Parathyroid adenoma presenting with spontaneous cervical and anterior mediastinal hemorrhage
A case report
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Abstract
Rationale: Spontaneous anterior cervical or mediastinal hemorrhage is a rare presentation of parathyroid adenoma.
Patient concerns: A 69-year-old woman presented with neck hematoma and dysphagia and was found to have a soft tissue mass adjacent to her thyroid gland as seen on MRI and neck ultrasound.
Diagnosis: Laboratory testing demonstrated elevated calcium and parathyroid hormone supporting diagnosis of parathyroid adenoma.
Interventions: She underwent right inferior parathyroidectomy and en bloc right hemithyroidectomy due to significant fibrosis.
Outcomes: Pathology confirmed hypercellular parathyroid and normal thyroid tissue. Postoperatively, patient’s calcium and parathyroid hormone levels had normalized.
Lessons: In conclusion, imaging may not always be specific in identifying the source of neck hematoma and so laboratory studies should be done to rule out parathyroid adenoma as the underlying etiology.

1. Introduction
Primary hyperparathyroidism has an incidence of approximately 2–4 per 10,000 with 80%–90% of cases due to a benign parathyroid adenoma.[1] The most common symptoms of hyperparathyroidism are chronic fatigue, body aches, difficulty sleeping, bone pain, memory loss, poor concentration, depression, and headaches. Parathyroid disease also frequently leads to osteoporosis, kidney stones, hypertension, cardiac arrhythmias, and kidney failure.[2] Spontaneous hemorrhage is a rare consequence of a parathyroid adenoma, first reported by Capps in 1934.[3] Possible complications associated with hemorrhage include chest pain, hemoptysis, dysphagia, and even airway obstruction.

2. Case presentation
A 69-year-old woman with no significant medical history initially presented with a sensation of globus which then evolved to mild dysphagia, neck swelling, and bruising extending down to her chest in April 2018 (Fig. 1A–C). Of note, she had been taking up to 12 naproxen (220mg) a day due to left hip pain during this time. Her primary care physician at an outside hospital ordered an MRI of her neck with and without contrast which showed a 1.4/C2 2.2/C2 2.3cm soft tissue mass posterior to the right lower pole of her thyroid with mild edema surrounding it (Fig. 2). A thyroid ultrasound was also done which helped characterize this mass as solid and vascular (Fig. 3).

She was then seen at Surgical Oncology clinic at UCSD. Physical examination showed some ecchymosis of her anterior neck and chest and a soft palpable mass in her right neck. Due to concern for a parathyroid adenoma resulting in spontaneous hemorrhage, her labs were drawn, which showed elevated calcium at 10.7 mg/dL and parathyroid hormone (PTH) at 90 pg/mL. No further biopsy of the mass was done and patient was scheduled for parathyroidectomy. The patient was taken to the operating room for parathyroidectomy. Because of dense scar tissue and no clear demarcation between the parathyroid adenoma and the right lower lobe of the thyroid, an en bloc right hemithyroidectomy was done as well.
The right lower parathyroid weighed 0.81g measuring 2.3 × 1.2 × 0.8 cm (Fig. 4). Pathology confirmed the frozen section as hypercellular parathyroid tissue with focal hemorrhage and scarring (Fig. 5A and B). Patient’s postoperative course was uncomplicated. On postoperative day 1, patient’s serum calcium had normalized to 8.6 mg/dL and PTH to 21 pg/mL. Patient had no further signs of neck hematoma or swelling upon follow up in clinic within 14 days of surgery. Patient’s serum calcium remained normal at 9.3 mg/dL 30 days after surgery.

3. Discussion
Spontaneous swelling and ecchymosis of the neck can be signs of life-threatening complications and require a broad differential[4-6]
including ruptured aneurysm, cervical hematoma, retropharyngeal abscess, mediastinitis, laryngocele, thyroid adenoma, and more. When secondary to a parathyroid adenoma, it has been hypothesized that the rapid growth of the parathyroid adenoma surpasses available vascular supply[7] which leads to spontaneous hemorrhage. Given the parathyroid tumor’s thin capsule,[8] hemorrhage is usually extracapsular expanding to cervical and mediastinal subcutaneous tissue compared to thyroid nodules whose hemorrhage is usually intracapsular given their thicker capsule.

This patient’s presentation was very similar to previous case reports of spontaneous hemorrhage secondary to a parathyroid adenoma[9]—mostly localized as seen with her cervical and mediastinal hemorrhage and symptoms of globus and dysphagia. Like previous studies, she was also of the common demographic of middle-aged women who are otherwise healthy and have relatively asymptomatic hyperparathyroidism.

Although imaging studies are first line in localizing such lesions, they are not always specific in identifying the anatomical source; for example, this patient’s MRI and ultrasound both indicated possible thyroid pathology. Hence, even though spontaneous hemorrhage may be a rare manifestation of parathyroid adenomas, lab studies are essential in ruling them out to avoid unnecessary procedures including thyroid biopsy.

Finally, this patient required a hemithyroidectomy as part of her procedure due to scar tissue formation and fibrosis. This was most likely secondary to spontaneous hemorrhage but could also inherently be due to the pathology of the parathyroid adenoma.[10] Although this patient did not suffer any further complications due to this, this additional procedure does have a risk of iatrogenic hyperparathyroidism, laryngeal nerve injury, bleeding, and infection.[11] Hence, further study can be done to determine risk factors or preventive measures of scar formation in tissue surrounding hemorrhage from a parathyroid adenoma.

4. Conclusions

Parathyroid adenomas rarely present as spontaneous cervical or mediastinal hemorrhage. Hence, there are currently no standardized recommendations for their diagnosis and management. Nonetheless, it should remain on the differential as it can be easily screened for with laboratory analysis as imaging may not always be specific. Further, additional study should be done to better define risk factors and indications for hospitalization as well as to optimize surgical approach given the risk of scar tissue and fibrosis of surrounding structures.

Author contributions

Funding acquisition: Michael Bouvet.
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References

[1] Hammett-Stabler CA, Maygarden SJ, Reisner HM. Pathology of the endocrine system. Pathology: A Modern Case Study New York, NY: McGraw-Hill; 2014.
[2] Fitzgerald PA, Papadakis MA. Endocrine disorders. Current Medical Diagnosis & Treatment New York, NY: McGraw-Hill; 2018.
[3] Capps RD. Multiple parathyroid tumors with massive mediastinal and subcutaneous hemorrhage. Am J Med Sci 1934;188:801–4.
[4] Cohen O, Yehuda M, Adi M, et al. Spontaneous neck hematoma in a patient with fibromuscular dysplasia: a case report and a review of the literature. Case Rep Otolaryngol 2013;2013:352830.
[5] Damodara M, Patil S, Saravanappa N. Spontaneous neck haematoma. BMJ Case Rep 2009;2009.
[6] Giotakis EI, Hildenbrand T, Dodenhof J. Sudden massive neck swelling due to hemorrhage of a thyroid adenoma: a case report. J Med Case Reports 2011;5:391.
[7] Yoshimura N, Mukaida H, Mimura T, et al. A case of an acute cervicomediastinal hematoma secondary to the spontaneous rupture of a parathyroid adenoma. Ann Thorac Cardiovasc Surg 2014;20(Suppl): 816–20.
[8] Rehman HU, Markovski M, Khalifa A. Spontaneous cervical hematoma associated with parathyroid adenoma. CMAJ 2010;182:E632.
[9] Ulrich L, Knee G, Todd C. Spontaneous cervical hemorrhage of a parathyroid adenoma. Endocrinol Diabetes Metab Case Rep 2015;2015:150034.
[10] Wieneke JA, Smith A. Parathyroid adenoma. Head Neck Pathol 2008;2:305-8.
[11] Ahn D, Sohn JH, Jeon JH. Hypothyroidism following hemithyroidec- tomy: incidence, risk factors, and clinical characteristics. J Clin Endocrinol Metab 2016;101:1429–36.