

Parathyroid Adenoma Presenting with Multiple Long Bone Lytic Lesions in An Adult Female: A Diagnostic Dilemma

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Abstract

Lytic bone lesion is one of the manifestations of Parathyroid adenoma due to osteoclastic bone resorption. It is also referred to as Brown's tumor and it is characterized by elevated serum calcium and serum parathyroid hormone. Primary hyperparathyroidism is due to parathyroid adenoma in 80-85% of cases, hyperplasia in 10-15% of cases and carcinoma in 1-5% of cases. Our case is a 38year old female with multiple long bone lytic lesions resembling malignancies for which the primary source could not be identified from the clinical examination. A possible diagnosis of primary and secondary bone malignancies was made. Postoperative histological diagnosis gave a conflicting finding of fibrous dysplasia and non-ossifying fibroma. She was eventually found to have parathyroid adenoma for which she had partial parathyroidectomy with subsequent healing of the bone lesions.

Keywords: Parathyroid Adenoma; Multiple Long Bone Lytic Lesions; Tumors; Lytic Bone Lesions.

Introduction

Lytic bone lesions are also known as osteitis fibrosa, cystica generalisata or Von Recklinghausen's disease of bone, is a metabolic bone disease that develop in primary, secondary or tertiary Hyperthyroidism [1]. With a 2.3–3% overall incidence, this tumor-like lesion is the last stage of the bone remodelling process in persistent hyperparathyroidism. [2,3]. Increased fibroblastic proliferation and osteoclastic activity may be the cause. Although brown tumors can develop in any area of the skeleton, they are most frequently detected in the pelvic girdle, ribs, jaws, clavicles, and extremities.

Although they can be aggressive, they are rarely malignant [4,5]. Clinical features include swelling, pathological fracture, and diffuse skeletal pain; when it involves involving multiple bones it can mimic a metastatic disease [3,6,7,8,9,10,11,12]. To make a diagnosis of BT, we need a detailed clinical history, clinical examination, laboratory results, and radiological imaging. [2,3,13]

Surgical biopsy and histology are the gold standard for a conclusive diagnosis of this tumor, but radiographic results and biochemical evaluation, including blood calcium, phosphorus, and parathyroid hormone (PTH) values, are crucial for the diagnosis as well. [2,3,13].

Parathyroid glands may be removed entirely or partially as part of the treatment for these tumors, which causes the tumor to suddenly recede. [14,15]

Here we report a case of a Multiple long bone lytic lesions with initial conflicting diagnosis of non-ossifying fibroma and fibrous dysplasia but who was subsequently discovered to have a parathyroid adenoma and then a partial parathyroidectomy with rapid resolution of symptoms.

Case Report

Patient is Mrs. A.M, a 38-year-old teacher who presented with pain in both thighs with difficulty in walking for 2 months duration. On examination there was generalized long bone tenderness in upper and lower limbs, nil deformities, Nil spine tenderness. No neck masses or Palpable breast masses, abdominal masses. X-rays of both thighs revealed multiple Osteolytic lesions in both femur with impending fractures. A possible diagnosis of primary bone malignancy and secondary bone Malignancy were entertained. However, no primary source could be identified from the clinical examination and laboratory and radiological investigations.

She was prepared for an open reduction and internal fixation with intramedullary (interlocked) nail plus bone biopsy which she had both femur in separate occasions. Initial histology showed fibrous dysplasia composed of curvilinear bony trabeculae, fibrous tissue made up of spindle shaped cells within fibrocollagenous stroma and focal areas of hemosiderin laden macrophage. Patient subsequently developed similar symptoms in both tibial and the upper limbs with similar features of Osteolytic lesions on x-ray. Both tibial and one humerus were also fixed with intramedullary nails.

Subsequent biopsies suggested non ossifying fibroma composed of bone fragments with no osteoblastic rimming dispersed within a cellular stroma which is made up of proliferating fibroblasts in storiform pattern with scattered osteoclast like giant cells.

She was then noticed to have hypocalcemia and then the General surgeons were invited. Subsequent Investigations which include neck ultrasound and neck CT scan showed large oval enhancing mass posterior to the right lobe of the thyroid gland measuring 2.9x1.4x1.2 presumably an enlarged parathyroid gland -parathyroid adenoma. There was generalized osteopenia with multiple lytic lesions on the demonstrable skull, bone, vertebral column, sternum and clavicle. Enlarged Pituitary fossa measuring 16.9cm in AP diameter.

Laboratory investigation revealed initial Serum Calcium of 3.5 mmol/L, then Alkaline phosphatase > 850 IU/L, Parathyroid hormone 3612 PG/ML (10-65 PG/ML), No Bence Jones proteins in urine...

A diagnosis of parathyroid adenoma with primary hyperparathyroidism was made.

Patient subsequently had partial parathyroidectomy with intraoperative findings of an enlarged right inferior parathyroid gland measuring 2x3x2 with cystic areas, with a Normal left inferior parathyroid gland. The skeletal lesions of the patient healed fairly rapidly following the surgery.



AP view of left leg showing osteolytic lesions with cortical disruption



AP view of left Humerus showing osteolytic lesions and cortical disruption

IM Nailing of left tibia



Discussion

Brown tumors or long bone lytic lesions are as a result of aberrant bone metabolism in hyperparathyroidism. About 3% of people with PHPT and 2% of those with secondary hyperparathyroidism have these rare tumors, which are the outcome of chronic hyperparathyroidism (primary, secondary, or tertiary) [16,17,18]. Radiologically, they show up as well-defined bordered osteolytic lesions.

Bone metastases, amyloid cysts, chondroma, aneurysmal bone cysts, osteosarcoma, giant cell tumors, or myeloplax tumors are the main conditions included in the differential diagnosis [16,18]. After thyroid disease and diabetes, primary hyperparathyroidism (PHPT) is the third most common endocrine disorder, with postmenopausal women having the highest incidence of the condition [19,32]. Eighty to eighty-five percent of patients have a single adenoma, five percent have multiple adenomas, ten to fifteen percent have parathyroid hyperplasia, and less than five percent have carcinoma [20].

Although PHPT seldom causes symptoms, it can be identified through routine biochemical screening when no overt clinical symptoms are present [21]. The primary tissue impacted by PHPT is the bone, which might change due to diffuse osteopenia, cysts, and bone reabsorption. When multiple bone lytic lesions are present, a diagnostic conundrum may arise, similar to our case where fibrous dysplasia and non-ossifying fibroma were the initial histology. The differential diagnosis included multiple myeloma, metabolic bone disorders, and metastatic bone lesions. Long lytic lesions that masquerade as malignant lesions or bone metastases have been described in a great deal of literature [1].

The serum calcium and parathyroid hormone levels of the patient were elevated, which are features of metabolic bone disease. Hypercalcemia with elevated PTH levels and radiological evidence of parathyroid gland tumours is clinical evidence, but this was the case in our index patient. However, the increased secretion of PTH from the parathyroid adenoma resulted in high osteoclast activity, leading to the multiple osteolytic bone lesions seen in our patient.

The different histologic diagnoses of our patient also posed a diagnostic dilemma for us. This reiterates that a histology diagnosis should not be considered the final diagnosis for a patient.

PHPT and Lytic bone lesions simulating giant cell tumours (GCT) have been mentioned in the literature [22, 23–24]. Pezzillo et al. [22] reported two cases of BT: an isolated GCT in the humerus and an aneurysmal bone cyst in the femur.

Jouan et al [23] reported a patient with clinical features of PHPT who underwent an unnecessary amputation of his right hand's fifth ray; and Vera et al. [24]. reported a patient who underwent surgical ablation of a costal mass on his third right rib that histology misdiagnosed as a GCT.

Given the clinical and radiological similarities between long bone lytic lesions and GCTs, a differential diagnosis can be extremely challenging. To avoid this diagnostic conundrum, the pathologist must have access to crucial clinical and laboratory data [1].

There is research on the coexistence of BTs for hyperparathyroidism with GCT [25, 33]. When the bone lesion does not regress after hyperparathyroidism treatments, GCT is a likely diagnosis [1]. The case of a 37-year-old white lady with simultaneous asymptomatic PHPT with parathyroid adenoma and GCT of her proximal left tibia was reported by Rossi et al. [33].

The presence of different diagnoses resulted in a diagnostic dilemma and affected the management of the patient. The parathyroid adenoma was first addressed, and nine months later she underwent curettage of a tibial GCT and received denosumab for 12 months [1].

The surgical excision of the hypersecreting parathyroid gland is the primary treatment for BTs. It is anticipated that BTs will either fully resolve or regress after the gland is removed. In our instance, this applied to our index patient. The bony lesions receded quickly. Orthopaedic treatments, such as fixing pathological fractures, are considered appropriate [1].

Using criteria, the likelihood of a pathologic fracture should be assessed.

We chose to perform a preventive fixation in this particular case since the osteolytic lesion on the left tibia and femur had a borderline Mirels score of 8 [1]. In our case, the diagnosis of lytic bone lesions was supported by the confluence of hyperparathyroidism, improvement, remission of all bony lytic lesions, and the following parathyroidectomy.

Conclusion

This case report serves as a reminder that for proper management of patients with osteolytic lesions and to prevent pathologic fractures and iatrogenic harm to the patient after surgical treatment, lytic bone lesions of primary hyperparathyroidism must be taken into consideration in the differential diagnosis of multifocal osteolytic bone lesions [1]. Early diagnosis of this disease requires a high index of suspicion; laboratory testing for serum phosphate, calcium, and PTH levels should be part of the routine evaluation of patients with multifocal osteolytic lesions; also, osteolytic metastases and multiple myeloma should be ruled out [1].

A multidisciplinary approach involving orthopedic surgeons, pathologists, and radiologists is important to make an accurate diagnosis and in management of the patient.

Conflict of Interest

The authors declare that they have no conflict of interest.

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