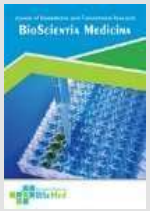
eISSN (Online): 2598-0580



Bioscientia Medicina: Journal of Biomedicine & Translational Research

Journal Homepage: www.bioscmed.com

Parathyroid Adenoma with Multiple Bone Fractures: A Case Report

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ARTICLE INFO

Keywords:
Bone fractures
Brown tumor
Hypercalcemia
Hyperparathyroidism
Parathyroid adenoma

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All authors have reviewed and approved the final version of the manuscript.

https://doi.org/10.37275/bsm.v8i5.987

A B S T R A C T

Background: Parathyroid adenoma is a benign neoplasm of the parathyroid glands and can result in hypercalcemia due to excessive secretion of parathyroid hormone (PTH). This elevated PTH can have various manifestations, including bone fractures. **Case presentation:** We report the case of Tn. AB is a 41-year-old man with a series of recurrent bone fractures following minor traumas. Within a span of three years, he suffered from fractures in the right femur, left femur, and left humerus, which led to multiple surgeries, including total hip replacements. Further assessments showed an increased level of PTH and a suspicious lesion in the neck region. A CT scan revealed a potential parathyroid adenoma in the peritracheal region of the right side. Moreover, a Sestamibi scan confirmed an adenoma in the postero-inferior lobe of the right thyroid, consistent with Perrier type C, alongside osteoblastic processes and brown tumours. A subsequent parathyroidectomy confirmed a 2 cm diameter adenoma in the right lower parathyroid lobe. Histopathology from the excised tissue confirmed the diagnosis of parathyroid adenoma. **Conclusion:** Parathyroid adenomas can have significant clinical manifestations, including recurrent bone fractures, due to their metabolic effects on bone integrity. Early detection and surgical intervention can prevent complications and improve the quality of life for these patients.

1. Introduction

Parathyroid adenoma represents a significant chapter in the spectrum of parathyroid gland disorders. It emerges from a succession of proliferative changes within the parathyroid gland, initiating from follicular hyperplasia and potentially culminating in parathyroid carcinoma.[1] For clinicians, the vagueness of its presenting symptoms poses a diagnostic challenge. A majority of patients may complain of seemingly unrelated symptoms such as loss of appetite, frequent urination, increased thirst, and an overall feeling of weariness. Furthermore, gastrointestinal manifestations might encompass appetite reduction, feelings of nausea, and abdominal bloating.[2] Delving deeper, musculoskeletal complaints emerge, most prominently seen as myalgia, pains localized to the bones, and even osteoporosis.[2] What this culminates in is a patient presenting with a mélange of gastrointestinal, renal, and musculoskeletal symptoms, sometimes all at once. To solidify the diagnosis, practitioners typically rely on biochemical markers. These include a noted increase in serum calcium levels, a decrease in phosphate levels, and a consequential increase in serum parathyroid levels.[3] It is worth noting that the recent surge in health consciousness and regular medical check-ups has led to incidental findings of hypercalcemia during routine biochemical evaluations.[4]

In terms of etiology, primary hyperparathyroidism predominantly has its roots in parathyroid adenomas. This is followed in prevalence by follicular hyperplasia and the more sinister parathyroid carcinoma.[5] Whereas follicular hyperplasia presents with a

generalized hyperplasia encompassing all four parathyroid glands, parathyroid adenoma is more localized, typically involving just a single gland. In their normal state, parathyroid glands maintain a diminutive size (around 5 mm × 3 mm × 1 mm), making them virtually undetectable on routine imaging studies. However, for visualizing pathologically altered parathyroid tissue, imaging modalities such as ultrasonography (USG) and 99mTc-sestamibi scintigraphy are the gold standards.[6] For increased sensitivity, Single Photon Emission Computed Tomography (SPECT) is preferred over the sestamibi scan, especially in visualizing a parathyroid adenoma. For the more elusive ectopic glands, advanced imaging like contrast-enhanced CT or MRI becomes essential.[6] In light of these non-specific symptoms, it becomes even rarer for clinicians to correlate an isolated bone fracture with the possibility of a parathyroid adenoma. Such rare presentations can easily mislead physicians, leading to misdiagnosis and inappropriate management. In this report, we emphasize one such unique case where an isolated fracture served as the initial presentation, which eventually unfolded the diagnosis of a parathyroid adenoma, prompting a comprehensive exploration of the patient's neck.

2. Case Presentation

A 41-year-old male, Mr. AB, with a history of multiple fractures, presented to our Outpatient Department (OPD). He had a history of falling off a motorcycle in June 2020, leading to a fracture of the right femur, which was managed with a total hip replacement. In July 2021, he fractured his left femur while repairing a motorcycle and subsequently underwent another total hip replacement at the same hospital. By July 2022, he sustained another fracture involving the left upper arm. Upon clinical examination, multiple swollen lymph nodes were palpable in the submandibular region. His thyroid was not palpably enlarged, and there was no evidence of vascular bruits (Figure 1).

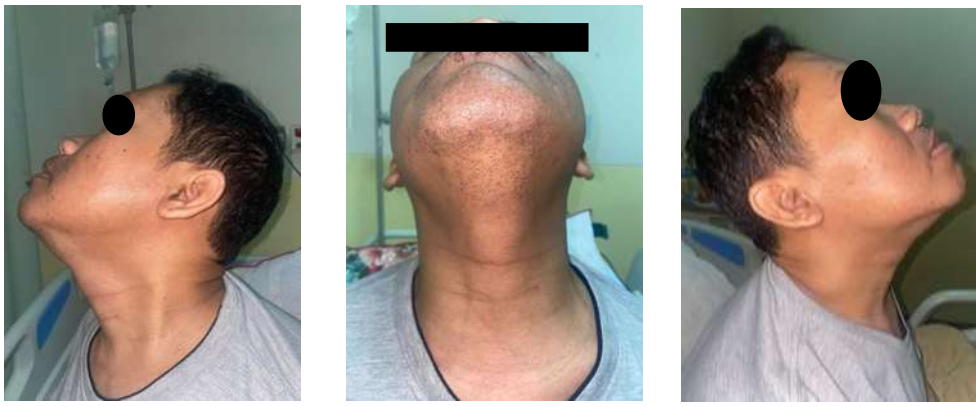

Figure 1. Clinical presentation.

Standard radiographs revealed a closed fracture of the left humerus. Initial laboratory investigations demonstrated raised Parathyroid Hormone (PTH) levels at 1243 pg/mL (Normal: 15-65 pg/mL), suggestive of primary hyperparathyroidism. This was further corroborated by a CT scan of the neck, which indicated a possible parathyroid adenoma. Sestamibi scan supported the diagnosis by showing an adenoma in the postero-inferior region of the right thyroid lobe (Perrier type C). The scan also displayed signs of osteoblastic processes and brown tumors in the calvaria and proximal humerus but no evidence of metastasis (Figure 2).

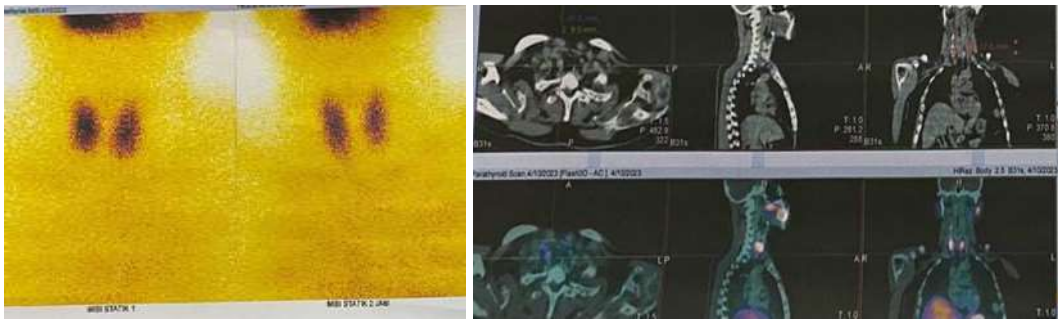

Figure 2. Sestamibi scan shows an adenoma in the region of the right thyroid lobe (Perrier type C) and osteoblastic processes and brown tumors in the calvaria and proximal humerus.

After consultation with our multidisciplinary team, a decision was made to proceed with surgery. Intraoperative findings confirmed an enlarged parathyroid gland in the inferior right lobe, measuring 2 cm in diameter with a yellow-gray appearance (Figure 3). The suspected adenomatous tissue was excised. The postoperative pathological examination of the resected tissue confirmed the diagnosis of parathyroid adenoma.

Figure 3. Enlarged parathyroid gland.

Following surgery, the patient was managed on a diet providing 1800 kcal/day, with a protein intake of 1.2 gr/kg body weight/day and a carbohydrate to lipid ratio of 60:40. Additionally, the patient was prescribed Alendronate 70mg once weekly and Calcitriol 0.25 mg twice daily orally. Regular follow-ups were planned, with the primary focus on clinicoradiological assessments and monitoring of biochemical parameters.

3. Discussion

Primary hyperparathyroidism (PHP) stands as a prevalent endocrine disorder predominantly seen in postmenopausal women. The typical diagnostic hallmarks for PHP are elevated levels of serum calcium and PTH.[7-10] Ultrasonography (USG) and sestamibi scans often provide the preliminary diagnosis of adenoma, but the confirmation usually comes from histopathological examination (HPE). The tendency of parathyroid adenoma to present as asymptomatic or minimally symptomatic makes its diagnosis a challenge.[11-15] Furthermore, a pathological fracture resulting from a parathyroid adenoma is an extreme rarity, further complicating diagnosis.[16-20] An extensive review on PubMed reveals less than ten documented cases of pathological fractures that can be attributed to parathyroid adenomas. Similar to the cases reported by Bennett et al., Buisset et al., and

Terro et al., who all described fractures associated with parathyroid adenomas,[8-10] we recount an exceptional case involving Mr. AB, a 41-year-old male. In June 2020, after a motorcycle accident, he underwent a total hip replacement for his right femur. Just a year later, he experienced a left femur fracture while repairing a motorcycle, once again requiring a total hip replacement at the same institution. By July 2022, he presented with a fractured left upper arm. Subsequent examinations highlighted a giant cell tumor.

A plethora of examinations, including a CT scan of the neck and a sestamibi scan, further supported the suspicion of a parathyroid adenoma. The pathological fracture, especially considering its recurrence and the correlation with the parathyroid adenoma, is a remarkable aspect of this case. Following a parathyroidectomy where an enlarged parathyroid in the lower right lobe was identified and removed, histopathological studies confirmed it as a parathyroid adenoma. Recent case reports and studies have illuminated the varied presentations and complexities of diagnosing parathyroid adenoma, reaffirming its rarity and the diagnostic challenges it poses. For instance, Shukla et al. reported a case of a giant parathyroid adenoma, emphasizing the importance of precise localization through Sestamibi scan and ultrasonography for effective surgical management.[11] Chen et al. described a parathyroid adenoma with rare severe osteolytic lesions, illustrating the potential for parathyroid adenomas to cause significant skeletal damage, aligning with the observations in our case.[12] Furthermore, Juhlin et al. presented a unique case of parathyroid adenoma with respiratory-like epithelium, a reminder of the histological variability of parathyroid adenomas and the need for careful histopathological examination.[13] Concluding, this case reinforces the imperative for heightened suspicion of underlying metabolic or endocrine disorders in patients who present with recurrent or unusual fractures. Though our observation period remains short, with Mr. AB being closely monitored for just over two years since the onset of symptoms, it's crucial to highlight the necessity for long-term follow-ups, as recurrence, though most commonly reported within the initial 2-3 years post-excision, can potentially manifest later.

4. Conclusion

Primary hyperparathyroidism, a well-documented endocrine disorder, can sometimes manifest in unusual ways, complicating diagnosis and management. In the instance of Mr. AB, an unexpected series of fractures, starting from bilateral hip fractures to a humerus fracture, underscored the importance of understanding the potential skeletal consequences of parathyroid abnormalities. The discovery of a parathyroid adenoma in Mr. AB further emphasized the necessity of a rigorous diagnostic process when confronted with seemingly unrelated clinical presentations. Post-parathyroidectomy, Mr. AB's clinical and biochemical parameters are anticipated to show improvement. It remains paramount for healthcare professionals to consider underlying metabolic or endocrine disorders, such as hyperparathyroidism, when encountering atypical bone fractures, even in the absence of other symptoms. Continuous monitoring and long-term follow-ups will be essential in ensuring Mr. AB's ongoing health and in detecting any potential complications or recurrences.

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