

Atypical non- ^{99m}Tc]Tc-MIBI avid parathyroid adenoma with a sternal brown tumor, mimicking ectopic parathyroid adenoma in planar and SPECT images: Importance of SPECT/CT acquisition

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ABSTRACT

Primary hyperparathyroidism (pHPT) is defined by the excess level of parathyroid hormone (PTH). Parathyroid adenoma is responsible for about 80% of all pHPT. Parathyroid adenoma with cystic feature and atypical pathology is recognized as a rare entity. Technetium-99m methoxy isobutyl isonitrile (^{99m}Tc]Tc-MIBI) scintigraphy has favorable detection rate for ectopic gland as well as the brown tumors. SPECT/CT plays critical role in localizing abnormal tracer uptake. Herein, we report a severely hypercalcemic patient with cystic atypical parathyroid adenoma and mediastinal uptake, which was confined to a sternal lytic lesion (suggestive of brown tumor), which highlights the importance of SPECT/CT in ^{99m}Tc]Tc-MIBI imaging.

Key words: Atypical parathyroid adenoma; Brown tumor; SPECT/CT; Technetium-99m; MIBI

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INTRODUCTION

Hyperparathyroidism is a relatively common medical condition and the patient can be asymptomatic for a long time [1, 2]. Primary hyperparathyroidism (pHPT) is defined by the excess level of parathyroid hormone (PTH) due to adenomatous, hyperplastic parathyroid tissue and rarely parathyroid adenocarcinoma [3]. Isolated parathyroid adenoma is responsible for about 80% of all pHPT cases [3, 4].

Atypical (solid and cystic) adenoma and adenocarcinoma is considered as a very uncommon aetiology of pHPT [4]. Parathyroid adenoma with cystic feature and atypical pathology is recognized as a rare presentation [5, 6].

Among various imaging tools for pre-operative evaluation of parathyroid adenoma, [^{99m}Tc]Tc-MIBI and ultrasonography (US) are indicated as modalities of choice [7]. [^{99m}Tc]Tc-MIBI scan has a very favorable detection rate for hyperfunctioning parathyroid tissues, ectopic gland and even brown tumors [8]. SPECT/CT acquisition plays a critical diagnostic role in discriminating and localizing abnormal tracer activity [9]. Herein, we report a patient presented with severe hypercalcemia, subsequently find to have a cystic atypical parathyroid adenoma along with mediastinal uptake, which was confined to a sternal lytic lesion (suggestive of brown tumor), which highlights the importance of SPECT/CT in [^{99m}Tc]Tc-MIBI imaging for localization of the parathyroid adenoma.

CASE PRESENTATION

A 39-year-old man with history of fatigue, mood swings and renal stone (Calcium Oxalate containing) in the past four years, presented with generalized musculoskeletal pain, polyuria and cognitive impairment with no other remarkable contributing personal or family history. Laboratory data revealed PTH level ranging from 866 to 1300 (pg/mL), calcium 14.5 (mg/dL), phosphorus level (2.2 mg/dL) and alkaline phosphatase 233 (IU/L). Additional lab data for evaluating MEN syndrome (e.g., prolactin, calcitonin and pheochromocytoma profile) were unremarkable. The patient was supposed to have pHPT thus BMD (Bone Mineral Density) measurement by DXA (dual-energy x-ray absorptiometry) was done and confirmed osteoporosis (lowest Z-score: -5.0). On physical examination, a thyroid nodule in the lower pole of the left thyroid lobe was palpated. Cervical ultrasound (US) showed cystic focus (22×16×46 mm) with suspicious microcalcification adjacent to the lower pole of the left thyroid lobe extending to the thoracic inlet. In view of clinical assumption of parathyroid adenoma and since the patient was a candidate for minimally invasive surgery, a [^{99m}Tc]Tc-MIBI scan was performed for further evaluation. The scan findings in planar images

revealed an area of tracer uptake in the mediastinum, which was highly suspicious for an ectopic mediastinal parathyroid adenoma (Figure 1).

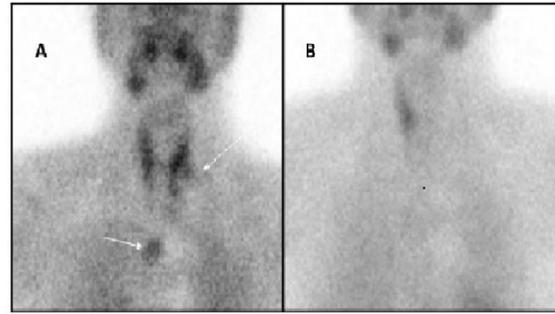


Fig 1. (A) Planar [^{99m}Tc]Tc-MIBI images depicted an area of tracer uptake in the mediastinum (solid arrow) with a focal zone of absent tracer activity in the lower pole of the left thyroid lobe (dotted arrow). (B) Six-month Follow-up scan showed no abnormal mediastinal uptake with evidence of left thyroid lobectomy.

In order to localize the exact location of the presumed ectopic parathyroid adenoma, SPECT/CT was performed from the cervico-thoracic region and a focal zone of absent tracer activity was noticed in the lower pole of the left thyroid lobe, which thought to be due to a [^{99m}Tc]Tc-MIBI cold thyroid nodule (Figure 2A). In addition, a focus of radiotracer uptake was surprisingly confined to a well-defined lytic lesion in the manubrium sterni (Figure 2B).

There was no other abnormal increased tracer activity throughout the neck and mediastinum in the SPECT/CT images. Due to severe hypercalcemia, the patient was admitted for proper management with calcitonin, IV infusion of normal saline hydration with addition of Pamidronate to achieve better treatment response. Considering patient history and paraclinical findings specially thyroid ultrasonography, the patient underwent left thyroid lobectomy by resection of cystic mass.

The pathology report was consistent with atypical parathyroid adenoma. After the lobectomy, calcium and PTH levels dropped to 8.5mg/dL and 160 pg/mL, respectively. After a six-month follow-up, he remained in continuous remission and the follow-up parathyroid scan (Figure 2C and 2D) revealed no uptake within the sternal lesion with evidence of osteoblastic reaction suggesting proper resolution of the assumed sternal brown tumor.

DISCUSSION

Parathyroid cysts are classified in two categories, functional and non-functional. Approximately 9% of parathyroid cysts entitled functional cystic adenoma [10]. Thus cystic parathyroid adenoma, particularly with atypical features, reflects a rare cause of pHPT [11].

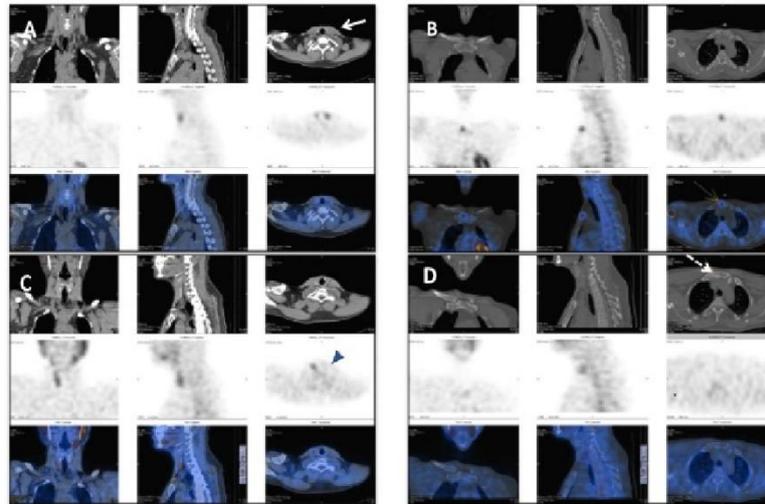


Fig 2. [^{99m}Tc]Tc-MIBI Hybrid SPECT/CT from thoraco-mediastinal region; (A) Showed focal photon-deficient area (white solid arrow) in the lower pole of the left thyroid lobe. (B) depicted a well-defined lytic lesion in the manubrium of sternum with uptake (colored solid arrow). (C, D) Six-month Follow-up [^{99m}Tc]Tc-MIBI scan revealed no uptake within the sternal lesion with evidence of osteoblastic reaction (arrow head, white dotted arrow).

The presumed pathogenesis of atypical cystic lesions in parathyroid is believed to be due to cystic degeneration of primary solid adenoma [12].

Although, pre-operative localization of parathyroid adenoma is still challenging and crucial to decrease surgical complications, a combination of SPECT/CT with planar [^{99m}Tc]Tc-MIBI scintigraphy can improve the accuracy of the scan [8, 13].

To the best of our knowledge, there are few cases of atypical cystic parathyroid adenoma with severe hypercalcemia and brown tumors in the literature.

This case highlights the importance of SPECT/CT acquisition. Performing SPECT/CT played a key role in the management of the patient. Since suspicious uptake in the planar images could be interpreted as an ectopic parathyroid gland in the mediastinum without further imaging, which is known as the most common place for ectopic parathyroid adenomas [14]. In our case, the activity in the mediastinal region localized to the brown tumor in the sternum, which was confirmed by application of SPECT/CT images, changed the surgical plan.

This case also illustrates the possibility of detecting cold parathyroid adenomas, which is not frequently encountered. Clinical and laboratory presentation of our patient made us to seek thoroughly for a responsible adenoma of the parathyroid gland and resulted in finding an atypical cystic parathyroid adenoma (despite being [^{99m}Tc]Tc-MIBI negative on the scan).

CONCLUSION

Technetium-99m methoxy isobutyl isonitrile ([^{99m}Tc]Tc-MIBI) scintigraphy has favorable detection rate for ectopic gland as well as the brown

tumors. SPECT/CT plays critical role in localizing abnormal tracer uptake.

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