DEKA IN MEDICINE

Case report

The role of parathyroid and bone scintigraphy in detecting multiple parathyroid adenomas with fibrous dysplasia: A case report

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ABSTRACT

BACKGROUND: Primary hyperparathyroidism is the leading cause of hypercalcemia, usually caused by a parathyroid adenoma and potentially leading to metabolic bone disorders. Fibrous dysplasia is a rare skeletal disorder that can coexist with hyperparathyroidism, although it is rarely found without McCune-Albright syndrome.

CASE: A 31-year-old woman with a history of hemodialysis presented with progressive swelling of the upper and lower jaw over the past two years, accompanied by bone pain and fatigue. Laboratory tests revealed elevated parathyroid hormone levels, serum creatinine, and hypocalcemia. Magnetic resonance imaging (MRI) of the neck identified an isointense lesion in the left thyroid gland but failed to localize the parathyroid adenoma. 99mTc-Sestamibi parathyroid scintigraphy showed multiple adenomas in the lower poles of both thyroid lobes. 99mTc-MDP bone scintigraphy demonstrated a metabolic superscan pattern, leading to a diagnosis of primary hyperparathyroidism with polyostotic fibrous dysplasia. The patient underwent minimally invasive parathyroidectomy, which was histopathologically confirmed as bilateral inferior parathyroid adenomas. Postoperatively, the patient experienced significant symptom improvement, including reduced bone pain and improved quality of life.

CONCLUSION: The coexistence of primary hyperparathyroidism and fibrous dysplasia without McCune-Albright syndrome is rare but important to recognize. Parathyroid and bone scintigraphy play a crucial role in diagnosis, assessing bone involvement, and planning appropriate therapy. A multimodal imaging approach enables early detection and more effective surgical strategies, improving clinical outcomes for patients.

KEYWORDS: Hyperparathyroidism; parathyroid adenoma; technetium tc 99m sestamibi; bone scintigraphy; fibrous dysplasia.

INTRODUCTION

Primary hyperparathyroidism, a hormonal disorder characterized by excessive secretion of parathyroid hormone (PTH) causing an increase in calcium levels in blood and disturbances in the metabolism of bones, usually has a single parathyroid adenoma as its cause, although, in some cases, there may be multiple adenomas or parathyroid hyperplasia.[1] If untreated, it may give rise to complications like nephrolithiasis, osteoporosis, or other metabolic bone diseases.[2]

Fibrous dysplasia is a rare bone disorder in which normal bone is replaced with structurally weaker fibrous tissue.[3] It can either be monostatic-involving single bone or polyostotic-multiple bones affected.[4] It may also be associated with McCune-Albright

syndrome, which shows features like early puberty and peculiar pigmentation of the skins in addition to endocrine irregularities.[5]

Primary hyperparathyroidism along with fibrous dysplasia is a rare case, particularly when one does not have McCune-Albright Syndrome.[6] Here, the patient had both: on one end, there was polyostotic fibrous dysplasia, whereas, on the other, she was suffering from primary hyperparathyroidism, without showing features of the McCune-Albright Syndrome.[7] Detailed imaging methods-parathyroid and bone scintigraphy-played a vital role in the diagnosis and treatment decision making.[8] This report highlights the impact of imaging in the detection of parathyroid adenomas with regard to evaluating bone involvement as well as the potential role of minimally invasive surgery in providing good patient outcomes.

CASE PRESENTATION

A 31-year-old woman presented with progressive swelling of the upper and lower jaw over the past two years, causing difficulty in eating and speaking. She also reported bone pain and fatigue. Her medical history revealed that she had been undergoing hemodialysis twice a week for the past five years. Menstruation began at the age of 12 but became irregular after starting dialysis. Physical examination showed significant swelling of the upper and lower jaw without tenderness. No café-au-lait spots were observed, and there was no family history of similar conditions. A biopsy performed in July 2023 confirmed fibrous dysplasia of the upper jaw. The patient had a history of taking amlodipine, bisoprolol, candesartan, calcium supplements, sodium bicarbonate, and folic acid. Laboratory findings revealed elevated parathyroid hormone and serum creatinine levels, along with hypocalcemia.

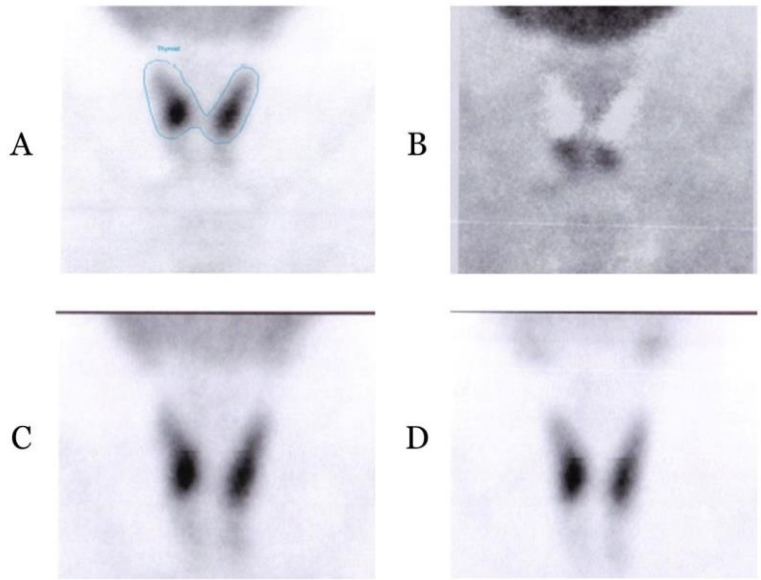

Figure 1. Parathyroid scintigraphy using dual tracer 99mTc-Sestamibi and 99mTc-Pertechnetate. A). The thyroid gland shows radiopharmaceutical uptake, indicating the possibility of parathyroid hyperplasia or adenoma at the lower poles of both thyroid lobes. B). An area with persistent uptake, suggesting the presence of a parathyroid lesion that is more active than the surrounding thyroid tissue. C). Early-phase parathyroid scintigraphy (15 minutes) with 99mTc-Sestamibi shows radiopharmaceutical uptake in the thyroid gland and the potential presence of parathyroid hyperplasia or adenoma. D). Delayed-phase (2 hours) results show decreased uptake in thyroid tissue, while the hyperplastic or adenomatous parathyroid areas retain radiopharmaceutical uptake.

Magnetic resonance imaging (MRI) of the neck showed an isointense lesion with minimal contrast enhancement in the posterior inferior part of the left thyroid gland, suggesting involvement of the left inferior parathyroid gland. Additionally, thickening of the maxillary, mandibular, and other cranial bones was observed, consistent with fibrous dysplasia. Parathyroid scintigraphy using dual tracers 99mTc-Sestamibi and 99mTc-Pertechnetate identified parathyroid hyperplasia or adenomas in the lower poles of both thyroid lobes. Increased radiopharmaceutical uptake in the thyroid gland suggested possible parathyroid hyperplasia or adenomas in the lower pole (Figure 1A). Areas with persistent uptake indicated more active parathyroid lesions compared to surrounding thyroid tissue (Figure 1B). The early-phase parathyroid scintigraphy (15 minutes) showed radiopharmaceutical uptake in the thyroid gland, suggesting potential parathyroid hyperplasia or adenoma (Figure 1C). The delayed-phase imaging (2 hours) demonstrated reduced uptake in thyroid tissue, while the hyperplastic or adenomatous parathyroid areas retained radiopharmaceutical uptake (Figure 1D).

Bone scintigraphy revealed a metabolic superscan pattern characteristic of hyperparathyroidism with fibrous dysplasia. The anterior whole-body view demonstrated a diffuse metabolic superscan pattern with increased radiopharmaceutical uptake, consistent with hyperparathyroidism (Figure 2A). The posterior whole-body view showed a similar pattern, further supporting the diagnosis (Figure 2B). The right lateral bone scintigraphy image exhibited prominent radiopharmaceutical accumulation in the maxillary and mandibular bones, indicating fibrous dysplasia of the facial bones (Figure 2C). The left lateral view showed a similar uptake pattern, confirming the presence of fibrous dysplasia in the maxillary and mandibular bones (Figure 2D).

The patient was diagnosed with multiple parathyroid adenomas and polyostotic fibrous dysplasia. She underwent minimally invasive parathyroidectomy, and histopathological examination confirmed bilateral inferior parathyroid adenomas as the cause of hyperparathyroidism. Postoperatively, the patient reported a significant improvement in symptoms, including reduced bone pain and better overall well-being.

DISCUSSION

Among asymptomatic outpatients, primary hyperparathyroidism is by far the most prevalent etiology of hypercalcemia.[9] Two or more adenomas and parathyroid gland hypertrophy are less frequent than graving a single parathyroid adenoma (90% of the time).[1] On the other hand, tertiary hyperparathyroidism occurs due to parathyroid hyperplasia induced by chronic hypocalcemia, and secondary hyperparathyroidism is mostly related to chronic kidney failure and vitamin D deficiency.[10] Parathyroidectomy is the sole effective intervention for hyperparathyroidism.[11] As for optimal results, exact preoperative localization and reference to a proficient surgeon is vital.[12] Previously, abnormal parathyroid glands had to be localized by bilateral neck exploration.[13] But because of advances in imaging technology, surgical techniques can now be more targeted, reducing operating times and lowering risks and improving patient outcomes.[14] Minimally invasive techniques for parathyroidectomy have advantages over traditional methods, including reduced incision size, less invasive tissue dissection, and a lighter complication rate.[15] This procedure can cure up to 95% of cases and complications are rare.[16]

In this case, MRI could not accurately localize the adenoma; however, parathyroid scintigraphy detected multiple adenomas or adenomatous hyperplasia in the inferior

aspect of both thyroid lobes.[17] Preoperative parathyroid gland localization can be done using a variety of imaging methods, including ultrasonography (US), computed tomography (CT), MRI, and 99mTc-Sestamibi scintigraphy with single-photon emission computed tomography (SPECT/CT).[18] In this context, US is the modality of choice for detecting parathyroid abnormalities before surgery as its rapid, non-invasive, safe, and inexpensive and it does not use radioactive or contrast substances.[19] The sensitivity of US in detecting enlarged parathyroid glands ranges from 51% to 96%, with a specificity of approximately 71%.[19] The sensitivity of US in detecting solitary adenomas, hyperplasia, and double adenomas is 79%, 35%, and 16%, respectively.[20]

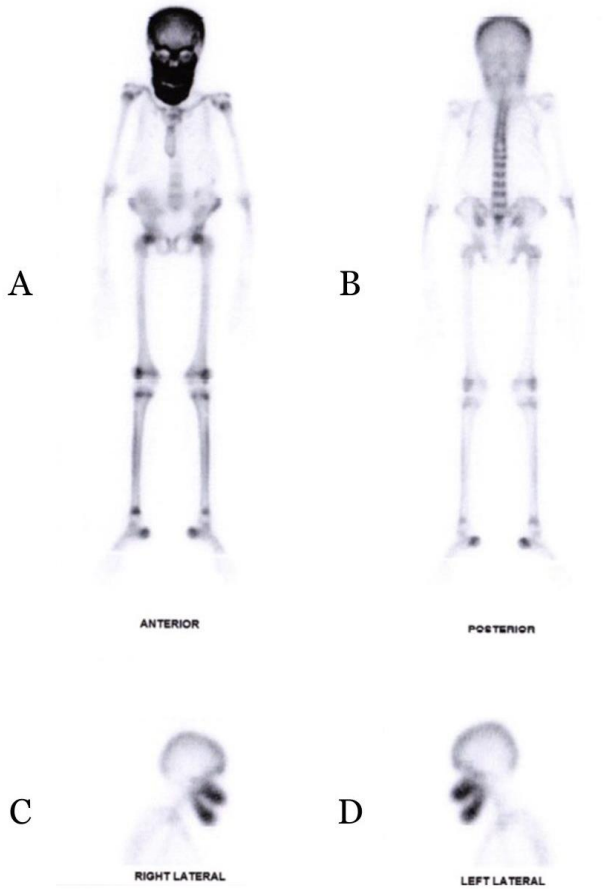

Figure 2. A). Whole-body anterior view of bone scintigraphy shows a metabolic superscan pattern with diffuse radiopharmaceutical uptake, consistent with hyperparathyroidism. B). Whole-body posterior view of bone scintigraphy shows a similar pattern with diffuse metabolic activity, supporting the diagnosis of hyperparathyroidism. C). Right lateral view of bone scintigraphy reveals prominent radiopharmaceutical accumulation in the maxilla and mandible, indicating the presence of fibrous dysplasia in the facial bones. D). Left lateral view of bone scintigraphy shows a similar radiopharmaceutical uptake pattern to the right lateral view, further confirming fibrous dysplasia in the maxillary and mandibular bones.

Contrast-enhanced CT is another imaging technique frequently used to detect parathyroid adenomas.[21] However, small adenomas are often difficult to identify as they may be misinterpreted as lymph nodes or blood vessels.[22] The reported sensitivity of CT in detecting parathyroid adenomas ranges from 46% to 87%.[19] MRI can also be used to detect parathyroid adenomas.[23] The sensitivity of 1.5 Tesla MRI is reported to range from 43% to 77%, while 3 Tesla MRI has higher sensitivity and specificity, at 97.8% and 97.5%, respectively.[24-27]

99mTc-Sestamibi scintigraphy is the most commonly used nuclear medicine imaging technique for the preoperative localization of parathyroid adenomas.[28] This technique is based on the preferential uptake of sestamibi in mitochondria-rich parathyroid adenoma cells, whereas normal parathyroid tissue has lower uptake.[29] There are two main image acquisition methods: subtraction imaging and dual-phase imaging.[30] Subtraction imaging uses a thyroid-specific radiotracer, such as 99mTc-pertechnetate, which is then subtracted from the 99mTc-Sestamibi scan to enhance the visualization of parathyroid adenomas.[21] Meanwhile, dual-phase imaging is performed in the early phase (10–15 minutes after injection) and the delayed phase (2–3 hours after injection).[31] Parathyroid adenomas retain radiotracer uptake for a longer duration, whereas normal thyroid tissue experiences rapid washout.[21]

The SPECT/CT method has advantages over other techniques because it combines functional and anatomical information.[32] The sensitivity of 99mTc-Sestamibi SPECT/CT scintigraphy ranges from 61% to 79%, with a specificity of approximately 71%.[19] However, its sensitivity in detecting hyperplasia and double adenomas is lower, at 44% and 30%, respectively.[33] Bone scintigraphy plays an important role in evaluating bone involvement in hyperparathyroidism, particularly in distinguishing fibrous dysplasia from other metabolic bone diseases.[34] In hyperparathyroid bone disease, increased osteoclastic activity leads to excessive bone turnover, which can be detected using 99mTc-methylene diphosphonate (MDP) bone scintigraphy.[35]

In this case, bone scintigraphy showed a "metabolic superscan" pattern, characterized by increased radiotracer uptake throughout the skeleton and a marked reduction in soft tissue background activity.[36] The kidneys appeared faint or even showed an absence of normal renal activity, producing a "superscan" effect where most of the radiotracer uptake occurs in the bones.[36] This pattern is often found in severe metabolic bone disorders, including hyperparathyroidism and fibrous dysplasia.[37] The ability of bone scintigraphy to detect polyostotic fibrous dysplasia makes it a valuable tool in assessing the extent of the disease.[38] Additionally, SPECT/CT can help evaluate lesion invasion more accurately, as it provides both functional and anatomical imaging.[39] Fibrous dysplasia is often misdiagnosed as a neoplastic process, which can lead to delays in diagnosis and inappropriate treatment.[34] Therefore, nuclear medicine imaging, particularly bone scintigraphy, plays a crucial role in distinguishing fibrous dysplasia from other metabolic bone diseases and malignancies.[40]

Fibrous dysplasia and primary hyperparathyroidism are often associated with McCune-Albright syndrome, a condition characterized by polyostotic fibrous dysplasia, precocious puberty, and café-au-lait spots (distinctive brown skin patches seen in certain genetic disorders).[41] However, in this case, the patient had hyperparathyroidism with fibrous dysplasia but did not meet the criteria for McCune-Albright syndrome. Although rare, the coexistence of fibrous dysplasia and primary hyperparathyroidism in the absence of McCune-Albright syndrome has been documented in the literature.[42] Therefore, a comprehensive evaluation and multimodal imaging are necessary to determine the appropriate treatment strategy for patients with complex parathyroid and bone disorders.[8]

CONCLUSION

In this case, the patient was diagnosed with primary hyperparathyroidism accompanied by fibrous dysplasia without McCune-Albright syndrome, a rare condition. Parathyroid scintigraphy played a crucial role in detecting two parathyroid adenomas, allowing for the planning of minimally invasive parathyroidectomy. Bone

scintigraphy helped identify fibrous dysplasia and assess the extent of bone involvement. After surgery, the patient showed significant symptom improvement, emphasizing the importance of multimodal imaging in the diagnosis and management of hyperparathyroidism with bone complications.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

Patient consent was waived due to no identification of patients

CONFLICTS OF INTEREST

We have no conflict of interest

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We have no source of funding

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AUTHOR CONTRIBUTION

Idea/concept: AK, HK. Design: AK, EA. Control/supervision: AHSK. Data collection/processing: AK. Analysis/interpretation: AK, EF. Literature review: AK, HK. Writing the article: AK. Critical review: HB, AHSK. All authors have critically reviewed and approved the final draft and are responsible for the content and similarity index of the manuscript.

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