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Case report

The role of unhenanced CT in a ruptured parathyroid adenoma: A case report. ☆,☆☆

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ABSTRACT

Acute bleeding is a rare and potentially life-threatening complication of a Parathyroid Adenoma described in just a few cases in literature. We describe the case of a healthy 53-years-old female patient without prior history of parathyroid pathology who presented with acute onset of neck and mediastinal hemorrhage. Ultrasound (US), Computed Tomography (CT) and Magnetic Resonance Imaging (MRI) combined with laboratory tests led to the diagnosis of a bleeding Parathyroid adenoma. This case is presented to sensitize both Radiologists and Clinicians about this rare presentation that should be put into differential diagnosis of acute neck swelling and pain.

Introduction

Parathyroid Adenoma (PA) is a benign neoplasm arising from Parathyroid glandular tissue.PA is the most common proliferative disorder of the parathyroid [1] and contributes to 75%-85% of cases of Primary Hyperparathyroidism (pHPT). It's usually asymptomatic and suspected on the basis of a laboratory finding of hypercalcemia, most of the times associated with increased levels of Parathyroid Hormone (PTH). Signs and symptoms, when present, are related to hypercalcemia and have a wide range of manifestations, most commonly bone pain (demineralization), abdominal pain (constipation, peptic ulcer disease, pancreatitis, nephrolithiasis), depression, lethargy. These lesions are usually treated with parathyroidectomy, curative in almost 95% of cases.

☆ *Competing Interest*: We confirm that this work is original and has not been published elsewhere nor is it currently under consideration for publication elsewhere.
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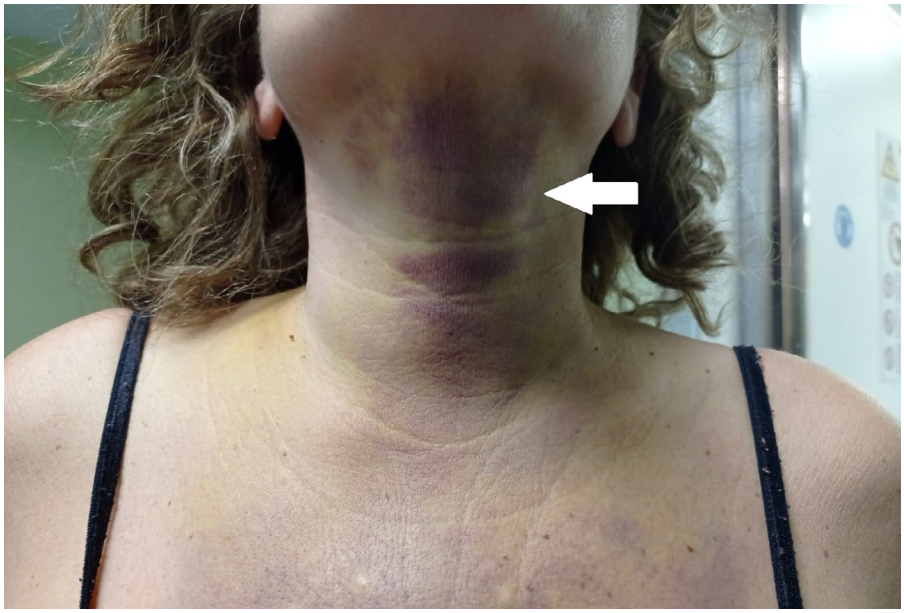

Fig. 1 – Image depicting swelling of the patient's neck (white arrow) caused by the underlying hemorrhage.

Primary bleeding from a PA is a rare complication and sometimes first manifestation of disease presenting in an Emergency Medicine setting, possibly leading to upper airways obstruction and urgent surgery [4,12,13].

We present the case of a spontaneous hemorrhage involving the neck and upper mediastinum from a previously unknown PA, successfully diagnosed and treated.

Case report

A 53-year-old woman patient come to the Emergency Room complaining progressive development of mild dysphagia and neck swelling in the last few days. The patient had no prior history of sign or symptoms related to PA or Primary Hyperthyroidism.

Physical examination confirmed the presence of a visible neck swelling with associated bruising extending from the chin to the upper anterior chest area (Fig. 1). No signs of airways compromission or hemodynamic instability was found so the patient underwent ultrasound examination of the neck as first line evaluation.

US with Color Doppler evaluation revealed the presence of a 2 cm focal nodular hypoechoic hyper vascular lesion in the right lateral area of the neck, below the right thyroid lobe associated with a surrounding dishomogeneous hyperechoic pseudo-mass (Fig. 2).

Contrast-enhanced Multi Detector Computed Tomography (MDCT) was immediately performed which better defined the right paratracheal 2 cm nodular lesion, showing moderately high HU on unenhanced CT (Fig. 3) and intense contrast enhancement, associated with a heterogeneous bleeding extending from the neck to the posterior mediastinal compartment resulting in a little mass effect on the trachea and esophagus, the latter showing reactive circumferential wall thickening (Fig. 4A, 4B, 4C).

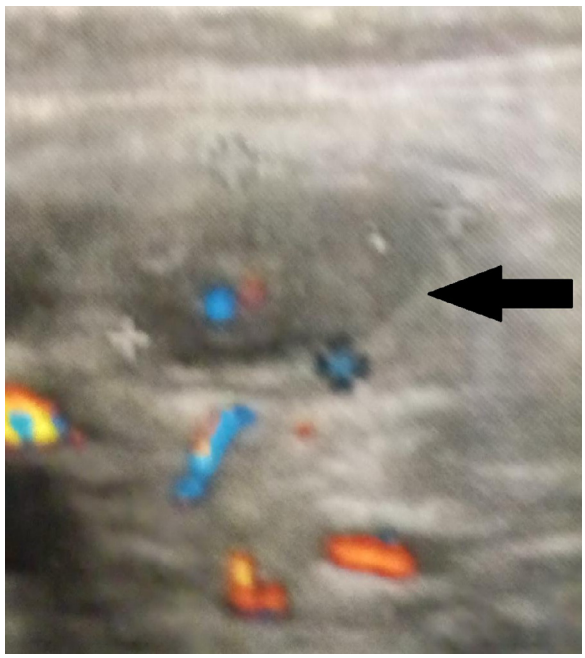

Fig. 2 – Ultrasound image shows, under the lower right lobe of the thyroid, a hypoechoic capsulated nodular formation (black arrow) of 22 × 18 mm, with intense color-Doppler signal.

Laboratory findings revealed the presence of hypercalcemia (serum calcium 12 mg/dl) associated with increased blood levels of Parathyroid Hormone (PTH 321 pg/ml), highly

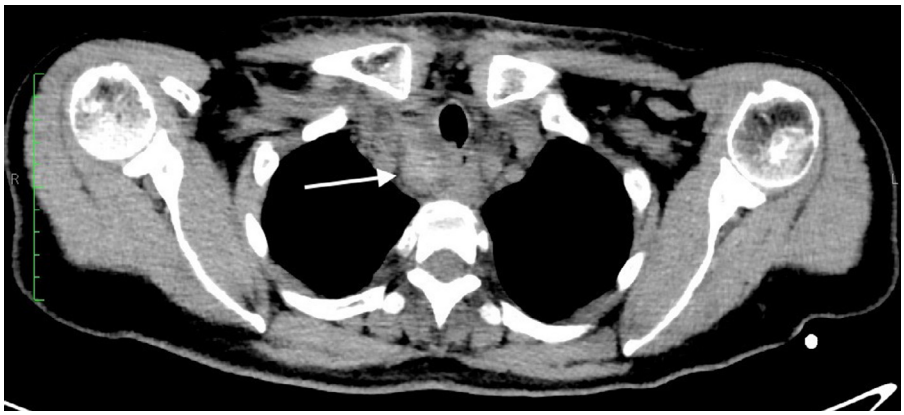

Fig. 3 – Unenhanced CT image shows dishomogeneous density in the upper and anterior mediastinum (hemorrhage). An ovalar hyperdense lesion on the right side of the upper trachea is shown (white arrow).

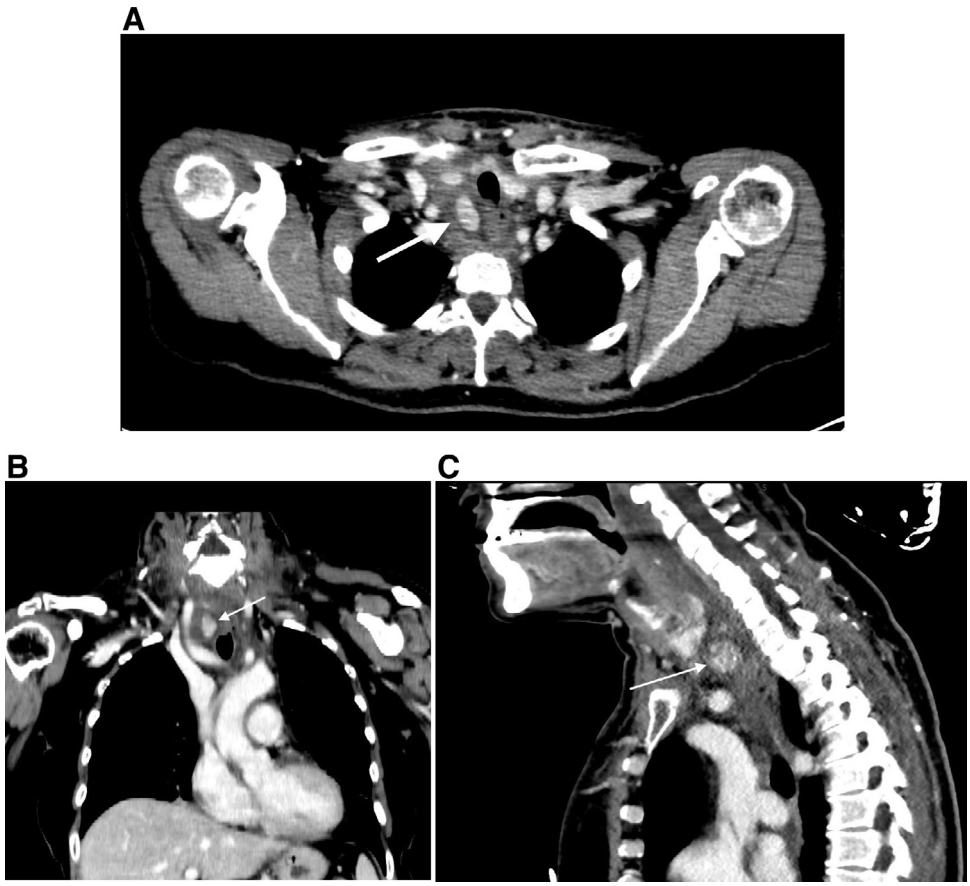

Fig. 4 – Contrast-enhanced CT images (A: axial, B coronal, C sagittal) shows homogeneous enhancement of the ovalar lesion (white arrow).

raising suspicion of a spontaneous bleeding from a parathyroid adenoma.

Clinical stability of the patient suggested a conservative treatment of the hemorrhage.

An elective surgical intervention for definitive treatment of the lesion was scheduled three weeks after the acute bleeding.

Preoperatively a 99mTc-Sestamibi Scan was performed which showed uptake of the tracer on the right side of the neck.

A Contrast-enhanced MRI of the neck was also performed. The MRI showed inhomogeneous bleeding in the prevertebral space, extended from the level of C1 vertebra to the

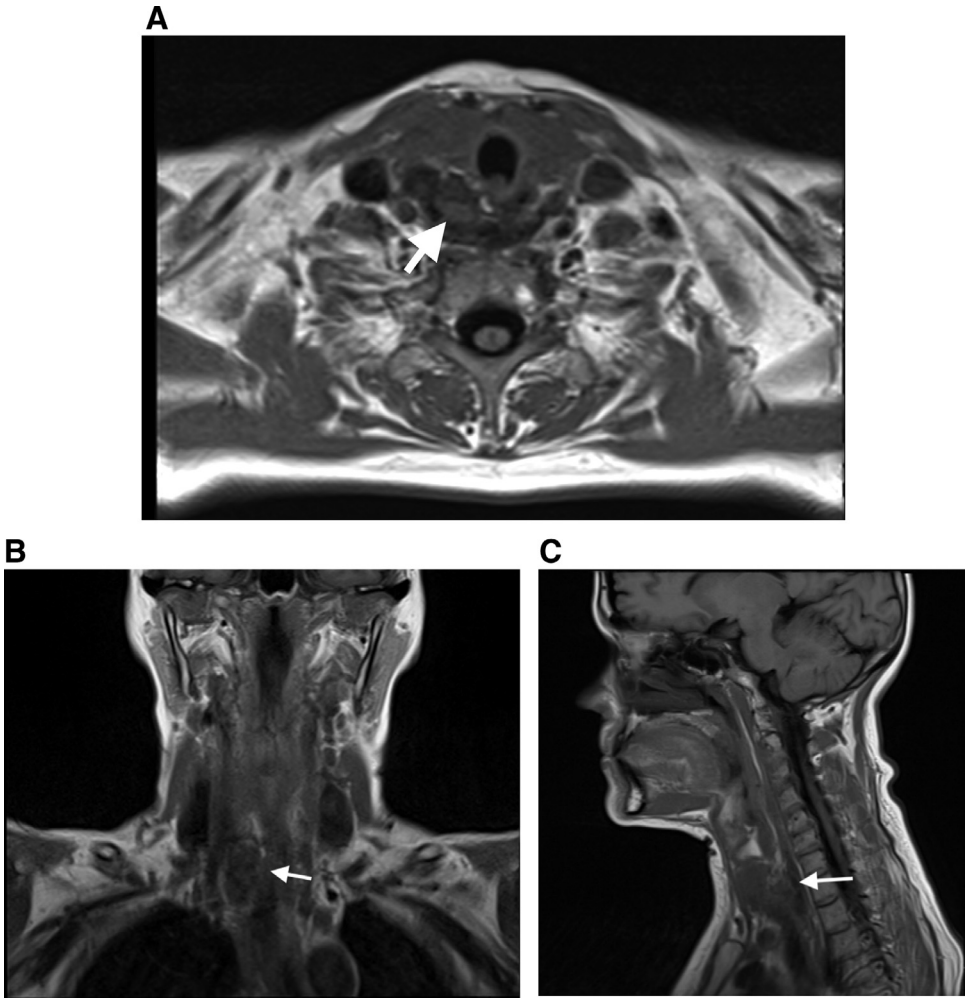

Fig. 5 – T1 TSE Dark-fluid images (A: axial, B coronal, C sagittal) shows a dishomogeneous tissue consistent with hemorrhage surrounding a poorly defined ovalar lesion on the right side of the upper trachea is shown (white arrow).

level of T3 vertebra, exerting compression on the right thyroid lobe anteriorly and the esophagus on the left side. After contrast administration an homogeneously enhancing nodular lesion was better defined on the right behind the thyroid lobe (Fig. 5A,5B,5C; Fig. 6).

Right hemithyroidectomy with right parathyroidectomy was performed due to extensive fibrous connections between the structures subsequent to the prior hemorrhage.

Histology showed a partially encapsulated proliferation of chief cells and oxyphil cells in the parathyroid with no signs of vascular invasion or necrosis, thus confirming the diagnosis of a Parathyroid Adenoma.

The patient had an unremarkable postoperative course and fully recovered.

Discussion

Parathyroid Adenoma is the most common proliferative disorder of the parathyroid, a spectrum of pathologies that includes parathyroid hyperplasia, parathyroid adenoma, and parathyroid carcinoma [1]. A solitary adenoma is the etiology of 75%-85 % of cases of Primary Hyperparathyroidism, a generalized disorder of calcium, phosphate, and bone metabolism due to an increased secretion of Parathyroid Hormone (PTH), with subsequent Hypercalcemia and Hypophosphatemia.

PA can occur in any age group, with the highest incidence in fifth and sixth decades and a female predilection (2:1 female/male ratio) [2].

PAs are well delineated and often encapsulated lesions usually circumscribed by a rim of normal parathyroid tissue. Some studies suggest a slight predominance in the lower rather than in the upper glands [3], even though they can be found in any location including ectopic and supernumerary parathyroid glands [4-9]. Most of the times only one gland is involved by proliferative disorders, but multiglandular disease has been reported in 6%-33% of cases [10].

Primary Hyperparathyroidism (pHPT) etiology may be sporadic or inherited, being sporadic in the vast majority of cases. Some cases of sporadic pHPT have been related to environmental factors, primarily radiation exposure [11-15] and lithium [16-18]. Many mutations and molecular alterations have been linked to both inherited and sporadic pHPT and

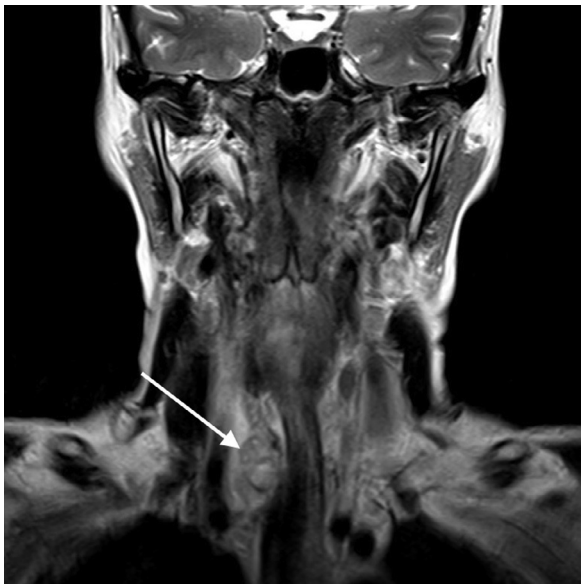

Fig. 6 – T2 TSE coronal image depicts a mildly hyperintense nodular lesion (white arrow) surrounded by hemorrhage exerting mass effect on the esophagus.

may affect the frequency of multiglandular disease, the most common involving MEN1, RET, PRAD1 and CDC73 genes [10]. pHPT is usually asymptomatic, however the occasional symptoms are often vague and nonspecific, thus diagnosis is based on the laboratory findings. Patients most of the times show increased levels of total and/or ionized calcium, but cases of normocalcemic pHPT have been reported [19]. PTH levels are usually high or inappropriately normal (not suppressed by hypercalcemia).

Parathyroidectomy is the only curative treatment and is indicated in both symptomatic and asymptomatic patients [10].

Imaging has no definite role in confirming or discharging the diagnosis of pHPT/PA, and even patients with negative imaging remain candidates for parathyroidectomy [10]. Imaging is usually performed after deciding for parathyroidectomy for surgical planning and is usually less accurate in the setting of multiglandular disease. Standard imaging modalities for the preoperative localization of a PA include Ultrasound and 99mTc-Sestamibi-SPECT scintigraphy.

Spontaneous rupture causing a neck hematoma is a rare and potentially life-threatening complication of a PA, first reported by Capps in 1934 [20]. Suggested pathogenesis is related to the rupture of a very thin capsule of the PA that allows blood to extend in the surrounding soft tissues of the neck with potential compression on trachea and esophagus [21].

Clinical presentation of a neck hematoma secondary to ruptured PA usually includes bruising and swelling of the neck, pain, dysphagia, and hoarseness. Hypercalcemia was associated in some cases and might be a key feature to suggest the parathyroid source of bleeding.

Diagnostic imaging work-up in the acute stage is not fully established. Ultrasound is commonly the first performed examination in the emergency setting, sometimes showing the typical hypoechoic nodular parathyroid adenoma associated with extensive hematoma in the surrounding soft tissue, but CT or MRI is usually performed for better characterization. In our opinion CT and MRI may better depict the hyperenhancing nodular lesion and are able to better define the extension of the hemorrhage and complications such as tracheal-esophageal compression or deviation possibly requiring surgical intervention.

Treatment of the acute phase aims to protection of the airways; surgical intervention is considered when continued bleeding causes hemodynamic compromise [21]. Parathyroidectomy is indicated as elective surgery after the complete resolution of the bleeding [21].

Disclosures

Publication is approved by all authors and by the responsible authorities where the work was carried out. Each author have participated sufficiently in any submission to take public responsibility for its content.

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