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Case Report

A rare case of a giant intrathyroidal parathyroid adenoma only localized with a 4D CT scan

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Abstract

Ectopic parathyroid adenomas (PTAs) are quite rare and are typically located within the thymus, retroesophageal space, retropharyngeal space, or within the thyroid gland. Likewise, intrathyroidal parathyroid adenomas (IPAs) and giant PTAs are also a rare subset of PTAs. To date, only six studies have investigated localizing techniques of IPAs using 4-dimensional computed tomography (4D CT) scan with only one study demonstrating two IPAs successfully localized using both 4D CT scan and 4-dimensional magnetic resonance imaging. We describe a single lesion in a patient which would be classified both as an IPA and a giant parathyroid adenoma (GPA). This lesion only successfully localised with a 4D CT scan which is not in keeping and unusual with the available literature.

Keywords: Intrathyroidal parathyroid adenoma, 4-Dimensional computed tomography scan, Technetium based scintigraphy

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Introduction

Primary hyperparathyroidism is the third most prevalent endocrine disorder after diabetes and hypothyroidism affecting 1 in 500 women and 1 in 20 000 men over the age of 40 (1). It is usually caused by a single adenoma in approximately 85% of cases, multiple gland hyperplasia in about 15% of cases, or rarely a parathyroid carcinoma (2). Parathyroid adenomas (PTAs) are usually small and weigh less than 1000 milligrams whereas giant PTAs are rare and defined as adenomas weighing more than 3500 milligrams (3).

Parathyroid adenomas are typically located within the parathyroid gland however, they can also be found at ectopic sites such as the thymus, retropharyngeal space, retroesophageal space or within thyroid (4). The incidence of ectopic PTAs ranges from 9%-22% (4). Intrathyroidal parathyroid adenoma (IPA) are PTAs which are partly or completely enveloped by thyroid gland and have an incidence rate ranging from 0.7% to 6% of PTAs (4). IPAs can be challenging to manage compared to non-ectopic PTAs.

This case report describes a young female patient with a rare giant IPA weighing 8000 mg. She presented with significant hypercalcemia and an acute kidney injury. First-line imaging modalities including ultrasound and sestamibi were unable to localise the lesion while the 4D CT successfully localised the lesion thereby assisting operative management. We also reviewed the published literature of clinical presentation, imaging localization and management of IPAs.

Case Presentation

A 33-year-old Vietnamese female was referred by her general practitioner to the emergency department with acute kidney injury and an elevated troponin. She reported a 2-week history of lethargy, poor oral intake, nausea and 15 kg weight loss over two months. Her past medical history included gestational hypertension and hypertension. She had been recently commenced on telmisartan and amlodipine. She did not take other medications. On physical examination, her blood pressure was 140/90 mm Hg with remaining vitals within normal limits. The physical examination was unremarkable and there was no neck swelling or palpable mass with no palpable lymphadenopathy. The remainder of the examination was unremarkable.

Laboratory results showed normal full blood count, liver function tests and thyroid function tests. Her corrected serum calcium was elevated at 4.63 mmol/L (normal range: 2.15-2.65 mmol/L) with a parathyroid level of greater than 212 pmol/L (normal range: 2.0–8.5 pmol/L). Vitamin D level was 24 nmol/L (normal range: > 50 nmol/L). Her serum creatinine level was also elevated at 129 µmol/L (normal range: 40-90 µmol/L) and 24-hour urine calcium was raised at 8.5 mmol/h (normal range: 2.0-7.5 mmol/h). Troponin level was elevated at peak of 48

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Implication for health policy/practice/research/medical education

Traditional imaging modalities such neck ultrasound and sestamibi scan have good evidence for localising intrathyroidal parathyroid adenomas (IPAs) whereas the 4-dimensional computed tomography (4D CT) does not. Where there is strong clinical and biochemical suspicion of a parathyroid adenoma (PTA) and first line imaging modalities are unsuccessful in localising a lesion, a 4D CT scan should be considered to localise the lesion and assist operative management.

ng/L (normal range: <11 ng/L) with no ECG changes. She received intravenous fluids therapy and zoledronic acid for initial management of hypercalcemia. Her calcium level improved to 2.42 mmol/L (normal range: 2.15 -2.65 mmol/L) within 5 days with the treatment.

Imaging investigations included a neck ultrasound (US) which revealed a dominant nodule within the left thyroid lobe with no detectable PTAs identified (Figure 1). A parathyroid sestamibi scan (Figure 2) demonstrated a left side hypofunctioning thyroid nodule with no evidence of PTA. Meanwhile, 4-dimensional computed tomography (4D CT) scan of neck was performed due to strong clinical suspicion of primary hyperparathyroidism with ultrasound and sestamibi scan unable to localise a parathyroid lesion. The CT (Figure 3) demonstrated a heterogeneously enhancing nodule arising within the inferior aspect of the left lobe of the thyroid, measuring 36 mm, with a prominence of the left inferior thyroid artery suspicious for an IPA.

This patient underwent left side hemithyroidectomy with parathyroid biopsy and neck exploration. Parathyroid hormone (PTH) monitoring demonstrated a drop in PTH from 14.8 pmol/L post excision to 4.2 pmol/L over 24 hours post operation. Intraoperative frozen sections were sent to pathology laboratory. The patient was discharged on the second day post operation without any surgical complications. Her corrected calcium level was 2.70 mmol/L on discharge. Final histopathology results showed intrathyroidal atypical PTA in the left lobe of thyroid, which was completely excised with an estimated weight of 8000 mg. The pattern of immunohistochemistry was parafibromin positive and PGP 9.5 (protein-encoding gene product 9.5) negative, with low likelihood of recurrence. The patient was followed up in clinic for a total of eight months post operation and remained asymptomatic with normal calcium levels.

Discussion

Giant parathyroid adenoma (GPA) is defined as an adenoma larger than 3 cm and/or greater than 3500 mg (95th percentile of PTAs) and is usually managed with a parathyroidectomy (3). Likewise, an IPA is a PTA located partially or completely within the thyroid gland and is also typically managed with a parathyroidectomy though in some cases, complete resection may require a complete thyroidectomy (4). Our patient had a single lesion which was located within the thyroid gland and weighed 8000 mg classifying it as both an IPA and a GPA.

Localization techniques for PTAs classically involve a neck ultrasound followed by a sestamibi scan (2). Where these imaging modalities are unable to successfully localise a lesion, a 4D CT scan may be used although careful consideration needs to be taken to limit side effects of radiation (2). Based on the available literature, the sensitivity of ultrasound to localise IPAs has been reported to range from 29% to 67% (4-6), with the largest cohort of 50 IPAs having sensitivity from 29% to 76% (6). Key characteristics on the ultrasound to look for include the presence of a solid nodule, absence of cystic components, hypoechogenicity and the presence of a single feeding artery on doppler (6). A sestamibi scan commonly referred as technetium-based scintigraphy (sestamibi) with variants including SPECT and SPECT/CT, have sensitivities ranging from 60%-83% in studies with sample sizes of more than 10 IPAs (5,7). These studies are associated with higher rates of false positives due to retention of radiotracer in benign thyroid nodules and malignant thyroid disease while false negatives may occur due to multiglandular disease and smaller adenomas (5,7). Regardless, an unremarkable scan should not be a reason to exclude a diagnosis if there is strong biochemical suspicion of an IPA as was the case in our patient.

Six studies have described the use of 4D CT scans for localising IPAs (5,8-12). Three of these studies had sample sizes less than 5 IPAs and the 4D CT did not localise IPAs in any of these cases (8-10). It should be noted that further details on any imaging features underlying these cases was not provided and therefore it cannot be conclusively stated that the 4D CT was completely negative. Two studies

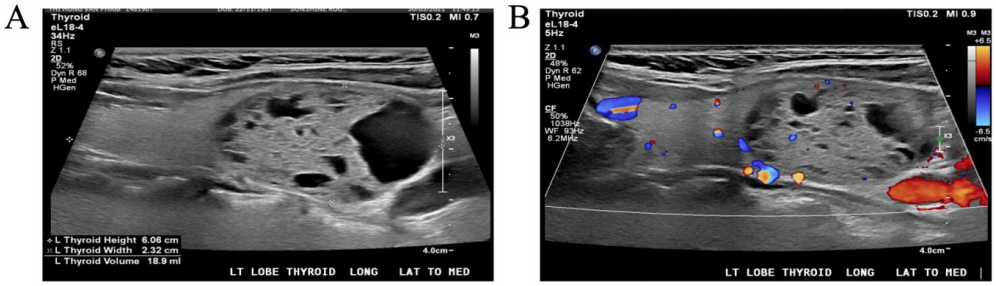

Figure 1. Ultrasound of neck showed a mixed solid/cystic, hyperechoic lesion within the left thyroid lobe (A), with normal vascularity (B).

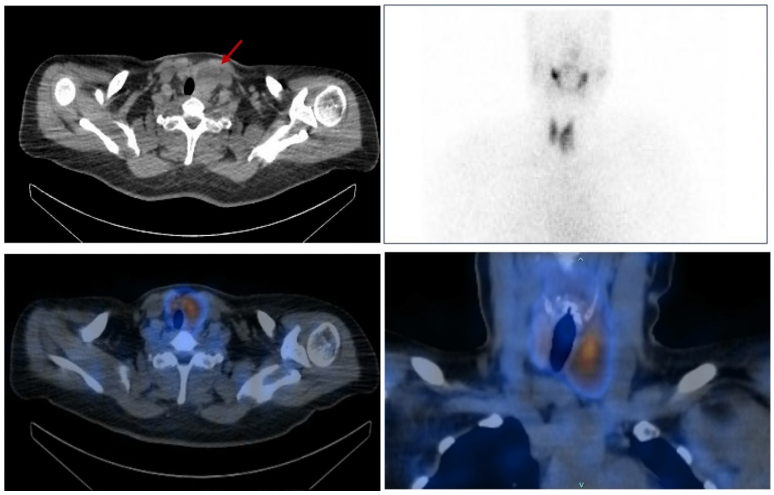

Figure 2. A parathyroid sestamibi scan demonstrated an enlarged left thyroid lobe with relative photopenia on its lateral mid to lower pole suggesting a hypofunctioning nodule. No discordance or relative retention is appreciated to suggest parathyroid adenoma.

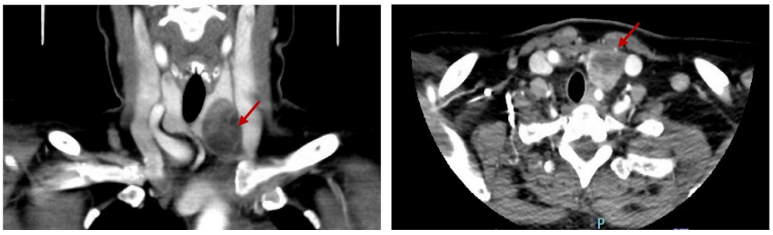

Figure 3. 4D neck CT demonstrated a heterogeneously enhancing nodule arising within the inferior aspect of the left lobe of the thyroid suggestive of an intrathyroidal parathyroid adenoma.

reported an approximately 65% success rate using the 4D CT however both studies had less than 10 patients with IPAs (11,12). Finally, one study looking into 10 patients with IPAs and 4D CT successfully identified 2 IPAs (5). However, it is important to note that these patients had 4D MRIs first prior to the 4D CT scans where the MRI successfully identified 3 lesions while the 4D CT scan only identified 2 lesions (5).

Conclusion

In our case, the ultrasound and sestamibi scans did not localise the IPA which is quite unusual in the literature. Given there was a strong clinical suspicion and biochemical evidence of a PTA, we proceeded with a 4D CT scan which was the only modality that successfully identified the lesion. From the available literature it is quite rare for a 4D CT scan to successfully localise an IPA and from all the literature available this would be the first case where a 4D CT scan successfully localised an IPA without concurrent magnetic resonance imaging (MRI) support. As a result, we recommend the use of 4D CT scan for localising PTA where there is strong clinical and biochemical suspicion without radiological localisation with ultrasound or sestamibi scans.

Authors' contribution

Conceptualization: Ibrahim Shahid.
Formal analysis: Ibrahim Shahid and Jing Ren.
Investigation: Ibrahim Shahid and Jing Ren.
Methodology: Ibrahim Shahid and Jing Ren.
Project administration: Ibrahim Shahid and Jing Ren.
Resources: Ibrahim Shahid and Jing Ren.
Supervision: Ibrahim Shahid.
Writing–original draft: Ibrahim Shahid and Jing Ren.
Writing–review & editing: Ibrahim Shahid.

Conflicts of interest

Both authors have no conflict of interest that could be perceived as prejudicing the impartiality of the research reported.

Ethical issues

This case report was conducted in accordance with the World Medical Association Declaration of Helsinki. Written informed consent was obtained from the patient's next of kin for publication of the submitted article and accompanying images. Verbal consent was obtained from the patient as the patient was unable to physically sign the document due to medical limitations. Ethical issues (including plagiarism, data fabrication, double publication) have been completely observed by all authors.

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