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Case Report



Cervical Retrieval of Giant Posterior Mediastinal Parathyroid Adenoma with Bizarre Morphology

Abstract

Parathyroid adenoma is the most common cause of primary hyperparathyroidism and rarely reaches huge sizes. As the gland enlarges it may exhibit atypical morphology and extension to the mediastinum which may complicate the excision of the tumor while preserving the capsular integrity. We present a 35-year-old male patient who was referred to our department with a complaint of severe hypercalcemia. Neck ultrasound and parathyroid scintigraphy revealed an adenoma of 45x25 mm in size in the left parathyroid lobe extending to the retrosternal area. The patient underwent an uneventful parathyroidectomy. The patient remained well with no evidence of biochemical recurrence during the 4-year follow-up. Here, we presented a very original case of one of the largest parathyroid adenomas (8 cm) with atypical configuration (having multiple spicular extensions from the main body) ever reported in the literature which was completely retrieved from the posterior mediastinum via cervical route.

Keywords: Giant adenoma, mediastinal parathyroid, hyperparathyroidism, parathyroidectomy

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Parathyroid adenoma (PA) is the most common cause of primary hyperparathyroidism (PHPT) and constitutes almost 80-85% of the PHPT.^[1] Although the majority of the PA's are smaller than 2 cm and lighter than 1 gram (g), rarely they may reach very huge dimensions and display atypical morphologic shape.^[2] Parathyroid adenomas heavier than 3.5 g are defined as giant parathyroid adenomas which had to be identified in such a subgroup since they display different clinical entities than non-giant adenomas and they are more likely to have higher serum calcium and parathormone levels which were usually attributed to parathyroid carcinoma.^[3]

The vast majority of the parathyroid adenomas are oval or rounded with margins that are smooth or slightly lobulated. Less common configurations of parathyroid adenomas are discoid, teardrop, tubular, and pyramidal. [4] As the gland enlarges it may exhibit atypical morphology which may complicate the excision of the tumor with preserving the capsular integrity. The spicular extensions from the main body of the adenoma have not been reported yet.

Herein we presented a very original case of one of the largest parathyroid adenomas with atypical configuration ever reported in the literature which was retrieved from the posterior mediastinum via cervical route.

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Case Report

A 35-year-old male patient with hypertension and nephrolithiasis was admitted to the hospital and during the investigations, severe hypercalcemia [15.4 (N=8.5-10.6) mg/ dL] and hypophosphatemia [1.9 (N=2, 3-4, 7) mg/dl] were detected. Further investigations revealed elevated serum parathormone level [1014 (18-75) pg/mL], and other biochemical parameters were unremarkable. The patient was referred to general surgery clinic for the treatment of PHPT. Neck ultrasound showed a part of 45x25 mm vascular echogenic mass in the left parathyroid lobe extending to the retrosternal area. Parathyroid scintigraphy using 99 mTc-MIBI and fusion images with single-photon emission computed tomography with CT (SPECT/CT) demonstrated increased uptake over the inferior posterior aspect of the left thyroid gland extending to the posterior mediastinum (Fig. 1). Initial management was hydration and forced diuresis with furosemide since the patient had severe hypercalcemia. The lowest serum calcium value achieved with diuresis was 13.6 mg/dL, so 4 mg intravenous zoledronate was administered to prepare the patient for anesthesia and surgery. After zoledronate administration, it was 11.9 mg/ dl on the morning of surgery.

In operation using a 5 cm cervical Kocher incision, a huge, dirty-yellowish bizarre shape mass was observed behind the left thyroid gland extending to the posterior mediastinum. There were minimal adhesions with the gland capsule and surrounding tissue and it had well-delineated margins from the thyroid capsule. Cautious stepwise sharp dissection and traction enabled the complete retrieval of the mass via transcervical approach in en bloc manner with the use of intermittent neuromonitoring of the left recurrent laryngeal nerve. The capsular integrity was preserved during the excision and atypical, small spicules of the main body were left on the gland. Eighty-five percent decline in intraoperative parathormone assay (997 pg/mL-147 pg/mL) was achieved 10 minutes post excision. The left lower parathyroid gland was seen in normal location with normal morphology. On the postoperative first day, parathormone was 6 pg/mL and serum calcium was 11.3 mg/dL. The patient was discharged on the postoperative second day without any complications with a serum calcium level of 9.6 mg/dL with an oral calcium supplement for on-demand use.

Macroscopic examination revealed a solitary parathyroid adenoma measured 80x40x26 mm in dimension and 33 g in weight with two main spicules to the side from the main body under the same capsule (Fig. 2). The gland sur-

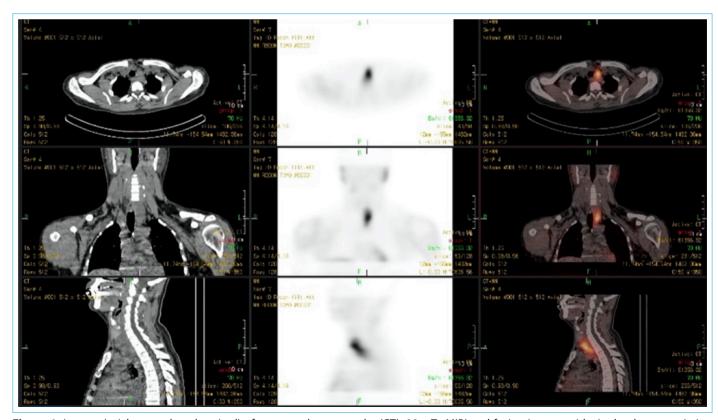


Figure 1. Images (axial, coronal, and sagittal) of computed tomography (CT), 99 mTc-MIBI and fusion images with single-photon emission computed tomography with CT (SPECT/CT) revealed an increased uptake at the inferior-posterior aspect of the left thyroid lobe and extended to posterior mediastinum.



Figure 2. A very unique shape of the parathyroid adenoma. The red arrow marks the spicules and the black arrow marks one of the septa on the surface.

face was septated to many small bumps under the capsule. Microscopically, intermittent areas of adipose tissue and parathyroid cells forming solid and trabecular layers in thin hyalinized stroma were observed (Fig. 3a). Parathormone showed a positive diffuse immunohistochemical staining pattern (Fig. 3b) whereas the Ki67 proliferation index was <1% in the parathyroid adenoma. The patient remained

well with no evidence of biochemical recurrence during the 4-year follow-up. Written informed consent was obtained from the patient who participated in this study.

Discussion

Severe hypercalcemia of PHPT requires a prompt intervention to reduce serum calcium and the patients should be prepared for elective parathyroidectomy as soon as possible. The severe hypercalcemia and extremely high serum parathormone levels traditionally were attributed to parathyroid carcinoma; however, most of those patients had giant adenomas rather than carcinoma. [5] As the gland size increases, the morphology may change and exhibit an unusual pattern: some budding or spicular multiple extensions from the main body of the gland have not been reported before and they could complicate the en bloc excision of the gland while preserving the capsular integrity. The capsular integrity during the parathyroidectomy is important because the capsular rupture and seeding of the parathyroid cells may result in recurrences after a parathyroidectomy which is more prominent in parathyroid carcinoma.[5]

A cervical incision is considered to be adequate for most mediastinal parathyroid adenomas while an extracervical approach is required for 1–2% of cases. [6] Some single case reports were reported describing giant mediastinal parathyroid adenomas excised via a transcervical / extracervical approach in the prior literature. [7-9] According to recently published case reports, the patients with larger and heavier parathyroid adenomas than ours all underwent extracervical interventions such as thoracotomy or sternotomy. [8,9] In our case, although deeply located and extended to

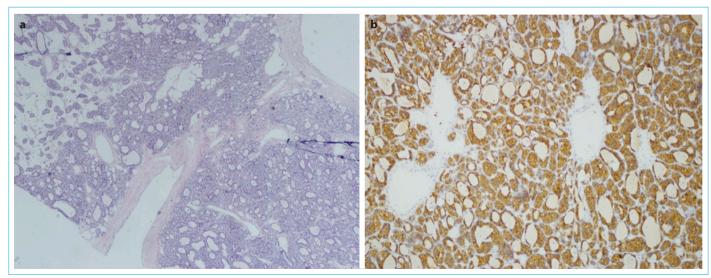


Figure 3. (a) Hypercellular parathyroid tissue with hyalinized stroma and minimal adipose tissue (x4, Hematoxylin&Eosin). **(b)** Diffuse cytoplasmic staining (x20, parathormone, immunohistochemistry).

the posterior mediastinum, the retrieval of the parathyroid mass via a transcervical approach could be achieved. It would probably be a superior adenoma prolapsed to posterior mediastinum since a normal lower gland was encountered in normal position. The adenoma was one of the largest to be excised via a transcervical route from the posterior mediastinum.

In conclusion, the case described is highly unusual in its size and shape. To our knowledge, it was one of the largest and heaviest posterior mediastinal solitary parathyroid adenomas ever reported in the literature which was retrieved via cervical route. It should be kept in mind that with cautious stepwise dissection, retrieval of giant mediastinal parathyroid tumors can be achieved via the cervical route preserving the gland's capsular integrity even for adenomas with atypical configuration.

Disclosures

Informed Consent: Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.

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Patient Informed Consent: Informed consent was obtained from participant for whom identifying information is included in this article.

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References

- 1. Duan K, Gomez Hernandez K, Mete O. Clinicopathological correlates of hyperparathyroidism. J Clin Pathol 2015;68:771–87. [CrossRef]
- Neagoe RM, Sala DT, Borda A, Mogoantă CA, Mühlfay G. Clinicopathologic and therapeutic aspects of giant parathyroid adenomas - Three case reports and short review of the literature. Rom J Morphol Embryol 2014;55:669–74.
- 3. Spanheimer PM, Stoltze AJ, Howe JR, Sugg SL, Lal G, Weigel RJ. Do giant parathyroid adenomas represent a distinct clinical entity? Surgery 2013;154:714–9. [CrossRef]
- 4. Hoang JK, Sung WK, Bahl M, Phillips CD. How to perform parathyroid 4D CT: tips and traps for technique and interpretation. Radiology 2014;270:15–24. [CrossRef]
- Gücek Haciyanli S, Acar N, Gür EÖ, Çelik SC, Karaıslı S, Dilek ON, et al. Severe hypercalcaemia of primary hyperparathyroidism: could giant adenoma be the real culprit rather than carcinoma? Ann R Coll Surg Engl 2020;102:363–8. [CrossRef]
- 6. Hu J, Ngiam KY, Parameswaran R. Mediastinal parathyroid adenomas and their surgical implications. Ann R Coll Surg Engl 2015;97:259–261. [CrossRef]
- 7. Miller BJ, Isaacs K, Khan E, Palazzo FF. Transcervical excision of a giant mediastinal parathyroid adenoma. BMJ Case Rep 2019;12:e228292. [CrossRef]
- 8. Migliore M, Pulvirenti G, Okatyeva V, Cannizzaro MA. Persistent hyperparathyroidism owing to a giant parathyroid adenoma in posterior mediastinum. Surgery 2013;154:132–133. [CrossRef]
- 9. Garuna Murthee K, Tay WL, Soo KL, Swee DS. A migratory mishap: giant mediastinal parathyroid adenoma. Am J Med 2018;131:512–6. [CrossRef]