

## CASE REPORT

# A rare case of mediastinal functioning parathyroid adenoma removed successfully with thoracoscopy

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## Abstract

We report here a rare case of primary hyperparathyroidism that was associated with an ectopic parathyroid adenoma located in the mediastinum. A 66-year-old woman suffering from primary hyperparathyroidism had been followed-up on an outpatient basis for over 10 years. She suffered from persistent urolithiasis and osteopenia due to hypercalcemia. After technetium-99-sestamibi ( $^{99m}\text{Tc}$ -MIBI) scintigraphy revealed an ectopic adenoma in the superior mediastinum, thoracoscopic resection of the tumor was performed. Subsequently, her serum parathyroid hormone (PTH) level decreased dramatically and her serum calcium concentration was restored to normal. Two years following surgery, her serum PTH and Ca levels remain stable.

## INTRODUCTION

Successful resection of mediastinal tumor is usually achieved with precise localization and estimation of the extension or invasive nature of the disease. Recently, video-assisted thoracoscopic surgery (VATS) is preferred to thoracotomy or sternotomy because it provides a magnified view with small skin incision and less pain.

Functioning adenoma is rare among mediastinal tumors. Due to its relatively small size and benign nature, surgeons often hesitate to operate. Therefore, accurate preoperative evaluation, histological confirmation during surgery and medical management for postsurgical complications are all essential for its successful treatment.

Here, we report a case of primary hyperparathyroidism due to a mediastinal parathyroid adenoma that was managed successfully by thoracoscopic resection. In this case, detailed preoperative information and magnified visual fields through thoracoscopy led to correct tumor identification during surgery,

despite dense adhesions due to pleurisy. Thus, VATS resection should be the recommended treatment for functioning mediastinal parathyroid adenoma.

## CASE REPORT

A 66-year-old woman with hypercalcemia had been followed-up on an outpatient basis for over 10 years. Her first presentation was abdominal pain due to ureteral stones, and she suffered from persistent urolithiasis and osteopenia afterwards. She had never smoked and had a medical history of appendicitis and pleuritis. Although her serum parathyroid hormone (PTH) level was high, cervical sonography and technetium-99-sestamibi ( $^{99m}\text{Tc}$ -MIBI) scan were normal at the first visit, and under the diagnosis of primary hyperparathyroidism, she had been under long-term treatment for her symptoms. Ten years after the initial visit, re-examination by  $^{99m}\text{Tc}$ -MIBI scan revealed a hot spot in the mediastinum (Fig. 1). Chest computed tomography (CT)

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and magnetic resonance imaging (MRI) confirmed a small nodule adjacent to her brachiocephalic artery (BCA), suggesting an ectopic PTH-secreting adenoma located in the superior mediastinum (Fig. 2). Because a transcervical approach was deemed impractical for surgical resection, she was referred to our department of thoracic surgery.

Preoperative evaluations of the patient were as follows: serum calcium concentration, 11.0 mg/dl (normal; 8.8–10.2 mg/dl); serum PTH level, 95 pg/ml (normal; 10–65 pg/ml); bone mineral ratio, ~75% of young adult mean; and estimated glomerular filtration rate (suggestive of creatinine clearance) from serum creatinine level, 51 ml/min (normal; >90 ml/min). Abdominal CT revealed right hydronephrosis with multiple ureteral stones (Fig. 3). She had hematuria with slight abdominal pain. Physical examinations were otherwise unremarkable.

Thoracoscopic tumor resection under general anesthesia with one-lung ventilation was performed. After dissociation of diffuse pleural adhesion due to a history of pleuritis, the

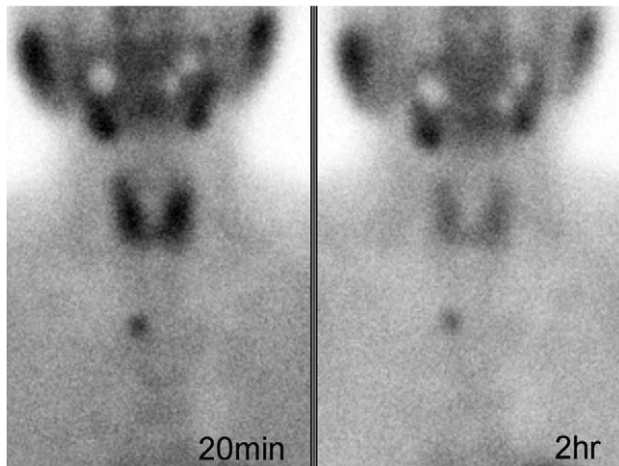


Figure 1: Technetium-99-sestamibi ( $^{99m}\text{Tc-MIBI}$ ) imaging showing focal uptake during early (left) and delayed (right) phases in the mediastinum.

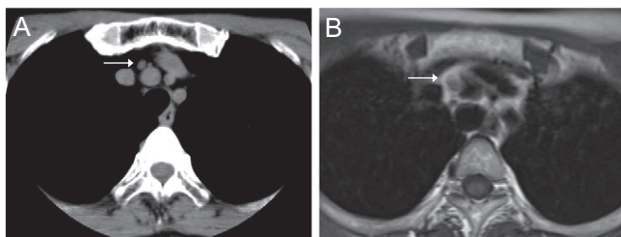


Figure 2: Chest CT (A) and MRI (B) revealed a mediastinal nodule adjoining the BCA.

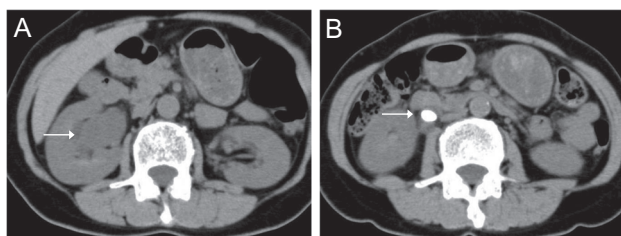


Figure 3: Abdominal CT indicated right hydronephrosis (A) and ureteral stones (B).

mediastinal pleura was excised and the right brachiocephalic vein (BCV) was identified and taped. A small nodule, 7 mm in diameter, was found between the right BCV and BCA, and was subsequently excised with surrounding fat tissue (Fig. 4). The diagnosis of ectopic adenoma was confirmed with intraoperative pathological consultation.

Her serum PTH concentration was monitored serially, which revealed a dramatic reduction immediately after tumor removal. Her serum PTH levels before surgery, and at 10 min, 20 min and 1 day after tumorectomy were 95, 14, 13 and 28 pg/ml, respectively (Fig. 5).

The patient experienced transient postoperative hypocalcemia (POD2: 7.5 mg/dl) without any clinical manifestations which was corrected with oral calcium supplementation for 4 days. The postoperative clinical course was otherwise unremarkable, and she was discharged 9 days after surgery. Histopathological examination confirmed parathyroid adenoma within adipose tissue with remains of thymic gland tissue (Fig. 6).

The patient subsequently underwent extracorporeal shock wave lithotripsy for the remaining ureteral stones and was given monthly administrations of oral bisphosphonates. Her abdominal pain disappeared, and serum calcium/PTH levels have been stable 2 years following surgery.

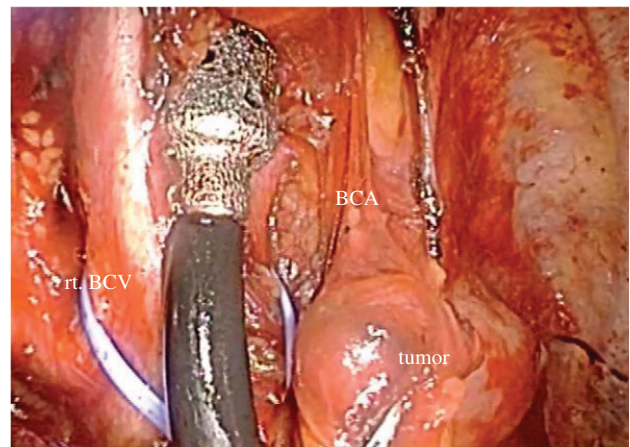


Figure 4: Thoracoscopic view of the mediastinal tumor.

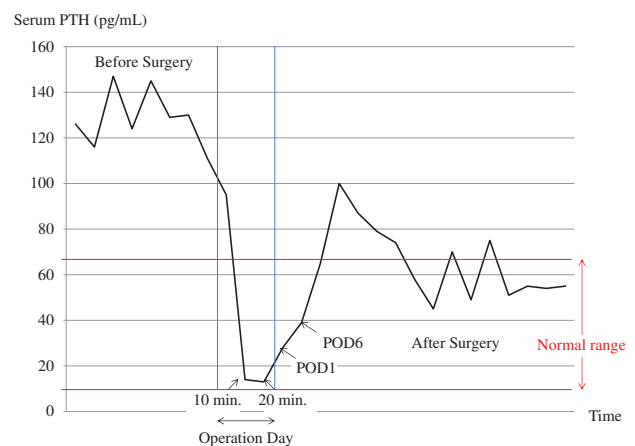
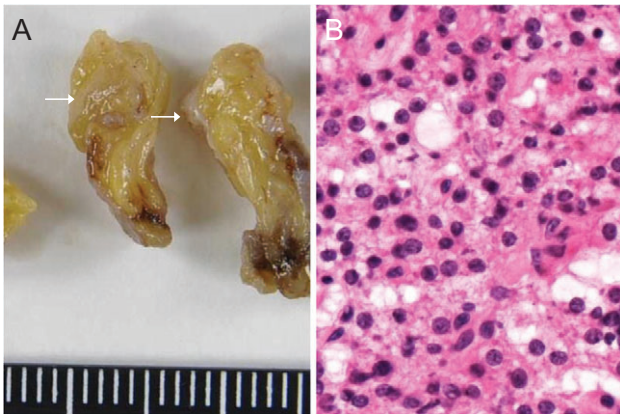


Figure 5: Serum PTH concentrations before and after surgery. After the immediate reduction during surgery, serum PTH level returned to normal range.



**Figure 6:** Macroscopic (A) and microscopic (B) views of the nodule confirmed the diagnosis of ectopic parathyroid adenoma.

## DISCUSSION

Hormone-secreting parathyroid adenoma is a common cause of hypercalcemia. Combinations of symptoms classically described as ‘stones, bones, moans, groans and psychic overtones’ are rarely observed, whereas usual clinical manifestations are ureteral stones and bone mineral loss [1]. Persistent risk of intractable urolithiasis or osteoporosis and cost of medical intervention favor surgical removal of the tumor.

Primary parathyroidism usually results from excessive secretion of PTH from the cervical parathyroid gland. In contrast, ectopic parathyroid adenomas can be located in any part of their embryonic migratory path [2]. Among them, functional ectopic mediastinal parathyroid adenomas are relatively rare which account for 1–2% of patients undergoing surgical intervention [3].

The precise localization of adenoma is vital for accurate diagnosis. Imaging modality includes cervical ultrasound, CT, MRI and parathyroid scintigraphy. Whereas high-resolution sonography is a reliable method for localizing cervical parathyroid glands,  $^{99m}\text{Tc}$ -MIBI scanning is the gold standard for initial discovery of ectopic adenomas. Other imaging techniques include positron emission topography CT/MRI and dual energy CT [4].

Management of hyperparathyroidism depends on clinical manifestations. Criteria for surgery in asymptomatic primary hyperparathyroidism suggested by the latest guidelines include: (i) serum calcium  $>1$  mg/dl above upper limit of normal; (ii) markedly reduced bone density or presence of vertebral fracture on spine imaging; (iii) creatinine clearance  $<60$  ml/min, presence of nephrolithiasis or nephrocalcinosis, or marked hypercalciuria ( $>400$  mg/dl) and (iv) under 50 years of age. Patients need to meet only one of these criteria to be recommended for surgery [5].

In general, surgical approach for mediastinal tumor depends on its location and extension, including cervical approach, thoracotomy, and median sternotomy. Thoracoscopy provides magnified visual fields with small skin incision, which is especially useful for localization of small nodules [6, 7].

PTH monitoring during operation is valuable as serum PTH concentration declines immediately after tumor resection [8]. Intraoperative pathological confirmation is also helpful because surrounding fat/thymic tissue or mediastinal lymph nodes can make small adenomas indistinguishable. Although postoperative hypocalcemia is reported to be mild and asymptomatic, close follow-up of serum calcium levels is also important. Oral calcium tablets may be required to prevent severe hypocalcemia.

In conclusion, a case of hyperparathyroidism due to an ectopic PTH-secreting adenoma in the mediastinum was successfully treated by thoracoscopic tumor resection.  $^{99m}\text{Tc}$ -MIBI scanning was useful in the initial detection of the adenoma. Intraoperative PTH level decreased immediately after tumor removal and has been stable thereafter. Precise localization, adequate preoperative evaluation, appropriate surgical approach and management of pre-/postoperative clinical manifestations are all essential for successful management of this disease.

## CONFLICT OF INTEREST STATEMENT

None declared.

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