The ‘sentinel clot’ sign in spontaneous retropharyngeal hematoma secondary to parathyroid apoplexy

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Abstract
Spontaneous retropharyngeal hemorrhage from a cervical parathyroid adenoma is a rare complication of primary hyperparathyroidism. Because of its rarity, it has seldom been documented in the radiologic or ENT literature. Patients may present with a variety of manifestations, ranging from dysphagia to dysphonia to life-threatening dyspnea or hemorrhage. Awareness of a possible thyroid or parathyroid etiology may expedite treatment and prevent unnecessary interventions. We present a case of spontaneous retropharyngeal hemorrhage in which the “sentinel clot” sign enabled us to identify the lesion of origin.

Introduction
Parathyroid adenomas may secrete small, unrecognized hemorrhages, possibly as a result of their rich vascular supply and areas of cellular necrosis.1,2 This is perhaps not surprising because thyroid adenomas, which share a common vascular supply with the parathyroid glands, also frequently hemorrhage. In all likelihood, a neoplasm that affects the parathyroid glands, even though benign, will demonstrate features of neovascularity.3 Numerous imaging modalities have been used to diagnose hemorrhage from a parathyroid mass, be it an adenoma, cyst, or adenocarcinoma.1 However, the mainstay of imaging in the emergency setting, is computed tomography (CT). Among the several advantages of CT is that it may detect a “sentinel clot,” a high-density focus of clotted blood. Such a finding can direct the radiologist to the source of the hemorrhage. We report a case of retropharyngeal hematoma in which an accurate diagnosis was obtained on CT by virtue of the detection of a sentinel clot adjacent to the area of the parathyroid and thyroid glands.

Case report
A 68-year-old woman presented to the Emergency Department with dysphonia and stridor that rapidly progressed to respiratory distress and necessitated insertion of a nasotracheal tube. Her symptoms had followed a 3-day prodrome of dyspnea, myalgia, and sore throat. Physical examination revealed that her neck was swollen, and plain radiographs demonstrated significant retropharyngeal edema. The patient’s clinical history was negative for recent trauma, anticoagulation, and significant illness. Laboratory blood analysis demonstrated that her serum calcium level was elevated (2.70 mmol/L; reference range: 2.12 to 2.65) and that she was anemic.

Contrast-enhanced spiral CT demonstrated an extensive retropharyngeal fluid collection, with the characteristic whorled, variable-density appearance typical of acute hemorrhage (figure). The hemorrhage extended from the soft palate well into the mediastinum. A focal area of higher density within this space, which was believed to represent a sentinel clot, had displaced the thyroid gland anteriorly and the carotid space laterally, and it had compressed the trachea, pharynx, and esophagus.

The patient recovered well following surgery and was successfully managed on medical therapy. No further surgery has been performed.

Discussion
Approximately 80% of all cases of primary hyperparathyroidism are attributable to parathyroid adenoma.4 Parathyroid adenoma usually occurs secondary to excessive parathyroid hormone production and the sequelae of hypercalcemia.1,2 Spontaneous, nontraumatic hemorrhage is a recognized, albeit rare, manifestation of a cervical or mediastinal parathyroid adenoma. It is attributable to...
the rich vascularity of this area, to the neovascularization that occurs with an underlying neoplastic process, and to the imbalance that occurs when uncontrolled cellular growth outstrips its blood supply. This form of "autoparathyroidectomy" or "parathyroid apoplexy" results in hemorrhagic necrosis of the gland that is similar to processes seen in other endocrine gland neoplasms, namely pituitary apoplexy and adrenal adenoma. Patients may be hypercalcemic as a result of the hyperfunctioning of the preexisting adenoma. This problem may be further compounded by a massive surge of parathyroid hormone that is released into the circulation as the gland necroses.

Intracapsular rupture of a parathyroid lesion is more common than extracapsular rupture, and it runs a benign, limited course; patients simply present with discomfort in the thyroid gland area. Most of these cases are probably misdiagnosed as hemorrhage into a thyroid cyst. Most hemorrhages within the thyroid gland are self-limited, owing to the firm and complete fibrous capsule in which the gland is located. The capsule encasing the parathyroid glands is not as dense, and it is often attenuated or incomplete. Therefore, the potential exists for extracapsular rupture with associated extensive hemorrhage into (1) adjacent soft tissues, potentially including the neck and mediastinum, or (2) if the gland is ectopic, primarily within the mediastinum.

Patients with extracapsular rupture of a parathyroid adenoma may present with diverse symptomatology. The clinical triad of hypercalcemia, neck swelling, and ecchymosis has been described. Other manifestations include pain, dysphagia, stridor, and respiratory distress. Mediastinal hemorrhage may also occur in the setting of an ectopic parathyroid gland. Mediastinal involvement may mislead the clinician and radiologist, who are often concerned that an aorta is responsible for the hemorrhage; this can result in a delayed diagnosis. Manifestations of hyperparathyroidism are likely to be evident sometime before this, and may be an important diagnostic clue.

It may be difficult to differentiate a parathyroid adenoma hemorrhage from other diagnoses—such as carcinoma, thyroiditis, trauma, and a hemorrhage from a thyroid and parathyroid cyst—or from a vascular structure. Identification of an elevated serum calcium level may help secure the preoperative diagnosis because cysts and thyroiditis are associated with normal calcium levels. Occasionally, an enlarged parathyroid gland will be visualized on CT, but it would be prudent not to over-rely on identification of the parathyroid lesion because most parathyroid adenomas are small and likely to be obscured in the setting of significant acute hemorrhage.

The first reported case of retropharyngeal hemorrhage was published by Capps in 1934. That patient presented with hemorrhagic shock, but the cause was not diagnosed until autopsy after the patient had been exsanguinated. Since then, several patients have been successfully managed both surgically and conservatively; the choice of treatment being made according to the severity of presenting symptoms, hemodynamic stability, and the presence or absence of active bleeding.

Detection of a sentinel clot reliably indicates the presence of an injury to an adjacent organ. Previous reports have documented the usefulness of this radiologic sign in localizing the source of hemorrhage in the abdomen. Various imaging modalities have been used to confirm retropharyngeal hemorrhage and to guide the clinician to the causative pathology. The mainstay of imaging, particularly in the emergency setting, is CT. Chest and lateral neck radiographs may demonstrate features of retropharyngeal hemorrhage—including anterior bulging of the retropharyngeal space, mediastinal widening, and...
displacement and distortion of the trachea, larynx, and pharynx—but they provide little in the way of identifying its source. On x-ray, the appearance of a hemorrhage is similar to that of an infection or a soft-tissue mass. CT provides a better characterization of the cause of the swelling, its extent, and the presence of complications. CT confirms the presence of retropharyngeal hemorrhage (and thereby excludes abscess), defines its extent, and localizes its epicenter. It may also detect a sentinel clot and thereby direct the radiologist to the source of the hemorrhage. Furthermore, because CT demonstrates adjacent structures, the radiologist can assess related complications such as tracheal, pharyngeal, and vascular compression and aspiration pneumonitis. For all these reasons, CT is the modality of choice.

Other imaging modalities are less likely to have a significant impact on management in the acute setting, although ultrasonography may be useful in assessing vessel caliber and hemorrhage and in detecting a cyst or adenoma. Technetium, thallium, and sestamibi isotope scans can localize the site of an adenoma, but they are not likely to be used in the acute setting. Angiography has been used in the context of traumatic hemorrhage from a ruptured parathyroid adenoma, and it resulted in the successful embolization of branches of the thyrocervical trunk; however, it has been reported in only a single patient, so extensive comment on its role is not warranted. The use of magnetic resonance imaging is not widespread in the emergency setting because of the acute nature of cases and the unlikely that movement-related artifact can be avoided.

References