

# A Rare Case of Primary Hyperparathyroidism due to Ectopic Parathyroid Adenoma

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## ABSTRACT

Primary hyperparathyroidism (PHPT) is the most frequent cause of hypercalcaemia. Surgical intervention with parathyroidectomy is the only curative option. Ectopic parathyroid adenomas (EPA) are challenging to localise before surgery and may result in persistent PHPT if not localised properly before surgery. A case of PHPT due to EPA located in the mediastinum, more specifically in the thymus gland, which was removed successfully with video-assisted thoracoscopic surgery (VATS) is hereby presented. A 16-year female patient was admitted to the endocrinology unit. She was referred by a general physician who noted abnormalities in an x-ray of her lower limbs, showing brown tumours, which were suggestive of PHPT. The 99m Tc-MIBI scintigraphy with Single Photon Emission Computed Tomography (SPECT) was suggestive of EPA in the mediastinum. Following surgery, there was a significant decline in serum calcium and parathyroid hormone levels. Postoperative hypocalcaemia was corrected with alpha-calcidol and calcium carbonate.

**Key Words:** *Primary hyperparathyroidism, Parathyroid adenoma, Video-assisted thoracoscopic surgery, Technetium Tc 99m sestamibi MIBI scan, Single photon emission computed tomography.*

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## INTRODUCTION

Primary hyperparathyroidism (PHPT) is caused by excessive secretion of parathyroid hormone (PTH) due to parathyroid adenoma (PA), glandular hyperplasia, or very rarely parathyroid carcinoma. The biochemical profile of PHPT includes hypercalcaemia and elevated or inappropriately normal PTH levels.<sup>1</sup> The treatment of PHPT is mainly surgical when indicated<sup>2</sup> and consists of the excision of the particular over-secreting parathyroid gland.

In around 6 to 16% cases of PHPT, hyper-functioning parathyroid glands are ectopically located.<sup>3</sup> Ectopic PAs (EPAs) arise from migratory abnormalities during embryogenesis.<sup>4</sup> The origin of the inferior parathyroid glands is from the third brachial arch, from which the thymus also originates. They accompany the thymus gland during its downward migration. Thus, abnormalities in their descent may result in their ectopic position anywhere in the mediastinum. Mediastinal PAs constitute 20% of all EPAs.<sup>5</sup>

EPAs are difficult to properly localise before surgery, thus making surgical cures difficult on the very first attempt.<sup>6</sup> Neck ultrasound (US) and Technetium-99 methoxy-isobutyl-isonitrile (99mTc-MIBI or Tc-sestamibi) scans are the first-line imaging modalities but have low sensitivity and specificity.<sup>7</sup> Ectopic PAs are better visualised with Tc-sestamibi, Single-Photon, Emission Computed Tomography (SPECT), combined with computerised tomography (Tc-sestamibi SPECT/CT).<sup>8</sup>

## CASE REPORT

A 16-year girl was referred to the Endocrinology Department at the Lady Reading Hospital, Peshawar, Pakistan. She was referred by a general physician, to whom she presented with generalised aches and pains, in particular, pain in both knees. Brown tumours were noted in her knee x-ray at that point.

At the endocrinology department, physical examination was otherwise unremarkable except for pallor. She had no palpable mass in her neck. Laboratory workup showed calcium of 2.96 mmol/l (NR = 2.2-2.6 mmol/l), intact PTH of 138 pmol/l (NR = 1.0 - 6.9 pmol/L), and 25-hydroxy Vitamin D 35 nmol/l (Adequate: 50-75 nmol/l). A repeat x-ray of both knee joints demonstrated a brown tumour in the distal end of the right femur and the proximal end of the left tibia (Figure 1).

The US of the neck, abdomen, and pelvis showed no significant abnormality. CT scan of the neck and chest was reported as generalised osteopenia, mixed lytic, and sclerotic lesions with bony expansion bilaterally in multiple ribs both anteriorly and posteriorly with associated rib cage abnormality. A similar lesion was also noted in the scapula and right humeral head.

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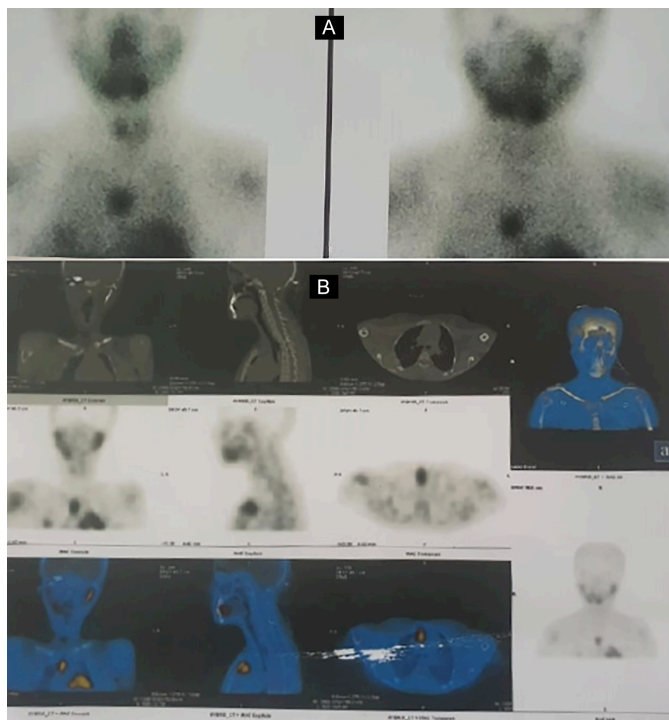
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**Figure 1: X-ray of both the knee joints.**



**Figure 2: (A) 99mTc-MIBI showing a possible ectopic parathyroid adenoma in the mediastinum and (B) Tc-sestamibi SPECT/CT showing tracer retention in upper mediastinum suggestive of parathyroid adenoma.**

The 99mTc-MIBI revealed a focal area of increased tracer uptake in the central region of the upper chest in the early phase. The rest of the thyroid gland showed more or less uniform tracer uptake. The delayed image showed persistence of focal area of increased tracer uptake in the region of the central mediastinum (Figure 2A). Tc-sestamibi SPECT/CT images showed tracer retention in the upper mediastinum, behind the manubrium, and slightly towards the left side of the central mediastinum, suggestive of PA (Figure 2B).

After a multidisciplinary team consultation among the departments of endocrinology, thoracic surgery, and radiology, the patient underwent VATS. Ectopic nodule excision with partial thymectomy was performed. After successful surgery, the patient complained of symptoms suggestive of hypocalcaemia such as paresthesia in the perioral region and extremities. This was associated with the dropping of serum calcium from preoperative level of 2.9 mmol/l to postoperative level of 1.9 mmol/l. Postoperative PTH was 12.4 pmol/L. A surgical cure was thus

achieved but along with hungry bone syndrome, most likely secondary to hypovitaminosis D. The patient was started on oral calcium carbonate with alpha-calcidol and intravenous Calcium Gluconate. Her serum calcium normalised after two days and the patient was safely discharged. Histopathology report received later suggested that the specimen was compatible with PA with adjacent benign thymic tissue.

## DISCUSSION

In clinical practice, there are various clinical scenarios in which EPA must be taken into consideration. These include patients with PHPT in whom either preoperative imaging is suggestive of an EPA, or the initial preoperative imaging is unable to identify a potential PA, or after parathyroidectomy, if there is persistent PHPT.<sup>6</sup> The present patient belonged to the first category of these three clinical scenarios.

Mediastinal PAs (MPAs) pose a significant challenge in both diagnosis and management. US is a very common imaging technique used for preoperative localisation of the usual PAs. However, because of the technical limitations of deploying the US in the mediastinum, it has a very low sensitivity for the identification of EPAs located in this region. In a previous study,<sup>3</sup> only 58% of retro-oesophageal and 48% of thymic EPAs, which were later surgically confirmed, were identified using US. Preoperative Sestamibi / 99mTc-MIBI has shown a sensitivity of around 89% and a positive predictive value (PPV) of approximately 90% for localising EPAs.<sup>3</sup> However, the limitation of sestamibi scan in EPAs is to miss PA that resides in a retro-pharyngeal location.<sup>8</sup> SPECT/CT has the advantage of combining the utilities of anatomical and functional imaging, thus improving sensitivity for localising EPAs. A study done in the United States on patients with PHPT which compared 99mTc-MIBI, SPECT, and SPECT/CT showed that SPECT/CT was superior in accurate identification of PAs (including EPA) as compared to SPECT (100% vs. 61%) in terms of their presence and location.<sup>9</sup> Wimmer *et al.* in their study reported that SPECT/CT was superior to SPECT in terms of exactly localising the abnormally functioning parathyroid gland (86% vs. 43%).<sup>10</sup> As mentioned earlier, there was a clinical suspicion of PHPT in the present patient because of radiological findings in her initial x-ray. The next step taken after biochemical confirmation of PHPT and a negative US of the neck was utilising 99mTc-MIBI combined with SPECT/CT. This approach helped in localising a mediastinal EPA in the patient confidently without causing a delay in the diagnosis.

VATS is an ideal choice of surgery for EPAs as it gives the benefits of minimally invasive surgery.<sup>4</sup> The benefits include reduced surgical trauma, morbidity associated with the procedure, and reduced hospital stay along with superior cosmetic results.<sup>11</sup> During the procedure, the surgeon did a partial thymectomy as the lesion was localised to the thymus gland in the preoperative imaging. In EPAs related to inferior parathyroid glands, the thymus has the highest frequency (31 - 38%) of harbouring the ectopic gland.<sup>3</sup>

In this case, the EPA even showed up on a planar Sestamibi scan. However, if there is any clinical scenario which is compatible with PHPT (high calcium and PTH levels) but the initial imaging does not show a PA in the neck, it should alert the physician to look for an ectopic source by combining different imaging modalities. SPECT/CT is a relatively cost-effective option in resource-limited countries such as Pakistan.

# PATIENT'S CONSENT:

Written informed consent was taken from the patient to publish the data concerning this case.

# COMPETING INTEREST:

The authors declared no conflict of interest.

# AUTHORS' CONTRIBUTION:

NW: Concept, design, interpretation of data, analysis, and drafting of the manuscript.

IA: Revision of the work critically.

SQ: Acquisition of data.

All authors approved the final version of the manuscript to be published.

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