Parathyroidectomy guided by intraoperative parathyroid hormone monitoring in a case of parathyroid hyperplasia

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ABSTRACT

Introduction: Primary hyperparathyroidism (PHPT) is a common endocrine disorder of the parathyroid glands, characterized by excessive production of parathyroid hormone by one or more of the parathyroid glands, causing hypercalcemia. The cause is usually an abnormal group of cells forming a benign parathyroid adenoma, hyperplasia, or, rarely, a carcinoma. Primary hyperparathyroidism is typically diagnosed by routine serum chemistry analysis showing hypercalcemia, hypophosphatemia, and elevated parathyroid hormone (PTH) level. Case Report: A case report of a 54-year-old female who presented with pain, swelling, and itching in the front of the neck for few months. Laboratory investigations revealed sustained hypercalcemia and gradually elevating PTH level. Computed tomography (CT) scan showed multiple hypo-enhancing thyroid nodules in both lobes. Ultrasonography (USG) showed an enlarged thyroid gland with multiple variable-sized cystic to spongiform nodules suggesting multinodular colloid goiter. Total thyroidectomy with right superior parathyroidectomy was done. Intraoperative PTH level monitoring showed markedly decreased level. Microscopically, fat tissue was present throughout the sections, no compressed tissue at the periphery, few chief and oxyphil (oncocytic) cells were seen.

Conclusion: These findings were most consistent with parathyroid hyperplasia. However, PTH level decreased immediately after surgery for >50% (Miami or “>50% intraoperative PTH drop” criterion). It can be either due to parathyroid adenoma or single gland hyperplasia. Thus, it could be challenging to distinguish between parathyroid adenoma and hyperplasia. Correct diagnosis is achieved only after correlating the pathologic findings with the clinical setting and biochemical results and it is advised for long-term follow-up with PTH level in cases of hyperplasia.

Keywords: Adenoma, Hypercalcemia, Hyperparathyroidism, Hyperplasia, Parathyroid hormone

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INTRODUCTION

Primary hyperparathyroidism (PHPT) is a common endocrine disorder of the parathyroid glands, characterized by excessive production of parathyroid hormone, causing hypercalcemia [1]. In this condition, one or more of the parathyroid glands are overactive. In 80% of patients with primary hyperparathyroidism, the underlying cause is an adenoma in which a single parathyroid gland is affected, whereas chief cell hyperplasia counts for 10–15% of cases, multiple adenomas for 5%, and rarely parathyroid carcinoma for <1% of cases [2].
Primary hyperparathyroidism is typically diagnosed based on the simultaneous presence of persistent hypercalcemia and elevated or inappropriately normal PTH level [3]. The differential diagnosis between parathyroid adenoma and hyperplasia is challenging as both are identical; one requires a good knowledge of the histopathology to distinguish parathyroid adenoma from hyperplasia [4]. Surgery is the most effective treatment up to 98% of the time. In parathyroid adenoma, PTH level decreases to normal level immediately after removing one of the parathyroid glands. Whereas in parathyroid hyperplasia, PTH level only decreases when all of four the parathyroid glands are removed. Medications that prevent calcium loss from the bone are also used as the only option for the patients who are unable to go for surgery [5].

CASE REPORT

A 54-year-old hypertensive female, presented with pain, swelling, and itching in the front of the neck for a few months. Physical examinations revealed a blood pressure of 130/80 mmHg, heart rate 82/min, temperature 36.5 °C, respiratory rate 20/min, clear chest, and soft non-tender abdomen. Laboratory investigations showed sustained hypercalcemia (10.7–11.4 mg/dL) and gradually elevating PTH level (159–168.4 pg/mL). Computed tomography scan showed multiple hypo-enhancing thyroid nodules in both lobes. No pulmonary consolidation, cavitation, or mediastinal lymphadenopathy was seen. Ultrasonography showed an enlarged thyroid gland with multiple variable-sized cystic to spongiform nodules. Some of these nodules showed curvilinear macro-calcifications and measured 1.5–2 cm in size. Thus, suggesting multinodular colloid goiter. Mild hepatomegaly with fatty changes, a borderline spleen (12.9 cm) and colonic gas distention were also detected.

A subsequent fine needle aspiration (FNA) from thyroid gland showed very sparse cellularity. Most of these cells are abundant cyst macrophages (pigmented and non-pigmented) with rare benign-appearing follicular epithelial cells. No malignant cells were seen in the smears examined. Thus, suggesting cystic benign follicular nodule. Intraoperative assessment and total thyroidectomy with right superior parathyroidectomy were done. Intraoperative parathormone level monitoring showed markedly decreased level (from 168.4 to 19.46 pg/mL; reference range 15–65 pg/mL).

The specimen was sent for histopathological examination. Grossly, a cut section of the right lobe showed multiple colloid filled areas. As for the left lobe, yellow areas were seen. Microscopically, sections from thyroid showed follicles of variable sizes and shapes (micro and macrofollicles), filled with homogenous colloid. Follicles were lined by simple cuboidal to flattened epithelium and arranged in nodular pattern intervened by areas of fibrosis. There was evidence of recent and old hemorrhage. Areas of cholesterol clefts surrounded by histiocytes and fibrosis were also seen. No evidence of malignancy was seen. Sections from the right superior parathyroid gland showed parathyroid tissue within a thin capsule. Fat tissue was present amidst the parathyroid cells (Figures 1 and 2). It was composed of hyperplastic chief cells and focal oxyphil (oncocytic) cells (Figure 3). Cells were mainly arranged in follicular architecture (Figure 4). No compressed, demarcated parathyroid tissue or atypia was seen. The findings were suggestive of multinodular goiter with degenerative changes in the thyroid and parathyroid hyperplasia. The postoperative phase was uneventful.

DISCUSSION

Primary hyperparathyroidism is a hypercalcemic condition generated due to excessive secretion of PTH by one or more of the parathyroid glands [6].
hyperparathyroidism can occur at any age, but most often affects people between 50 and 60 years of age. Women are of 3–4 times more affected than men [7]. Approximately 80% of patients with primary hyperparathyroidism are asymptomatic at diagnosis or have nonspecific symptoms, such as weakness, fatigue, anxiety, depression, or bone pains. A patient diagnosed in later stages may have symptoms like severe bone disease, nephrolithiasis, neuromuscular dysfunction, gastrointestinal problems, or cardiovascular disease [8]. A single parathyroid adenoma takes up to 80–85% of hyperparathyroidism, while parathyroid hyperplasia is responsible for only 10–12% [9].

Parathyroid adenoma is the most common cause of hyperparathyroidism. It is a benign neoplasm, characterized by enlargement of a single gland. Typically, in parathyroid adenoma, the remaining parathyroid glands have a normal or atrophic appearance [10]. Microscopically, the adenomas are encapsulated, cellular, mostly composed of chief cells and a small percentage of oxyphil (oncocytic) cells that are rarely seen in the case of hyperplasia. The nuclei appear round and densely stained, hyperchromatic, and larger than those present in normal parathyroid tissue. Furthermore, the presence of normal parathyroid tissue at the periphery of an adenoma helps to distinguish an adenoma from hyperplasia [11]. In contrast, chief cell hyperplasia involves all four glands that appear to be enlarged and hypercellular. In some cases, only one gland is visibly enlarged whereas the other is normal in size which can be confused with an adenoma. In other cases, all four glands appear normal in size but appear hyperplastic on histological examination. Enlargement can be either symmetrical or asymmetrical. The pattern of growth is usually diffuse or nodular [12]. The amount of stromal fat is remarkably decreased or absent, which results in an increase in the proportion of parenchymal cells to stromal fat cells [13].

In our case, a single parathyroid gland was enlarged. Microscopically, fat tissue was present throughout the sections, no compressed tissue at the periphery, few chief and oxyphil (oncocytic) cells were also seen. These findings are most consistent with parathyroid hyperplasia. However, PTH level decreased immediately after surgery for more than 50% which is according to Miami or “>50% intraoperative PTH drop” criterion and can be either due to parathyroid adenoma or single hyperfunctioning parathyroid hyperplasia [14]. Miami criterion is a protocol that uses a 50% decline from either the highest pre-incision or pre-excision intraoperative PTH level in a blood sample taken 10 min after excision indicating the completion and the success of the parathyroidectomy. The parathyroid hyperplasia is mostly identified surgically and by biochemical findings.

CONCLUSION

It could be challenging to distinguish between parathyroid adenoma and hyperplasia. Correct diagnosis is achieved only after correlating the pathological findings with the clinical setting and biochemical results, which is often assessed intraoperatively. Hyperplasia can be distinguished from adenoma by microscopy, while biochemical results help in determining whether it is a hyperfunctioning gland or not. It is advised to have a long-term follow-up with PTH level in cases of hyperplasia.

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**Author Contributions**

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Guarantor of Submission

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Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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