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Case Report
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Giant Double Parathyroid Adenoma: A Case Report and a Mini Review

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Abstract

We report the extremely rare case of a giant double parathyroid adenoma. A 56- year-old female was admitted to hospital due to primary hyperparathyroidism. During the typical investigation a giant tumor of parathyroid origin was detected behind the right thyroid lobe. The patient underwent neck exploration with double parathyroidectomy and right thyroid lobectomy. Based on the histological findings, the diagnosis of giant double adenomas of the right superior and right inferior parathyroid glands was set, while the thyroid gland showed nodular goiter lesions. Primary hyperparathyroidism may be difficult to be recognized since most cases are asymptomatic or mildly symptomatic. Preoperative investigation can contribute to define the appropriate surgical strategy.

Keywords: Primary hyperparathyroidism; Double parathyroid adenoma; Giant double parathyroid adenoma; Neck exploration; Parathyroidectomy

Introduction

Nearly 2% to 11% of primary hyperparathyroidism cases are caused due to double parathyroid disease with only four cases being attributed to giant lesions [1-6]. Here we present the fifth case of Giant Double Parathyroid Adenoma (GDPA). GDPA may pose a differential diagnosis dilemma since the high PTH levels and their size can be mistaken for a malignancy. Preoperative imaging is crucial for diagnosis. Surgical intervention is curative, while histopathology sets the definite diagnosis.

Case Presentation

A 56 year old female patient presented to the pathologic outpatient department due to generalized fatigue, muscle weakness and reduced bone mass. Upon investigation a PTH of 1056.80 pg/ml with serum calcium levels of 12.56 mg/dl and normal vitamin D3 were detected. Primary hyperparathyroidism was diagnosed and the patient was referred for further imaging investigation. Neck ultrasonography showed two distinct hyperechoic masses 7.7 cm × 3.8 cm × 2.8 cm and 3.4 cm × 3.1 cm × 2.1 cm behind the right thyroid lobe (Figure 1). Dual-phase Tc-99m parathyroid scintigraphy with early and delayed images was also performed. The results showed the presence of a 6 cm × 2 cm parathyroid tumor, indicated by the increased and persistent retention of the radiotracer uptake near the lower right thyroid gland on the early image that remained on the delayed image (Figure 2). Taking into consideration the malignant potential of the lesion(s), the patient was scheduled for neck exploration. Two giant parathyroid masses were recognized and excised (Figure 3). En-block right thyroid lobectomy was performed when removing the right inferior parathyroid gland. The remaining parathyroid glands appeared normal. Postoperative course was uneventful and the patient was discharged on the 2nd day. PTH levels, serum calcium, and serum phosphate was within normal range on the 7th postoperative day and on the 6th postoperative month. Patient's symptoms were also resolved.

Two specimens were sent for histopathological examination. Macroscopically, the first specimen, labeled as "right superior parathyroid gland", was ovoid in shape, measuring 8 cm × 4 cm × 2.5 cm and weighing 30.274 gram. On cross-section, it had solid consistency, a yellow-tan and brown-tan color and areas of hemorrhage (Figure 4A). The second specimen labeled as "right inferior parathyroid gland" which was attached to the lower pole of the thyroid lobe, with a band of fatty and connective tissue had a weight of 20 gm and size of 5.3 cm × 3 cm × 2.5 cm. Parallel sections revealed a yellow-tan/pinkish-tan surface. The thyroid specimen was composed by the right lobe (4.2 cm × 2.5 cm × 0.8 cm) and the isthmus with the pyramidal lobe, (0.5 cm × 0.5 cm × 0.2 cm). Microscopic examination of the first parathyroid gland revealed an encapsulated neoplasm

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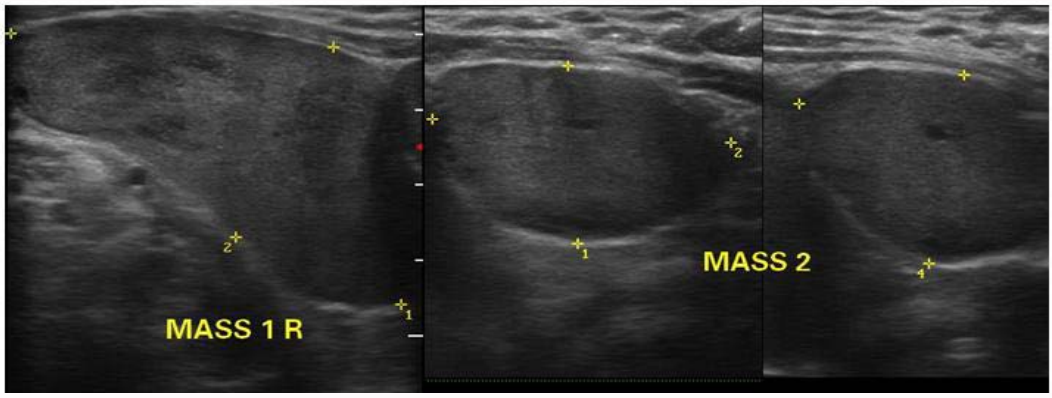

Figure 1: Preoperative ultrasonography depicted two masses behind the right thyroid lobe.

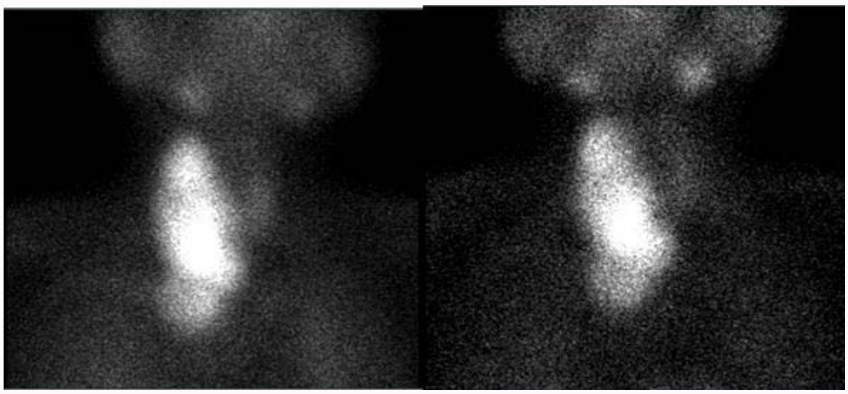

Figure 2: Early image Increased thyroid gland dimensions. 6 cm × 2 cm area of increased and persistent retention of the radiotracer uptake behind the lower right thyroid gland.
Delayed image: Persistent retention of the radiotracer uptake near the lower right thyroid gland.

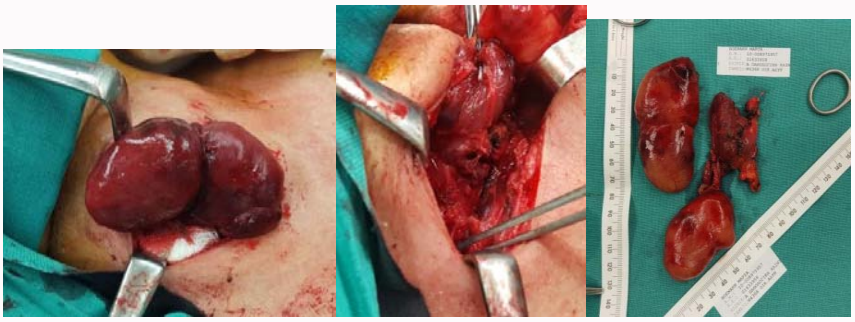

Figure 3: a) Intraoperative images showing resection of both the parathyroid adenomas and b) the right thyroid lobe, c) Parathyroid adenomas and right thyroid lobe after excision.

composed of hyperplastic chief and clear cells, demonstrating minimal nuclear polymorphism, atypia and mitoses (Figure 4B). The Ki67 proliferation index was low, approximately 3% (Figure 4C). The neoplastic cells were arranged in nests, adenoid structures or rarely in trabecula. Areas of fibrosis in the form of thin diaphragms (Figure 4D), edema and hemorrhage were observed in the stroma, as well as a focus of numerous foamy macrophages. No broad bands of collagen were present and no invasion of the capsule or capsular blood vessels were seen. Remnants of thymic tissue were visible at the extracapsular area, but no apparent normal parathyroid tissue. Hematoxylin and eosin-stained sections of the second parathyroid gland specimen revealed similar findings. The constituent chief and clear cells showed negligible nuclear polymorphism and atypia (Figure 4E). Small foci of fibrosis separated infrequently the neoplastic cells into solid clusters, nests and adenoid formations. The neoplasm was well demarcated with a thin fibrous capsule. No invasion of the capsule or its blood vessels was noted. At the periphery of the capsule, non-neoplastic parathyroid tissue and thymic tissue were recognized (Figure 4F). Sections from the thyroid parenchyma showed variable sized follicles, foci of fresh hemorrhage and limited lymphocytic infiltrates in the stroma. A band of fibrous connective tissue linked the parathyroid neoplasm to the lower pole of the thyroid lobe. The diagnosis of giant

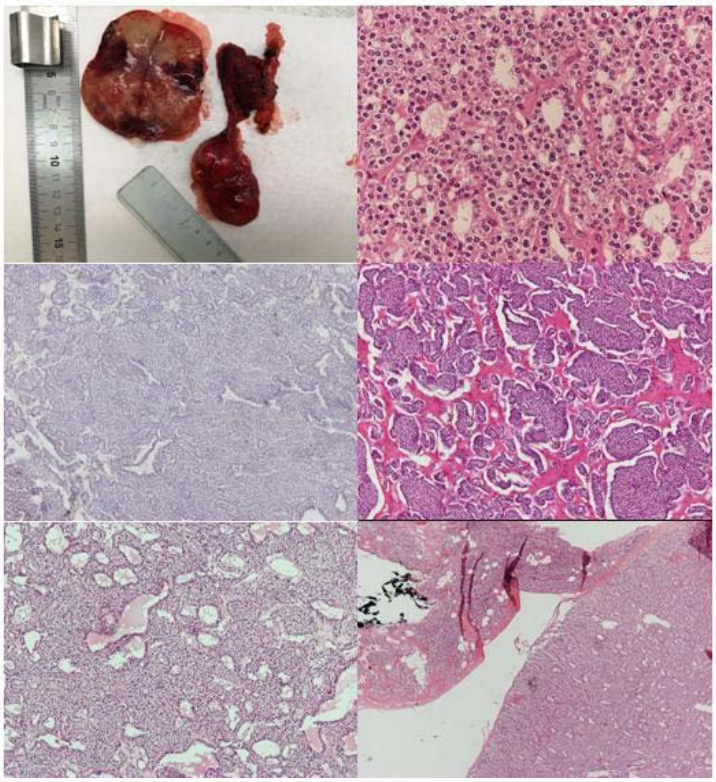

Figure 4: Macroscopic image of giant double adenomas, one of them attached with the lower pole of the right thyroid lobe. A. Histopathological findings of the largest adenoma showing chief and clear cells without pleomorphism, atypia or mitotic activity (B), a low proliferative index (C), and areas with thin fibrous septae (D). The second parathyroid adenoma presents similar morphological features E. and a rim of non neoplastic parathyroid tissue outside the capsule of the adenoma (F). (B, D: Hematoxylin and eosin x400; C: Ki67/MIB1 x400; E: Hematoxylin and eosin x100; F: Hematoxylin and eosin x40).

Table 1: Patients demographic and clinical data.

Date	Author	Sex	Age	Ca PTH
1995	Fahey	M	62	4.54 mmol/L 2.11 ng/L
2004	Younes	M	48	11.9 mg/dl 490pg/ml
2010	Sanoop	F	29	11.3 mg/dl 678 pg/ml
2016	Krishnamurthy	M	34	15.2 mg/dl 1789 pg/ml

double parathyroid adenoma was set.

Discussion

Giant parathyroid adenomas are defined as adenomas weighing >3.5 g [7]. Reporting a giant adenoma is expected. Reporting a giant double parathyroid adenoma is a scarce condition, though. Here we present the 5th case in the literature. Table 1 shows the demographic and clinical data of the other 4 cases. All patients, including ours, had remarkably elevated serum calcium and PTH levels. Their clinical presentation varied from atypical symptoms like generalized weakness and fatigue to severe symptoms with bone and renal involvement. However, no statistical significance was observed between patients with double adenomas and other patients with primary hyperparathyroidism regarding demographics, symptomatology, preoperative serum calcium and PTH levels as well [8,9]. Current localization imaging modalities include a combination of scintigraphy with high-resolution Ultrasonography (US), or scintigraphy with thin-section Computed Tomography (CT) [10,11]. In all of the reviewed cases preoperative investigation included parathyroid us and scintigraphy. However, sensitivity falls significantly in the case of multiple gland disease [12-14]. Interestingly, in 2 of the cases imaging failed to reveal the underlying pathology due to lesions overlapping [3,6]. As GDPA could mimic an atypical parathyroid neoplasm or a parathyroid carcinoma, neck exploration through a cervical approach should be the procedure of choice as it was performed in all cases. The affected glands were recognized and excised. In our case right thyroid lobe was also excised. Immediately postoperatively calcium and vitamin D3 supplementation is necessary so as to avoid "hungry bone syndrome" symptomatology. All patients seem to remain free of disease till nowadays.

Conclusion

Giant double parathyroid adenomas are an extremely rare finding. Severe hypercalcemia and extremely high PTH levels should activate further localizing investigation, most commonly with scintigraphy plus neck ultrasonography. Preoperative localization will allow optimal surgery planning.

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