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Case Report

Life-threatening intrathyroidal parathyroid adenoma

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Abstract: Acute primary hyperparathyroidism and parathyroid crisis are characterized by life-threatening hypercalcemia, a rare disorder. A 69-year-old female patient presented at our hospital's neurology clinic with weakness, nausea, vomiting, depression, and hypercalcemia. Treatment of hypercalcemia resulted in no improvement in neurological symptoms, indicating resistance to treatment. Thyroid ultrasonography and parathyroid scintigraphy revealed hypoechoic nodules in the right lobe, pieces of nodules in the left lobe, and high serum calcium and parathyroid hormone levels. After provision of intensive medical treatment including hydration, diuresis, and bisphosphonate infusion resulted in only minimal decrease in the calcium level, urgent surgical treatment was performed. Frozen biopsy of the right intrathyroidal giant parathyroid adenoma in the right lobe confirmed initial diagnosis of primary hyperparathyroidism. Based on the biopsy findings, right parathyroidectomy and right total and left subtotal thyroidectomy were performed. Histopathologic examination revealed a parathyroid adenoma localized inside large thyroid nodules. Review of the findings resulted in diagnosis of intrathyroidal parathyroid adenoma. Symptoms of hypercalcemia improved rapidly during the postoperative period.

Keywords: Acute primary hyperparathyroidism, hypercalcemia, hyperplasia, intrathyroidal parathyroid adenoma, parathyroid crisis, parathyroidectomy

Introduction and summary

Hyperparathyroidism is an endocrine disease caused by excessive function of the parathyroid glands. The third most common of the endocrine diseases, hyperparathyroidism is classified as primary, secondary, and tertiary hyperparathyroidism [1]. Primary hyperparathyroidism is caused by increased production of parathyroid hormone from abnormal parathyroid glands. Secondary hyperparathyroidism is caused by an increase in parathyroid hormone (PTH) levels as a compensatory response to the hypocalcemic conditions resulting from chronic renal failure or degradation of gastrointestinal calcium absorption. Tertiary hyperparathyroidism is caused by chronic stimulation of the glands resulting in acquisition of autonomic features.

Of all cases of primary hyperparathyroidism, 85% are parathyroid adenoma, 15% parathyroid hyperplasia, and 3% to 4% carcinoma [2]. Primary hyperparathyroidism is often asymptomatic and seldom continues to manifest as parathyroid crisis. A sudden increase in calcium and PTH levels can be life threatening [3]. Here we describe our evaluation and treatment of a case of primary hyperparathyroidism that presented with neurological symptoms as the most prominent features along with life-threatening hypercalcemia. After observing no response to initial medical treatment, urgent surgical treatment was performed, resulting in successful treatment.

Case report

A 69-year-old female patient presented at our hospital's neurology clinic with weakness, nausea, vomiting, depression, and hypercalcemia. Despite treatment of hypercalcemia, no improvement in neurological symptoms was observed, indicating resistance to treatment. Thyroid ultrasonography revealed hypoechoic nodules in the right lobe, the largest of which was 23 × 15 mm and consistent with parathyroid adenoma. Several pieces of nodules, the largest of which was 5 × 3 mm, were also observed in the left lobe. Tc99m-MIBI (20 mCi)

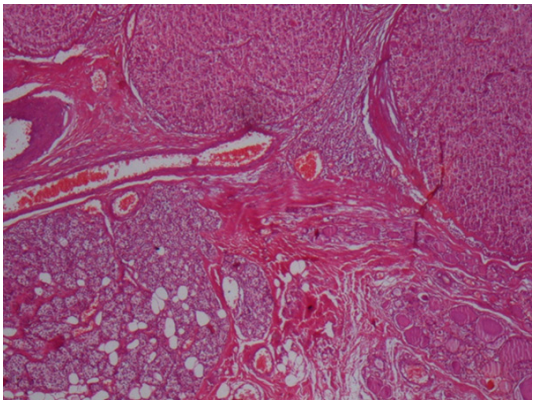

Figure 1. Microscopic view of intrathyroidal Parathyroid Adenoma, He × 2.

parathyroid scintigraphy of the focal lesion showed more intense activity than that of normal thyroid tissue in the sub-pole of the right lobe of the thyroid gland. While the serum calcium level (mean 13.7 mg/dL, range 8.2-10.9) and the intact parathyroid hormone level (mean 507 pg/ml, range 15-88) were high, all thyroid values (mean TSH level 1.14 uIU/ml, range 0.36-5.8; mean T4 level 0.89 ng/dl, range 0.61-1.12; and mean T3 level 3.62 pg/mL, range 2.5-3.9) were normal. Systemic examination revealed the presence of cholelithiasis, nephrolithiasis, eye keratitis, and corneal calcification.

Despite intensive medical treatment that included hydration, diuresis, and bisphosphonate infusion, the calcium level could only be reduced to 11 mg/dL, and urgent surgical treatment was performed. Frozen biopsy of the right intrathyroidal giant parathyroid adenoma in the right lobe confirmed diagnosis of primary hyperparathyroidism. Based on the biopsy findings, right parathyroidectomy and right total and left subtotal thyroidectomy were performed. Histopathologic examination revealed a parathyroid adenoma 2. 8 × 1.5 cm in diameter localized inside large thyroid nodules measuring 3.5 × 4 × 2 cm (**Figure 1**). Immunohistochemical analysis revealed the nodular structure to be thyroglobulin and galectin-3 negative, to contain large focal areas positive for progesterone receptor and heat shock protein 27, and to be characterized by a Ki-67 proliferation index below 1%. The patient was diagnosed with parathyroid adenoma localized inside the thyroid. Examination on the first postoperative day revealed a serum calcium level of 8.5 mg/dL and intact parathyroid hormone level of 3 pg/ml. Symptoms of hypercalcemia improved rapidly during the postoperative period. In the first year follow-up, serum calcium level was 9.4 mg/dL and intact parathyroid hormone level 52 pg/ml.

Discussion

Primary hyperparathyroidism occurs in 0.1% to 0.3% of the general population and is more common in women of advanced age. The disease is often asymptomatic, and parathyroid crisis is rarely observed. However, sudden onset of hypercalcemia, caused by an increase in vitamin D3 production, a decrease in renal calcium clearance, and an increase in PTH production increasing gastrointestinal calcium absorption, can be life-threatening. In these cases, urgent parathyroidectomy is recommended. The cause of primary hyperparathyroidism is growth of a single gland or parathyroid adenoma in 80% of cases, multiple adenomas or hyperplasia in 15% to 20% of cases, and parathyroid carcinoma in 1% of cases. Parathyroid hyperplasia should be suspected in cases of detection of multiple abnormal parathyroid glands before or during surgery until proven otherwise [4]. Although most cases of primary hyperparathyroidism are sporadic, it may be associated with inherited disorders, such as multiple endocrine neoplasia type 1 (MEN 1) syndrome, MEN 2A syndrome, or isolated familial hyperparathyroidism, all of which are autosomal dominant transitive.

In past years, patients often presented with kidney stones, bone pain, abdominal complaints, psychic symptoms, and extreme fatigue, a group of symptoms referred to as the “classic quintet”. However, patients today more often present with no or minimal symptoms. The most common symptoms of patients who do present with complaints are weakness, polyuria, polydipsia, bone and joint pain, constipation, itching, heartburn, depression, memory loss. Psychosis, depression, and anxiety are also often observed. Patients are usually admitted for elective surgery after preparation with medical treatment [5].

Recent studies have found the incidence of intrathyroidal parathyroid adenoma to be between 1.4% and 6% [6]. Regarding location, Cheng et al. found that 70% of adenomas are

located in the right and bottom third of the thyroid lobe [7]. Regarding weight, Lalanne-Mistrih et al. found that adenomas heavier than 3.5 g are responsible for 7.2% of primary hyperparathyroidism cases [8], while Gasparrini et al. identified a correlation between serum calcium level and parathyroid adenoma weight [9]. Although parathyroid carcinoma, which is detected in less than 1% of cases of surgically treated primary hyperparathyroidism, is rare, hyperparathyroidism crisis related to parathyroid carcinoma is much more severe compared to adenoma-related hyperparathyroidism crisis [10]. Intrathyroidal parathyroid adenoma is a rare cause of acute primary hyperparathyroidism or parathyroid crisis, a rare clinical condition characterized by life-threatening hypercalcemia that requires urgent surgical treatment.

Conclusion

The case described here presented with neurological symptoms and medically resistant life-threatening hypercalcemia. Evaluation of the symptoms and biopsy findings resulted in the decision to perform immediate surgical parathyroidectomy, which led to rapid decrease in postoperative calcium and parathyroid hormone levels and dramatic improvement in clinical symptoms. The findings and successful treatment of this case indicate that large intrathyroidal parathyroidal adenoma should be considered in patients presenting with neurological symptoms and severe hypercalcemia.

Disclosure of conflict of interest

None.

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