



Case report

## Neck emergency due to parathyroid adenoma bleeding: a case report

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### Abstract

**Introduction:** The spontaneous rupture of a parathyroid adenoma accompanied by extracapsular hemorrhage is a rare, potentially fatal, condition and is a cervicomedial surgical emergency.

**Case presentation:** This report describes an atypical two-step spontaneous rupture of an asymptomatic parathyroid adenoma in a 56-year-old Caucasian woman who presented with a painful mass in the right side of her neck.

**Conclusion:** Based on this case report and similar cases reported in the medical literature, a diagnosis of extracapsular parathyroid hemorrhage should be considered when a non-traumatic sudden neck swelling coexists with hypercalcemia and regional ecchymosis.

### Introduction

Hypercalcemia is the most common clinical sign of a parathyroid adenoma [1]. Hemorrhagic infarction may occur both in a parathyroid adenoma and in hyperplastic parathyroid glands, whereas extracapsular hemorrhage due to hyperplasia, adenoma, or cancer is an uncommon but threatening occurrence, resulting in a cervicomedial hematoma and is often associated with severe blood calcium imbalance. To date, 27 cases have been reported in the literature (usually as single cases or small case series) and none of them describe a two-step clinical picture of bleeding from the parathyroid gland (Table 1) [1-25].

Patients usually present with a palpable lateral neck mass with signs of ecchymosis, appearing slowly 24 to 48 hours after the sudden onset of neck discomfort, dysphagia, dyspnea, or hoarseness [19,24]. Such an emergency requires immediate surgical treatment and the prognosis depends on the extent and location of the hematoma.

This case report describes a patient who experienced a two-step spontaneous rupture (with extracapsular bleeding) of a large (probably long-standing) asymptomatic parathyroid adenoma. To the best of our knowledge, this is the first report of such an atypical modality of parathyroid adenoma rupture.

**Table 1.** Clinicopathological features in case reports of extracapsular parathyroid hemorrhages

Author (year)	#	Clinical features	Ca <sup>2+</sup> (normal values)	T	Histology (Ø = cm)
Capps R (1934) [2]	1	Weakness, sore throat, cervical swelling/ ecchymosis, dysphagia, dyspnea	NA	5 weeks	Adenoma (Ø = 7)
Berry BE (1974) [3]	1	Weakness, retrosternal pain, signs of superior vena caval compression	NA	1 day	Adenoma (Ø = 5)
Santos GH (1975) [4]	1	Retrosternal pain, dizziness, hypotension, hypercalcemia	NA	1 day	Adenoma (Ø = 8)
Jordan FT (1981) [5]	1	Anterior cervical pain, swelling and ecchymosis, dysphagia	13.3 mg/dL	1 month	Hyperplasia (Ø = 6)
Roma J (1985) [6]	1	Hoarseness, dysphagia, cervical swelling, cervical-thoracic ecchymosis	3.00 mmol/L	1 day	Hyperplasia
Simcic KJ (1989) [7]	1	Painful cervical swelling, dysphagia, dyspnea	4 mmol/L (2.14-2.52)	2 weeks	Adenoma (Ø = 4.5)
Massard JL (1989) [8]	1	Difficulty swallowing, dysphonia, cervical pain, cervical ecchymosis	3.15 mmol/L	10 hours	Adenoma
Hotes LS (1989) [9]	1	Hoarseness, dysphagia, discomfort in the anterior area, ecchymosis	Normal	3 days	Adenoma
Alame A (1990) [10]	1	Difficulty swallowing, dysphonia, cervical-thoracic ecchymosis	2.86 mmol/L	1 day	Adenoma
Mantion G (1990) [11]	1	Dysphagia, dyspnea, ecchymosis	2.67 mmol/L	2 days	Adenoma (Ø = 2.5)
Amano Y (1993) [12]	1	Hoarseness, dysphagia, cervical swelling and ecchymosis	Normal	2 days	Adenoma (Ø = 5.6)
Korkis AM (1993) [13]	1	Hoarseness, dysphagia, cervical swelling and cervical-thoracic ecchymosis	11.6 mg/dL	1 day	Adenoma (Ø = 4)
Jougon J (1994) [14]	1	Dysphagia, cervical swelling and cervical-thoracic ecchymosis	2.9 mmol/L	1 day	Adenoma (Ø = 3)
Menegaux F (1997) [15]	1	Cervical pain, dysphagia, dyspnea	2.62 mmol/L	1 day	Adenoma
Hellier WPL (1997) [16]	1	Dysphagia, dysphasia, cervical-thoracic ecchymosis	Elevated	1 day	Adenoma
Ku P (1997) [17]	1	Hoarseness, cervical pain and ecchymosis	3.15 mg/dL	1 day	Adenoma (Ø = 2)
Kihara M (2001) [18]	1	Painful cervical swelling and cervical-thoracic ecchymosis	Normal	1 month	Adenoma (Ø = 2)
Kozlow (2001) [19]	1	Dysphagia, odynophagia, cervical swelling	11.3 mg/dL (8.4-10.2)	7 days	Adenoma
Nakajima J (2002) [20]	1	Retrosternal pain, cervical-thoracic ecchymosis	Normal	3 weeks	Adenoma (Ø = 3.5)
Govindaraj S (2003) [21]	1	Hyper-normo-calcemia, right-sided headaches, severe throat pain	13 mg/dL	2 weeks	Adenoma
Taniguchi I (2003) [22]	1	Cervical swelling, pain, dysphagia, cervical-thoracic ecchymosis	NA	1 month	Cyst (Ø = 6)
Maweja S (2003) [23]	2	Painful cervical swelling and cervical-thoracic ecchymosis	2.57; 2.80 mmol/L	2 days	Adenoma
Tonerini M (2004) [24]	1	Painful cervical swelling	NA	1 day	Adenoma
Akimoto T (2005) [25]	1	Left pleural effusion in IPT II at chest X-ray and computed tomography scan	Elevated	1 day	Hyperplasia
Devezè A (2006) [1]	2	1. Dysphagia cervical hematoma, hypercalcemia 2. Latero-cervical pain and ecchymosis	2.57 mmol/L	1 week 5 days	1. Adenoma 2. Adenoma
Merant-Boschin I (2009)	1	Painful cervical swelling, dysphonia, dyspnea	3.18 mmol/L (2.10-2.55)	2 days	Adenoma (Ø = 4.0)

**Notes:** #, number of cases described; [Ca<sup>2+</sup>]: calcium level; T: time of onset; Normal: in the normal range; NA: Not assessed.

### Case presentation

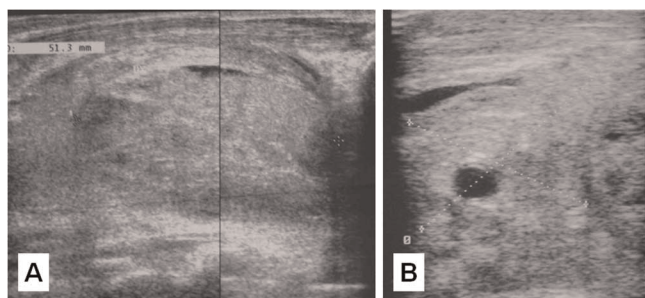
In April 2007, a 56-year-old Caucasian woman with a painful, right-sided neck mass presented to a private practitioner. Ultrasound (US) suggested a clinical diagnosis of subacute thyroiditis, which was not supported by subsequent laboratory tests (C-reactive protein 1.9 mg/L; leukocytes 9700/ $\mu$ L; thyroid hormones within normal range; antithyroid auto-antibodies negative). Two days later, the patient had an exacerbation of the latero-cervical pain which prompted a repeat US of the neck, which

revealed an iso-echoic lesion (51.3 mm in size), apparently included within an enlarged right thyroid lobe (83.5 mm). The lesion was interpreted as an intrathyroid hematoma (Figure 1A,B) and the opinion of a neck surgeon (MRP) was requested. The patient's medical history was collected at this time and included a severely diminished bone mass treated with bisphosphonate, though no information on bone metabolism was provided. History ruled out any regional traumatic event. The patient seemed quite anxious and dysphonic but not

dyspnoeic. Physical examination revealed a tender right-sided cervical mass, extending from the right mandibular arch to the thoracic inlet.

The patient was referred to the Special Surgical Pathology Department at Padova University Hospital, where computed tomography (CT) showed a laterocervical hemorrhagic lesion, extending from the lateral neck to the right prevertebral/paratracheal spaces (Figure 2); a distinct midline shift and compression of both the hypopharynx and the trachea were also documented. During the CT procedure, the patient suffered from severe respiratory distress with dyspnea and she was immediately referred for surgical treatment, where an ovoid, hemorrhagic mass (4.0 × 2.4 × 1.3 cm, weight 8.1 g) was revealed posterior to the right thyroid lobe. Laboratory tests (conducted during the surgical procedure) demonstrated severe hypercalcemia (3.18 mmol/L; normal range: 2.10 to 2.55 mmol/L) with a decrease in hemoglobin level (12.0 g/dL). Surgery consisted of hematoma evacuation, parathyroidectomy and "en bloc" right thyroid lobectomy (Figure 3A). There was no evidence of regional lymph node involvement. The surgery was curative and both serum calcium and parathyroid hormone (PTH) levels quickly dropped to within the normal range (at discharge: calcium 2.29 mg/dL; PTH 52 pg/mL).

Gross section of the surgical specimen revealed a three-layered lesion consisting of peripheral areas of (partially fluid) hemorrhagic material, invading a more internal, compact (partially organized) zone around the core of the specimen, which consisted of necrotic parathyroid remnants (Figure 3B). Multiple gross samples were obtained for histological assessment, which showed an extensively hemorrhagic chief cell parathyroid adenoma surrounded by a loosely organized hemorrhagic and fibrous reaction,



**Figure 1.** Neck ultrasound on admission. Longitudinal and transverse views demonstrating a 51.3 mm nodular iso-echoic lesion, of dyshomogeneous structure and hemorrhagic pattern (A). The lesion surrounds the right common carotid artery and internal jugular vein, and is located posterior to the right lobe of the thyroid, with ill-defined posterior margins (B).

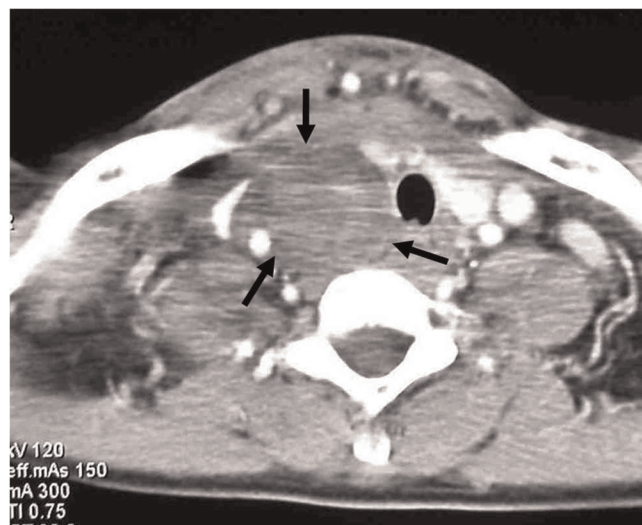
which became frankly hemorrhagic in the tissue samples obtained from the periphery of the resected mass.

A 9-month follow-up including clinical evaluation, serology and US, revealed no clinical abnormalities.

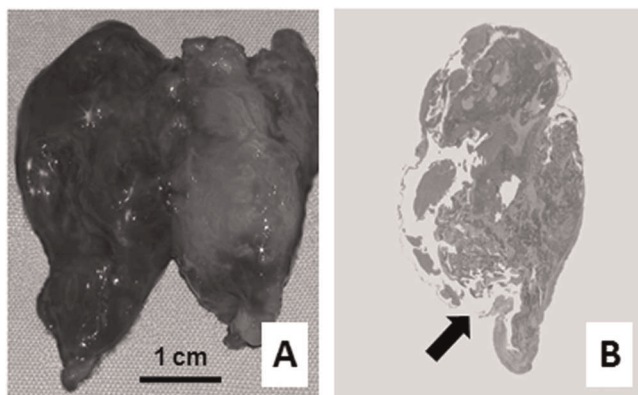
## Discussion

Spontaneous neck hemorrhage is a rare, frightening surgical emergency, usually resulting from the traumatic rupture of vessels or from generally spontaneous thyroid or parathyroid extraglandular bleeding. A parathyroid intra- and extracapsular hemorrhage is a serious, potentially fatal, complication of parathyroid gland enlargement due to hyperplasia, adenoma or cancer. The physiopathological mechanisms behind such non-traumatic bleeding are not known. They probably stem from an imbalance between cell growth and blood supply, a situation prone to the onset of necrotic-hemorrhagic foci, which may ultimately spread outside the glandular structure; this mechanism has been considered similar to the apoplexy seen in other endocrine neoplasia [26].

Capps first documented a fatal case of spontaneous massive parathyroid hemorrhage with cervical/mediastinal infarction in 1934, which was only assessed at post-mortem examination [2]. To date, 27 cases have been reported in the literature, with variable clinical presentations [1-25], the variability concerning the endocrine clinical syndrome at presentation (usually a



**Figure 2.** Computed tomography scan on admission (at thoracic inlet level) showing signs of cervical-mediastinal hematoma (black arrows) in the right prevertebral and paratracheal space. A marked midline shifting and compression of the trachea is evident.



**Figure 3.** Gross specimen consisting of red-brown parathyroid adenoma of the upper-right parathyroid gland (left) and right thyroid lobe (right) (A). Gross histological section showing the whole parathyroid gland and the rupture of its capsule (arrow) (B). Original magnification 2×.

hypoparathyroidism of sudden onset), the size of the hemorrhagic mass and the timing of the cervical/thoracic bleeding. According to Simic *et al.*, however, a clinical triad consisting of acute neck swelling, hypercalcemia, and neck and/or chest ecchymosis strongly point to this clinical hypothesis [7]. Table 1 summarizes the cases reported in the literature as at December 2007. The table refers strictly to cases featuring extracapsular parathyroid bleeding, confirmed on histology and excluding cases relating to neck traumas. The group of cases considered shows a significant variability in both clinical presentation including symptoms, time of onset and laboratory data such as calcium levels.

The differential diagnosis of non-traumatic lateral-neck bleeding involves thyroid lesions, cyst or nodular goiter, subacute thyroiditis, and parathyroid conditions, such as adenoma, hyperplasia or cancer [9,14,24]. As in this patient, it may be difficult, if not impossible, to distinguish clinically between a thyroid and parathyroid origin of the problem, and even imaging techniques such as CT and US may be bewildering. In this respect, a primary parathyroid involvement should be considered when a clinical syndrome centered in the lateral area of the neck, such as cervical swelling, cervical-thoracic ecchymosis, dysphagia and dyspnea, coexists with blood calcium imbalance.

In our patient, the clinical history of a bland presentation, quiescent interval and final emergency, and the pathological features of the resected mass are both consistent with a spontaneous rupture of a parathyroid adenoma in two successive stages. As described in many other parenchymal organs here too we can assume that an initial episode of

paucisymptomatic intracapsular bleeding progressed to a capsular rupture resulting in a massive cervical and/or mediastinal infarction.

### Conclusion

This case should alert physicians that parathyroid extracapsular hemorrhage needs to be considered among the list of non-traumatic surgical neck emergencies and, in line with the current literature, any neck swelling, variably associated with "mass" symptoms such as dysphagia and/or dyspnoea, in association with hypercalcemia and regional ecchymosis, should strongly point to this clinical hypothesis.

### Competing interests

The authors declare that they have no competing interests.

### Authors' contributions

All authors of this paper have participated directly in the planning, execution, or analysis of the study, and have read and approved the final version submitted.

### Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

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