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Case Report

Excision of an Elusive Tiny Ectopic Parathyroid Adenoma

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Abstract
An elderly lady was diagnosed with primary hyperparathyroidism after being admitted to hospital with pyelonephritis and constipation. A sestamibi parathyroid scan demonstrated increased uptake in the upper mediastinum, suggesting an ectopic mediastinal parathyroid adenoma. The 4-mm adenoma was successfully removed through a mini-sternotomy incision using a gamma probe to identify the gland from the surrounding tissue. The patient made an uneventful recovery.

Introduction

Ectopic parathyroid glands are relatively common, with authors quoting an incidence of up to 25% [1]. Although the first procedure for mediastinal parathyroid gland adenoma removal was performed by Churchill in 1932 [2], there is still disagreement on the definition of

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what constitutes a mediastinal parathyroid gland. While certain authors consider all glands inferior to the superior border of the manubrium to be mediastinal [3], others classify mediastinal parathyroid glands as those which cannot be accessed through cervical incisions [4, 5]. Therefore, the reported incidence of mediastinal parathyroid glands varies between 1 and 2%.

The inferior and superior parathyroid glands develop from the 3rd and 4th pharyngeal pouches, respectively. As the inferior parathyroid glands are derived from the dorsal part of the 3rd pharyngeal pouch and the thymus arises from the ventral part of the 3rd pharyngeal pouch, both the inferior parathyroid glands and the thymus migrate together downwards towards the mediastinum. Usually, the inferior parathyroid glands rest at the inferior poles of the thyroid, while the thymus continues to migrate further downwards. The superior parathyroid glands also migrate downwards together with the ultimobranchial bodies which likewise develop from the 4th pharyngeal pouch, however, the distance travelled is shorter, and, therefore, they have a more constant position in the neck posterior to the middle 3rd of the thyroid lobes. Eighty percent of all deep parathyroid tumours are ectopic inferior parathyroid glands, usually located in the anterior mediastinum in close contact with the thymus [3].

Primary hyperparathyroidism is characterised by excessive secretion of parathyroid hormone (PTH), resulting in a high plasma calcium ion and low phosphate ion concentration. PTH secretion by the parathyroid glands is normally controlled by negative feedback by the plasma calcium ion concentration. Vitamin D levels are reduced in patients with primary hyperparathyroidism to combat hypercalcaemia. Symptomatic mediastinal parathyroid glands are exceedingly rare, accounting for only 1% of operations for parathyroid disease [6]. The commonest parathyroid pathologies leading to symptomatic disease are adenomas, and these account for 80–85% of cases. Benign hyperplasia makes 15% of all cases, while parathyroid carcinomas account for <1% [7].

Case Presentation

An 80-year-old lady presented to the accident and emergency department complaining of severe right upper quadrant and epigastric pain associated with multiple episodes of vomiting. She also noted increasing constipation over the previous days. She was diagnosed with right-sided pyelonephritis, from which she made an uneventful recovery after admission for intravenous antibiotics and rehydration. Blood results showed persistently high calcium and low phosphate levels, and she was, therefore, referred to the endocrinologist to investigate her for primary hyperparathyroidism. In retrospect, the constipation was probably attributable to the high calcium levels.

The patient had high plasma PTH levels and low vitamin D levels (Table 1), but ultrasound (US) and computed tomography (CT) of the neck showed no detectable abnormality. However, a sestamibi parathyroid scan showed a mildly increased uptake in the upper mediastinum slightly to the left of the midline in keeping with an ectopic parathyroid tissue as shown in Figure 1.

The patient successfully underwent removal of the ectopic parathyroid adenoma through a mini-sternotomy by splitting the manubrium in the middle and the sternum down

to the 3rd intercostal space just above the 4th rib. The right hemi-sternum was then divided to give more access. On inspection and dissection, there was no visible parathyroid or adenoma. A radioactive tracer (technetium-99m) was administered 15 min prior to the procedure, and a gamma probe (inserted in a sterile sheet) was used to identify an area of fat near the ectopic parathyroid tissue. At the location of the parathyroid adenoma, the gamma probe gave a 4 times higher count per min (CPM) than in the surrounding fat tissue. The area of fat in question was excised, and the gamma probe was used ex vivo showing increased CPM. There was no increased CPM in the remaining tissue in situ. Before closure of the wound, there was haemorrhage from underneath the sternum that could not be controlled adequately, and the sternotomy had to be completed. The culprit bleeder was an intercostal artery in the right 4th space. Post-operatively, the patient was monitored with serial serum calcium levels, and no hypocalcaemia occurred. Histology showed a completely excised parathyroid adenoma 4 mm in size embedded in fat.

PTH levels, vitamin D levels and serum calcium levels returned to normal levels post-operatively (Table 1), and the patient was fit to be discharged home 4 days following the procedure. No complications occurred.

Discussion

This case highlights the salient steps in the management of mediastinal parathyroid adenomas. Exploration for mediastinal parathyroid adenomas without prior imaging has a reported failure rate of 30–36% [4, 8]. Therefore, with improved and targeted imaging techniques, surgeons demand pre-operative imaging for parathyroid adenomas to provide targeted treatment. CT has low sensitivity (45–55%) for parathyroid disease, although it is slightly better in detecting mediastinal parathyroid disease [9]. Sensitivity of US varies and is operator dependent (36–76%) [9]. While US is effective in the detection of upper cervical parathyroid adenomas, it is not very useful in the detection of mediastinal parathyroid adenomas. Technetium-99m sestamibi has multiple advantages over thallium-technetium subtraction scintigraphy in that it allows dual phase acquisition and 3-dimensional imaging through single-photon emission CT [10]. Dual-phase sestamibi scan, as used in this case, has a sensitivity of 80–91% depending on the gland size [11]. Magnetic resonance imaging (MRI) has an overall sensitivity of 78% [9]. MRI sensitivity goes up to 88% in mediastinal parathyroid glands. However, MRI was not used in this case. Selective venous PTH sampling and selective angiography are seldom used in view of their invasive nature. The adenoma was very small, and, therefore, it is no surprise that it was not detected by CT and US and only showed very little positivity with the isotope scan.

Medical treatment for primary hyperparathyroidism with resorptive agents and calcimimetics is still controversial [12]. Surgery is the definitive treatment for this condition because it achieves symptom relief and biochemical homeostasis and prevents the sequelae of the disease [13].

The surgical approach is dependent on the position of the gland. Most anterior mediastinal tumours can be reached through a cervical incision, from which up to 10 cm of the thymus may be teased out. However, in elderly patients, such as the one described in this report, traction on the thymus is virtually impossible in view of the fragility of the capsule of

the thymic remnant. This surgical procedure also gives access to the para-oesophageal and retro-oesophageal glands and may be modified by making the incision inside the border of the sternocleidomastoid to gain access for posterior mediastinal tumours.

Tumours that cannot be reached via the trans-cervical approach have traditionally been handled with median sternotomies and thoracotomies. With minimally invasive surgery, the patient is saved from a full-length median sternotomy. Ectopic parathyroidectomy may also be performed using video-assisted thoracoscopic surgical techniques [14]; however, this is only considered when the ectopic parathyroid adenoma has been properly localised. Peri-operative technetium-99m has been used before in the detection of ectopic parathyroid glands or adenomas [15, 16].

This case shows that mini-sternotomy allowed the successful excision of a 4-mm invisible ectopic parathyroid adenoma with the use of a gamma probe. It also demonstrates the ease of converting a mini-sternotomy into a complete one if the need arises.

Statement of Ethics

Informed consent was obtained from the patient.

Disclosure Statement

There are no conflicts of interest.

References

1 Shields T: General thoracic Surgery. Philadelphia, Wolters Kluwer Health/Lippincott Williams and Wilkins, 2009.
2 Bauer W: Hyperparathyroidism epitomized: the case of Captain Charles E. Martell. Metabolism 1962;11:21–29.
3 Dubost C, Bouteloup PY: Explorations mediastinales par sternotomie dans la chirurgie de l'hyperparatyroidie. J Chir (Paris) 1988;125:631.
4 Rothmund M, Diethelm L, Brünner H, Kümmerle F: Diagnosis and surgical treatment of mediastinal parathyroid tumors. Ann Surg 1976;183:139–145.
5 Nathaniels EK, Nathaniels AM, Wang C-A: Mediastinal parathyroid tumors: a clinical and pathological study of 84 cases. Ann Surg 1970;171:165–170.
6 Russell CF, Edis AJ, Scholz DA, Sheedy PF, Van Heerden JA: Mediastinal parathyroid tumors: experience with 38 tumors requiring mediastinotomy for removal. Ann Surg 1981;193:805–809.
7 DeLellis RA: Parathyroid tumors and related disorders. Mod Pathol 2011;24:S78–S93.
8 Wang C, Gaz RD, Moncure AC: Mediastinal parathyroid exploration: a clinical and pathologic study of 47 cases. World J Surg 1986;10:687–695.
9 Shen W, Düren M, Morita E, Higgins C, Duh Q-Y, Siperstein AE, et al: Reoperation for persistent or recurrent primary hyperparathyroidism. Arch Surg 1996;131:861–869.
10 Nguyen BD: Parathyroid imaging with Tc-99m sestamibi planar and SPECT scintigraphy. Radiographics 1999;19:601–614.
11 Norton KS, Johnson LW, Griffen FD, Burke J, et al: The sestamibi scan as a preoperative screening tool. Am Surg 2002;68:812–815.
12 Leere JS, Karmisholt J, Robaczyk M, Vestergaard P: Contemporary medical management of primary hyperparathyroidism: a systematic review. Front Endocrinol (Lausanne) 2017;8:79.

13 Samuel A, Dowthwaite SA, Young JE, Pasternak JD, Yoo J: Surgical management of primary hyperparathyroidism. J Clin Densitom 2013;16:48–53.

14 Prinz RA, Lonchyna V, Carnaille B, Wurtz A, Proye C: Thoracoscopic excision of enlarged mediastinal parathyroid glands. Surgery 1994;116:999–1005.

15 Daliakopoulos SI, Chatzoulis G, Lampridis S, et al: Gamma probe-assisted excision of an ectopic parathyroid adenoma located within the thymus: case report and review of the literature. J Cardiothorac Surg 2014;9:62.

16 Buicko JL, Kichler KM, Amundson JR, Scurci S, Kozol RA: The sestamibi paradox: improving intraoperative localization of parathyroid adenomas. Am Surg 2017;83:832–835.

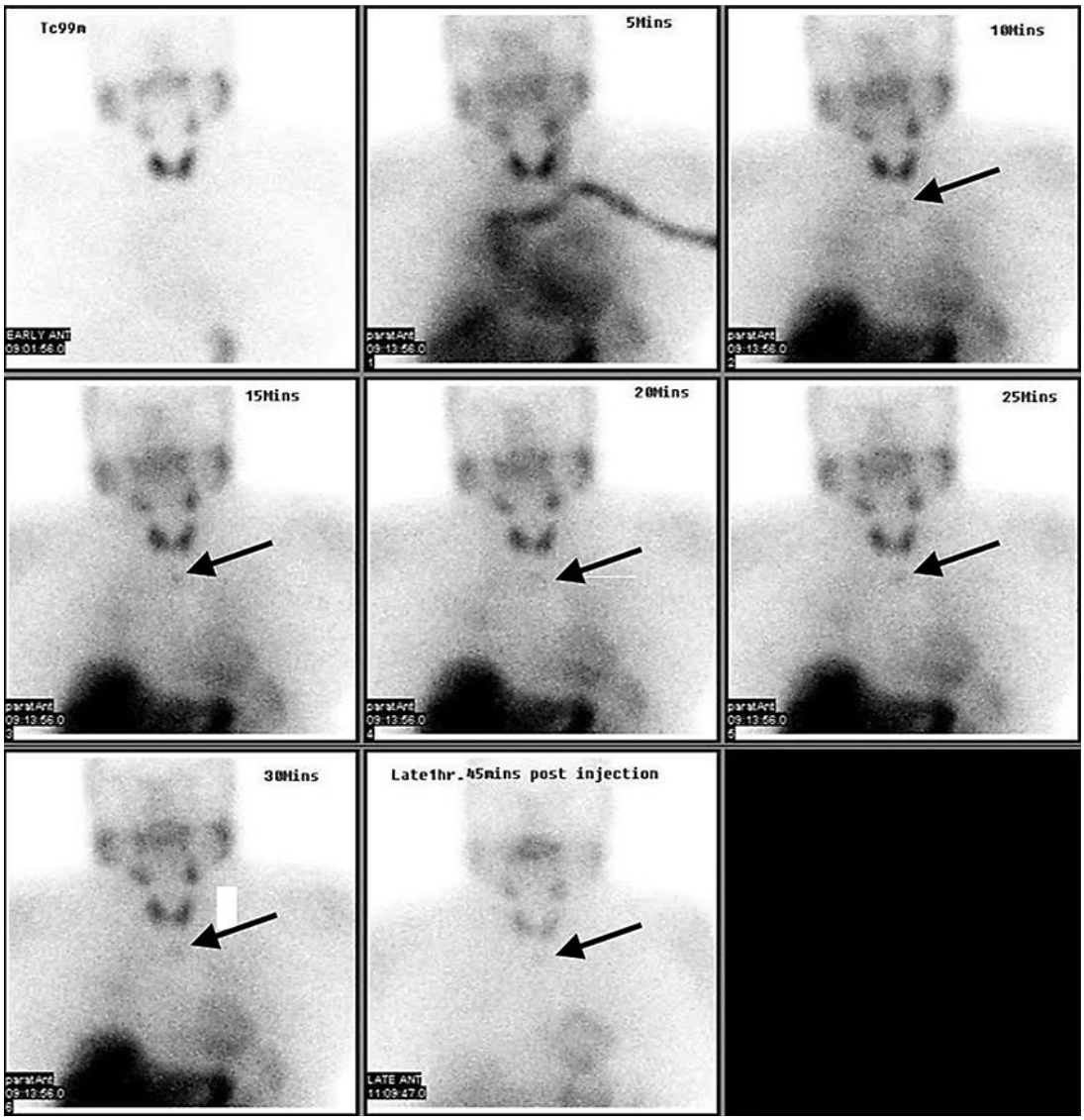

Fig. 1. Ectopic parathyroid tissue faintly visible (arrow) using technetium-99m sestamibi parathyroid scan.

Table 1. Pre- and post-operative values for plasma PTH, serum calcium, serum phosphate and serum vitamin D

	Pre-operative level	Post-operative level	Normal range
Plasma PTH, pg/mL	127	36	15–65
Serum calcium, mmol/L	2.68	2.17	2.15–2.55
Serum phosphate, mmol/L	0.74	1.27	0.8–1.5
Serum vitamin D, ng/mL	24	36	30–100

PTH, parathyroid hormone.