The Giant Parathyroid Adenoma Reaching Mediastinum: A Case Report

Rahim Mahmodlou
Urmia University of Medical Sciences
Amin Sedokani (a.sedokani@gmail.com)
Urmia University of Medical Sciences https://orcid.org/0000-0003-1325-3309
Bita Najafinejad
Urmia University of Medical Sciences

Case report

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Abstract

Background: The primary hyperparathyroidism is the most common disease of parathyroid glands and the third common endocrine disease. Giant parathyroid adenoma is defined as weight > 3.5^{gr} and size of > 2^{cm}, however, the normal weight of the parathyroid gland is about 70^{mg} to 1^{gr}.

Case Presentation: This report describes the most giant diagnosed primary parathyroid adenoma due to literature. The case is a 48-year-old Persian man with past medical history of mitral valve replacement and several episodes of bilateral nephrolithiasis. The chief complaints were knee and lower back pain. After complete assessment, the neck mass with possible origin of thyroid were detected and resected by surgery. The size of resected mass was $9\times6\times4^{cm}$ weighting 122^{gr}. The histopathological assessment revealed giant parathyroid adenoma.

Conclusion: There are some reports of giant parathyroid adenoma describing the weight of up to 110^{gr} leading to significant hypercalcemia with a size of 8^{cm}. despite the size of the giant parathyroid adenomas and parathormone high levels, the calcium crisis may not always occur in these patients and the masses may be misdiagnosed with thyroid mass at primary assessments.

Background

The most common disease of parathyroid glands is primary hyperparathyroidism (PHPT), simultaneously, it`s the third common endocrine disease [1, 2]. The incidence of PHPT in the general population is 22 per 100,000 but in postmenopausal women, it reaches 1 per 500 [3]. In 80%-90% of PHPT cases Parathyroid adenoma is the cause of that disease and the remaining 10%-20% are due to parathyroid hyperplasia and < 1% due to parathyroid carcinoma [4]. Giant parathyroid adenoma (GPA) is defined as weight > 3.5^{gr} and size of > 2^{cm}, however, the normal weight of the parathyroid gland is about 70^{mg} to 1^{gr} [1, 5]. The mainstay of the pathophysiology of the hyperparathyroidism is increased secretion of parathormone (PTH), that cause releasing calcium from bone cells by inhibiting osteoblasts and stimulating osteoclast activity. In the kidney, PTH increases calcium reabsorption and also decreases phosphate reabsorption, and on the other hand besides by stimulating the conversion of 25-hydroxy Vit D to 1,25 hydroxy vit D, indirectly stimulate calcium absorption from the gut [5].

Clinical presentation of PHP is related to organs that are involved in calcium metabolism namely kidney (nephrolithiasis), skeletal system (osteoporosis-osteopenia), gastrointestinal tract (pancreatitis), nervous system (depression and cognitive disorders) and etc. The severity of symptoms is correlated with adenoma weight and PTH level [1, 5]. On the other hand, hypercalcemic patients may be presented with life-threatening conditions namely hypercalcemic crisis due to the high level of PTH secondary to parathyroid carcinoma or giant parathyroid adenoma [3, 6]. Nowadays, most of PHPT patients are diagnosed by screening tests by measuring serum calcium level and elevated intact parathormone, because the most

common cause of PHPT is adenoma and most adenomas are very small to diagnose, although PHPT have no specific symptoms [4, 5].

There are some reports of GPA describing the weight of up to 110^{gr} leading to significant hypercalcemia (3.21$^{mmol/L}$ equivaling to 12.84$^{mg/dL}$) with a size of 8×5×3.5^{cm} [7]. This report describes the most giant diagnosed primary parathyroid adenoma due to literature.

Case Presentation

The case is a 48-year-old Persian man, from Urmia, Iran that presented at the Urmia's Imam hospital with bone pain in knee and lower back pain that were started from two months ago and also constitutional symptoms such as fatigue and dizziness. In past surgical and medical history, the patient has undergone mitral valve replacement operation, 30 years ago, nephrolithotripsy several times due to bilateral nephrolithiasis, and also diabetes mellitus treating with oral agents. In physical examination vital signs were normal, however, the patient had a large nodule in left lobe of lower and middle neck (zone 2, 3) that migrate with his swallowing with soft consistent on palpation. An old scar due to previous cardiac surgery was seen in chest. Other physical examinations were normal. Due to evaluate differential diagnosis, the color Doppler ultrasonography of thyroid and parathyroid gland was done. The report revealed a solo isoechoic nodule in right lobe of thyroid (12×9.5^{mm}) and two solid cystic nodules in left lobe of thyroid (40×23^{mm} and 30×16^{mm}). The neck and chest CT scan with intravenous contrast showed giant parathyroid adenoma with solid and cystic sides in inferior and left lateral lobe (Fig. 1). In the primary laboratory tests CBC was normal. The biochemical tests are shown in Table 1.

Table 1
Biochemical and Urine Analysis tests of the patient.

Variable (Serum)	Patient's values	Reference range	Unit
Calcium	14.6	8.5–10.5	mg/dL
Na (Sodium)	138	136–145	mEq/L
K (Potassium)	4.3	3.5–5.5	mEq/L
Mg (Magnesium)	2.04	1.8–2.6	mg/dL
P (Phosphorous)	2.14	2.8–4.5	mg/dL
PTH (Parathyroid hormone)	2702	14–65	pg/mL
Macroscopic U/A			
Color	Yellow	Yellow	—
Appearance	Clear	Clear	—
PH	7	4.6–8	—
Protein	trace	Negative	—
Glucose	Negative	Negative	—
Blood / Hb	trace	Negative	—
Microscopic (U/A)			
WBC	1–2	0–2	per HPF
RBC	4–5	0–2	per HPF
Epithelial	2–3	0–1	per HPF
Mucus	Negative	Negative	per HPF

Due to confirmation of IV-contrasted CT scan and size of the mass, the plan was excisional surgical biopsy of whole mass. The operation was done under general anesthesia and after putting a roll between his shoulders (exposing the neck towards surgeon). A large collar incision was done (two finger width) upper suprasternal notch. Subplatysmal flaps created as routine thyroidectomy, the raphe between strap muscles were opened and on the left side, these muscles were incised as superior as possible. At first glance, the surgeons faced a thyroid mass that pushed the carotid sheet laterally and extended to the mediastinum. But evaluation of the mass revealed that the left lobe of the thyroid has been pushed superiorly and the mass that replaced the thyroid is separate from the thyroid and extended to the mediastinum. An artery was traversing the mass towards the carotid artery (inferior thyroidal artery). This finding gave the surgeons the clue that the mass was parathyroid mass, most probably originating from inferior parathyroid gland. With concerning recurrent laryngeal nerve after sharp dissection of carotid artery, surgeons ligated this branch where emerging from under of carotid artery. Then, with sharp and

blunt dissection, the lateral side of the tumor was released. Considering the posterior extension of tumor, by elevating with narrow deaver, anterior side of mass released sharply as distal as possible and the tumor delivered to the neck (Fig. 2). In the second step the recurrent laryngeal nerve was explored sharply that was located between the trachea and the medial side of the tumor, continued towards the larynx that in its way was traveling under the inferior thyroid artery. The surgeons removed the mass with adjacent lymph nodes without saving the other parathyroid gland, then resected the left lobe and isthmus of the thyroid. According to the preoperative investigation, surgeons did not explore the right side parathyroid. After inserting Hemovac drain, neck wound was closed. The size of resected mass was $9\times6\times4^{cm}$ weighting 122^{gr}.

We believe that it originated from the superior parathyroid gland because it had descended to retroesophageal space and it was posterior to the recurrent laryngeal nerve. Then after operative, for prophylaxis of bone hunger syndrome, we started intravenous infusion of 3000^{mg} calcium gluconate every 8^{h} ($9^{gr/24h}$).

At the first post-operative day liquid diet was started and the patient discharged on second post-operative day with oral $CaCO_3$ and Calcitriol pearls. The histopathological assessment of resected mass revealed a parathyroid adenoma and multinodular goiter for the resected thyroid lobe (Fig. 3). The patient was followed for six months, gradually the doses of calcium and calcitriol were reduced and eventually were discontinued. The patient did not have any further problem after six months.

Discussion

Parathyroid adenoma is an unusual, benign, and functional tumor of parathyroid gland. The primary symptoms are related to high levels of PTH causing increasing the reabsorption of Ca from bones, renal tubules and gut, leading to hypercalcemia signs and symptoms [1, 4, 8]. However, giant parathyroid adenoma can cause other symptoms for the mass size and location as dysphagia, sore throat, and dyspnea [9, 10]. Studies have revealed that in the most of the parathyroid masses, the serum level of Ca increases and due to increase in PTH levels and parathyroid function, combined use of ultrasonography and ^{99m}Tc-MIBI scintigraphy can help for diagnosis [11].

Our patient has suffered bilateral nephrolithiasis for multiple times. Considering back and bone pain, it can be interpreted that the adenoma was chronically functional and the patient has not experienced Ca creases symptoms. Our diagnosis of parathyroid mass in this case was primary with laboratory data of Ca and PTH. On the laboratory tests CBC were normal but we observed a very high level of PTH (up to 50 times of normal upper limit). However, the Ca raised less than the PTH. This could be due to chronic high levels of PTH or genetic resistance to PTH [12], but, we are not sure and we had no genetic or receptor

evidence to prove it. After surgery and follow up of the patient with treatment of calcium and calcitriol for six months (that eventually was tapered and discontinued), the patient showed no signs or symptoms of hypo or hyperparathyroidism that disproved any genetic disorder of Ca metabolism.

Eventually, giant hyperparathyroidism should be considered as a serious differential diagnosis of neck and cervical masses despite its rareness.

Conclusion

This case report described the most giant parathyroid adenoma have been diagnosed ever. The size of the mass, location, and the tissue view in the imaging may lead to misdiagnosis of the mass origin. Also, the very high levels of parathormone in these patients may not always lead to calcium crisis signs and symptoms.

Declarations

Ethics approval and consent to participate

The patient’s identity is secret and preserved unknown in the article and the patient received oral and written permission form that was approved by ethics committee of both Imam Hospital and Urmia University of Medical Sciences.

Consent for publication

The consent was obtained from the study participant prior to study commencement and the study participants gave consent to publish.

Availability of data and materials

Not applicable.

Competing interests

The authors report no kind of conflict of interest in this case report.

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Authors' contributions

In the present case report, the chief surgeon and supervisor was RM, AS and BN contributed in data collection and manuscript writing. All of authors have read and approved final revision of the manuscript.

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Figures

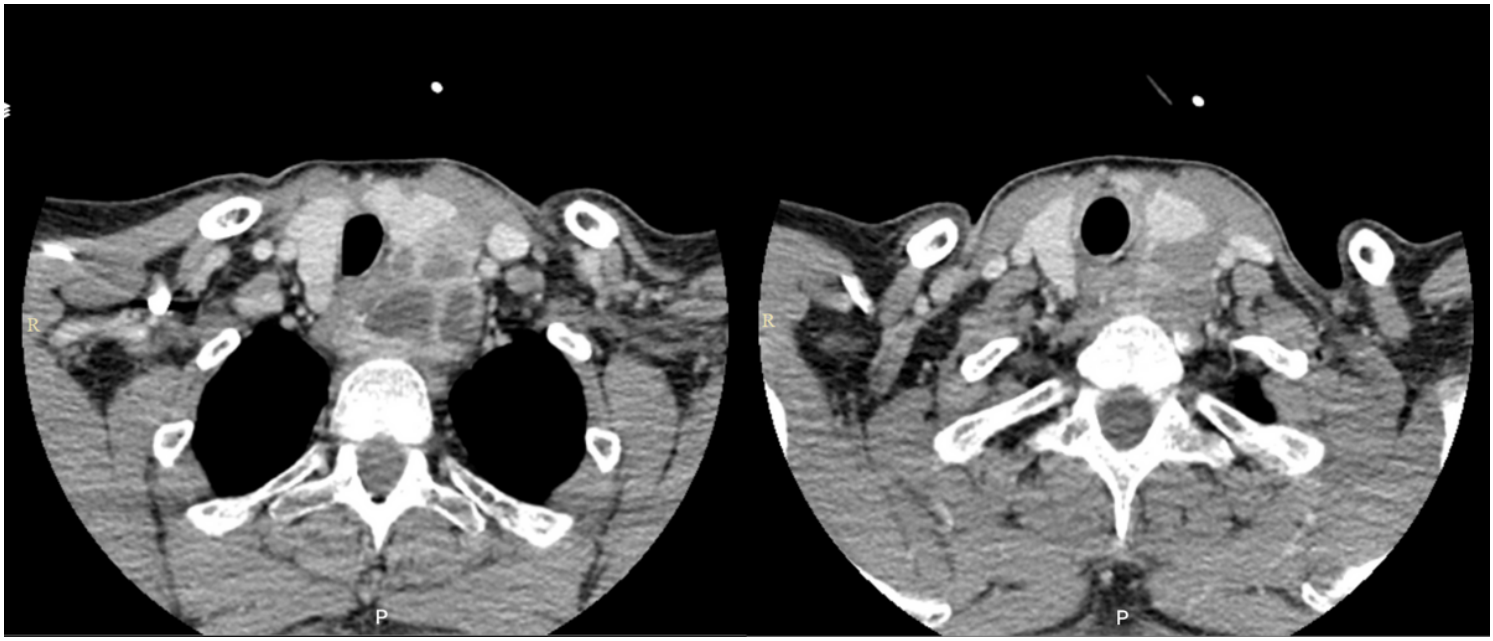

Figure 1

Histopathological view of the resected mass in favor of parathyroid adenoma.

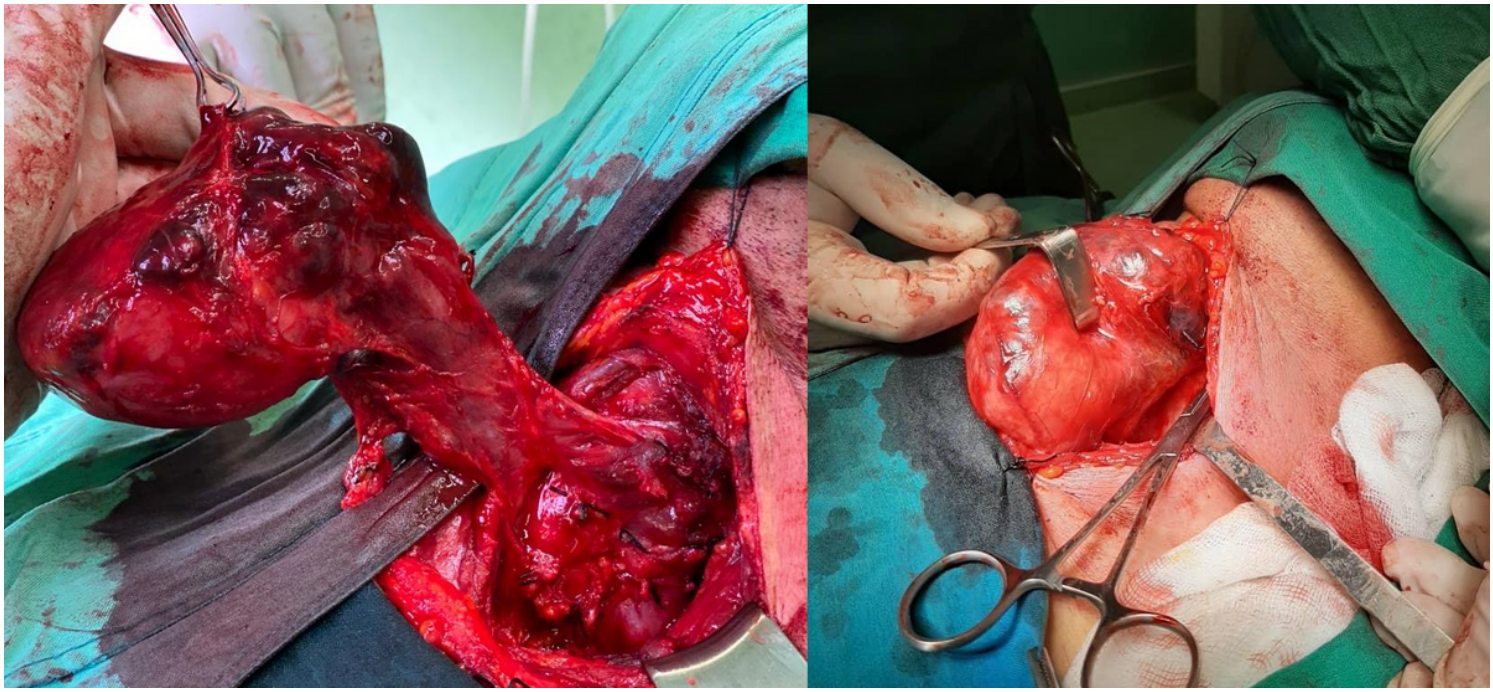

Figure 2

The CT scan of lower neck (right) and upper mediastinum (left) shows a huge mass pushing trachea out of midline.

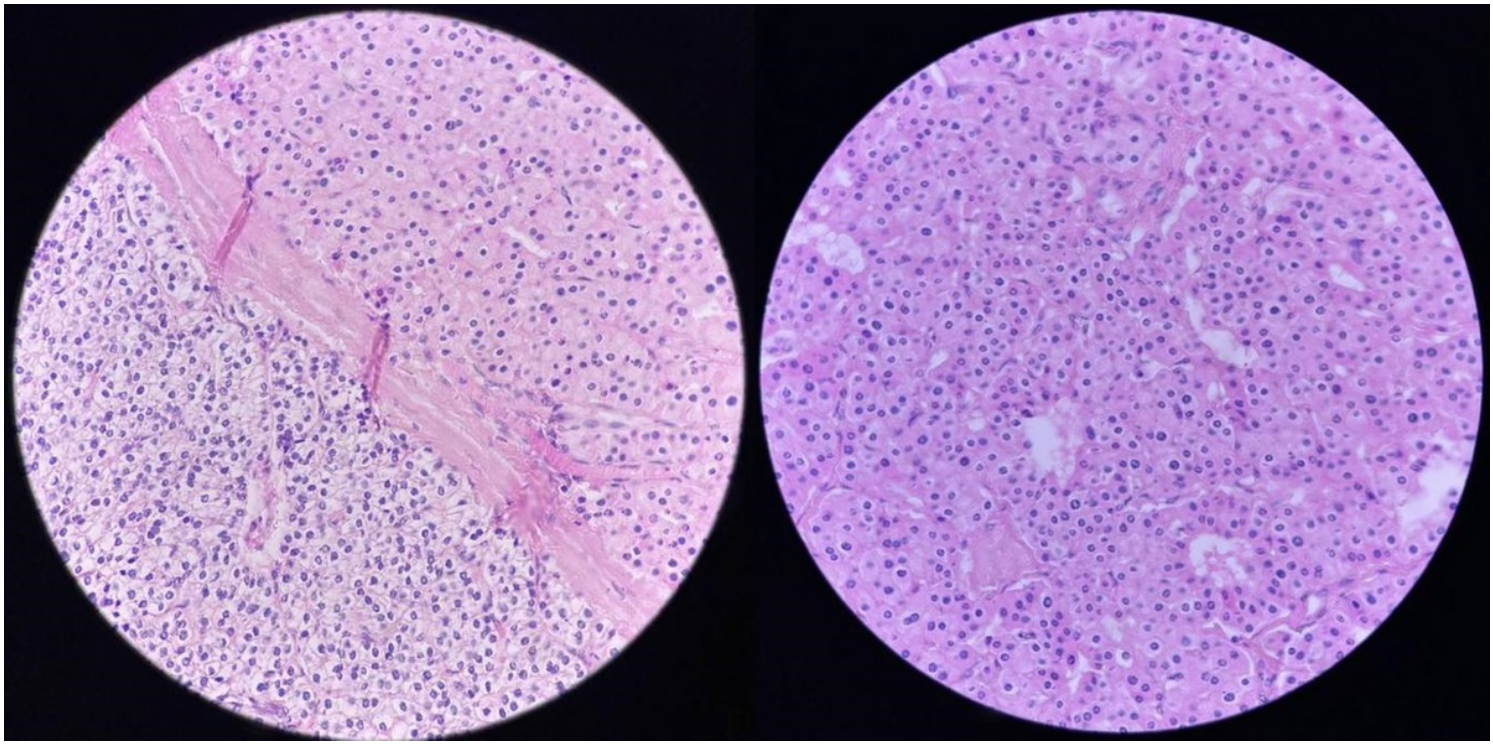

Figure 3

Surgical view of the mass obscuring the thyroid gland (right) and dissection of the mass (left).