

A Case of Parathyroid Adenoma with Bilateral Subtrochanteric Pathological Femur Fracture – A Rare Case Report

Siddharth Gunay¹, Tanmay Avhad², Nihar Modi², Harshal Patil²

Learning Point of the Article:

This case report highlights the importance of considering primary hyperparathyroidism as a potential cause for pathological fractures, even in cases where the patient is asymptomatic. A specific protocol for diagnosing parathyroid adenoma was followed in a planned and staged manner, demonstrating the need for a multidisciplinary approach in such rare presentations

Abstract

Introduction: Primary hyperparathyroidism is a commonly occurring endocrine disorder that is characterized by elevated calcium levels, decreased phosphate levels, and high levels of parathyroid hormone (PTH). The condition can lead to significant bone resorption and pathological fractures.

Case Report: We report a case of a 44-year-old female who presented with bilateral thigh pain after a trivial fall at home. Radiological investigations revealed a subtrochanteric fracture of the bilateral femur that was deemed pathological. Biochemical testing indicated severe hypercalcemia and hypophosphatemia with elevated levels of serum PTH and an increased alkaline phosphatase level. Ultrasound and computed tomography scans confirmed a parathyroid adenoma, which was treated through excision and histopathological examination. The patient underwent orthopedic intervention for bilateral subtrochanteric femur fracture, and follow-up investigations showed normal biochemical markers and fracture union within 6 months.

Conclusion: Primary hyperparathyroidism should be kept in mind when dealing with bone lesions connected to hypercalcemia, even in asymptomatic individuals and individuals presenting with a trivial mode of trauma. The diagnosis of parathyroid adenoma requires a combination of radiological and biochemical investigations, and a multidisciplinary approach is recommended for the best possible outcome.

Keywords: Primary hyperparathyroidism, pathological fracture, parathyroid adenoma, orthopedic intervention, multidisciplinary approach.

Introduction

Hyperparathyroidism is a condition that can be categorized as primary, secondary, or tertiary and is characterized by elevated calcium levels, decreased phosphate levels, and high levels of parathyroid hormone (PTH). Primary hyperparathyroidism can lead to significant bone resorption, resulting in bone pain and pathological fractures. It is the third most common endocrine disorder, after diabetes and thyroid disease. While not frequently documented, there have been instances of pathological fractures

uncovering primary hyperparathyroidism.

The major cause of primary hyperparathyroidism is parathyroid adenoma followed by follicular hyperplasia followed by parathyroid carcinoma [1]. Parathyroid adenoma is a progression of proliferative pathogenesis impacting the parathyroid gland, beginning with hyperplasia of parathyroid follicles and potentially leading to parathyroid carcinoma [2]. A majority of patients exhibit non-specific symptoms, such as fatigue, loss of appetite, polyuria, and polydipsia. Loss of

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Author's Photo Gallery



Dr. Siddharth Gunay



Dr. Tanmay Avhad



Dr. Nihar Modi



Dr. Harshal Patil

¹Department of Orthopaedics, HBT Medical College and Dr. R.N Cooper Hospital, Mumbai, Maharashtra, India,
²Department of Orthopaedics, B.Y.L Nair Charitable Hospital, Mumbai, Maharashtra, India.

Address of Correspondence:

Dr. Siddharth Gunay,
Department of Orthopaedics, HBT Medical College and Dr. R.N Cooper Hospital, Mumbai Maharashtra, India.
E-mail: siddharthgunay@gmail.com

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Figure 1: Radiographs of left hip at presentation (Antero-posterior and lateral views) showing pathological left subtrochanteric femur fracture.

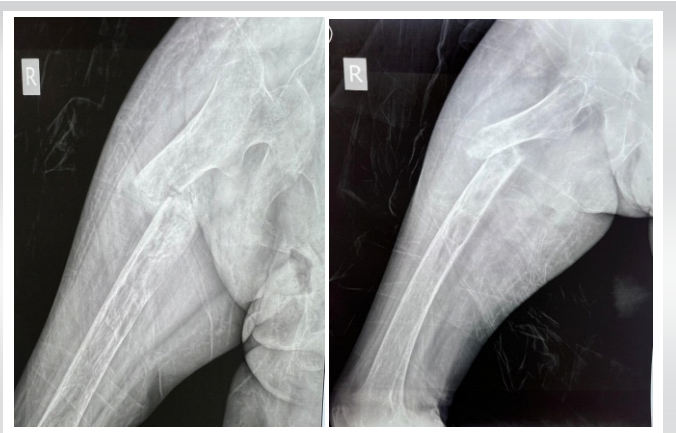


Figure 2: Radiographs of right hip at presentation (Antero-posterior and lateral views) showing pathological right subtrochanteric femur fracture.

appetite, bloating, and nausea are common intestinal symptoms, while musculoskeletal symptoms include osteoporosis, myalgia, and bone pain [3]. Confirmation of the diagnosis typically involves biochemical screening to detect increased levels of serum calcium, decreased levels of phosphate, and elevated levels of serum parathyroid [4].

As a result of the non-specific symptoms, it is unlikely for a parathyroid adenoma to be

diagnosed solely based on the occurrence of an isolated fracture. This scenario can lead to mismanagement of the condition. The case presented involves a fracture of the bilateral femur whose cause was later identified as a parathyroid adenoma, leading to the exploration of the neck.

Case Report

A 44-year-old female with insignificant past medical or surgical history presented to our outpatient department (OPD) with bilateral thigh pain after a trivial fall at home. Upon clinical

examination, she was having pain and swelling over both thighs. Tenderness could be elicited over a proximal aspect of both thighs. The distal pulses and neurological examination were normal. The radiographs taken of the pelvis displayed a subtrochanteric fracture of the bilateral femur that was deemed pathological. (Fig. 1 & 2) As the fall was deemed insignificant, biochemical investigations were conducted to eliminate any likelihood of a metabolic disorder.

The patient's biochemical reports indicated severe hypercalcemia (12.6 mg/dL, with normal levels ranging from 8.6 to 10.3 mg/dL), hypophosphatemia (2.3 mg/dL, with normal levels ranging from 2.8 to 4.5 mg/dL), elevated levels of serum PTH (907.7 pg/mL, with normal levels

ranging from 11 to 51 pg/mL), hypercalciuria (234 nmol/L, with normal levels ranging from 3 to 8 nmol/L), and an increased alkaline phosphatase level (1550 IU/L, with normal levels ranging from 30 to 300 IU/L). Based on the biochemical findings, the patient was diagnosed with hyperparathyroidism. The bone densitometry results indicated values below -2.5 standard deviation, which suggested the presence of severe osteoporosis.

To rule out parathyroid disorder, ultrasonography (USG) of the neck was done which was suggestive of a hypoechoic lesion posterior to the thyroid (10×6 mm). (Fig. 3) To gather more information, we conducted a 4D computed tomography (CT) scan of the neck which showed a

$1.1 \times 1.1 \times 0.6$ cm lesion in the posterior aspect of the left hemithyroid suggestive of parathyroid adenoma. After consultation with the endocrinology and the surgery department, the patient underwent right and left inferior parathyroidectomy. Following the removal of the mass, there was a significant decrease in the

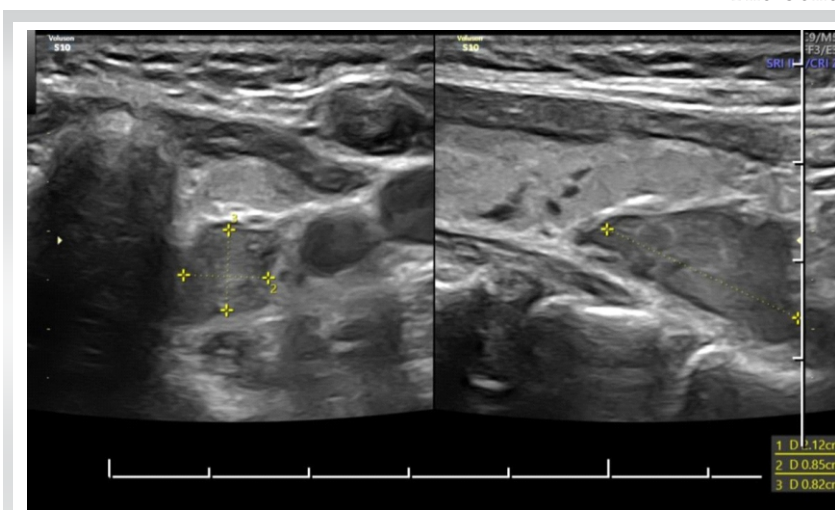


Figure 3: Ultrasonography of neck showing presence of parathyroid adenoma.

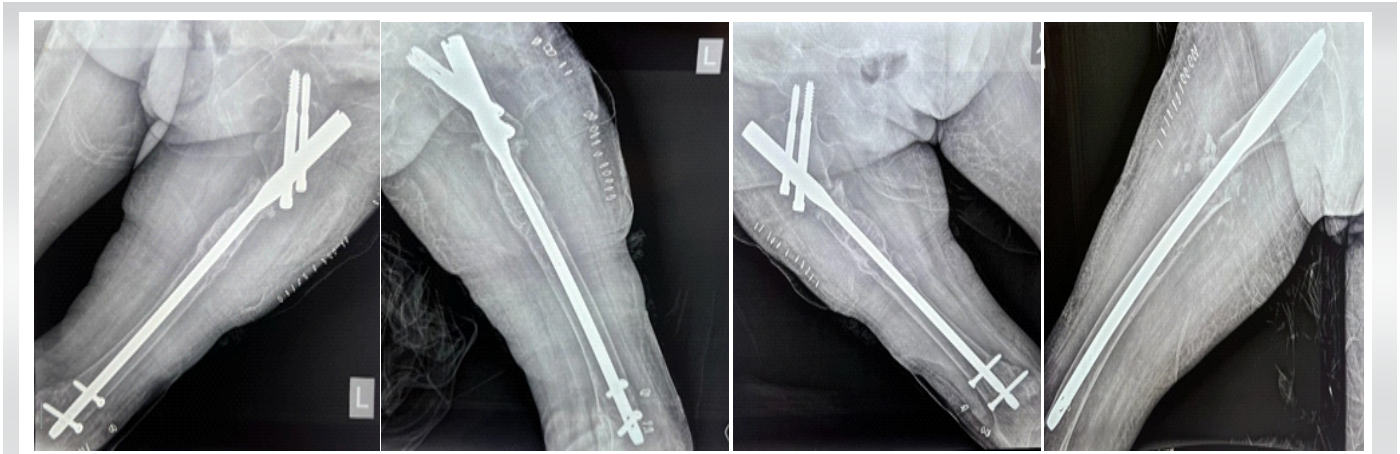


Figure 4: Immediate Post operative radiographs of bilateral femur operated with cephalomedullary nail fixation.

patient's PTH levels. By the 1st post-operative day, PTH levels fell down to 65.4 pg/mL. To prevent hypocalcemia, the patient was given oral calcium and Vitamin D supplements after the operation. Subsequently, the removed mass was sent for histopathological examination (HPE), which indicated the presence of a parathyroid adenoma. The patient then underwent cephalomedullary nail fixation for bilateral subtrochanteric femur

fracture in the same setting. (Fig. 4) Following this, the patient underwent regular clinicoradiological examinations in the OPD, with a 1-month interval between each assessment. Following the excision of the parathyroid, the patient underwent regular clinicoradiological examinations in the OPD, with a 1-month interval between each assessment. The fracture was united by a 3-month follow-up, and repeat biochemical investigations at that time showed normal results. The patient's follow-up was continued for a total of 6 months,

during which there were no indications of recurrence, whether based on clinical or biochemical assessments. Upon completing a 6-month follow-up, the patient displayed a favorable functional outcome with fracture union. (Fig. 5) She reported no pain and was able to walk unassisted.

Discussion

Hyperparathyroidism is a frequently occurring endocrine ailment that primarily affects postmenopausal women. The condition is usually detected by elevated levels of serum calcium and PTH in the body [5]. The initial diagnosis of adenoma generally involves USG and 4D CT scans, followed by confirmation through HPE. Due to the generally asymptomatic nature of parathyroid adenoma, diagnosis can be difficult. Furthermore, the occurrence of an isolated bone

fracture in cases of parathyroid adenoma is rare, which can pose an additional challenge for diagnosis. After conducting an

extensive search of PubMed, it was found that there are fewer than 10 recorded cases of pathological fractures resulting from parathyroid adenoma. In one such case, Bennett et al. (Table 1) reported a fracture sustained by an adolescent female in the right femoral neck [6]. This was managed by closed reduction, internal fixation using pinning, and the application of a hip spica cast. In one case reported by Buisset et al., (Table 1) a 40-year-old female with a pathological left forearm fracture was managed conservatively using a cast [7]. It was later discovered that the patient had a right inferior parathyroid mass, which was treated through en bloc excision. Another similar case was reported by Terro et

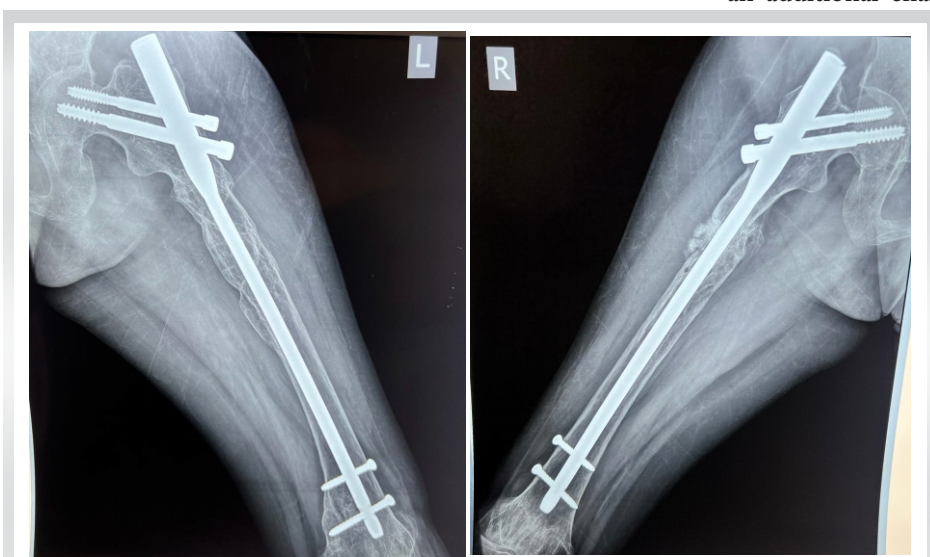


Figure 5: 6 month follow-up radiographs of bilateral femur operated with cephalomedullary nail fixation showing fracture union.

| Case number | Author | Clinical presentation | Diagnosis | Management | Clinical outcome |
|-------------|----------------|---|--|--|----------------------------|
| 1 | Bennett et al. | Right hip pain since 2 weeks in a 14-year-old female | Right pathological neck of femur | Closed reduction, internal fixation using pinning with an application of a hip spica cast with parathyroidectomy | Fracture union at 24 weeks |
| 2 | Buisset et al. | Left forearm pain for 10 days in a 40-year-old female | Left pathological both bone forearm fracture | Managed conservatively with Parathyroidectomy | Fracture union at 20 weeks |
| 3 | Terro et al. | Left clavicle pain for 10 days | Left pathological clavicle fracture | Managed conservatively with Parathyroidectomy | Fracture union at 20 weeks |

Table 1: Showing similar cases of pathological fractures due to underlying parathyroid disorder

al.,(Table 1) where a 41-year-old male with a left clavicle fracture was treated conservatively [8]. The patient was found to have a giant parathyroid adenoma, which was managed through right lower parathyroidectomy. Upon the diagnosis of a pathological fracture, it is important to investigate metabolic bone diseases such as primary hyperparathyroidism and osteomalacia, along with conditions, such as multiple myeloma and bone metastasis [9]. Even mild cases of primary hyperparathyroidism can increase the risk of non-vertebral and vertebral fractures due to a decrease in volumetric densities within both the cortical and trabecular compartments of bones. This increased risk is supported by research findings [10, 11]. To make a conclusive diagnosis, one must consider the patient's clinical history, radiological examination findings, and biochemical tests. These tests may include measuring serum levels of PTH, alkaline phosphatase, calcium, phosphate, and Vitamin D [12]. In cases of pathological fractures or significant cortical involvement, orthopedic interventions should be considered [13, 14].

We report an uncommon case of a pathological fracture resulting in a bilateral subtrochanteric femur fracture in a female patient who was previously asymptomatic. Despite the trivial nature of the fall that resulted in the fracture, a specific protocol for diagnosing parathyroid adenoma was followed in a planned and staged manner. Once the adenoma was excised and the fracture was fixed, normal biochemical markers were observed and the fracture united within 6 months of the follow-up.

However, the main limitation of the study is the relatively short 6-month duration of follow-up since adenoma recurrence typically occurs within 2–3 years after excision. Therefore, a longer-term assessment may be necessary to determine the overall outcome.

Conclusion

Given the rarity of parathyroid adenoma, it is critical to maintain a high level of suspicion to diagnose bone fractures that present in an extremely uncommon manner. It is important to keep primary hyperparathyroidism in mind when dealing with bone lesions connected to hypercalcemia since this disorder is typically asymptomatic. The best way to diagnose and treat such conditions is to combine clinicoradiological and biochemical studies with the expertise of specialists in handling pathological fractures. To achieve the best possible outcome for such rare presentations, a multidisciplinary approach is recommended.

Clinical Message

A diagnosis of the underlying pathology can be achieved by utilizing a combination of clinicoradiological and biochemical studies. It is important to maintain a high index of suspicion to ensure an accurate diagnosis.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given the consent for his/ her images and other clinical information to be reported in the journal. The patient understands that his/ her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict of interest: Nil **Source of support:** None

References

- Hussain A, Mahmood H, Geddoa E, Hamish M, Reda AH. Parathyroid adenoma and hypercalcemia in a patient presenting with multiple pathological fractures of the long bones. *Int J Surg* 2008;6:465-8.
- Wieneke JA, Smith A. Parathyroid adenoma. *Head Neck Pathol* 2008;2:305-8.
- Yigit B, Tanal M, Citgez B. Giant parathyroid adenoma diagnosed by brown tumour, a clinical manifestation of primary hyperparathyroidism: A case report. *J Pak Med Assoc* 2021;71:1266-9.
- Adegoke OO, Ajani MA, Awosusi BL, Onakpoma FA, Saiki O, Daniel A. Parathyroid adenoma with unusual presentations of rib bone and thoracic vertebrae fractures in a premenopausal female in Ibadan, Nigeria. *Niger Med J* 2020;61:273-5.
- Pappu R, Jabbour SA, Reginato AM, Reginato AJ. Musculoskeletal manifestations of primary hyperparathyroidism. *Clin Rheumatol* 2016;35:3081-7.
- Bennett JT, Alexander HH, Morrissy RT. Parathyroid adenoma presenting as a pathologic fracture of the femoral neck in an adolescent. *J Pediatr Orthop* 1986;6:473-6.
- Buisset C, Demarquet L, Raynal M, Busby H, Nominé-Criqui C, Brunaud L. When a pathological forearm fracture led to explore the neck: About a case. *Head Neck Pathol* 2020;14:828-32.
- Terro JJ, El-Helou E, El-Khoury E, Lakkis RS, Shibli A, Al Raishouni MA, et al. Giant parathyroid adenoma presenting with a pathological left clavicular fracture: An extremely rare case report. *Int J Surg Case Rep* 2020;75:311-6.
- Vanitcharoenkul E, Singsampun N, Unnanuntana A, Sirinvaravong S. Osteitis fibrosa cystica and pathological fractures-the classic but neglected skeletal manifestation of primary hyperparathyroidism: A case report. *BMC Musculoskelet Disord* 2021;22:443
- Narayanan N, Palui R, Merugu C, Kar SS, Kamalanathan S, Sahoo J, et al. The risk of fractures in primary hyperparathyroidism: A meta-analysis. *JBMR Plus* 2021;5:e10482.
- Ejlsmark-Svensson H, Rolighed L, Harsløf T, Rejnmark L. Risk of fractures in primary hyperparathyroidism: A systematic review and meta-analysis. *Osteoporos Int* 2021;32:1053-60.
- Olvi LG, Da Cunha IW, Santini-Araujo E, Kalil RK. Brown tumor of hyperparathyroidism. In: Santini-Araujo E, Kalil RK, Bertoni F, Park YK, editors. *Tumors and Tumor-like Lesions of Bone*. Germany: Springer; 2020.
- Panagopoulos A, Tatani I, Kourea HP, Kokkalis ZT, Panagopoulos K, Megas P. Osteolytic lesions (brown tumors) of primary hyperparathyroidism misdiagnosed as multifocal giant cell tumor of the distal ulna and radius: A case report. *J Med Case Rep* 2018;12:176.
- Sybil Biermann J. *Orthopaedic Knowledge Update: Musculoskeletal Tumors*. Vol. 3. United States: Wolters Kluwer Health; 2018.

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