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Multifocal brown tumour associated with parathyroid adenoma

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Abstract

Brown tumors are focal lytic bone lesions associated with primary or secondary hyperparathyroidism. In the present era, brown tumors causing multiple lytic bone lesions are rare due to the early detection of hyperparathyroidism through biochemical investigations and effective treatment.

We report a case of a 30-year-old woman initially thought to have a bone tumor in her tibia, but further radiological and biochemical investigations revealed primary hyperparathyroidism with multiple brown tumors. This case emphasizes the importance of using imaging techniques to accurately diagnose and treat uncommon conditions like brown tumors in cases of primary hyperparathyroidism.

Keywords: hyperparathyroidism, parathyroid adenoma, parathyroid ultrasound, osteitis fibrosa cystica, sestamibi

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Introduction

The term brown tumor was coined due to its gross pathological appearance and is focal reactive osteolysis secondary to elevated parathyroid hormone that can cause bone pain or fractures.[1] Contrary to what the name implies, brown tumors are not indicative of a tumor growth; rather they result from bone restructuring due to hyperparathyroidism or paraneoplastic syndrome, often caused by a parathyroid adenoma.[2] The incidence of brown tumors is approximately 1.5-4.5% in primary or secondary hyperparathyroidism. Symptoms include weakness, weight loss, and recurrent stone formation. More common in women in their fifth to sixth decades, brown tumors are found incidentally on imaging scans in areas like the mandible, clavicle, ribs, and pelvis.[3,4] Hypercalcemia and hyperparathyroidism should also be assessed, as distinguishing between brown tumors and other giant cell-containing lesions can be challenging from a histologic standpoint.[3,4]

Case Report

A 30-year-old female presented at the orthopedic clinic complaining of persistent pain and swelling in her left knee following a fall injury. X-ray showed a multiloculated osteolytic lesion in her tibia with pathological fracture, Figure 1a. Magnetic Resonance Imaging (MRI) showed an expansile enhancing well-defined lobulated lesion involving the proximal tibial diaphysis, consisting of multi-cystic areas with a fluid level suggestive of blood and associated with cortical breach, Figure 1b.

A targeted biopsy revealed the presence of spindle-shaped mononuclear cells, as well as osteoclast-like giant cells and hemorrhage. Subsequent tests indicated elevated parathyroid hormone (PTH) levels. Based on suspicion of primary hyperparathyroidism, the patient was referred to the otorhinolaryngology department for further evaluation. During the physical examination, a firm, immovable lump measuring 3 cm was palpated in her neck. Ultrasonography (USG) of the patient's neck showed a well-defined, oval mass with a uniform hypoechoicity, measuring 2.5 x 1.8 cm in the lower pole of the left thyroid gland, Figure 1c. Contrast-enhanced Computed Tomography (CECT) scans of the neck and chest revealed a heterogeneously enhancing left paratracheal soft-tissue density mass lesion immediately caudal to the left lobe of the thyroid gland, Figure 1d.

Furthermore, 99mTc-MIBI parathyroid scintigraphy (Sestamibi) revealed tracer uptake just below the lower pole of the left thyroid lobe with tracer retention in delayed images, suggesting it was a parathyroid adenoma, Figure 1e. The lateral view of the skull radiograph showed multiple tiny osteolytic lesions resulting in a "pepper pot skull" appearance, Figure 2a. The patient's chest radiograph showed an expansile lytic/ sclerotic lesion in the lateral aspect of the left clavicle, Figure 2b. The hand radiograph displayed a lytic lesion in the medial aspect of the radial head, Figure 2c. The lytic lesions were also observed on the right side of the mandible. Abdominal USG revealed multiple bilateral nephrolithiasis, Figure 2d.

Following the radiological and biochemical findings, a diagnosis of primary hyperparathyroidism secondary to parathyroid neoplasm was made. The patient underwent a left-focused parathyroidectomy with removal of a red fleshy mass under general anesthesia, Figure 3a-b. Her pre-incision PTH was 3777 pg/ml, and her PTH level 20 minutes post removal was 467 pg/ml. Ionized serum calcium sent on the 1st postoperative day was 6.72 mg/dl, and 9.3 mg/dl on the 3rd postoperative day. Histopathology showed tumor cells composed of chief cells with round nuclei and slightly granular cytoplasm, and a thin fibrous capsule surrounding the tumor, confirming the diagnosis of parathyroid adenoma, Figure 3c-d. The postoperative period was uneventful, and the patient was subsequently discharged and is under regular follow-up.

Discussion

Osteitis fibrosa cystica, also known as Von Recklinghausen's disease of bone, is a rare condition accounting for only 4.5% of primary hyperparathyroidism (HPT) cases and 2% in secondary hyperparathyroidism.[1,2] The main cause of primary HPT is solitary parathyroid adenoma, accounting for 80% of cases. Symptoms of primary hyperthyroidism include bone pain, kidney stones, and gastrointestinal and neurological issues, known as "bones, stones, groans, and moans."[3]

Radiographically, brown tumors appear as distinct lytic lesions in the medullary region, with or without sclerotic borders. Progressive osteoclastic activity can lead to cortical bone destruction, pathological fractures, and soft-tissue components resembling metastatic disease. It is crucial to review basic radiographs, such as chest and pelvic radiographs, and a skeletal survey, before making a final diagnosis.

Contrast-enhanced Computed Tomography (CECT) is useful for defining lesions in complex bones like the pelvis and spine, while Magnetic Resonance Imaging (MRI) shows T1 and T2 iso to hypointense solid or mixed solid/cystic mass with fluid-fluid levels secondary to repeated hemorrhages and hemosiderin deposits. Enhancement is seen in solid components and septae after contrast administration.[4]

Adenomas can be accurately identified using ultrasonography (USG) as a distinct, hypoechoic lesion at the lower part of the thyroid gland. In cases of small adenomas, a thorough examination of the lower and upper poles is necessary, as they may not be visible. Adenoma presence and location can be confirmed with 99mTc-MIBI parathyroid scintigraphy (Sestamibi), delayed images of which show continuous

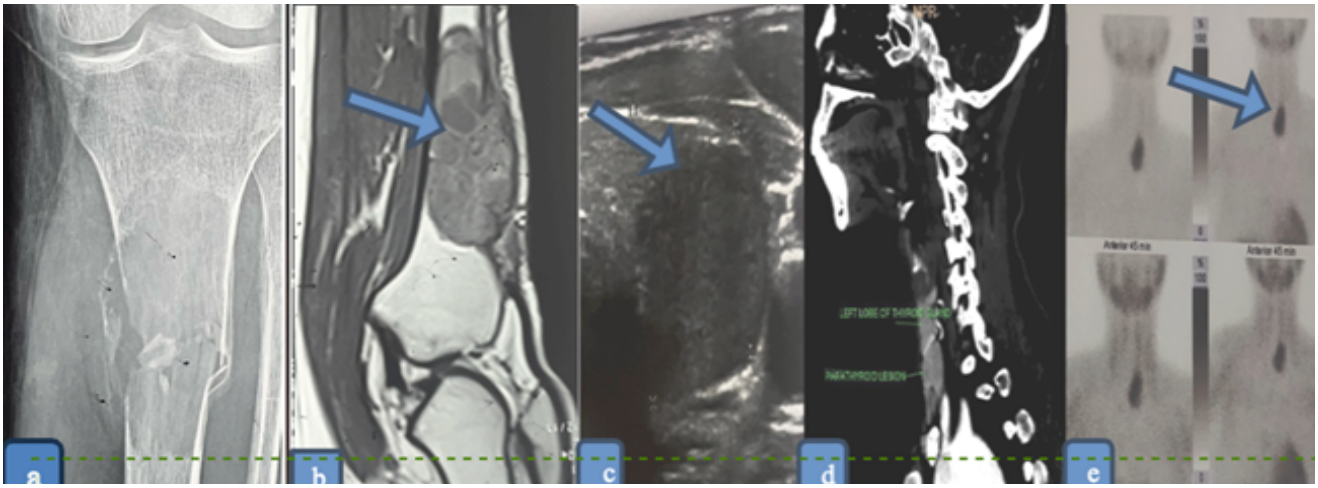

Figure 1. Expansile lytic lesion with pathological fracture of tibia (a). The lesion shows heterogeneous enhancement with enhancing septa (b). Ultrasonography of neck shows well defined homogenously hypoechoic lesion at inferior pole of thyroid gland (c). Sagittal (d) contrast-enhanced computerized tomography neck showing well-defined heterogeneously enhancing lesion inferior to left lobe of thyroid gland. 99 m Tc - MIBI Parathyroid Scintigraphy showing uptake of radiotracer at the inferior aspect of left thyroid lobe.

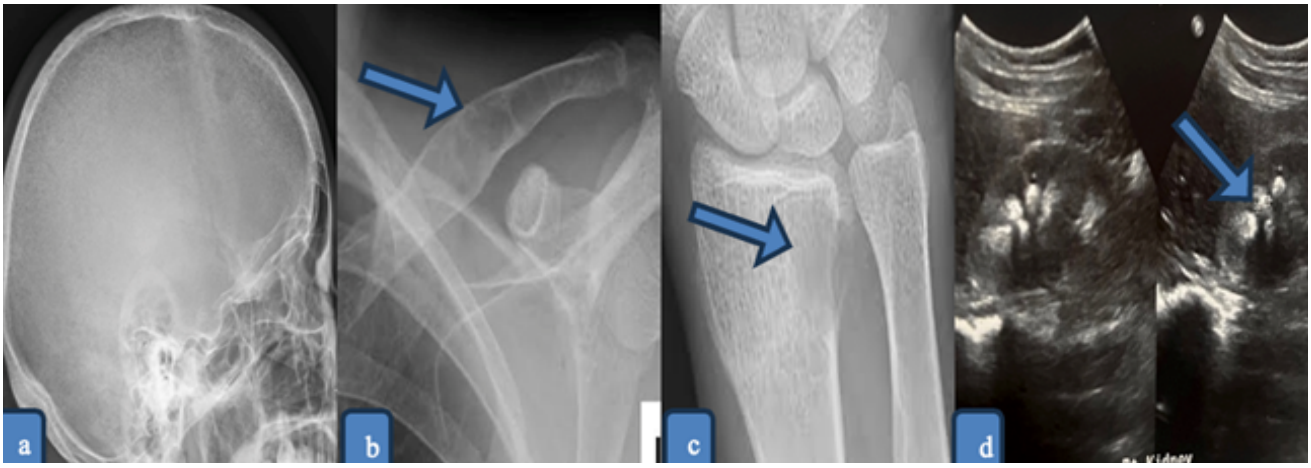

Figure 2. Skull radiograph lateral view(a) showing multiple tiny osteolytic lesions giving classical appearance of pepper pot skull. Expansile lytic lesion in left clavicle without bony erosion(b) Subcortical lytic lesion in medial aspect of radius(c) USG abdomen(d) showing bilateral nephrolithiasis.

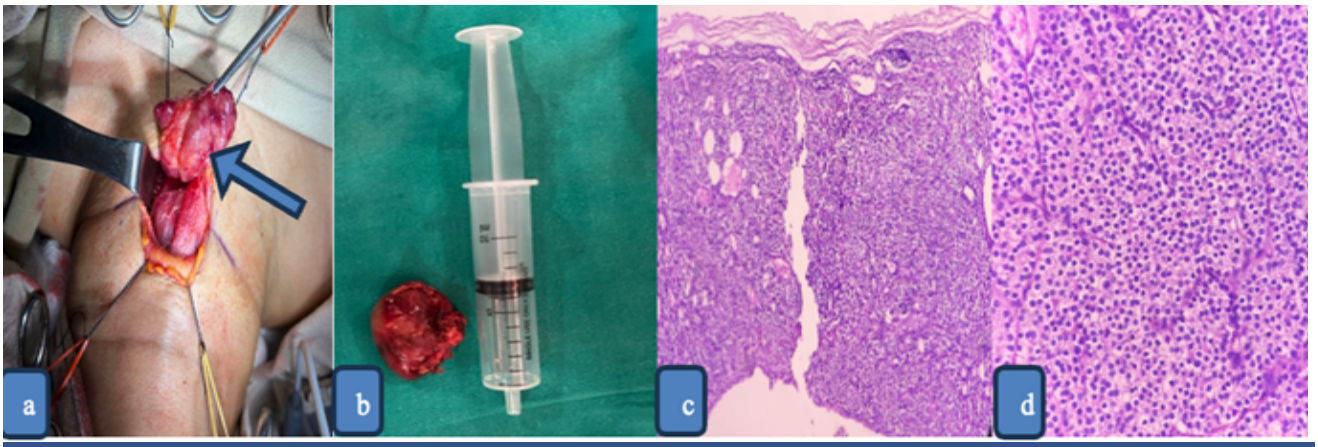

Figure 3. Intraoperative image (a) showing solid lesion (thick arrow head). HPE showing tumor cells composed of chief cells with round nuclei and slightly granular cytoplasm (c). Thin fibrous capsule surrounding the tumor (d).

tracer uptake in adenomas compared to the thyroid gland.[5] 4-dimensional Computed Tomography (4D-CT) is increasingly used for pre-operative identification of parathyroid adenomas, providing detailed anatomical and functional information and aiding in differentiation from nearby tissues. The "polar vessels" sign on 4D-CT can assist in localization. Studies have shown that 4D-CT is superior to Sestamibi for localizing parathyroid adenomas, especially in cases of solitary adenomas and multi-gland disease.[6]

Distinguishing between brown tumors and malignant tumors can be challenging due to their mixed radiological appearance and raised levels of PTH, requiring histological confirmation. Brown tumors appear brown due to vascularity, hemorrhage, and hemosiderin deposits. They are similar to giant cell tumors of the bone, with hypervascular fibroblastic stroma and osteoclastic giant cells.[7]

Treatment typically involves parathyroidectomy, leading to healing and tumor disappearance. Removing the parathyroid tumor can aid in healing via. osteoblastic activity and remineralization, leading to sclerotic lesions in the healed bone.[8] Close monitoring is important for patients opting for conservative management to prevent neurological complications. Our patient showed significant improvement after adenoma resection, leading to notable improvement after one year. Following parathyroid adenoma resection, patients may develop hungry bone syndrome due to severe hypocalcemia lasting more than 4 days. This condition is more common in patients with multiple brown tumors and is typically treated with high-dose calcium supplementation for up to a year.[9]

Conclusion

Accurate detection of brown tumour caused by hyperparathyroidism requires various bone imaging techniques and biochemical tests. Ultrasonography (USG), 99mTc-MIBI parathyroid scintigraphy (Sestamibi), and 4-dimensioal Computed Tomographhy (4D-CT) can help locate the parathyroid adenoma. Treatment involves parathyroidectomy, with a multi-disciplinary team for comprehensive management.

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Conflict of Interest

None

Author's Contribution

Concept, design, planning: SKC, MP; Literature review: SKC; Draft manuscript: SKC, MP, DY; Revision of draft: SKC, MP, DY, MS; Final manuscript: SKC, MP, DY, MS; Accountability of the work: SKC, MP, DY, MS.

References

1. Choi JH, Kim KJ, Lee YJ, Kim SH, Kim SG, Jung KY, et al. Primary hyperparathyroidism with extensive brown tumors and multiple fractures in a 20-year-old woman. Endocrinol Metab (Seoul). 2015;30:614-9. DOI
2. Misiorowski W, Bilezikian JP. Osteitis fibrosa cystica. JBMR Plus. 2020 Sep 7;4(9):e10403. DOI
3. Lala M. Management of primary hyperparathyroidism. Indian J Surg Oncol. 2022 Mar;13(1):143-51. DOI
4. Xie C, Tsakok M, Taylor N, Partington K. Imaging of brown tumors: a pictorial review. Insights Imaging. 2019 Jul 29;10(1):75. Full Text
5. Tawfik AI, Kamr WH, Mahmoud W, Abo Shady IA, Mohamed MH. Added value of ultrasonography and Tc-99m MIBI SPECT/CT combined protocol in preoperative evaluation of parathyroid adenoma. Eur J Radiol Open. 2019 Nov 15; 6:336-42. DOI
6. Yeh R, Tay YD, Tabacco G, Dercle L, Kuo JH, Bandeira L, et al. Diagnostic performance of 4D CT and Sestamibi SPECT/CT in localizing parathyroid adenomas in primary hyperparathyroidism. Radiology. 2019 May;291(2):469-76. DOI
7. Majumdar S, Uppala D, Kotina S, Alekhya B. Brown tumor of hyperparathyroidism with multiple lesions. J Oral Maxillofac Pathol. 2022 Feb;26(Suppl 1): S111-5. DOI
8. Ameerudden S, He X. Management and surgical treatment of parathyroid crisis secondary to parathyroid tumors: report of four cases. Int Med Case Rep J. 2011 Aug 15; 4:59-66. DOI
9. Anwar F, Abraham J, Nakshabandi A, Lee E. Treatment of hypocalcemia in hungry bone syndrome: A case report. Int J Surg Case Rep. 2018; 51:335-9. DOI