Acute Hypercalcemia Coincidental with Intracapsular Parathyroid Hemorrhage

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A thirty-two-year-old female presented with acute hypercalcemia and elevated Parathyroid Hormone (PTH) as the only finding. She had a history of progressive generalized muscle and bone aches, weakness with inability to carry objects, insomnia, dysphonia, dysphagia, difficulty in walking and feeling depressed. Neck MRI revealed intra-capsular hemorrhage in a large parathyroid adenoma. Hypercalcemia from intracapsular parathyroid hemorrhage is a rare clinical presentation. Two months earlier, she had completely normal blood chemistry which was performed during her regular thyroid replacement follow-up, the calcium level was 9.5 mg/dl (normal reference range: 8.40-10.20).

The investigations at presentation revealed elevated total calcium 11.2 mg/dl (8.40-10.20 mg/dl) and PTH 115 pg/ml (15-65 pg/ml) with normal albumin and phosphorus levels. Three days later, the patient had developed dysphonia, dysphagia, odynophagia and difficulty in walking; her repeat calcium level had increased to 12.7 mg/dl. Neck MRI revealed a right inferior parathyroid (2.2x1.7x0.7 cm) adenoma with internal hemorrhage. Her surgical pathology confirmed the presence of intra-capsular hemorrhage in 2 cm parathyroid adenoma.

Clinicians should be aware of the potential for occurrence of intracapsular parathyroid hemorrhage manifested as acute symptomatic hypercalcemia.

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Primary hyperparathyroidism is the most common cause of hypercalcemia in the ambulatory setting. Approximately 75% to 80% of cases are attributable to a solitary parathyroid adenoma. Less common causes of hypercalcemia include parathyroid hyperplasia, multiple parathyroid adenomas, functional parathyroid cyst and parathyroid carcinoma[1,2]. Although it can occur at any age, it commonly affects people over the age of 50 years and postmenopausal women.

Over the past few decades, the presentation changed from a condition usually causing symptoms to a condition of asymptomatic hypercalcemia discovered on routine screening tests[3]. Spontaneous extracapsular hemorrhage is a rare clinical entity presenting as acute neck pain, neck masses or swelling, ecchymosis of the neck area, and dysphagia[4]. Spontaneous intracapsular hemorrhage is less reported than extracapsular hemorrhage.

The aim of this case report is to present a patient with acute symptomatic hypercalcemia with intracapsular parathyroid hemorrhage.

THE CASE

A thirty-two-year-old female was admitted with symptomatic hypercalcemia. She had a one-week history of progressive generalized muscle and bone aches, weakness with inability to carry objects, insomnia, and feeling depressed. She had no prior history of neck radiation and no history of fractures or nephrolithiasis. The patient's blood calcium performed at thyroid hormone replacement follow-up was normal two months preceding presentation. On physical examination, the patient's vital signs were unremarkable and she had no thyromegaly or palpable neck masses. She was otherwise fit and well, except for long standing hypothyroidism which was being treated with 25 mcg of Levothyroxine.

Investigations revealed elevated total calcium of 11.2 mg/dl (8.40-10.20 mg/dl) and PTH of 115 pg/ml (15-65 pg/ml) with normal albumin and phosphorus levels. Three days later, the patient had developed dysphonia, dysphagia, odynophagia and difficulty in walking; her repeat calcium level had increased to 12.7 mg/dl.

A neck MRI revealed 2.2x1.7x0.7 cm oval-shaped mixed T2 and T1 signal lesion with predominant high signal and moderate enhancement attached to the inferior lobe of the thyroid gland highly suggestive of a parathyroid adenoma with internal hemorrhage, see figures 1 and 2. She was admitted for hydration, IV bisphosphonate and surgical parathyroidectomy. The patient had immediate relief of all her symptoms with normalization of calcium and parathyroid hormone levels. Histopathology revealed intra-capsular hemorrhage in a parathyroid adenoma.

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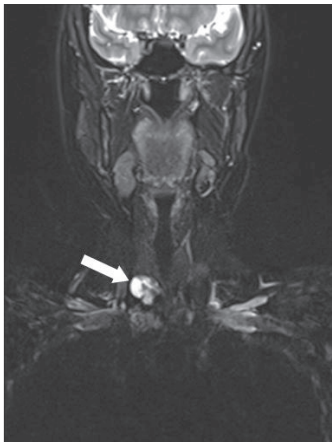

Figure 1: Coronal T2 Weighted Image of the Neck with Fat Saturation Showing an Oval Shaped Predominant High Signal Parathyroid Adenoma at the Lower Right Lobe of the Thyroid Gland (Arrow)

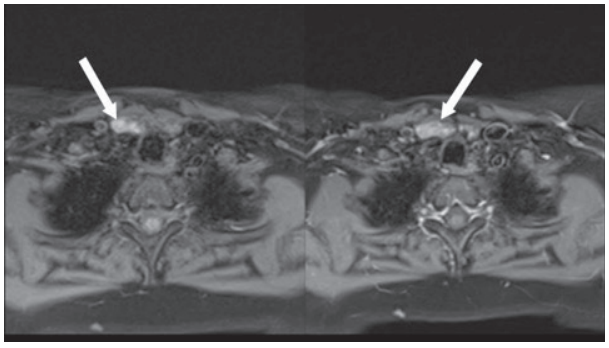

Figure 2: Axial T1 Weighted Image with Fat Saturation (Pre-contrast on the Left and Post Contrast on the Right) at the Center of the Lesion Shows High Signal Foci (Arrow) Indicative of Hemorrhage within the Adenoma in Addition to Moderate Enhancement Seen as High Signal along its Anterior Aspect (Arrow)

The provisional diagnosis was of a primary hyperparathyroidism with hemorrhage in a parathyroid adenoma, which was confirmed by MRI Neck and later by histopathology.

DISCUSSION

Asymptomatic hypercalcemia, detected on routine laboratory tests is the most common presentation of primary hyperparathyroidism (PHPT)[1,2]. In some cases, the presentation may be atypical and could range from parathyroid crises to normocalcemic PHPT. The vast majority of PHPT are asymptomatic with mild elevation of less than 1.0 mg/dl (0.25mmol/L) above the normal range[4-7]. However, approximately 30% percent of patients with asymptomatic PHPT may develop clinical manifestations of PHPT including skeletal manifestations, nephrocalcinosis, or kidney stones[8-9].

A hypercalcemic hyperparathyroid crisis (parathyroid crisis) is a rare life-threatening complication of primary hyperparathyroidism characterized by a serum calcium level exceeding 3.5 mmol/l (or 14 mg/dl) due to a marked elevation of PTH resulting in severe signs and marked symptoms of hypercalcemia, in particular, central nervous system dysfunction. However, some cases have previously documented mild PHPT. While in others, it is the first indication of parathyroid disorder[10].

Several cases of extracapsular parathyroid hemorrhage have been described; this type of extracapsular hemorrhage occurs mainly in parathyroid adenomas, parathyroid hyperplasia and rarely in parathyroid cysts[11-12]. Although many were previously known cases with primary hyperparathyroidism, some were diagnosed at the onset of symptoms only. More than half of the cases were above 50 years of age and mostly in females[13]. Some cases persisted with hypercalcemia after the hemorrhage but some became hypocalcemic or eucalcemic[14]. The most common signs and symptoms of extracapsular hemorrhage include neck mass or swelling, bruises, hematoma, dysphagia, neck pain, hoarseness and dyspnea. Rarely, cases may mimic aortic dissection, superior vena cava syndrome, or even death. Some cases rarely present with spontaneous rupture of the parathyroid adenoma with extracapsular hemorrhage leading to potentially fatal consequences[15-18].

Intracapsular parathyroid hemorrhage had been rarely reported. Parathyroid infarction is referred to as parathyroid apoplexy with remission. Hemorrhage into a parathyroid cyst with intermittent hyperparathyroidism/hypercalcemia and rarely hypercalcemic crises are coincidental with parathyroid hemorrhage. Infarction or hemorrhage can be classified as necrosis without hemorrhage, intracapsular hemorrhage and necrosis, or extracapsular hemorrhage[19-20]. Although no mechanism for hemorrhage is known; the predisposing factors include anticoagulants, non-steroidal anti-inflammatory drugs, trauma, or perhaps imbalance between adenoma growth and vascular supply. The variation in the location of the parathyroid adenoma accounts for the different signs and symptoms at presentation. Hoarseness could be due to the compression of the recurrent laryngeal nerve.

Our case was managed promptly and operated within days of initial presentation, which may have prevented spontaneous rupture with serious consequences.

CONCLUSION

This is a case of a young female presenting with acute symptomatic primary hyperparathyroidism found to have a coincidental hemorrhage in a large parathyroid adenoma. Clinicians should be aware of this condition especially in the asymptomatic elderly patients generally being managed conservatively.

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