

## Case Report

# Parathyroid adenoma causing a spontaneous cervical and mediastinal massive hematoma

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**Abstract:** Cervical and mediastinal hemorrhaging caused by a spontaneous rupture of a parathyroid adenoma has been rarely reported. Herein, we report a case of a 52-year-old woman who experienced a sudden onset of cervical and mediastinal bleeding related to a parathyroid adenoma. The patient had a history of trauma to the left thorax with a fracture of two ribs. Ultrasonography and computer tomography revealed a nodule in the retrotracheal space and hematoma of the bilateral trachea and thorax. Laboratory examinations revealed significant hypercalcemia, hypophosphatemia, and a high level of intact parathyroid hormone. Surgery was performed and a dark-red, soft tumor was found behind the trachea, which adhered to the surrounding tissues. The pathological diagnosis was a parathyroid adenoma with hemosiderin deposition and fibrosis.

**Keywords:** Parathyroid adenoma, hemorrhage, ultrasound, parathyroidectomy

## Introduction

Parathyroid adenomas commonly present with hyperparathyroidism, but spontaneous rupturing of a parathyroid adenoma causing an extracapsular hemorrhage is rare, as only 29 cases have been reported in the literature to date [1-8]. Here, we report a case of sudden neck swelling with dyspnea, hypercalcemia, and skin ecchymosis caused by spontaneous extracapsular bleeding of a parathyroid adenoma

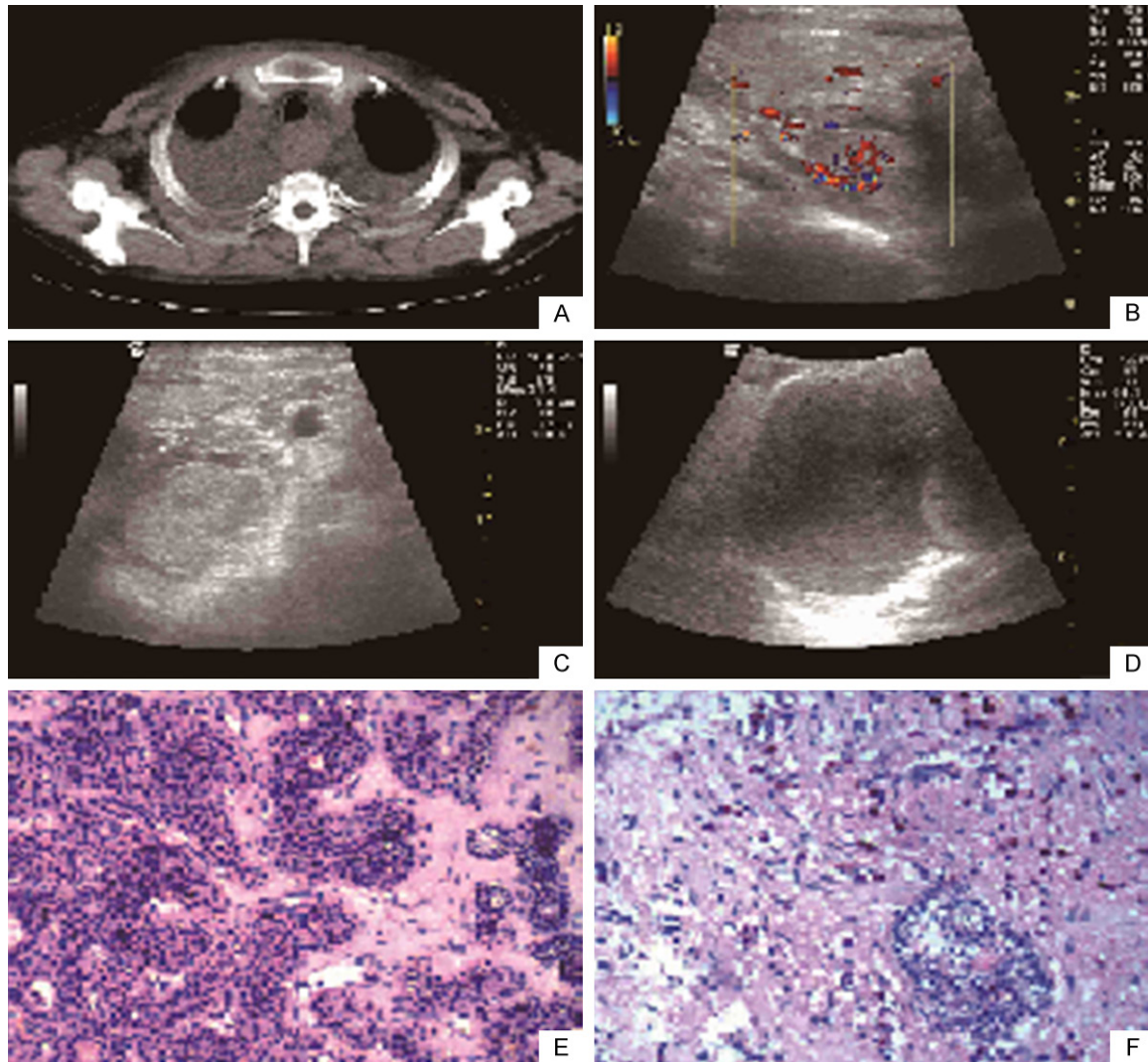
## Case report

A 52-year-old woman experienced a sudden onset of pain and swelling in her neck for about 12 h. Four hours later, she presented to the emergency department of our hospital with an increasing degree of dyspnea and chest and neck pain on December 10, 2014. The patient reported a history of trauma to her left thorax with a fracture of two ribs 11 days before. A physical examination revealed diffuse and ecchymotic swelling and tenderness in the patient's anterior neck and upper chest wall, but no distinct palpable masses in her neck. Her blood pressure was 110/65 mmHg and heart rate 79

beats/min. She suffered from stomach ulceration 6 months before and a coccyx fracture 4 years before, but did not undergo surgery. A computed tomography (CT) scan of the patient's neck and chest revealed a hematoma in the peritracheal space, which extended caudally to the superior mediastinum and bilateral thoracic cavity, and an ill-defined, soft tissue density was suspected as a hematoma (**Figure 1A**). No abnormal findings were observed in the thoracic aorta or its branches via chest angiography. A diagnostic aspiration was performed and 10 cc of fresh blood was aspirated from the right chest cavity.

To rule out parathyroid abnormalities, blood chemical analysis was performed to assess parathyroid function, and the results were as follows: serum calcium level, 2.92 mM (normal range, 2.0-2.8 pg/mL); serum phosphate level, 0.7 mM (normal range, 0.8-1.6 mM); intact parathyroid hormone (PTH<sub>i</sub>) level, 216.40 mM (normal range, 15-65 mM). A diagnosis of a spontaneous parathyroid adenoma rupture was made on the basis of the patient's medical history and the examination results. Ultrasonography confirmed the diagnosis 4 days later.

## Parathyroid adenoma causing massive hematoma



**Figure 1.** Image and pathological features of the parathyroid adenoma. A. The first computed tomography (CT) image of the patient's neck and chest, revealed a hematoma with an ill-defined, dense soft tissue mass. B. Color Doppler flow imaging (CDFI) revealed affluent blood in the nodule. C. A hypoechoic nodule was partly detected bilaterally from the trachea and a linear effusion was shown around part of the mass using a high-frequency probe. D. A massive hematocoele was detected in the bilateral thoracic cavity. E and F. A parathyroid adenoma composed of round nuclei and acidophilic cytoplasmic cells with fibrosis and hemosiderin deposition in the stroma (hematoxylin and eosin staining; magnification,  $\times 100$ ).

Sonography using a low-frequency probe (Logiq 9; GE Healthcare, Wauwatosa, WI, USA) at a frequency of 2-4 MHz showed an intact retrotracheal mass ( $4.0 \times 3.3$  cm) adjacent to the superior mediastinum with ill-defined margins. A conversion to a high-frequency probe (9-14 MHz) revealed a hypoechoic nodule with affluent blood, which was partly detected on both sides of the trachea (**Figure 1B**). A linear effuse hematocoele was detected around the mass (**Figure 1C**), whereas a massive hematocoele was detected in the patient's neck and bilateral thoracic cavity (**Figure 1D**). The sonography

confirmed the diagnosis of a spontaneous rupture of an atopic parathyroid adenoma. About 3 weeks later, in January 2011, a follow-up enhanced CT scan and second ultrasound clearly revealed a mass within the posterior mediastinum, which was slightly smaller than the previous mass, and the linear effusion surrounding the mass disappeared. A parathyroidectomy was performed the next day and a dark-red, soft tumor was located behind the trachea, adhering to the surrounding tissue. The pathological diagnosis was a parathyroid adenoma with hemosiderin deposition and fibrosis, which

confirmed the diagnosis of a tumor (**Figure 1E** and **1F**). The patient's serum calcium and PTH levels normalized postoperatively.

### Discussion

The first case of a spontaneous parathyroid gland hemorrhage was reported by Capps in 1934, in which the patient died of massive blood loss. 1 A parathyroid intra- and extracapsular hemorrhage is a serious, potentially fatal complication of parathyroid gland enlargement due to hyperplasia, adenoma, or other cancers. The hemorrhage may be localized within the parathyroid gland, but often presents as extracapsular and the clinical symptoms vary with the size and location of the hematoma. 2 A cervical and mediastinal hematoma, as a result of parathyroid tumors, is characterized by painful swallowing, dysphagia, dyspnea, hoarseness, swelling of the anterior neck, or ecchymosis of the neck or chest. Severe compression of the pharynx or larynx leads to a narrowing of the airway and may require emergency surgical intervention, such as endotracheal intubation or tracheostomy. Hemorrhaging of an intrathoracic parathyroid tumor leads to the development of a mediastinal hematoma or hemothorax characterized by chest pain, cough, shortness of breath, or respiratory distress [3]. A differential diagnosis of parathyroid hematoma includes bleeding due to thyroid lesions, cysts, or a nodular goiter and subacute thyroiditis [2, 4, 5]. Although the thyroid gland is less likely to rupture, it is more likely to produce a cervical or mediastinal extracapsular hematoma than the parathyroid gland, partly because of the more complete, thicker capsule of the thyroid gland [6].

Subacute thyroiditis frequently follows a viral illness and has many systemic symptoms and, although calcium levels remain normal, a sonogram will indicate a characteristic change in the thyroid. Bleeding may also result from dissection of an aortic aneurysm, penetrating aortic ulcers, or rupture of a mediastinal mass or cyst. However, neck and chest trauma and vascular causes must also be considered [7]. Although the pathophysiological mechanisms of spontaneous bleeding from a parathyroid adenoma remain unknown, it has been suggested that they probably stem from an imbalance between cell growth and blood supply. Endocrine adenomata can undergo remission by spontaneous necrosis, a situation prone to the onset

of necrotic-hemorrhagic foci, which may ultimately spread outside the glandular structure via a mechanism considered similar to that of apoplexy seen in other endocrine neoplasias.

The parathyroid glands can vary in number and location in normal subjects. Most people have 4 parathyroid glands located on the posterior medial surface of the thyroid gland; however, in as many as 25% of normal subjects, more or less than 4 glands are present [9, 10]. The inferior parathyroid glands are much more variable in their distribution than the superior glands [11] and may be located on the lower pole of the thyroid or in a thymic tongue or may be juxtathyroidal or mediastinal [12]. The echo texture of normal parathyroid glands is similar to that of the overlying thyroid gland, thus normal-sized glands are barely visible by ultrasound. However, enlarged glands have a decreased echo texture and high-resolution ultrasonography performed by experienced clinicians is highly sensitive for a preoperative diagnosis of parathyroid adenoma in patients with primary hyperparathyroidism. Detecting the feeding vessels by color Doppler flow imaging (CDFI) is highly suggested for parathyroid lesions, as enlarged parathyroids are hypervascular compared to nodular lesions of the thyroid gland [13]. In our case, ultrasound with CDFI clearly showed the mass and provided diagnostic evidence to the clinician.

In conclusion, a spontaneous rupture of a parathyroid adenoma causing cervical and mediastinal hemorrhaging is uncommon and may require a differential diagnosis of extracapsular hemorrhage of cervical and mediastinal lesions. The authors believe that high-resolution ultrasonography with CDFI performed by experienced clinicians presents an accurate and cost effective diagnostic tool.

### Disclosure of conflict of interest

None.

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