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Intrathymic Parathyroid Adenoma

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Abstract

Introduction: Mediastinal parathyroid adenoma localization is a rare entity. We report a case of excision by manubriotomy of intrathymic parathyroid adenoma detected by Computed Tomography scan of the chest and neck and confirmed by Technetium-99 m-sestamibi scan (99 mTc-MIBI). Case Presentation: A 68 years old woman with history of hypercalcaemia, PTH elevation and operation for pathological fracture of the left femur 7 days before was presented to our service. The patient underwent manubriotomy and the adenoma was found within the right lobe of the thymus gland. Conclusion: The Technetium-99 m-sestamibi scan evaluation can be useful in the preoperative localization of ectopic parathyroid adenomas. The surgical approach by manubriotomy is privileged when the ectopic adenoma is in the upper part of anterior mediastinum.

Keywords

Intrathymic Parathyroid Adenoma, Technetium-99 m-Sestamibi Scan, Manubriotomy

1. Introduction

Primary Hyperparathyroidism (PHPT) is usually caused by an adenoma of the parathyroid gland located near the thyroid gland in the neck. Parathyroid glands may also have an ectopic position, especially at mediastinal site, up to 25% of cases [1]. The hyper-functioning adenoma is ectopically placed in the mediastinum in 1% - 2% of cases [2]. This location may be a risk condition for unsuccessful surgery. In fact, mediastinal parathyroid glands represent 25% - 38% of causes of reoperations because of persistent PHPT [3] [4]. In addition 54% of patients are asymptomatic at diagnosis, 58% of all patients exhibit some signs of bone disease [2] (most of these patients have only radiological signs but no symptoms). We present the case of an ectopic parathyroid adenoma located

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inside the thymus gland which is successfully managed surgically.

2. Case Report

A 68-year-old woman was referred to our institution with a mass of the anterior superior mediastinum, and complaining of bone pains and spontaneous fracture of left femur which was managed by intramedullary nail 7 days ago. Physical examination including the neck was normal. Routine laboratory investigations were also normal.

Serum calcium was elevated at 11 mg/dl, phosphorous was 2.4 mg/dl and serum proteins were normal with albumin. Serum Parathormone (PTH) was 45.3 pg/dl.

Cervical ultrasound revealed some bilateral nodular lesions of the thyroid and the renal one was normal. CT scan of the chest (**Figure 1(a)** and **Figure 1(b)**) showed a well-defined soft tissue lesion in the anterior mediastinum just behind the upper part of the sternum next to the superior vena cava about 3 cm × 1 cm. A Technetium-99 m-sestamibi scan (99 mTc-MIBI) (**Figure 2**) successfully localized a persistent abnormal tracer uptake in the superior mediastinum suggestive of mediastinal parathyroid adenoma. A manubriotomy was performed and a right thymic lobar parathyroid adenoma was excised (**Figure 3**) which was confirmed on histology: clear cells parathyroid adenoma (**Figure 4**). The postoperative course was uneventful, and the patient was discharged on day 5 with normocalcemia (8.4 mg/dl) and normal parathormone rate (1.5 pg/dl).

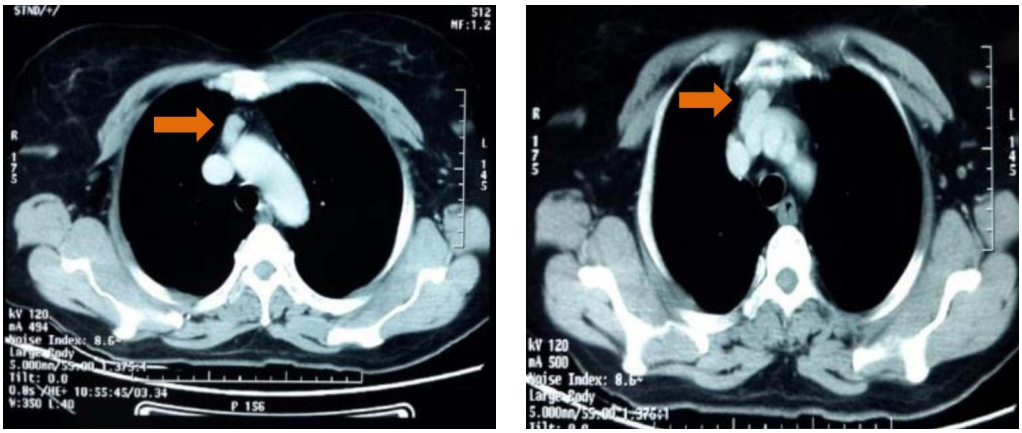
3. Discussion

Ectopic mediastinal parathyroid adenomas producing primary hyperparathyroidism (PHPT) appear to be an uncommon entity. Mc Henry *et al.* reported an incidence of 1.3% amongst 522 surgically treated patients with hyperparathyroidism [5].

The PHPT is caused by a solitary adenoma accounts for over 80% of cases. During embryogenesis, the upper and lower parathyroid glands descend into the neck and, as a result, 20% of parathyroid glands are found in ectopic locations [6]. Most ectopic glands are found in close proximation to the thymus with the inferior parathyroid glands originating from the third bronchial pouch with the thymus. Possible positions of the embryologically ectopic adenoma is the deep anterior mediastinum, the aortopulmonary window, the posterior mediastinum, or within the substance of the thyroid gland. However, paraesophageal and retroesophageal parathyroid tumors are not considered ectopic since they have normal blood supply from a branch of the inferior thyroid artery and they arise from superior parathyroid glands [7].

Generally the symptoms of primary hyperparathyroidism are characterized by recurrent nephrolithiasis and hypercalcemia with low level of blood phosphorus, bony defect, weakness, and low life expectancy. However mild hyperalcemia is combined with less severe symptoms [7].

The preoperative diagnostic tools to detect parathyroid adenomas consist of ultrasound (US), CT, and MRI



(a) (b)

Figure 1. Chest CT showed a well defined soft tissue (red arrows) in the anterior mediastinum.

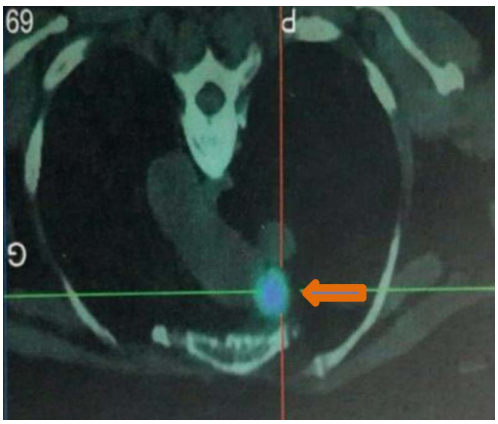


Figure 2. Persistent abnormal tracer uptake in the mediastinum suggestive of médiastinal parathyroid adenoma (red arrow).

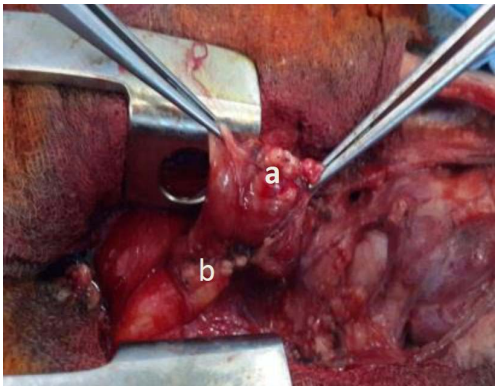


Figure 3. Intraoperative image of an ectopicadenoma (a) inside the thymus gland (b).

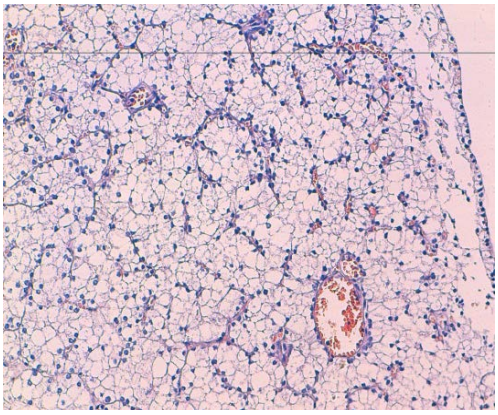


Figure 4. Parathyroid parenchyma tumor proliferation with clear cytoplasm and cell nuclei without a typia limited by a fibrous capsule.

with accuracy 57% - 68% depending on the size and location of these adenomas [8]. But also the 99 mTc sestamibi (MIBI) scintigraphy can detect parathyroid adenomas with a diagnostic accuracy of 85% - 95%. The combination of CT and 99 mTc sestamibi has 100% sensitivity and 97.4% positive predictive value for the detection of ectopic parathyroid adenomas [9].

The standard approaches to deep médiastinalparathyroids have been sternotomy or thoracotomy.

At our patient, the adenoma excision was performed by manubriotomy. This approach has never been reported in the literature. It is a minimally invasive procedure, better tolerated than sternotomy in the presence of bone fragility and risk of sternotomy complications.

Those complications include pleural effusions, sternal wound infection, mediastinitis and recurrent laryngeal nerve palsies (18.7%) [1].

To reduce the invasiveness of the procedure, some authors have proposed an anterior mediastinotomy by parasternal approach (Chamberlain technique), with an overall success rate of 88.9%, especially for immediately retrosternal and laterally located glands [10]. Thoracoscopy has been suggested as the technique of choice for ectopic glands located to the middle and lower mediastinal regions but some limitations have been reported in case of immediately retrosternal parathyroids in the upper part of the anterior mediastinum, with an increased risk of convertion [1].

4. Conclusion

Preoperative location of an ectopic parathyroid adenoma with the use of CT and 99 mTc sestamibi (MIBI) scintigraphy is accurate. The surgical approach by manubriotomy is privileged when the ectopic adenoma is in the upper part of anterior mediastinum like the case of our patient. The long term metabolic and rehabilitative management is crucial for effective management of these chronically ill patients. It is important that a multi-disciplinary team involving the endocrinologist, surgeons, radiologists, physical therapists and others as needed manage such patients.

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Abbreviations

CT, Computer Tomography; MIBI, Methoxyisobutylisonitrile; PHPT, Primary Hyperparathyroidism; PTH, Parathormone.