

Primary Hyperparathyroidism due to Ectopic Mediastinal Parathyroid Adenoma**P Rajendra Prasad¹, Shrinivas B. Somalwar^{2*}, Neelaveni³**¹Associate Professor, Osmania Medical College/Osmania General Hospital, Hyderabad, Telangana State, India²Assistant Professor, Department of Pathology, Osmania Medical College/Osmania General Hospital, Hyderabad, Telangana State, India³ Associate Ptofessor, Department of Endocrinology, Osmania Medical College/Osmania General Hospital, Hyderabad, Telangana State, India***Corresponding Author:****Name:** Shrinivas B. Somalwar**Email:** somalwar73@gmail.com

Abstract: Parathyroid Adenoma is the commonest cause of primary hyperparathyroidism. Majority of adenomas arise from eutopically positioned parathyroid glands. However in 10% of the cases they can be from ectopic parathyroid glands. Aberrant migration during development may lead to ectopic locations of parathyroid glands. Proper pre operative localization is crucial for successful surgical outcome. We describe a case of primary hyperparathyroidism due to parathyroid adenoma in mediastinum, which was excised using sternotomy.**Keywords:** Parathyroid gland, Aberrant migration, Mediastinum, Parathyroid adenoma.

INTRODUCTION

Primary hyperparathyroidism (PHPT) is due to adenoma in majority of the cases. About 85% of these cases are due to eutopic adenoma [1] but multigland disease can occur in 10-15% of cases [2]. However in 10% of the cases these adenomas can be in ectopic position. The ineffectiveness of surgical exploration without preoperative localization is known [3]. Various imaging modalities like USG,CT,MRI,MIBI scintigraphy are commonly used [4], where as PET and SPECT with CT fusion imaging are performed in difficult cases [4-6], to localize parathyroid adenoma either eutopic or ectopic.

However use of TC99 MIBI scan with CT scan is sufficient in majority of the cases to localize ectopic adenoma [3, 7].

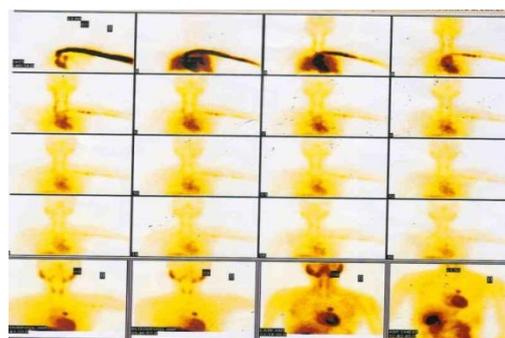
Here we are reporting one such case of primary hyperparathyroidism due to ectopic mediastinal adenoma. The purpose of this case report is to demonstrate the usefulness of preoperative imaging.

CASE REPORT

A 35 yrs old woman presented with symptoms of epigastric pain, occasional episodes of vomiting, muscle aches and bony pains of six years duration. She gradually developed anorexia and insomnia. She also had difficulty in walking and getting up from squatting position one year prior to presentation. She was admitted for these complaints elsewhere and was found to have hypercalcemia and renal calculi, hence had been referred to our centre for further evaluation and

management. She was not on any medication which can cause hypercalcemia like lithium or excess doses of vit D. None of the family members had similar problem. She was treated for pulmonary Koch's 8 yrs ago. Clinical examination revealed mild pallor, had no goiter, no other obvious neck swelling, no skeletal deformities. She was normotensive and except for painful proximal myopathy other systemic examination was normal.

Investigations- Routine biochemistry was normal except for anemia with hemoglobin of 9 gm/dl. Her serum calcium (corrected) 12.4 mg/dl, phosphorus-2.4 mg/dl, alkaline phosphatase-PTH-338pg/ml (15-65), 24 hr urinary calcium 424 mg. USG abdomen showed nephrocalcinosis, cholelithiasis, and no pancreatic calcifications. CECT chest revealed small enhancing lesion in anterior superior mediastinum, which was confirmed by TC99 sestamibi (Fig. 1).

**Fig. 1: Radionuclide imaging of the parathyroid glands**

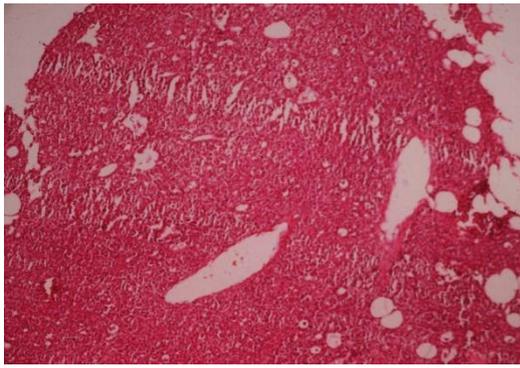


Fig. 2: Low power view of tumor tissue comprising of cribriform architecture and uniform population of cells

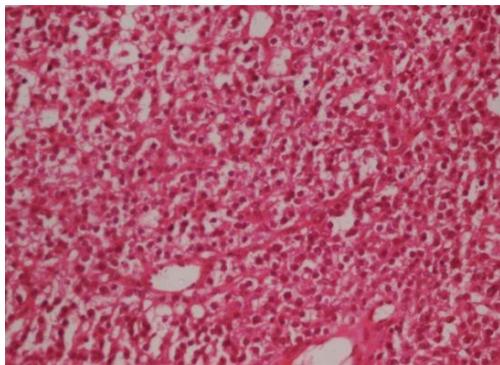


Fig. 3: High power view showing uniform population of polygonal cells with clear to amphophilic cytoplasm and round nuclei with vesicular chromatin

Median sternotomy was done and pea nut size yellow colored tumor found, which was excised without difficulty. The histopathological examination showed the presence of a well circumscribed nodule comprising of highly cellular monomorphic tumor cells with round nuclei, abundant granular eosinophilic cytoplasm- chief cells arranged in acini (Fig. 2, 3). Post operatively her calcium levels normalized but parathormone levels failed to demonstrate adequate fall. Hypovitaminosis D was found (vit D-8ng/ml), accordingly replacement was advised. One month follow up showed normal calcium, phosphorus, alkaline phosphatase and parathormone levels.

DISCUSSION

Primary hyperparathyroidism with its non specific varied manifestations and indolent course, poses a diagnostic challenge to the clinician [8]. Though uncommon, detection of ectopically located hyperfunctioning parathyroid gland is equally challenging and if done pre-operatively, is associated with successful surgical outcome.

Diagnosis of primary hyperparathyroidism in a clinically suspected case is suggested by hypercalcemia,

hypophosphatemia, elevated bone specific alkaline phosphatase and intact parathormone levels [8].

During embryogenesis the superior and inferior parathyroid glands originate from the 4th and 3rd branchial clefts respectively and migrate caudally to their normal positions in relation to the thyroid gland. Any aberrancy during this descent may lead to ectopic location of these glands [8, 9]. The ectopic parathyroid glands are more often derived from inferior glands, because of their long migration path, providing them a higher probability of being ectopic from the angle of mandible to pericardium [10]. They may be located in the mediastinum either anterior or posterior, in the thymus (intra thymic), in the trachea esophageal groove and very rarely in the thyroid parenchyma [8, 9]

Pre operative 99m TC sestamibi scan helps in localizing the tumor in almost 90% of cases [11]. It has an important role for the localizution of ectopic gland that helps the surgeon in order to plan the surgical approach as in our case. MIBI scintigraphy is one of the most widely used investigating technique for pre-operative localization [4, 12]. MIBI localizes in both thyroid and parathyroid glands initially. On the delayed images, the MIBI washes off from the thyroid and the normal parathyroid glands allowing persistent radio activity in the hyperfunctioning parathyroid gland [4].

MIBI is less sensitive in detecting hyperplastic parathyroid glands and also multigland disease [13].

CONCLUSION

We described a case of primary hyperparathyroidism due to parathyroid adenoma in mediastinum, which was excised using sternotomy

Preoperative scintigraphy helped in confirming the location of the adenoma in our case and simplified the surgical management which would have been a challenging task otherwise.

REFERENCES

1. Bienvenu M, Amar L, Vignaux O, Fulla Y, Bonnichon P, Richard B *et al.*; Diagnosis of ectopic mediastinal parathyroidadenomas: value of cardiac MRI. *J Radiol.*, 2003; 84(12 Pt 1): 1969–1973.
2. Feder JM, Sirrs S, Anderson D, Sharif J, Khan A; Primary hyperparathyroidism: An overview. *Int J Endocrinol.*, 2011 (2011), Article ID 251410, 8 pages. Available from <http://www.hindawi.com/journals/ije/2011/251410/>
3. Dsouza C, Bhagavan KR, Gopalakrishna, Rakesh K; Ectopic parathyroid adenoma. *Thyroid Reserch and Practice*, 2012; 9(2): 68-70.

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4. Vijayakumar V, Anderson ME; Detection of ectopic parathyroid adenoma by early Tc-99m sestamibi imaging. *Annals of Nuclear Medicine*, 2005; 19(2): 157–159.
 5. Ng P, Lenzo NP, McCarthy MC, Thompson I, Leeman PJ; Ectopic parathyroid adenoma localized with sestamibi SPECT and image-fused computed tomography. *Med J Aust.*, 2003; 179(9): 485–487.
 6. Otto D, Boerner AR, Hofmann M, Brunkhorst T, Meyer GJ, Petrick T *et al.*; Preoperative localization of hyperfunctional parathyroid tissue with (11)C-methionine PET. *Eur J Nucl Med Mol Imaging*, 2004; 31(10): 1405–1412.
 7. Berna L, Caixas A, Piera J, Gomez G, Matias-Guiu X, Estorch M *et al.*; Technetium-99m methoxyisobutylisonitrile in localization of ectopic parathyroid adenoma. *J Nucl Med.*, 1996; 37(4): 631–633.
 8. Muthukrishnan J, Verma A, Modi KD, Kumaresan K, Jha A; Ectopic parathyroid adenoma - The hidden culprit. *J Assoc Physicians India*, 2007; 55: 515-518.
 9. Mitlak B, Daly M, Potts JJ, Schoenfeld D, Neer RM; Asymptomatic primary hyperparathyroidism. *J Bone Miner Res.*, 1991; 6(S2): S103-S110.
 10. Gouveia S, Rodrigues D, Barros L, Ribeiro C, Albuquerque A, Costa G *et al.*; Persistent primary hyperparathyroidism: an uncommon location for an ectopic gland - Case report and review. *Arq Bras Endocrinol Metabol.*, 2012; 56(6): 393-403.
 11. Gogas J, Kouskos E, Mantas D, Markopoulos C, Kyriaki D, Tseleni-Balafouta S *et al.*; Pre-operative Tc-99m-sestamibi scanning and intra-operative nuclear mapping: are they accurate in localizing parathyroid adenoma? *Acta Chir Belg.*, 2003; 103(6): 626-630.
 12. Lo Y, Lang BH, Chan WF, Kung AW, Lam KS; A prospective evaluation of preoperative localization by technetium-99m sestamibi scintigraphy and ultrasonography in primary hyperparathyroidism. *American Journal of Surgery*, 2007; 193(2): 155- 159.
 13. Palestro CJ, Tomas MB, Tronco GG; Radionuclide imaging of the parathyroid glands. *Semin Nucl Med.*, 2005; 35(4): 266-276.