

Video Assisted Thoracoscopic Surgery (VATS) for Excision of an Ectopic Anterior Mediastinal Intra-Thymic Parathyroid Adenoma

DUKHABANDHU NAIK¹, KUMARADOSS FELIX JEBASINGH², RAMPRASATH³, GNANAMUTHU BIRLA ROY⁴, MAZHUVANCHARY JACOB PAUL⁵

ABSTRACT

Ectopic anterior mediastinal parathyroid adenoma is a rare cause of Primary Hyperparathyroidism (PHPT). Imaging studies such as Technetium-99m (^{99m}Tc) sestamibi parathyroid scan along with a Single Photon Emission Computerized Tomogram (SPECT), and contrast enhanced Computerized Tomogram (CT) of the neck and thorax can precisely localize the ectopic mediastinal parathyroid adenoma. We report a 40-year-old gentleman who presented with persistent pain in the right shoulder following trivial trauma. His biochemical investigations showed an elevated serum calcium of 11.6mg% (Normal: 8.3-10.4 mg%) along with an elevated iPTH of 1443 pg/ml (normal: 8-70 pg/ml) which were suggestive of primary hyperparathyroidism. The localization studies revealed an ectopic cystic parathyroid adenoma in the anterior mediastinum that was not accessible from the neck. He underwent a Video Assisted Thoracoscopic (VAT) excision procedure with normalization of serum calcium and an uncomplicated recovery. The VAT approach is a successful minimally invasive technique for mediastinal parathyroidectomy.

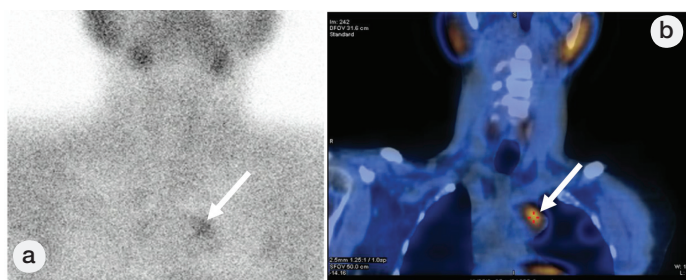
Keywords: Minimally invasive technique, Primary hyperparathyroidism, Sestamibi parathyroid scan

CASE REPORT

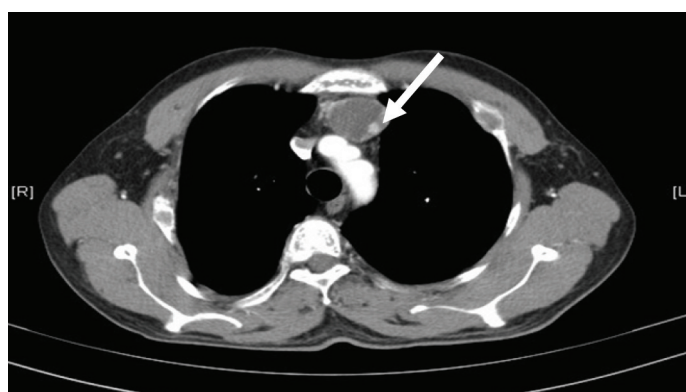
A 40-year-old male presented to the orthopedic department with complaints of pain in the both the lower limbs and generalized somatic pain since the past four years. Six months earlier, he had presented to the emergency department with shoulder pain following minor trauma when the radiographs of the shoulder region excluded a fracture; he defaulted orthopedic referral till the second visit when he was detected to have an elevated serum calcium level of 11.6mg% (Normal: 8.3-10.4 mg %) with normal Vitamin D level of 25ng/dl (normal 10-30ng/dl). He was referred to the Department of Endocrinology to evaluate the possibility of metabolic bone disease.

His biochemical investigations showed a persistently elevated serum calcium level with a raised Intact Parathyroid Hormone (iPTH) of 1443 pg/ml (normal: 8-70 pg/ml) and a low serum phosphorous of 2.0mg% (normal: 2.5-4.6 mg%) suggesting primary hyperparathyroidism. Further investigations revealed a low renal tubular maximum re-absorption rate of phosphate to glomerular filtration rate (TmPO₄/GFR) of 0.226mg/dl (normal: 2.6-4.4mg/dl), and bone mineral density showing osteoporosis (T-score at spine -Minus 4.3, Hip -Minus 3 and Forearm -minus 4.5). The ultrasonography of the abdomen did not reveal nephrocalcinosis or renal calculi. The ultrasound examination of the neck did not detect parathyroid enlargement. However, the Technetium-99m(^{99m}Tc)sestamibi parathyroid scan along with SPECT [Table/Fig-1a,b] and CT of the neck and thorax [Table/Fig-2] revealed an ectopic cystic parathyroid adenoma in the anterior mediastinum which appeared surgically inaccessible from the neck. He was assessed by the thoracic and endocrine surgical teams and it was decided appropriate to proceed with a VAT excision of the anterior mediastinal parathyroid adenoma.

The patient was positioned in the left lateral position with 30 degree tilt to the right using one lung ventilation. One 11mm camera port was placed in the 6th intercostal space in the mid axillary line. Under guidance, another 10mm port was placed in the 4th intercostal space in the anterior axillary line. A third 10mm

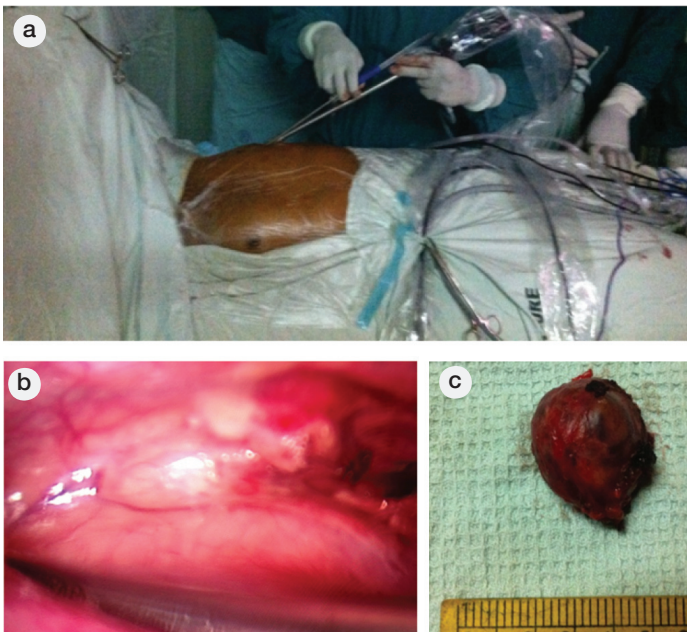


[Table/Fig-1a,b]: Showing an ectopic mediastinal parathyroid adenoma in ^{99m}Tc sestamibi parathyroid scintigraphy along with a Single Photon Emission Computerized Tomogram (SPECT) respectively (Indicated with the arrows).

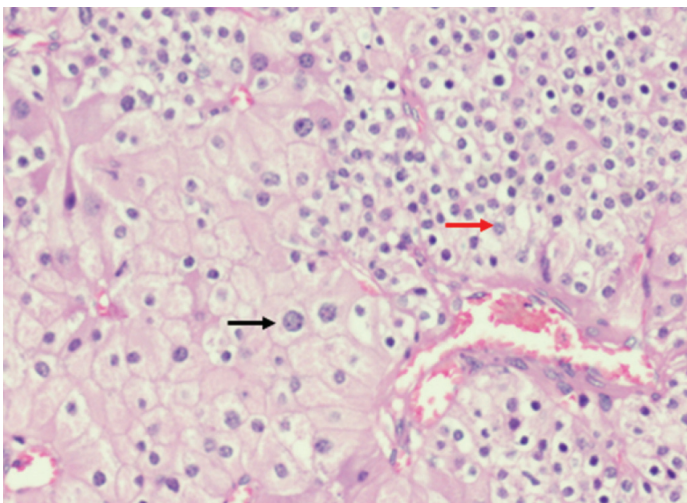


[Table/Fig-2]: Showing an ectopic mediastinal parathyroid adenoma in contrast enhancing Computerized Tomogram (CT) of the neck and thorax (Shown with an arrow).

port was placed in the 4th intercostal space in the posterior axillary line to permit access to the mediastinum without a CO₂ insufflation [Table/Fig-3a]. The mediastinal bulge of parathyroid tumour in the body of thymus was identified and the pleura incised to enter the mediastinum [Table/Fig-3b]. The tumour in the thymus was dissected all around and its vascular supplies were interrupted avoiding injury to the great vessels and phrenic nerve. The excised tumour was retrieved intact using an endo-bag [Table/Fig-3c]. The



[Table/Fig-3a-c]: (a) Showing operating position and transthoracic port positioning; (b) Showing endoscopic view of mediastinum; (c) Showing excised parathyroid tumour specimen.



[Table/Fig-4]: Haematoxylin and Eosin stain (200X magnification) showing chief cells (indicated with a red arrow) and focal clusters of oncocytic cells (indicated with a black arrow) suggesting parathyroid adenoma.

histopathological specimen was consistent with the parathyroid adenoma [Table/Fig-4]. After securing homeostasis, a 28F chest tube was placed and the wound was sutured. The patient was extubated at the end of the procedure and the recovery was uneventful. The drainage tube was removed after 48 hours and the patient was discharged. At the time of discharge, his corrected serum calcium level was 8.37mg % with intact PTH of 15.6 pg/ml, indicating a cure.

DISCUSSION

Parathyroid adenomas are the most common cause of primary hyperparathyroidism. Most parathyroid adenomas are located in the orthotopic position in close relation with the thyroid. However, approximately 10-20% of parathyroid adenomas are located ectopically [1]. The majority of ectopic parathyroid adenomas are located in the anterior mediastinum, in the thymus; other locations include para- or retro-oesophageal position, aorto-pulmonary window, along the carotid sheath and rarely intra-thyroidal. Ectopic parathyroid adenomas are the most frequent cause of a failed cervical exploration [2]. An accurate pre-operative localization is the key to successful surgery for an ectopic parathyroid adenoma. Modern imaging techniques such as ^{99m}Tc sestamibi parathyroid scan coupled with a SPECT, and CT neck and thorax will

precisely localize the ectopic parathyroid adenoma [3,4]. Magnetic resonance imaging, four dimensional CT scanning, Selective venous sampling or C11- Methionine PET can also be used to localize the tumour [5].

Resection of an ectopic Mediastinal Parathyroid Adenoma (MPA) is traditionally done through median sternotomy or thoracotomy. These conventional procedures are associated with a risk of injury to phrenic and recurrent laryngeal nerve, laceration of brachiocephalic vein, delayed wound healing, wound infection, mediastinitis and even death. The size of skin incisions is often large and disfiguring [6].

Prinz et al., first performed the thoracoscopic excision of MPA in 1994. Subsequently, several case series have reported VAT to be a feasible and safe approach for MPA with an overall cure rate of 98-100% [7]. Amer et al., reported seven cases of VATS resection of mediastinal parathyroid adenoma from Southampton General Hospital, UK between 2004 to 2009. The median hospital stay was two days and six out of seven patients had normalized serum calcium level. There was no mortality; however the surgical approach was converted to open thoracotomy in one patient on account of bleeding from the azygos vein due to excessive traction [8]. Tchervenjakov et al., reported the successful removal of parathyroid adenoma in four cases of ectopic MPA using VATS without any complications; all four cases had previous unsuccessful exploration through the cervical route [9]. Mohamed et al., (2013) reported the successful resection of parathyroid adenoma by the VAT procedure using radio-guided gamma probe combined with an intraoperative PTH testing in three cases of persistent hyperparathyroidism [10]. Similarly there are individual case reports of successful resection of MPA via VAT approach from Korea by Kim et al., Okagawa et al., from Japan and by Pecheva et al., from UK [11-13].

VATS was initially designed for diagnostic and therapeutic procedures of lungs, pleura and mediastinum such as biopsy, empyema irrigation, and pleurodesis and for foreign body removal. In the last three decades, VATS has become increasingly popular for the resection of several pathological lesions of the lung, pleura, mediastinum and even chest wall. Examples include extra lobar sequestration in children, lobectomy or segmental resection of tumours in primary lung malignancies, mediastinal lymph node resection, thymectomy for myasthenia gravis [14].

The VATS procedure has several advantages over median sternotomy or thoracotomy such as lesser invasiveness and pain, shorter operative time, better visualization of tumour and reduction in the duration of hospital stay. VATS also had a superior cosmetic result with a much smaller scar. In view of several advantages of VATS over the conventional procedures, it should be considered as the procedure of choice for anterior mediastinal parathyroid adenomas where surgical expertise is available. The rare side effects of VATS reported include hemorrhage, subcutaneous emphysema, recurrent pneumothorax and pulmonary oedema [15,16].

CONCLUSION

The ectopic mediastinal parathyroid adenoma is an uncommon cause of a rare disease that can be well localized using modern cross-sectional imaging. VATS is a safe and effective minimally invasive procedure for the resection of ectopic mediastinal parathyroid adenoma that is recommended where surgical expertise is available.

REFERENCES

- [1] Phitayakorn R, McHenry CR. Incidence and location of ectopic abnormal parathyroid glands. *Am J Surg.* 2006;191:418-23.
- [2] Richards ML, Thompson GB, Farley DR, Grant CS. Re-operative parathyroidectomy in 228 patients during the era of minimal-access surgery and intra-operative parathyroid hormone monitoring. *Am J Surg.* 2008;196:937-43.

- [3] Lavelly WC, Goetze S, Friedman KP, Leal JP, Zhang Z, Garret-Mayer E, et al. Comparison of SPECT/CT, SPECT, and planar imaging with single- and dual-phase (99m)Tc-sestamibi parathyroid scintigraphy. *J Nucl Med*. 2007;48(7):1084-89.
- [4] Giron J, Ouhayoun E, Dahan M, Berjaud J, Esquerre JP, Senac JP, et al. Imaging of hyperparathyroidism: US, CT, MRI and MIBI scintigraphy. *Eur J Radiol*. 1996;21(3):167-73.
- [5] Otto D, Boerner AR, Hofmann M, Brunkhorst T, Meyer GJ, Petrich, et al. Pre-operative localization of hyperfunctional parathyroid tissue with 11C-methionine PET. *Eur J Nucl Med Mol Imaging*. 2004;31(10):1405-12.
- [6] Hu J, Ngiam KY, Parameswaran R. Mediastinal parathyroid adenomas and their surgical implications. *Ann R Coll Surg Engl*. 2015 8;97(4):259-61.
- [7] Alesina PF, Moka D, Mahlstedt J, Walz MK. Thoracoscopic removal of mediastinal hyperfunctioning parathyroid glands: personal experience and review of the literature. *World J Surg*. 2008;32(2):224-31.
- [8] Amer K, Khan AZ, Rew D, Lagattolla N, Singh N. Video assisted thoracoscopic excision of mediastinal ectopic parathyroid adenomas: a UK regional experience. *Ann Cardiothorac Surg*. 2015;4(6):527-34.
- [9] Tchervenjakov P, Menon A, Milton R, Papagiannopoulos K, Lansdown M, Thorpe JA. Video-assisted mediastinoscopy (VAM) for surgical resection of ectopic parathyroid adenoma. *J Cardiothorac Surg*. 2007;2:41.
- [10] Mohamed SE, Li X, Khadra H, Saeed A, Mohamed H, Kandil E. Different surgical approaches in parathyroid adenoma resections. *Gland Surg*. 2013;2(4):227-29.
- [11] Kim YS, Kim J, Shin S. Thoracoscopic removal of ectopic mediastinal parathyroid adenoma. *Korean J Thorac Cardiovasc Surg*. 2014;47(3):317-19.
- [12] Okagawa T, Hiramatsu Y. Ectopic mediastinal parathyroid tumor resected by video-assisted thoracic surgery with intraoperative methylene blue infusion; report of a case. *Kyobu Geka*. 2014 ;67(3):255-57.
- [13] Pecheva M, Mahendran K, Kadlec J, Lofthouse M, Van Tornout. Mediastinal giant parathyroid adenoma-a minimally invasive mediastinal surgical approach for an emergency presentation. *Ann Cardiothorac Surg*. 2016;5(1):70-73.
- [14] Melfi FM, Fanucchi O, Mussi A. Minimally invasive mediastinal surgery. *Ann Cardiothorac Surg*. 2016;5(1):10-17.
- [15] Imperatori A, Rotolo N, Gatti M, Nardecchia E, De Monte L, Conti V, et al. Peri-operative complications of video-assisted thoracoscopic surgery (VATS). *Int J Surg*. 2008;6 Suppl 1:S78-81.
- [16] Jancovici R, Lang-Lazdunski L, Pons F, Cadot L, Dujon A, Dahan M, et al. Complications of video-assisted thoracic surgery: a five-year experience. *Ann Thorac Surg*. 1996;61(2):533-37.

PARTICULARS OF CONTRIBUTORS:

1. Associate Professor, Department of Endocrinology, Diabetes and Metabolism, Christian Medical College, Vellore, India.
2. Senior Registrar, Department of Endocrinology, Diabetes and Metabolism, Christian Medical College, Vellore, India.
3. Assistant Professor, Department of Thoracic surgery, Christian Medical College, Vellore, India.
4. Professor, Department of Thoracic Surgery, Christian Medical College, Vellore, India.
5. Professor and Head, Department of Endocrine Surgery, Christian Medical College, Vellore, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:

Dr. Mazhuvanchary Jacob Paul,
Professor and Head, Department of Endocrinology Surgery,
Christian Medical College Hospital, Vellore-632004, India.
E-mail: mjipaul@cmcvellore.ac.in

FINANCIAL OR OTHER COMPETING INTERESTS: None.

Date of Submission: **Dec 01, 2015**

Date of Peer Review: **Feb 11, 2016**

Date of Acceptance: **Apr 27, 2016**

Date of Publishing: **Jun 01, 2016**