Case report

Importance of serum calcium in spontaneous neck haematoma

Matthew Zammit, Richard Siau, Alessandro Panarese

SUMMARY

We present an unusual case of spontaneous cervical haemorrhage secondary to extra-capsular bleeding from a parathyroid adenoma. Signs and symptoms on presentation included sore throat, dysphagia and anterior chest ecchymosis. While CT confirmed active cervical haemorrhage, elevated serum calcium and parathyroid hormone raised suspicion of possible parathyroid pathology. This case report and literature review highlight the diagnostic value of serum calcium in presentations of acute spontaneous neck haematoma. This should be considered especially in the acute phase, where imaging may not identify the source of haemorrhage. Initial observation and deferred surgery is the treatment of choice, with emergency operative management reserved for respiratory distress and worsening compressive symptoms.

BACKGROUND

Primary hyperparathyroidism (PHPT) is becoming increasingly common due to a rise in biochemical screening for hypercalcaemia. The prevalence of PHPT is estimated to be 1:1000, with solitary parathyroid adenomas contributing to 85% of cases of PHPT.1

Infrequently, spontaneous extra-capsular bleeding from parathyroid adenomas may instigate retropharyngeal haemorrhage. This was first reported by Capps, with a parathyroid adenoma being the culprit for a fatal cervical bleed.2 However, international guidelines for PHPT fail to discuss the symptomatology and management of this lethal complication.3

This case describes the unprompted rupture of a parathyroid adenoma, highlighting the diagnostic importance of serum calcium in spontaneous mediastinal haemorrhage. Its management is further discussed through a review of 39 confirmed cases between 2001 and 2020 (cases with histological/radiological confirmation that haemorrhage is of a parathyroid origin), summarised in table 1.

CASE PRESENTATION

A 65-year-old woman presented to the emergency department with a 4-day history of a painful right anterior neck swelling, hoarse voice, dysphagia and anterior chest ecchymosis. She was seen by her general practitioner a few days prior with clarithromycin prescribed for suspected tonsillitis. She denied any recent trauma. Her medical history consisted of hypothyroidism, taking levothyroxine and was not on any anticoagulants or antiplatelets.

INVESTIGATIONS

A contrast-enhanced CT scan of the neck and thorax revealed active haemorrhage with a haematoma in superior mediastinum confined to the retropharyngeal space (figures 1 and 2). Bilateral haemathoraces measuring up to 22 mm axially were also reported. This prompted an immediate patient transfer to a district general hospital with a cardiothoracic and otolaryngology unit.

On arrival, oxygen saturations were 97% on room air, with a heart rate of 91 beats/min and blood pressure of 138/86 mm Hg. Examination of the oropharynx revealed bruising of the right tonsil and soft palate. Flexible nasendoscopy (FNE) showed submucosal blood in the nasopharynx, hypopharynx and a haematoma within the right aryepiglottic fold, obscuring the right laryngeal inlet. While unable to fully visualise the right vocal cord (VC), full movement of the left VC was confirmed.

Blood investigations showed unremarkable inflammatory markers, liver function tests and clotting screen, with a haemoglobin of 112 g/L (baseline 130 g/L, measured 6 months previously). A serum-corrected calcium (Ca2+) of 2.77 mmol/L (normal range 2.20–2.60 mmol/L) and a parathyroid hormone (PTH) level of 12.3 pmol/L (normal range 1.1–6.9 pmol/L) were detected on admission and monitored. Thyroid function tests were within normal limits.

Tranexamic acid was administered with an initial working diagnosis being a retropharyngeal bleed; however, no airway compromise exhibited. Cardio-thoracic assessment deemed that haemothoraces did not require invasive intervention, in view of their limited size.

TREATMENT

Critical care admission for observation was the initial management. A repeat FNE the following day showed bruising along the posterior pharyngeal wall; however, no pulsation, bulge or active bleeding was seen. The patient was stepped down to ward-level care 2 days later following improved neck movement, resolving ecchymosis and oropharyngeal swelling. Five days after admission, patient was discharged home on five further days of oral tranexamic acid (1 g orally, three times per day).
Lessons learned

Anterior neck ultrasound (US) was performed urgently as an outpatient 7 days after discharge. A normal appearance of the thyroid gland was reported, with no cysts or nodules present. Repeated Ca²⁺ and PTH remained elevated, and the patient was referred for further endocrinology assessment. Single-photon emission with computed tomography scans showed focal uptake inferior to the right lobe of the thyroid, in keeping with a solitary parathyroid adenoma (figure 3). Focused parathyroidectomy was performed successfully 5 months later, with a postoperative PTH of 3.3 pmol/L and Ca²⁺ of 2.30 mmol/L. Histology confirmed a parathyroid oncocytic adenoma, with siderophages seen within and around the tumour.

OUTCOME AND FOLLOW-UP

Neither hoarseness nor symptoms of hypocalcaemia were noted on follow-up, with normal Ca²⁺ and PTH levels 2 months after surgery. An outpatient appointment was arranged with the endocrinology team in 6 months’ time for follow-up of her osteopenia detected in her lumbar spine.

DISCUSSION

Atraumatic cervical haematomas are potentially life-threatening occurrences, and diagnosis of the underlying cause can be challenging. Typical symptoms at presentation include neck swelling and ecchymosis, which should raise clinical suspicion. The primary differential includes a ruptured aneurysm or mediastinal mass, aortic dissection and descending necrotising mediastinitis, meritting urgent radiological imaging. Contrast-enhanced CT, MRI, US and nuclear medicine all assist in delineating the source of bleeding. However, these may fail to identify a parathyroid culprit obscured by acute haemorrhage, as seen in our case

Table 1  Summary of data obtained from 39 case reports of spontaneous parathyroid haemorrhage between 2001 and 2020

<table>
<thead>
<tr>
<th>Patient demographics</th>
<th>Age (mean±SD)</th>
<th>56.56±12.67</th>
</tr>
</thead>
<tbody>
<tr>
<td>Gender</td>
<td>Males</td>
<td>12</td>
</tr>
<tr>
<td></td>
<td>Females</td>
<td>27</td>
</tr>
</tbody>
</table>

| Clinical presentation and investigations | Presenting symptoms (39/39 cases reported) | Pain | 92.3% (36/39) |
|                                          | Ecchymosis   | 52.3% (20/39) |
|                                          | Neck swelling| 61.5% (24/39) |
|                                          | Hoarseness   | 25.6% (10/39) |
|                                          | Dyspnoea     | 18.0% (7/39) |
|                                          | Dysphagia    | 46.2% (18/39) |

| Time of onset (mean days±SD) (31/39 cases reported) | 12.19±26.41 |
| Serum calcium (35 cases reported) | Elevated | 88.6% (31/35) |
|                          | Normal | 11.4% (4/35) |
| Serum parathyroid hormone (32 cases reported) | Elevated | 93.8% (30/32) |
|                          | Normal | 6.3% (2/32) |

| Management | Management approach (35/39 cases reported) | Deferred surgery | 19/35 |
|           | Emergency surgery | 2/35 |
|           | Immediate surgery | 14/35 |

| Operative timing (mean days±SD) (28/39 cases reported) | 55.36±66.67 |
| Histology (38 cases reported) | Adenoma | 24/38 |
|                          | Hyperplasia | 2/38 |
|                          | Carcinoma | 1/38 |
|                          | Cyst | 1/38 |

Below each criterion, the number of cases reporting the specified data is documented.
report. Shundo et al also remarked that MRI and US may falsely localise the source of bleeding to the thyroid gland.8

While an uncommon aetiology, parathyroid adenomas have been reported to instigate cervical haematomas.2 From a review of similar cases, it was commonly seen to occur in middle-aged women, in keeping with the increased incidence of PHPT3 in postmenopausal women.7 This is hypothesised to occur due to an imbalance of cell proliferation in relation to its blood supply,8–10 with a thin capsule allowing for easy rupture of a bleeding parathyroid tumour.8

In the acute setting where radiological testing may not reveal the underlying aetiology, raised serum Ca2+ is a strong diagnostic indicator of a parathyroid bleed. From our review of the current worldwide literature, only five case reports denied taking Ca2+ on work-up, 1,4–15–13 with 88.6% (31/33) showing an elevated Ca2+.3 15–7–10 14–10

In the four cases with normal Ca2+, 630–12 PTH was found to be elevated in one scenario.7 Shim et al hypothesised that such scenarios may represent either a non-functioning parathyroid adenoma or a necrotic lesion from the pressure effects of the haematoma.22

Simcic et al attempted to further delineate a diagnostic triad,33 considering a combination of neck swelling, ecchymosis and hypercalcaemia as pathognomonic for a haemorrhaging parathyroid tumour.3 33 While this is true for our reported case, the triad was witnessed in only 37.1% (13/35) of the cases with a Ca2+ result, due to a limited representation of swelling (61.5%) and ecchymosis (52.3%) in the reviewed reports. A summary of the initial symptoms on presentation can be found in table 1.

No clear guidance exists for managing such presentations. Chaffanjon et al proposed a 3-month interval period prior to definitive surgery in the absence of VC involvement or respiratory distress, avoiding a potentially hostile environment.16 This allows for reabsorption of the haematoma, improved localisation of the lesion and preoperative planning.3 16 26 Sixty per cent (19/35) of cases with documented surgical timing adopted this conservative strategy, with surgery delayed by a minimum of 3 months.3 11 14–17 22 24 25 27 30–12 Two cases required emergency interventions (cervical drainage,4 tracheostomy21) to relieve compressive symptoms, with definitive surgery deferred to a later date.4 23 Curative surgery was performed within the same inpatient admission in 40% (14/35),5 6 10 13 16 18 19 23 26 28 29 with worsening compressive distress (11/14)5 10 13 16 18 26 28 29 and deterioration in haemoglobin (2/14)22 being the chief concerns. Two authors acknowledged that immediate interventions resulted in difficult operative experiences due to large number of adhesions, necessitating hemithyroidectomies in both instances.3 7

Ulrich et al argued that patients with VC involvement should be operated within a month, minimising the risk of re-bleeding and recurrent laryngeal nerve injury secondary to tethering.5 From our review, VC immobility was described in six patients on initial FNE.5 9 12 16 32 Four patients underwent surgery within 4 weeks, of initial presentation7 9 16 26 32, with VC paralysis persisting in two cases.26 32 VC paralysis was also seen to persist in a further case from the literature review,12 although timing of surgery was not documented.

Our case study and literature review highlight the importance of serum Ca2+ in spontaneous neck haemorrhage, with parathyroid adenoma being a possible culprit. In such scenarios, definitive operative management is best deferred in the absence of airway compromise or VC paresis, achieving better surgical outcomes when compared with immediate curative procedures.2 16


