An Unusual Mass of Posterior Mediastinum: A Case of Retrotracheal Parathyroid Adenoma Presenting With Primary Hyperparathyroidism

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ABSTRACT: Although parathyroid ectopy in the mediastinum has been the subject of several publications, its location in the posterior mediastinum is very rarely reported. We report a case of a 69-year-old patient who presented with clinical symptoms of malignant hypercalcemia due to a retrotracheal mediastinal parathyroid adenoma. The surgical excision leads to a quick normalisation of the phosphocalcic balance with improvement of the clinical symptoms. Ectopic hypersecreting parathyroid adenoma with life-threatening hypercalcemia should prompt radiological assessment and appropriate surgical management to prevent further clinical complications.

KEYWORDS: mediastinal mass, parathyroid adenoma, hypercalcemia, surgery

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Introduction

Around 80% of patients treated for primary hyperparathyroidism have an associated parathyroid adenoma. It can rarely be hyperplasia of the parathyroid gland and exceptionally a parathyroid carcinoma.[1] Parathyroid adenomas usually develop in the lower parathyroid glands. Although ectopic localization within the mediastinum has been reported, a retrotracheal parathyroid adenoma remains exceptional.[2,3] Herein, through this clinical case report of hypersecreting ectopic parathyroid adenoma, we would like to discuss the importance of a precise topographic assessment to provide appropriate surgical management.

Case Presentation

A 69-year-old patient, who presented with asthenia, shortness of breath, palpitations, and evolving for 2 weeks, was referred to our thoracic surgery service after a fortuitous discovery of a mediastinal mass as part of the etiological assessment of his malignant hypercalcemia. The patient was found to have a malignant hypercalcemia (corrected calcemia 3.39 mmol/L, normal range of 1.15-1.35 mmol/L) and an elevated parathyroid hormone (56.4 pmol/L, normal range of 1.4-6.8 pmol/L). His blood pressure was 149/93 mmHg with a heart rate of 113 beats/minute. On the electrocardiogram, the patient's corrected QT interval was 335 ms. A thoracic computed tomography (CT) with contrast injection revealed a 3.8 cm enhanced retrotracheal lesion located in the posterior mediastinum (Figure 1A). The context of hyperparathyroidism with hypercalcemia, along with the CT-scan findings, prompted us to perform a technetium (^{99m}Tc) sestamibi scan. It revealed a significant hyperfixation by the lesion initially discovered on CT-scan, suggesting an ectopic parathyroid adenoma of the posterior mediastinum (Figure 1B). Surgical exploration after incision of the mediastinal pleura by thoracotomy found a well-encapsulated mass of the posterior and superior mediastinum behind the trachea (Figure 2A). The histopathological examination of the resected specimen disclosed the diagnosis of a parathyroid adenoma (Figure 2B). The patient's postoperative course was uneventful with normalisation of phosphocalcic metabolism and improvement of clinical symptoms. The consecutive postoperative calcium values of the patient were as follow: 1.01 mmol/L just after the surgery; 1.08 mmol/L 2 days after the surgery; and 1.19 mmol/L 4 days postoperatively.

After 6 months from surgery, the patient had no signs of the disease, his serum calcium and parathyroid hormone levels were within normal limits.

Discussion

The incidence of ectopic intrathymic parathyroid adenomas varies according to authors.[3,4] In the Phitayakorn and Mc Henry[5] series of 231 patients operated for hyperparathyroidism, 16% had ectopic lesions; of which, 3% were intrathymic. In fact, the lower parathyroids and the thymus have a common embryological origin from the third branchial pouch before their respective cervical and mediastinal migration.[6] This migration explains the possibility of supernumerary and ectopic parathyroid glands. Although intra-thoracic localization has been the subject of numerous publications, the posterior mediastinal retrotracheal parathyroid adenoma remains an exceptional localization. Primary hyperparathyroidism

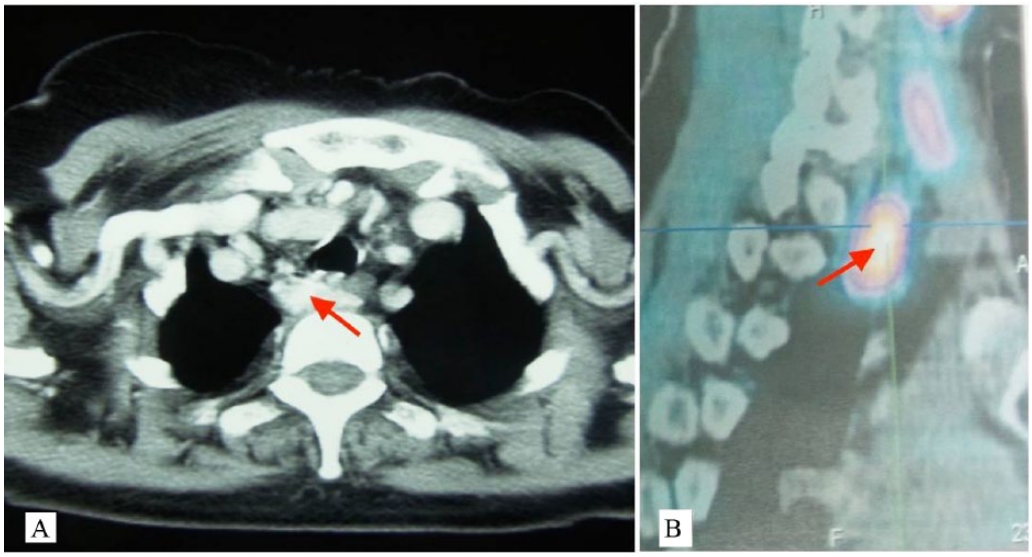

Figure 1. (A) Injected thoracic CT-scan shows an enhanced tissular mass of the posterior and superior mediastinum (arrow). (B) Hyperfixation of the lesion seen on the technetium (^{99m}Tc) sestamibi scan (arrow).

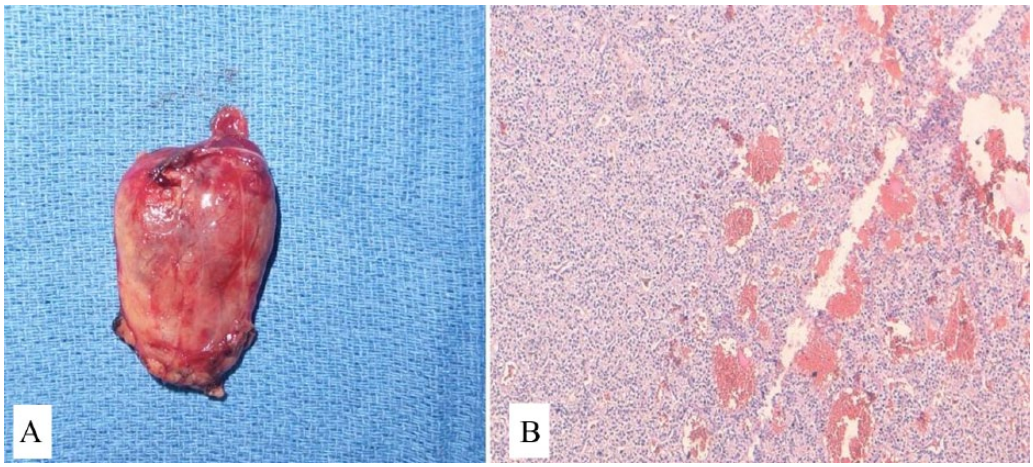

Figure 2. (A) The resected specimen showing a well-encapsulated solid mass. (B) The histological view showing a solid nested architecture with benign-looking cells arranged in a delicate vascular and hemorrhagic stroma (mainly parathyroid chief cells) (H&E ×50).

remains the most common finding. Symptoms are usually characterised by recurrent lithiasis of the urinary tract, calcium metabolism disorders such as hypercalcemia, muscle fatigability, cardiac arrhythmias, or spontaneous bone fractures.[7] Apart from these clinical signs secondary to metabolic disturbances, the clinical examination is generally non-specific. It is the persistence of signs of clinical and biological hypercalcemia that suggests the diagnosis of primary hyperparathyroidism, leading to radiological examinations such as ultrasonography and cervico-thoracic CT. In the specific case of our patient, ultrasound of the cervical region was not contributory, and we completed the assessment with an injected thoracic CT-scan which leads us to discover a mass of the posterior and superior mediastinum, localised in the retrotracheal region. The mass was enhanced after the injection of the contrast product. To support the diagnosis, a technetium (^{99m}Tc) sestamibi scan should be performed when a diagnosis of ectopic hypersecreting parathyroid gland is suspected. In fact, scintigraphy is currently considered the technique of choice, as it has a sensitivity of 95% and offers better image quality than CT with a lower irradiation rate.[8,9] In the Mitchell report, scintigraphy identified 16 adenomas of 17 cases and 19 glandular hyperplasia of 21 cases.[10] From a therapeutic point of view, complete resection of hypersecreting tissue is the appropriate management. Surgery is recommended for asymptomatic patients who have measurable markers of disease that is more severe at the outset or more likely to progress: young age (<50 years), moderate or severe hypercalcemia, osteoporosis or fragility fractures indicating substantial bone loss, or renal disease.[11] The morbid or even fatal consequences of hyperparathyroidism such as increased risk of premature death, blood hypertension, myocardial hypertrophy, and bone loss especially in elderly women have been reported in several studies, justifying the indication of the surgical management.[12–14] In symptomatic patients, parathyroidectomy leads to normalisation of

phosphocalcic metabolism with increase in bone density.[15] Although the surgical modalities are still controversial, the thoracic approach by sternotomy remains the classical approach for the ectopic parathyroid adenomas of mediastinal localization. In 1994, an alternative by video-assisted thoracoscopy was proposed with the aim to reduce the extension of the classical approach and reduce its morbidity.[16,17] This mini-invasive approach offers a good exploration of the lesion within the mediastinum. Its disadvantages are the lack of three-dimensional vision and the loss of dexterity of the surgeon compared to conventional surgery.[18] Hypocalcemia has been reported as the most common postoperative complication. The absence of this hypocalcaemia is a predictive element of incomplete resection of the entire hypersecreting tumoural tissue. Hypocalcemia is moderate in patients operated for primary hyperparathyroidism. Hypocalcemia should be treated regardless of symptoms (tetany, perioral numbness/tingling, and muscle cramping) as untreated hypocalcemia from transient hypoparathyroidism can potentially lead to cardiac arrhythmias. The persistence of hypocalcemia beyond 4 to 5 days after the surgery is most often due to an avidity of bone for calcium, thus requiring phosphocalcic supplementation and vitamin D therapy.[19]

Conclusion

The ectopic parathyroid adenoma is a rare lesion due to a defect in the migration of the parathyroid gland during embryogenesis. Its mediastinal retrotracheal localization is exceptional. An adequate topographic assessment must be conducted to perform a suitable surgical management leading to a complete resection of the hypersecreting tissue.

Authors' Note

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Author Contributions

All the authors contributed substantially to the authorship of this case report. S.R., H.H., M.L., Y.O., and M.S. were directly involved in the patient's care.

Informed Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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