Giant Cell Granuloma and Aneurysmal Bone Cyst like Rare Skeletal Manifestations of Hyperparathyroidism Single Center Experience in Bangladesh

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Annual Meeting of ARCCNM & KSNM

Background

Giant cell granuloma or brown tumor and aneurysmal bone cyst are rare skeletal manifestations of hyperparathyroidism with overall incidence of about 2-3%.

These lesions can be located in any part of the skeleton, but most frequently they are found in the maxillofacial regions, jaw, skull, ribs, clavicles, extremities, and rarely pelvic girdle; may be invasive in some cases, but lack of malignant potential; however frequently misdiagnosed as primary neoplastic bony lesion.

Objectives

This retrospective study was designed to assess the prevalence, clinical presentations, role of different imaging modalities and treatment outcome in hyperparathyroidism patients having giant cell like lesions or aneurysmal bone cyst.

Materials and Methods

In this retrospecpive study, a total 100 patients of hyperparathyroidism were enrolled who underwent 99mTc-MIBI SPECT-CT, from July 2019 to December 2021.

Results

Out of 100 patients, 4% had tumor-like bony lesions, primary vs. secendary hyperparathyroidism was 3:1. Three had histologically proven giant cell granuloma/ brown tumor involving maxillofacial regions with positive bony uptake on both 99mTc-MIBI SPECT-CT and 99mTc-MDP whole body bone scan. One had large aneurysmal bone cyst arising from sacral vertebrae with photopenic area on 99mTc-MIBI & 99mTc-MDP bone scan.

Only one had 99mTc-MIBI SPECT-CT positive parathyroid adenoma; further confirmed by postoperative histopathology. Two were false negative on MIBI parathyroid scan due to smallerer gland size and cystic degeneration of parathyroid adenoma on post operative histopathology respectively . Remaining patient of secondary hyperparathyroidism (SHPT) was true negative on parathyroid scan.

Spontaneous regression of brown tumor was seen in one case after parathyroid surgery without local curratage of brown tumor. Two patients need to underwent local curettage even after parathyroid surgery. Remaining case of SHPT was managed by biochemical correction of vitamin D deficiency and local curettage.

Representative Case (M/44 yrs)

Pain & mild swelling at left lower jaw for 2 months.

X-ray: ill defined radiolucent lesion at left mandible.

CT scan of Maxillo-facial region: multifocal expansile lytic lesions involving ramus & body of left mandible, right side of frontal bone with overlying soft tissue swelling. (Figure: 1).

Incisional biopsy revealed Irregular grayish-white pieces of soft tissue having osteoclast-like giant cells in a fibrovascular stroma –consistent with giant cell granuloma.

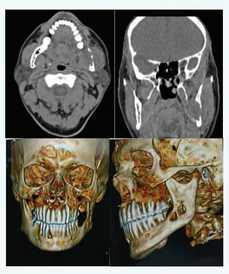
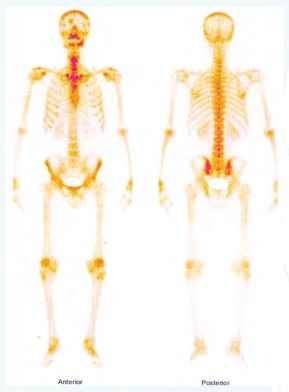


Figure 1: CT scan

Biochemical Parameters
Parathyroid Hormone (PTH): 2000 pg/mL (↑↑↑)
Serum Calcium: 11.8 mg/dL (↑)
Alkaline Phosphatase: 285 U/L (↑)
Serum Inorganic Phosphate: 1.7 mg/dL (↓)
Serum vitamin D level: 14.64 ng/mL
Serum Creatinine: 0.8 mg/dL (normal)

↓

Primary hyperparathyroidism (PHPT)



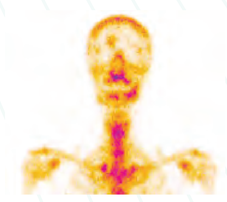


Figure 2: 99mTc-MDP whole body bone scan showing focal increased rediotracer uptake at left mandible & right frontal bone.

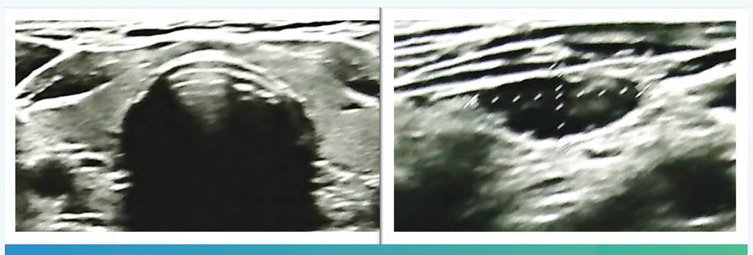


Figure 3: Suspected enlarged left lower parathyroid gland on neck ultrasound.

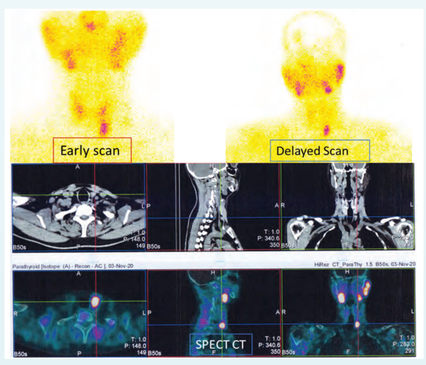


Figure 4:
99mTc-MIBI Parathyroid scan - early, delayed planar & delayed SPECT CT showing positive uptake for left lower parathyroid adenoma.

Patient underwent focused parathyroidectomy. Histopathology revealed parathyroid adenoma.

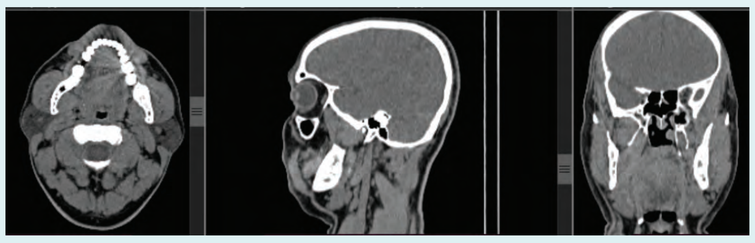


Figure 5: Follow-up CT scan of same patient one year after parathyroid surgery revealed complete regression of lytic lesion in left mandible with resorption of cortical bony destruction & remodeling of the bone by hyperdense sclerotic tissue.

Representative Case (M/50 yrs)

Slowly growing painless swelling at left maxilla for last one year.

Maxillo-facial CT scan: expansile lytic lesion having stipple calcifications invading the maxillary sinus (Fig:6).

Incisional biopsy: consistent with giant cell tumor.

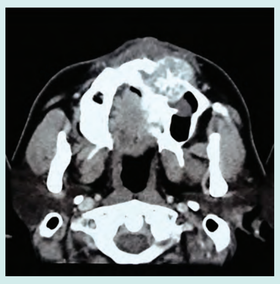
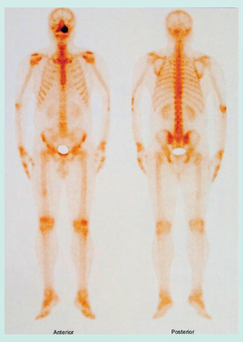


Figure: 6.



Biochemical Parameters
S. PTH level: 143.60 pg/mL (↑)
S. Calcium: 11.40 mg/dL (↑)
S. In. Phosphate: 2.40 mg/dL (↓)
ALP: 123.0 U/L (normal)
S. vitamin D level: 16.21 ng/mL (↓)
S. Creatinine: 1.2 mg/dL (normal)

↓

Primary hyperparathyroidism

Figure 7: 99mTc-MDP Bone scan: ↑radiotracer activity at left maxilla. Faint tracer activity at lower shaft of right femur which corresponds to old traumatic fracture.

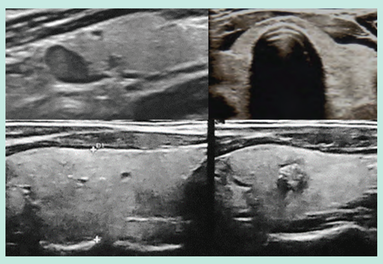


Figure 8:
Neck USG showing suspected left lower parathyroid gland
(5 x4 mm).
Tiny calcified nodule in right lobe.

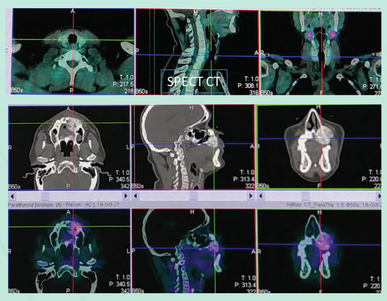


Figure 9 :
99mTc-MIBI Parathyroid SPECT CT was negative for parathyroid adenoma (upper row).
Focal increased radiotracer uptake at the bony lesion of left maxilla (lower row).

Patient underwent parathyroid surgery and local curettage of the lesion.

Representative Case (F/26 yrs)

Severe back pain radiating to right leg.

MRI L/S spine: a mixed signal intensity expansile lesion in presacral region arising from right sacral ala having multiple fluid-fluid level resembling aneurysmal bone cyst (ABC).

CT guided FNAC: fibroblastic tissue, no malignant cell.

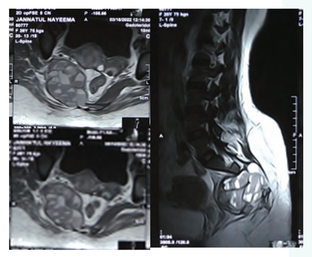


Figure 10: MRI L/S spine

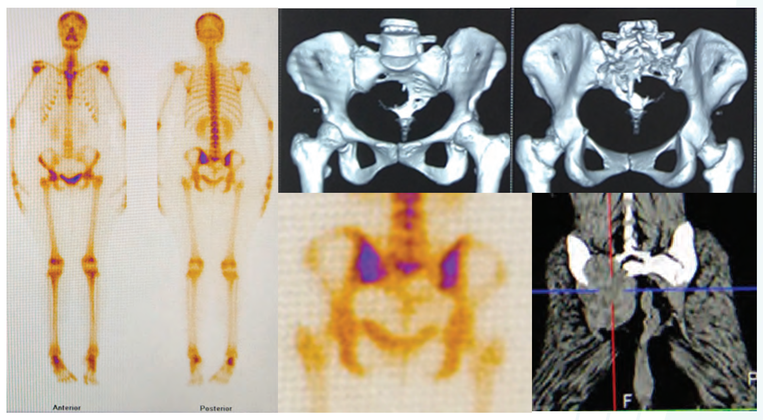


Figure 11: 99mTc-MDP whole body bone scan and spot image of pelvis showing photopenic area at right sacral ala; expansile osteolytic lesion with soft tissue mass in corresponding CT images.

However, biochemical parameter revealed PHPT.
Raised PTH (182.1 pg/ml, raised S. calcium level (12.9 mg/dl).

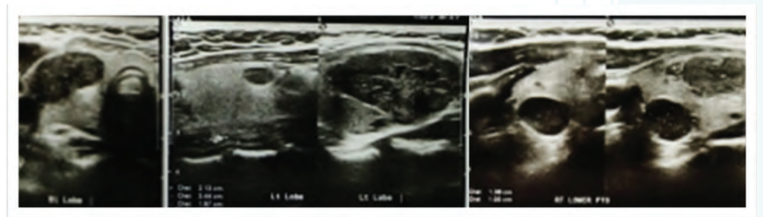


Figure 12: on neck USG, enlarged thyroid gland with multiple nodules; bilateral suspected parathyroid adenomas.

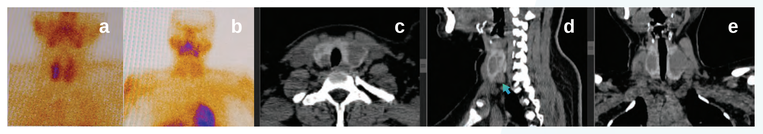


Figure 13: (a,b) 99mTc-MIBI Parathyroid scan was negative for parathyroid adenoma. (c-d) CT images showing thyroid nodules and suspected parathyroid adenoma (arrow).

Discussion

The skeletal changes in hyperparathyroidism usually triggered by diffuse demineralization in the early stage of the disease. Rarely it may become a lytic lesion similar to a Giant Cell Tumor (GCT); known as osteitis fibrosa cystica and brown tumor. Sometimes a focal absorption can produce a cyst like appearance on plain radiograph resembling a primary neoplasm of bone. Moreover, fibroblastic tissue may fill the defect that contour of the bone bulges, suggesting more strongly that a neoplasm is present. They are commonly misdiagnosed due to similar pattern of lucent bone tumors like aneurysmal bone cyst, GCT, myeloma, and metastatic carcinoma.

In most of the cases, definitive treatment option for these brown tumor or aneurysmal bone cyst includes excision of adenoma which facilitates normalization of PTH level and spontaneous regression of bony lesions. Additional surgery may be needed if there is slow regression of tumor or evidence of recurrence or fractures or extensive deformity.

Conclusion

Any bony lesion having raised PTH needs to be evaluated by systemic multidisciplinary approach involving physician, dentists, maxillofacial surgeon, endocrinologist, radiologist & nuclear medicine specialist to facilitates early diagnosis and proper management.

References

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