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Case Report

Atypical Giant Parathyroid Adenoma, Case Report

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Abstract

Atypical giant parathyroid adenoma is a rare tumor with unusual histological features, an unusual presentation of primary hyperparathyroidism but more elevated laboratory findings and more severe clinical presentation due to larger tissue mass, and defined as weighing>3.5 g, leading to diagnostic and therapeutic challenges. Herein, we report a case of a 56-year-old woman who presented with a palpable neck mass, fatigue, persistent hypercalcemia, osteoporosis and asymptomatic renal stones, Ultrasound showed a left-sided solid nodule, and Sestamibi nuclear scan showed a giant parathyroid adenoma. The diagnosis of giant atypical parathyroid adenoma was confirmed after surgical excision size (3.5*1.5cm). We discuss the investigations, treatment, and outcome of this rare case and highlight the importance of long-term follow-up care.

Keywords: Atypical parathyroid adenoma, hypercalcemia, surgery, Osteoporosis, kidney stones.

INTRODUCTION

Parathyroid adenoma is the most common cause of primary hyperparathyroidism and the third most common endocrine disorder after diabetes mellitus and hypothyroidism [2] and is characterised by excessive secretion of parathyroid hormone, leading to hypercalcemia, bone loss, and other complications. Atypical parathyroid adenoma is a rare variant of the parathyroid tumor with some features associated with parathyroid carcinoma but lacking unequivocal evidence of malignant involvement such as unusual histological features, including mitotic figures, nuclear atypia, and invasion of adjacent tissues. Also, those patients usually present with calcium levels intermediate between those of adenomas and carcinomas [1]. Studies reported that atypical parathyroid adenomas pursue a benign clinical course. The diagnosis and management of atypical parathyroid adenomas requires a multidisciplinary approach, including endocrinologists, radiologists, and surgeons. Herein, we report a case of a 56-year-old female patient with atypical parathyroid adenoma, highlighting the diagnostic and therapeutic challenges as well as the importance of long-term follow-up care.

CASE REPORT

A 56-year-old woman presented with fatigue, constipation, abdominal discomforts, urinary symptoms with dysuria related to her kidney stones back pain and osteoporosis. She denied any history of bone fractures. She had no family history of thyroid or parathyroid disorders. Her laboratory results showed hypercalcemia (serum calcium level, 12.8 mg/dL), and an elevated parathyroid hormone (PTH) level (925 pg/mL). A neck ultrasound revealed a left parathyroid mass, and a 99mTc-sestamibi scan confirmed a left inferior parathyroid adenoma. The patient underwent surgical excision of the adenoma, which was histologically confirmed to be an atypical parathyroid adenoma.

Investigations

Further investigations were performed to evaluate the extent and characteristics of these tumors. A Tc-99m Sestamibi parathyroid SPECT-CT scan showed a left inferior parathyroid mass measuring 3.5*1.5 cm in diameter without evidence of invasion or lymphadenopathy (Figure 1). The patient's bone mineral density (BMD) was also evaluated, and a T-score of -2.4 at the lumbar spine indicated osteoporosis. Her laboratory results showed hypercalcemia (serum calcium level, 12.8 mg/dL), hypophosphatemia, and an elevated parathyroid hormone (PTH) level (925 pg/mL).

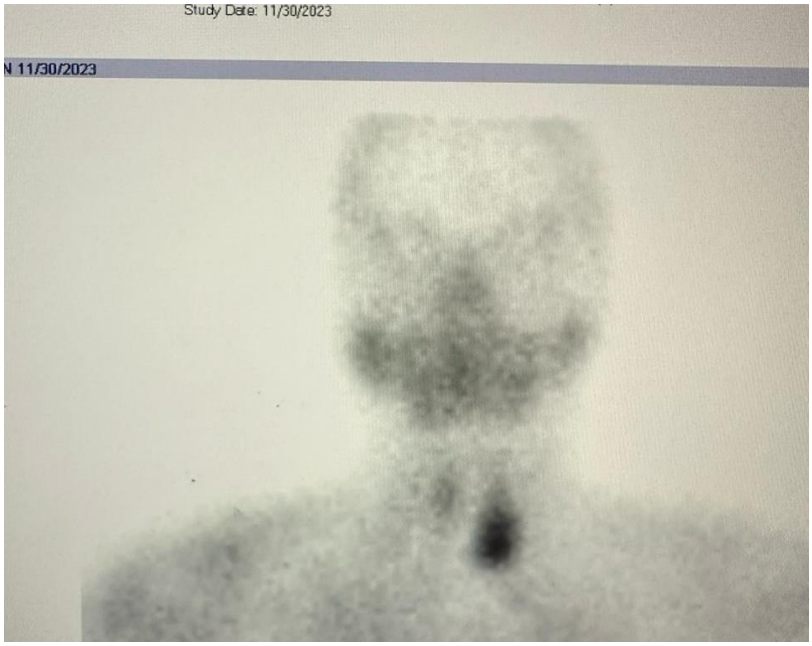

Figure-1

Treatment and Outcome

The patient underwent open left inferior Para-thyroidectomy, A Giant parathyroid adenoma was identified (Figure 2), and atypical parathyroid adenoma was confirmed histopathologically, the microscopic examination showed a nodular tumor with thick fibrous septae, composed predominantly of the proliferation of oncocytic cells with abundant granular cytoplasm. An area of proliferation of cells with clear cytoplasm is also seen.

Moderate nuclear atypia was also seen, and foci of fibro-fatty tissue with foci of fat necrosis and chronic inflammation were also seen.

Acinnary studies:

- CD56, GATA3: positive in tumor cells.
- Synaptophysin: weak positive.
- TTFI, thyroglobulin, chromogranin A, Calcitonin, cyclin D1: negative.
- Ki 67 shows a proliferative index of 1-2%.

The patient's postoperative course was uneventful, and she was discharged on the second postoperative day. Her serum calcium and PTH levels normalized, and she did not require calcium or vitamin D supplementation. Follow-up neck ultrasound and serum calcium levels were normal three months postoperatively. The patient continued to undergo regular follow-ups with serial measurements of serum calcium, PTH, and BMD.

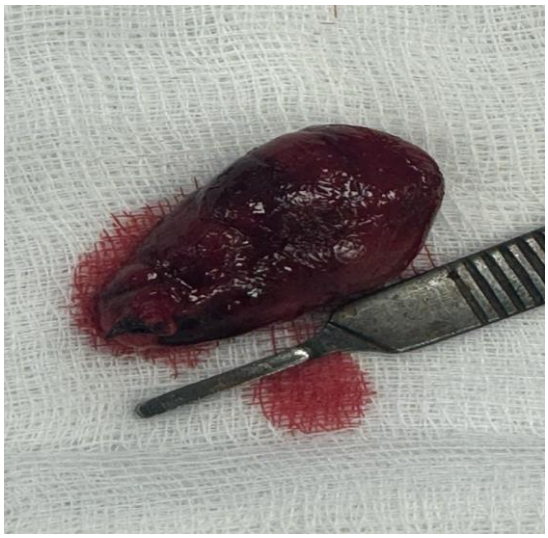

Figure-2

DISCUSSION

Atypical parathyroid adenoma is a rare tumor that accounts for less than 1% [3] of all parathyroid tumors. Among them about 1.2%-1.3% have malignant potential and are responsible for primary hyperparathyroidism [4], It has been described as a variant of parathyroid adenoma with unusual histological features including mitotic figures, nuclear atypia, and capsular or vascular invasion. The diagnosis is reached by excluding the presence of malignancy criteria.

The clinical picture is related to severe hypercalcemia, including anxiety, anorexia constipation, nausea and vomiting, bone pain, kidney stones, and osteoporosis.

The aetiology of the atypical parathyroid adenoma remains unknown, and only limited studies tried to explain it.

Regarding radiological assessment, the initial imaging modality of choice for evaluating the neck is ultrasound. This allows for a detailed description of the mass. Including whether it is cystic or solid, its size, and its relationship with nearby anatomical structures.

Technetium 99m Sestamibi Parathyroid scan is a procedure in nuclear medicine which is performed to localize parathyroid adenoma and is the preferred radiological imaging technique for preoperative tumor localization.

In terms of tumor management, the treatment is only surgical with en-bloc resection, Subsequently, the histopathological examination plays a crucial role in verifying the diagnosis of atypical parathyroid adenoma and distinguishing it from other potential conditions like a benign typical parathyroid adenoma and parathyroid carcinoma. According to the World Health Organization, the Atypical Parathyroid Adenoma is defined as a parathyroid tumor that does not show locally advanced growth or metastases but may show cell atypia, fibrotic tissue, trabecular growth, fibrotic capsular involvement, and increased mitotic rate [5].

Occasionally, even after conducting a histopathological analysis of the tumor, distinguishing between an Atypical Parathyroid Adenoma and Parathyroid Carcinoma can prove to be challenging, with only ongoing surveillance being able to confirm the diagnosis.

Parathyroid carcinoma is known to have a recurrence rate exceeding 50%, with the majority of recurrence happening 2-3 years post-surgery [6].

CONCLUSION

Typical Parathyroid Adenoma is a rare type of parathyroid tumors and may pose diagnostic and therapeutic difficulties. Early diagnosis and proper evaluation, which include imaging tests and histopathological analysis, are essential for effective management. Surgery is the preferred option, nevertheless, the size and features of the tumor might necessitate preoperative localization and meticulous intraoperative monitoring. because those tumors share similarities with both typical adenomas and parathyroid carcinoma, continued long-term monitoring and strict follow-up are vital to monitor for any recurrence and potential complications.

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