Case Report

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Pathology Section

Ectopic Parathyroid Adenoma Presenting as a Mediastinal Mass

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ABSTRACT

A parathyroid adenoma present in an ectopic site, in the anterior mediastinum, is a rare cause of persistent hyperparathyroidism. Though its occurrence in the mediastinum is unusual, existence has been noted in literature for more than a century. We describe a case of a 35-year-old male presenting with complaints of pain abdomen, clinically diagnosed as acute necrotizing pancreatitis, with raised serum calcium. Patient received symptomatic treatment for the pancreatitis which subsided. However, high levels of serum calcium persisted. Suspecting hyperparathyroidism, ultrasound neck was done, revealing apical thoracic mass. CT scan of neck revealed a large heterogeneous enhancing mass in superior mediastinum. Fine Needle Aspiration (FNA) of the mass done endoscopically was confusing as it showed features suggestive of a cystic teratoma. However, persistently raised calcium levels along with raised parathormone warranted a Technetium – 99 (Tc-99m) sestamibi scan which revealed positive uptake involving right inferior parathyroid extending to superior mediastinum. The mass was surgically excised and it was diagnosed as a parathyroid adenoma. This case helps bring to light the necessity to consider ectopic parathyroid adenoma as an important differential diagnosis in mediastinal tumour with persistent hypercalcaemia, and as a cause of hyperparathyroidism.

Keywords: Hypercalcaemia, Mediastinum, Necrotizing pancreatitis

CASE REPORT

We report a case of a 35-year-old male, who presented with complaints of epigastric pain of two weeks duration, which was persistent and associated with non-bilious vomiting. Patient was evaluated for the above complaints and on initial investigations, was found to have deranged levels of serum calcium, vitamin D, serum amylase and serum lipase [Table/Fig-1].

The findings on ultrasound abdomen were suggestive of acute necrotizing pancreatitis. The patient was given supportive treatment for the pancreatitis which led to a reduction in symptoms.

However, serum calcium levels continued to remain high, rising upto 14.55mg/dl. Ultrasound neck was performed to investigate for hyperparathyroidism. It revealed a mass posterior to the right common carotid artery in the apex of the right hemithorax. Further, CT scan of neck and thorax showed a well- defined heterogenous mass in the superior mediastinum in the right paratracheal region extending to the neck [Table/Fig-2,3].

Subsequently, Fine Needle Aspiration Cytology (FNAC) and Biopsy (FNAB) were done from the mediastinal mass. The mass was approached by a transesophageal endoscope. The FNAC report concluded the mass to have features suggestive of a cystic teratoma, with the presence of mature squamous epithelial cells [Table/Fig-4], occasional clusters of benign cuboidal to oval epithelial cells [Table/Fig-5] and areas of necrosis. The FNAB report was also in favor of cystic teratoma, where acellular homogenous material was taken as keratin [Table/Fig-6], along with epithelial cells suggestive of respiratory epithelium [Table/Fig-7].

Parameter	Patient	Normal range
Serum Calcium	14.3 mg/dl	8.5- 10.5 mg/dl
Vitamin D	4.14 ng/ml	25-80 ng/ml
Serum amylase	1063 U/L	40-140 U/L
Serum Lipase	1250 U/L	30-210 U/L

[Table/Fig-1]: Serum biochemistry on initial presentation.

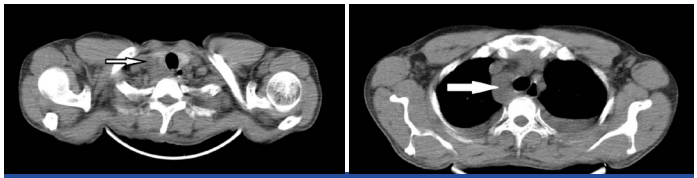
It may be considered that, the lesional tissue was missed during the procedure, and procurement of normal tissue arising from the respiratory tract and surrounding structures in the mediastinum is attributable to the skewed diagnosis here. Since no representative cells were included in the material, diagnosis was missed and the report was based on the material available on the slide.

The persistence of elevated serum calcium levels prompted the clinician to ask for a parathormone assay. Parathormone levels were found to be markedly raised at 1000 pg/ml (Normal value – 15-68.3 pg/ml).

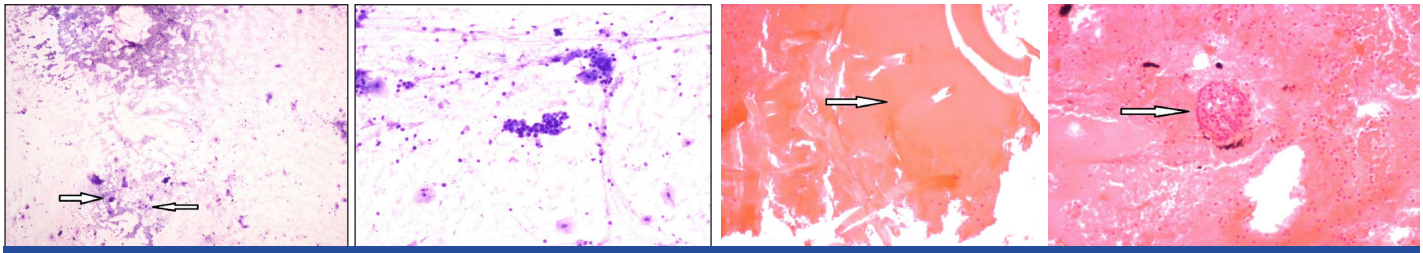
Hence, a prior history of pancreatitis, presence of hypercalcaemia, and raised parathormone level pointed in the direction of hyperparathyroidism, due to a parathyroid mass, probably ectopic. A Tc-99m sestamibi scan was ordered to investigate for parathyroid adenoma. The scan showed positive uptake in the right inferior parathyroid gland, extending to the mediastinum [Table/Fig-8].

Patient was thus given a working diagnosis of parathyroid adenoma, and a right parathyroidectomy was performed. The specimen was sent for histopathological examination. Histopathology revealed a tumour composed of uniform, bland cells arranged in follicular pattern, trabeculae and cords. The cells were round to oval with moderate amount of clear to eosinophilic cytoplasm and bland round to oval nuclei. The above features confirmed the suspicion of parathyroid adenoma [Table/Fig-9,10].

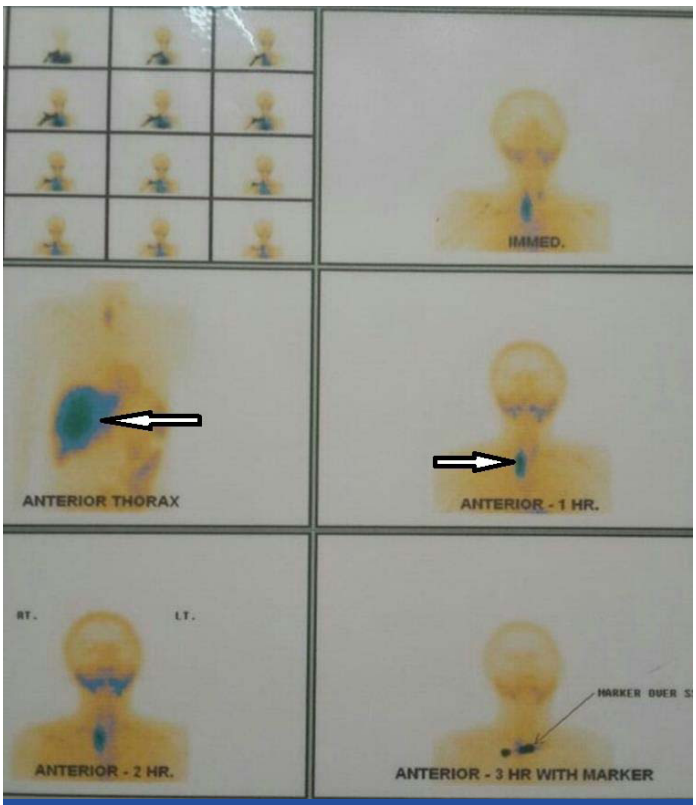
Following removal of the adenoma, patient's symptoms improved and serum calcium levels came back to normal value of 8.2 mg/dl.



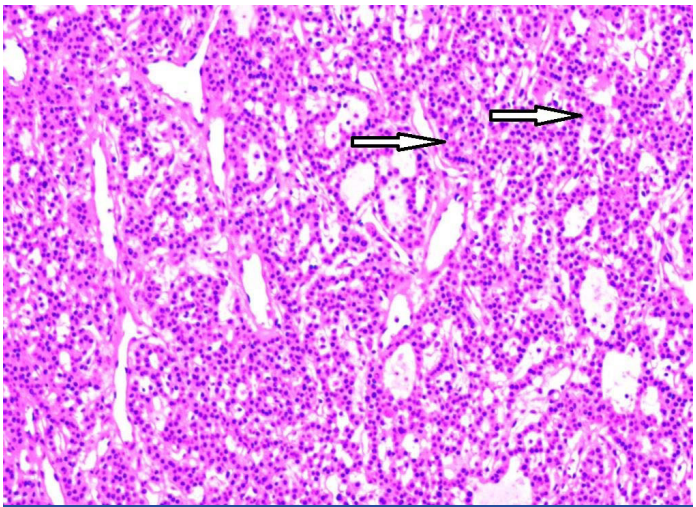
[Table/Fig-2]: CT scan of neck and thorax showing a heterogeneous mass abutting the thyroid anteriorly, and vertebral bodies posteriorly. **[Table/Fig-3]:** A large heterogeneous enhancing mass noted in right para tracheal region.



[Table/Fig-4]: FNAC of mediastinal mass showing area of necrosis and mature squamous epithelial cells (arrow) (H& E stain, 10X). **[Table/Fig-5]:** Benign looking cuboidal epithelium (H&E stain, 10X). **[Table/Fig-6]:** FNAB showing acellular homogenous material (arrow) (? Keratin) (H&E stain, 40X). **[Table/Fig-7]:** Cluster of benign looking cuboidal cells arranged in glandular pattern (Arrow) (H&E stain, 10X).



[Table/Fig-8]: Tc 99m Sestamibi scan: Positive uptake in right inferior parathyroid gland, extending up to the mediastinum.

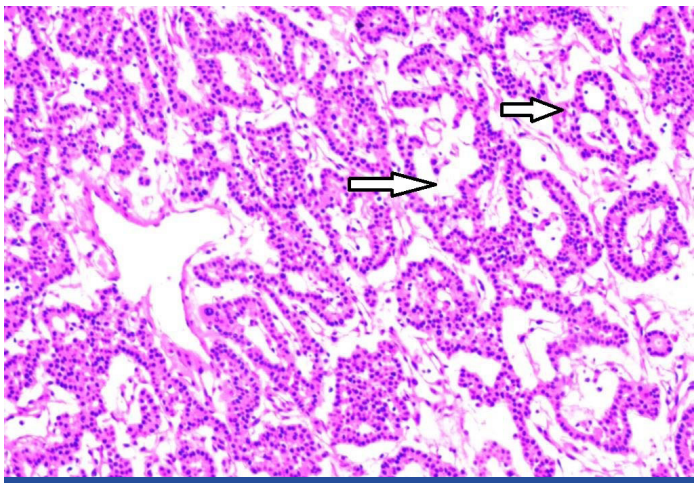


[Table/Fig-9]: Tumour cells arranged in trabeculae and cords (arrow) (H&E stain, 10X).

Follow up after six months also revealed serum calcium value of 8 mg/dl.

DISCUSSION

A solitary parathyroid adenoma is a common cause of primary hyperparathyroidism, and is usually, located in a juxtathyroid position in the neck [1]. Primary hyperparathyroidism often presents in an unusual manner, with 0.3-8% of the cases occurring due to an ectopically located parathyroid gland adenoma [2]. An inability to locate the adenoma may delay the diagnosis of these cases and lead to further complications of hyperparathyroidism.



[Table/Fig-10]: Tumour cells arranged in follicular pattern (arrow) in a variable stroma (H&E stain, 10X).

There are various mechanisms involved in development of tumours in ectopic sites such as the mediastinum. Two of the most likely explanations deal with an embryologic developmental misplacement or with the enlargement of the tumour in the neck that plunges into the mediastinal compartment [3].

The parathyroid glands are arranged in two pairs, upper and lower. The lower pair originates from the dorsal wing of the third pharyngeal pouch, along with the thymus which originates from the ventral wing. Hence, the common descending route of the inferior parathyroid with the thymus, can explain their aberrant situation in the mediastinum [4]. Ectopic parathyroid glands have been found to occur in various anatomic locations anywhere from the angle of the mandible to the mediastinum. The most common sites are the mediastinum, along the path of the vagus nerve and recurrent laryngeal nerve and within the thyroid parenchyma. Uncommon sites can include the hypoglossal nerve, posterior triangle of the neck, axilla and pericardium [5]. It is essential to consider parathyroid adenoma as a differential diagnosis when a patient presents with a mediastinal mass. Though teratomas are an important differential in anterior mediastinal masses, parathyroid adenoma should be considered if the patient presents with clinical symptoms suggestive of hyperparathyroidism.

Hypercalcaemia with raised parathormone levels, or isolated hypercalcaemia, are diagnostic of primary hyperparathyroidism [2]. Moran CA et al., described cases where patients were being reviewed for primary parathyroid tumour in the mediastinum, showed metabolic disturbances such as hypercalcaemia and hypophosphatemia [3]. Thus, these derangements help to serve as a clue to suspect a parathyroid mass.

Preoperative localization of the adenoma is critical, for which a Tc-99m-Methoxy Isobutyl Isonitrile (MIBI) scan has shown sensitivity of 80%-90% [6]. The modality helps to reduce the requirement of repeated neck explorations, as was reported in a study by Abbas S

et al., and proved useful in detecting the ectopic nature of the lesion in our case [1].

A cytological diagnosis of parathyroid gland adenoma can be considered reliable in 96% of cases, if there is correct localization of the nodule and adequate material for the biopsy [7]. Cytological features for diagnosis include anisokaryosis, stippled chromatin, a well-defined cell border, and oxyphilic cytoplasm [8].

According to Kitada M et al., median sternotomy is a popular surgical technique for surgical excision of mediastinal parathyroid adenomas that are difficult to excise using the cervical approach [9]. Less invasive approaches have gained popularity as well, as these tumours are benign. Doherty GM et al., suggested angiographic ablation be attempted as an initial procedure for patients with an angiographically identified mediastinal parathyroid adenoma [10]. Regardless of the method, it is a general consensus that preoperative localization is essential.

CONCLUSION

An unusual location for a parathyroid adenoma is the mediastinum, and it can be a cause of persistent hyperparathyroidism. This case helps bring to light the necessity to consider the diagnosis of an ectopic parathyroid adenoma in a case of persistent hypercalcaemia and absence of neck swelling. We report this case for it's rarity and unusual presentation.

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