focal giant-cell lesions that arise as a direct result of the effect of parathyroid hormone on bone tissue in some patients with hyperparathyroidism. Brown tumours can affect the mandible, maxilla, clavicle, ribs, and pelvic bones. Therefore, diagnosis requires a systemic investigation for lesion differentiation. In this case there was a 28 yr old woman who presented with solitary symptom of pain and swelling over left knee joint which was diagnosed as primary hyperparathyroidism with Brown’s tumour. Primary hyperparathyroidism is caused by a solitary adenoma in most of the cases and comparatively very less by glandular hyperplasia.

**Keywords:** Parathyroid adenoma, hypercalcemia, brown’s tumour

1. Introduction

Hyperparathyroidism results in disorders of bone and mineral metabolism. Diffuse and focal lesions may arise in multiple bones. The skeletal effects include massive bone resorption, bone fractures, and bone pain, as well as diffuse osteopenia, or circumscribed lytic lesions. On occasion, a patient with undiagnosed hyperparathyroidism presents with a lytic lesion that may be mistaken for a tumour. These lesions are termed "Brown’s Tumours" due to the presence of old haemorrhage in the lesion. Primary hyperparathyroidism is caused by a parathyroid adenoma, hyperplasia or carcinoma. This disease is found more often in women than men and its incidence increases with age. Primary hyperparathyroidism with bone disease has become rare in developed countries due to increased use of routine screening laboratory examinations. However, in less developed countries, patients may still present with primary hyperparathyroidism and advanced bone disease.

When primary hyperparathyroidism is associated with bone involvement it is termed a “Brown’s Tumour”, otherwise known as Osteitis Fibrosa Cystica. Clinically, hyperparathyroidism presents as "stones, bones and groans". The stones refer to recurrent kidney stones. Bones refer to the bone lesions that occur in severe or prolonged cases. Groans is meant to describe the gastrointestinal symptoms of nausea, vomiting, peptic ulcers and pancreatitis as well as the obtundation that occurs with hyper-calcemia. This condition is more common in females and incidence rises with age.

Radiologically, hyperparathyroidism presents as diffuse osteopenia as well as circumscribed lucent areas. Erosion of the tufts of the phalanges is an associated finding and is more pronounced on the radial aspect than the ulnar. Other characteristic areas of resorption include symphysis pubis, distal clavicle, vertebral bodies and lamina dura (bone at base of teeth). The calvaria may have a granular appearance called "salt and pepper" skull. Hyperparathyroidism has patchy increased activity on bone scan.

On investigating there is hypercalcemia due to increased calcium absorption in the gut, increased renal tubular resorption, and increased osteoclastic activity. Any of the Serum phosphate, alkaline phosphatase and urate levels may be elevated.

Treatment of hyperparathyroidism depends on the etiology of the condition. The tumours resolve once the abnormal metabolic condition is controlled. Most patients are treated before orthopaedic problems develop. In severe cases, bone fragility may require surgical stabilization. Primary hyperparathyroidism must be treated by surgical removal of the parathyroid neoplasm.

2. Case Report

A 28 years old female came with the complaints of pain and swelling over left knee joint for 6 months and unable to bare weight on left lower limb for last 2 weeks.

On physical examination of the patient, painful swelling was determined over the left knee joint which was firm to hard in consistency. On the right inferior thyroid lobe a 4 x 3 cm sized palpable nodule with soft consistency was noticed. Other systemic examinations revealed no abnormality.

2.1 Investigations

All routine blood investigations were within normal limits except serum calcium was 11.7 mg/dl (normal 8.4-10.7 mg/dl), serum alkaline phosphatase level was 200 IU/l (normal 50-240 IU/L), and serum parathyroid level was 1047 pg/ml (normal 8.7-79.6 pg/ml).

2.2 Radiological findings

2.2.1 X-ray of hip joint, bilateral knee joint and chest x-ray

There were multiple expansive osteolytic lesions eccentrically situated with sharp sclerosed margin with septations involving the left iliac bone, sacrum, lower ends of metaphyseal region of left femur and on the lateral end of right clavicle on. There was paper pot appearance seen involving the skull vault.
2.3 CT scan
CT scan examination showed the presence of a right cystic peripherally enhancing mass of size (coronal 4.6x2.5cms, axial 2.5x2.6 cms, saggital 5.0x2.3 cms.) which was thought to be compatible with parathyroid adenoma.

2.3 Operative notes
As planned, a right para-thyroidectomy was done in which the mass was taken out as a whole, which was clearly demarcated from the surrounding tissues by a pseudo-membrane. ORIF (open reduction and internal fixation) with condylar buttress plating with bone grafting in the left femur was done.

Histopathological report confirmed the diagnosis of parathyroid adenoma and bone biopsy revealed Brown’s tumour.
2.4 Macroscopic and microscopic findings

Macroscopic examination of the excised gland revealed greyish brown soft tissue mass measuring 5x3x2.5 cm and on cut section cystic area were identified from which thick brownish haemorrhagic material came out.

Microscopic examination showed the presence of oxyphilic cells and chief cells arranged in nests, follicular and pseudo-papillary patterns, separated by sinusoidal cells. Individual cells showed mild degree of pleomorphism, hyper-chromatism and crowding of nucleus.

2.5 Post operative status and follow up

Post operatively patient’s serum level of calcium was 8.6 mg/dl and serum level of parathyroid was 274pg/ml, patient was given oral calcium and vitamin D and above knee cast was given and advised bed rest for 3 months. We followed up the patient every monthly for 3 months and patient was now able to bare weight on left leg and there were no other complications post operatively till the last follow up.

3. Discussion

Although Brown tumour is generally seen more frequently in the seriously secondary hyperparathyroidism, it is fairly characteristic for primary hyperparathyroidism. Brown tumours may be observed on facial bones, pelvis, costa, manus bones and femur. In the hyperparathyroidism early findings are seen on hands. They may be multiple in the terminal stage of hyperparathyroidism or in the carcinomas. They may cause swelling, pathological fracture and bone pain.

Generally, high resolution USG, radionuclide imaging, CT and MRI combinations are used for imaging of parathyroid pathology. High resolution USG is one of the most common imaging methods and also the first imaging step in some private institutes used for neck evaluation. But the mainstay of confirmation of biochemical analysis remains the same. On USG, parathyroid adenoma is seen typically as round or oval homogenous, hypoecoic nodule localized behind the thyroid gland and at the lower aspect of paratracheal or paraoesophageal region. It is clearly separated from thyroid gland due to its capsule. Morphological differences such as hypercoic component, cystic changes and calcification may be seen particularly in large adenomas. More than 90% of parathyroid adenomas include intra-parenchymal hypervascular pattern in the colour flow imaging

In the radionuclide parathyroid imaging; Technetium (Tc) 99m marked 2-methoxyisobutyl- isonitrile (sestamibi) scintigraphy and 201-thallium and Tc 99m pertechnetat are predominant to subtraction imaging due to their short half time, giving more image quality and short half time, giving more image quality and scintigraphic findings is low in lesions that are smaller than 1 cm.

Thin section contrast CT is usable for localization of parathyroid adenoma. Its sensitivity range changes between 46-87%. One of the advantages of CT on USG is its ability to determine particularly ectopic parathyroid adenomas in the mediastinum.

Sensitivity of MR in the determination of parathyroid adenoma varies between 65-80%. Most common appearance of hyper functional parathyroid gland in T1 weighted images is isodens and increased intensity after intravenous gadolinium injection.

Brown tumour may cause various complications. Pathological fracture risk is higher in the brown tumours with hemorrhagic and/or cystic component in weight-bearing bones. MRI is important for determination of hemorrhage, cystic component and indirectly estimation of fracture risk in brown tumour.

Multiple Brown tumours and other multiple bone changes depending on primary hyperparathyroidism also may appear in the parathyroid adenomas like in our case. Modern imaging methods play an important role in the diagnosis of primary adenoma. Radiological findings in the skeletal system cannot differentiate parathyroid adenoma and parathyroid cancer. Determination of preoperative localization is important for security, surgical efficiency and particularly for invasive surgery. Even if USG is used for beginning evaluation, radionuclide imaging combination performed by usage of USG and Tc 99m-sestamibi is useful for determination of preoperative localization of parathyroid gland with hyper function. When parathyroid gland with hyper function. When doubtful results found in the USG and Tc 99m- sestamibi combination, CT and MRI may be practical. Radiological images are insufficient to differentiate hyperplasia, adenoma and carcinoma. Histopathology is necessary for definitive diagnosis.

References

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