

UBI EST Morbus-Where is the Disease?

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Abstract: Primary hyperparathyroidism is a disease characterized by excessive secretion of parathormone. During the course of this disease, bone loss occurs, particularly depending on resorption of the skeletal system. One of the complications of primary hyperparathyroidism is fibrotic, cystic bony changes which is called Brown tumor. Skeletal manifestations in the form of Brown tumors are rare and occur in less than 2% of patients suffering from any form of hyperparathyroidism. Such rare and multiple benign lesions may simulate a malignant neoplasm and pose a real challenge for the clinician in its differential diagnosis. We are presenting two such cases that were evaluated for multiple lytic expansile lesions with a strong suspicion of malignancy and fibrous dysplasia but turned out to be cases of primary hyperparathyroidism due to parathyroid adenoma

Keywords: bone pain, brown tumors, parathyroid adenoma, Parathyroidectomy

1. Introduction

Primary hyperparathyroidism is an uncommon disease, with a reported incidence of approximately 21 cases per 100,000 persons per year [1]. Solitary parathyroid adenoma accounts for approximately 85% of these cases. Less commonly, it may be caused by multiple adenomas or parathyroid hyperplasia [2]. The condition may be familial or sporadic and is characterized by Hypercalcemia with raised serum Parathyroid hormone.

In parathyroid adenomas along with hyper secretion, there is also an increase in urinary excretion of phosphorus and calcium and an increase in plasma calcium and alkaline phosphatase levels, which indicate osteoblastic activity.

One of the complications of primary hyperparathyroidism is fibrotic, cystic bony change which is called Brown tumor. Skeletal manifestations in the form of Brown tumors are rare and occur in less than 2% of patients suffering from any form of hyperparathyroidism. Such rare and multiple benign lesions may simulate a malignant neoplasm and pose a real challenge for the clinician in its differential diagnosis.

Very few cases have been reported in the English medical literature.[3-7]

We are presenting 2 cases of parathyroid adenoma with primary hyperparathyroidism presenting with lytic bone lesions.

Case 1

A 40 year old female patient of Asian origin presented to our casualty with sudden onset of swelling and pain in the left thigh after attempting to lift a heavy weight. An x-ray was done showing Fracture of the upper part of the shaft of the femur.

She had a past history of lower back pain radiating to the lower limb since two years even at rest relieved temporarily by analgesics. X-ray pelvis showed several lytic lesions and an MRI done 1 year later was suggestive of? Metastatic lesions of left sacral ala and bilateral iliac bones. The patient

underwent open reduction and internal fixation for the fracture femur.

With the above history a pathological fracture was suspected and surgery reference was given. Detailed History was taken. On physical examination the patient was found to have a spherical swelling of 1.5x1.5 cms at the lower part of the medial border of the right Sternocleidomastoid, with a smooth surface and firm consistency showing movement with deglutition.No other swellings were found in the neck

Differential diagnosis included-? Cervicallymphadenopathy? Solitary nodule Thyroid? Parathyroidadenoma. On investigation patient was found to have serum PTH levels of 731.8 pg/m(NI-11-54mg/dl)l and S. Calcium of 15.7mg/dl (NI 9-10.5 mg/dl) Serum Phosphate : 4.8 (NI : 2.5 - 4.5 mg/dL)Serum 25OH D3 : 12.6ng/dl (NI : 10 - 55 ng/mL),Urea: 17.0 mg/dlCreatinine: 0.6 mg/dl. Routine Blood parameters: Normal

USG neck showed a hypoechocic solid mass 18x14mm noted below the inferior pole of the right lobe of the thyroid with internal vascularity. Thyroid normal and other 3 parathyroids not visualized. CT neck showed a Homogenous enhancing mass 19x15x12mm noted below Right Lobe of thyroid suggestive of Parathyroid Adenoma, other 3 parathyroids were not visualized.Visualized bones show multiple osteolytic lesions.

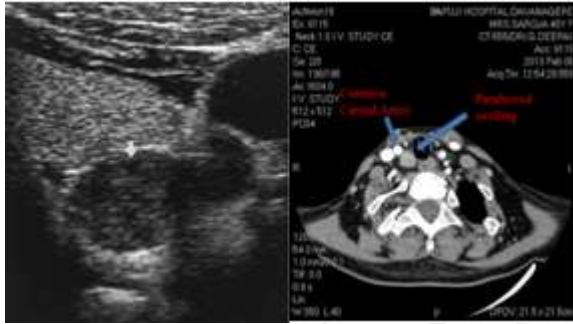
A final diagnosis of Right inferior parathyroid adenoma was made and the patient underwent Right Inferior Parathyroidectomy under general anesthesia. Postoperatively the S. Parathyroid hormone and S. Calcium were estimated 48hrs later and found to be 14pg/dl and 9.8mg/dl respectively. The drain was removed on the 2nd post-operative day.

Postoperative recovery was uneventful. Patient was discharged on the 4th postoperative day. Histopathological Examination revealed characteristic nesting pattern of chief cells confirming a parathyroid adenoma.

Case 1



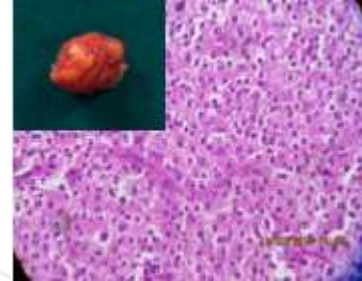
X-ray pelvis showing multiple lytic lesions.
 X-ray of the left femur.



USG and CT neck-hypoechoic mass



Parathyroid dissected



Wt. =30 Gms, Size= 2x1.8x1.2 Cms
 Nesting Pattern S/O Adenoma

Case 2

A 35 year old female patient was referred from the Dental College with C/o swelling on the right side of the lower jaw since 8 months. Swelling gradually increased, with no other associated complaints. X-ray of the jaw showed osteolytic lesion on the right side of the lower mandible

Patient was admitted under our surgery unit and evaluated. The patient underwent a CT scan of the neck showing a well defined soft tissue density enhancing lesion posterior to the left thyroid lobe at the level of C7 vertebra of 24x9mm. X-ray lumbosacral spine showed anterior marginal osteophytes from 11-13 with pencil thin cortices of the vertebral body suggestive of hyperparathyroidism

The serum PTH level were 649.3pg/ml, S.Calcium was 13.2 mg/dl and ALP was 500IU/L

Patient underwent Left Inferior Parathyroidectomy with frozen section of the swelling showing features suggestive of parathyroid adenoma. 48hrs post operatively the S.PTH was 3.4pg/ml, S calcium was 8.5 mg/ml and ALP was 415 U/L. Postoperative recovery was uneventful. Patient was discharged on the 4th post-operative day. Histopathological examination confirmed a parathyroid adenoma.



Skin Crease Incision, Along, Venous Line



Flaps Being Raised



Right Lobe of The Thyroid Retracted

Case 3



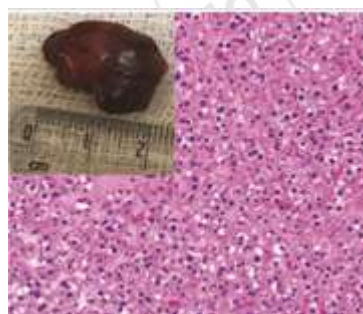
X-ray features of hyperparathyroidism



Skin incised and Lobes of the thyroid exposed



Thyroid and Parathyroid Exposed



Wt. = 20 gm,

2. Discussion

Primary hyperparathyroidism is characterized by the hyper functioning of parathyroid glands. The female: male ratio is 5:2. According to our literature reviews, multiple Brown tumor cases associated with primary hyperparathyroidism were initially reported by Joyce et al. in 1994. Very few cases have been observed since then. [3-7].

Imaging findings in hyperparathyroidism may include osteopenia, multiple osteolytic lesions from brown tumors, bone cysts, a salt-and-pepper appearance of the skull, and the characteristic radiographic findings of erosions and sub periosteal bone resorption most commonly involving the

radial aspect of the middle phalanges of the index and middle fingers.

Lytic lesions caused by hyperparathyroidism are called Brown tumors. The term "Brown tumor" is a misnomer because it is not a true neoplasm. Although Brown tumor is generally seen more frequently in the cases of serious secondary hyperparathyroidism, it is fairly characteristic for primary hyperparathyroidism. They consist of multinucleated osteoclasts (giant cells) and spindle-shaped stromal cells in a background of fibrous matrix and poorly mineralized woven bone. Their brown color is due to hemosiderin deposition. These tumors are known to regress after correction of hyperparathyroidism. Brown tumors may be observed in the facial bones, pelvis, ribs and femur. They may cause swelling, pathological fracture, and bone pain.

A diagnostic confusion arises mainly when the clinician encounters multiple lytic lesions involving different areas of the skeleton as was the case in our patient. Differential Diagnosis of Lytic bone lesions include:

- Skeletal Metastasis- 90% [6]
- Fibrous Dysplasia
- Hyperparathyroidism(brown tumours)
- Giant Cell Tumour
- Multiple Myeloma
- Chondroblastoma
- Hemangioma
- Osteomyelitis

3. Conclusion

In the case of Hypercalcemia and radiographic evidence of multiple lytic lesions, primary hyperparathyroidism should always be kept in differential diagnosis and should be looked into once more common causes such as malignancy have been excluded.

A combination of the clinical presentation, imaging findings, and the biochemical parameters of primary hyperthyroidism in relation to a parathyroid adenoma can lead to the accurate diagnosis of multiple skeletal brown tumors. Thus, the extensive work-up usually done for suspected bone metastases can be avoided. The above presented cases highlight the need to maintain a high degree of suspicion and suggest that surgery is curative in hyperparathyroidism due to parathyroid tumors.

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