

Intrathymic Parathyroid Adenoma

Abdelrahman Mahmood Radaideh, Duraid Jaradat, Fares Haddad

Department of Medicine, Division of Endocrinology, Department of Radiology, Princess Basma Teaching Hospital, Irbid, Jordan and King Hussein Medical Center, Amman, Jordan

ABSTRACT

The most common cause of primary hyperparathyroidism is parathyroid adenoma. Up to 20% of parathyroid adenomas has been reported to be in ectopic localization. Ectopic parathyroid glands can also become adenomatous, causing a diagnostic dilemma when they are difficult to localize preoperatively.

We report a case of intrathymic parathyroid adenoma that was localized preoperatively by Technetium MIBI SPECT and treated surgically via assisted Thoracoscopic surgery and developing hungry bone syndrome after surgery. (Rawal Med J 2013;38:314-316).

Key words: Primary hyperparathyroidism, parathyroid adenoma, Intrathymic.

INTRODUCTION

Primary hyperparathyroidism (PHPT) is the most common cause of hypercalcemia in patients seen in outpatient clinics.¹ There is excessive secretion of parathyroid hormone.² PHPT affects people of any age; however those older than 50 years of age and post menopausal women are more commonly affected.¹ 85-90% of PHPT is caused by parathyroid adenoma and 5-15% by parathyroid hyperplasia.^{1,2} Parathyroid carcinoma accounts for less than 1% of PHPT.^{1, 3} Ectopic location of adenomatous parathyroid tissue as a cause of PHPT accounts for 11-25% of all patients with PHPT.^{1,2}

Ectopic locations are related to the migratory pathways of embryologic tissue to the adult position.⁴ Sites of ectopic localization include thymus, tracheoesophageal groove, carotid sheath, intrathymic and paraesophageal location.⁵ The most frequent location of the ectopic parathyroid adenomas is the mediastinum and 70% of the ectopic parathyroid adenomas are located in the thymus.⁴

The treatment consists of surgical excision via cervical incision. In 2% of surgical candidates, the ectopic parathyroid tissue is not accessible via this approach as the gland would be deeply embedded in the superior aspect of the anterior or posterior mediastinum and in closed within the thymus. These cases have been treated with median sternotomy, thoracotomy and less invasively via video assisted

thoracoscopic surgery (VATS).²

Here we report a case of intrathymic parathyroid adenoma that was treated with VATS. The informed consent was obtained for publication.

CASE PRESENTATION

A 41-year-old female complained of general bone pain and lethargy with inability to stand up from sitting position unaided for over a year. She was referred after she was found to have high PTH 1075(15-65 pg/ml), high ALP 2190(0-240 units/L), normal calcium 2.47(2.1-2.6 mmol/L) and low phosphorus 0.58(0.8-1.8mmol/l) in March 2010. She had polyuria, polydipsia and loin pain. She is married with 3 children. General physical examination was unremarkable. Vital signs were stable. Blood pressure: 100/60 mm Hg, Pulse rate: 80 BPM, she was afebrile. Weight: 55 kgs, Height: 158 cm. Systemic examination showed no goiter and lymph node enlargement. Normal cardiovascular system, chest and abdomen. Neurological examination showed severe proximal muscle weakness, with power 3/5 and waddling gait. ECG showed no abnormalities. Laboratory examination showed high levels of intact parathormone (iPTH) with high normal calcium (CA++) and low phosphate (Po4). Urea was 2.6 mmol/l and creatinine 56 umol/l. CBC and thyroid Function test were normal. FBS was 93mg/dl. Vit.D3 level was 18 (20-40 ng/ml). Liver Function

test and urine were normal. Celiac disease was excluded by doing Anti-Tissue Trans glutaminase IgA, IgG and Anti-Endomysial IgA and IgG Abs which were negative. Upper endoscopy and distal intestinal biopsy were normal.

Bone x-ray of both hands showed periosteal absorption of the phalangeal bones. (Figure 1) DEXA Scan T-Score -2.5 confirmed osteoporosis. INM whole body scan was consistent with metabolic bone disease. NM parathyroid isotope scan no scintigraphic evidence of parathyroid adenoma. Neck CT- Scan showed 5mm low attenuation area in the posterior thyroid gland representing small cystic degeneration with thyroid gland. As the patient has clinical and biomedical picture of severe primary hyperparathyroidism associated with low normal Vit. D3 and normocalcemia further investigations were done to localize the parathyroid adenoma.

Neck and chest CT scans showed that the thymus gland appeared prominent regarding the age with small hypodensity within it which was suggestive of parathyroid adenoma. (Figure 2) To confirm the presence of adenoma, Sismibi SPECT-Scan was done which showed moderately increased uptake in the mid to lower mediastinum toward the right side with delayed retention strongly suggestive of mediastinal parathyroid adenoma (Figure 3).

The patient was referred to another institution for surgery. Investigations before surgery were as follows: PO_4^- 2.0 (2.7-4.5mg/dl), Ca^{++} 10.6(8.4-10.4mg/dl), Albumin 45 (34-48 g/l), ALP 1783 (0-240units/l), PTH>2500(15-65 pg/ml), CBC, blood sugar KFT,TFT and electrolytes were within normal limits. Ultrasonic of abdomen was normal. Repeat chest CT Scan showed 30 x 24 x 20mm, low attenuation mass in the right side of anterior mediastinum at the level of pulmonary trunk in contact with pericardium, which suspected ectopic parathyroid adenoma (Figure 2).

On 04 April 2010, under general anesthesia, Video Assisted Thoracoscopy (VAT) was done and a right thymic cyst was detected. She underwent VAT Thymectomy. She was given $CaCO_3$ 600mg tid and alfacalcidol(one alfa)1mcg supplementation to

avoid developing hungry bone syndrome. On the second day of surgery, she complained of perioral and all extremities parasthesia and numbness. Positive Trousseau's and Chovestek's signs were noted with drop of serum Ca^{++} 7.9 (8.6-10.6). She was treated with IV Calcium replacement and was also kept on oral Calcium supplementation. Ca^{++} values during hospitalizations were as follows: 7.9, 7.3, 7.7, 7.8, 8.0, 8.2 and 8.6mg/dl. PTH measurement postoperatively decreased from >2500 to 48.6 pg/mL.

On pathological examination, three congested pieces of soft tissue were found. They weighed 41 grams all together with the largest piece contained a 5 x 3 x 2cm cystic lesion filled with blood clots. Sections revealed an intrathymic parathyroid adenoma. It was composed of chief cells in solid and trabecular patterns, cystic degeneration and hemorrhage were also seen. There was no evidence of invasion. Ki67 by immunohistochemistry was 0-1%. The surrounding thymic tissue was unremarkable (Figure 4).

The patient was discharged on 20 April 2010 with good general condition. She did not show up for follow up. One and half year later, she was recalled for visit by myself on 13 September 2011. She was asymptomatic, healthy without any treatment. All lab investigations were normal. DEXA-Scan was normal. She does not need for further follow up.

DISCUSSION

Ectopic parathyroid adenomas are frequently located in thymic horns intrathymic.⁶ In most cases, they can be removed via cervicotomy. In only 2% of patients, intrathymic adenomas are located deeply in the mediastinum and require thoracic approach.⁶ The embryological origin of parathyroid glands is the endodermis of third and fourth pharyngeal pouches. From there, these glands migrate to their usual position behind the thyroid gland. If migration proceeds too far, the parathyroid may be found in cervical thymic horns, or inside or along the intrathoracic thymus.⁶

These anatomical variations are well known to surgeons who choose to approach ectopic

parathyroid glands by sternotomy, thoracotomy or thoracoscopy when preoperative imaging demonstrates the gland to be located deeply in the mediastinum.^{6,7} The accuracy of CT-Scan and CT-Scan MIBI-Scan now allow the location of the parathyroid adenoma within the thymus to be prechecked.^{3,6,7,8}

In this case, the patient has an overt picture of hyperparathyroidism, clinically and biochemically. The initial SISTAMIBI-Scan which was done did not reveal the location of the parathyroid adenoma. The patient underwent neck and chest CT-Scan which raised the suspicion of mediastinal parathyroid adenoma because the thymus gland was large for her age. SISTAMIBI-SPECT was proceeded and ectopic mediastinal adenoma intrathymic was detected. For further confirmation, neck and chest MRI were done.⁵

The patient underwent VAT and adenoma was not found but there was a visible right thymic cyst, as mediastinal parathyroid glands are more commonly an intrathymic than parathymic.⁹ Postoperative measurement of PTH showed a drop and as surgery was successful the drop of PTH >50% was from the pre-excisional level.^{3,6} Postoperative clinical picture was suggestive of hungry bone syndrome that may develop after abrupt decrease of PTH level toward normal which causes a decrease in bone resorption as well as increase in bone formation and the uptake of minerals such as calcium, phosphate and magnesium. The degree of fall in serum calcium level is related to preoperative bone turnover and the degree of PTH level.^{10,11}

Intrathymic location of parathyroid adenoma was described in 1941 and since that time the experience and the new surgical techniques were accumulated.¹² In only 2% of patients intrathymic adenomas are located deeply in the middle mediastinum and require thoracic approach,⁵ as in our patient. This is due to the parathyroid gland and thymus migration during fetal formation.¹³ In conclusion, this case illustrates how to approach ectopic located parathyroid adenoma. We have to localize and excise these kinds of adenoma by lowest invasive technique using new methods of imaging and surgery.

Author Contributions:

Conception and design: Abdelrahman Mahmood Radaideh
 Collection and assembly of data: Abdelrahman Mahmood Radaideh, Duraid Jaradat, Fares Haddad
 Analysis and interpretation of the data: Abdelrahman Mahmood Radaideh
 Drafting of the article: Abdelrahman Mahmood Radaideh
 Critical revision of the article for important intellectual content: Fares Haddad
 Final approval and guarantor of the article: Abdelrahman Mahmood Radaideh, Duraid Jaradat, Fares Haddad
Corresponding author email: radaideha@yahoo.com
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