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## **Case Report**

# Nontraumatic adrenal hemorrhage: the adrenal stress

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#### ABSTRACT

Bilateral adrenal hemorrhage is a rare condition, which is burdened by potentially life-threatening consequences related to the development of acute adrenal insufficiency. Despite treatment with stress-dose glucocorticoids, a mortality rate of 15% has been reported, which varies according to the severity of underlying predisposing illness and could be much more higher if the adrenal insufficiency is not promptly recognized. An early diagnosis is crucial, though, because of nonspecific clinical and laboratory findings, adrenal hemorrhage is rarely suspected. Therefore, imaging has a pivotal role for the diagnosis of this uncommon condition but, despite adrenal hematomas characteristically appear round or oval with peripheral fat stranding, their initial presentation could be ambiguous. The authors describe a case of postoperative bilateral adrenal hemorrhage initially presenting at computed tomography scan as thickening of both glands surrounded by fat stranding, which led to close monitoring of adrenal function before unequivocal hemorrhage developed.

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#### Introduction

Bilateral adrenal hemorrhage is a life-threatening condition leading to adrenal crisis and potentially to death if not early treated [1,2]. The exact pathogenesis is unknown but is thought to be related to the physiologic increase of vascularization of adrenal glands in response to stressful events [2,3]. Vascular congestion may occasionally cause hemorrhage, very rare bilateral. Several conditions determine such stress response, among which the most important risk factors seem

to be postoperative status, thromboembolic disease, and coagulopathy [3]. Most of patients presenting with adrenal hemorrhage are already affected by severe conditions; therefore, it is difficult to recognize signs and symptoms of an incoming adrenal crisis [2–6]. When suspected, the diagnosis is confirmed whit adrenocorticotropic hormone (ACTH) test, which however is not always feasible in unstable patients, and computed tomography (CT), which shows the adrenal injury. CT is considered pivotal to early diagnose adrenal hemorrhage and start a prompt corticosteroid replacement therapy [2,5,7].

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#### Case report

A 76-year-old woman underwent a pancreaticoduodenectomy with Whipple procedure for a malignant ampulloma. On the fifth postoperative day, the patient developed an acute epigastric pain with nausea, vomiting, and pyrexia (38.6°C) suggestive of postoperative complications. Therefore, a contrast-enhanced CT scan of the abdomen was performed and revealed nonspecific inflammatory stranding surrounding mildly enlarged adrenal glands (Fig. 1), which appeared normal at preoperative CT examination (Fig. 2). No anastomotic leak or other postsurgical complications were detected. Despite leukocyte count was within normal range, clinical suspect of systemic infection led to begin empiric antibiotic therapy, waiting for specific bacterial cultures. However, the patient continued suffering from epigastric pain, and in addition, on the 13th postoperative day, she presented with hemoglobin drop (120 g/dL), hypotension, hypovolemia, and tachycardia (130 bpm) with subtle electrolyte abnormalities (sodium 126 mmol/L; potassium 6.9 mmol/L) and low serum cortisol level (5 mcg/dL) with ACTH >300 ng/L, suggestive of adrenal crisis.

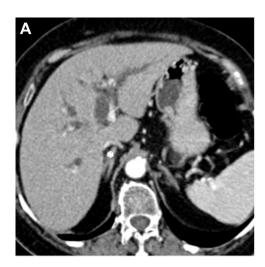
A new CT scan revealed bilateral mainly hypodense ovoid adrenal masses, which did not change after contrast administration, in keeping with organizing adrenal hemorrhages (Fig 3). Consequently, diagnosis of adrenocortical insufficiency secondary to bilateral adrenal hemorrhage related to postsurgical stress was made. Medical treatment, with intravenous hydrocortisone (initially, an intravenous bolus of 100-mg hydrocortisone followed by 200 mg over 48-hour continuous infusion) and correction of hypovolemia with isotonic saline (1000-mL isotonic saline within the first hour), resulted in a rapid improvement in her electrolyte imbalance with satisfactory outcome and no further abdominal symptoms. The patient made a good recovery and was discharged after 2 weeks on a reducing dose of steroids. After 6 months, the patient did not report any episode of adrenal insufficiency and a magnetic resonance imaging showed normal appearance of adrenal glands (Fig. 4).

#### Discussion

Bilateral adrenal hemorrhage is a rare condition with an estimated incidence of 0.14%-1.8% based on postmortem studies [1]. This infrequent clinical entity is burdened by potentially life-threatening consequences related to the development of acute adrenal insufficiency, when at least 90% of glands are injured [2]. Despite treatment with stress-dose glucocorticoids, some patients with adrenal hemorrhage may die because of underlying disease or diseases associated with the adrenal hemorrhage itself. Overall, a mortality rate of 15% has been reported; however, it varies according to the severity of the underlying predisposing illness and could be much more higher if the adrenal insufficiency is not promptly diagnosed [1,4,5].

The exact pathophysiology of bilateral adrenal hemorrhage is still uncertain. Cortisol has many important metabolic and endocrine functions, during stressful events in particular [6]. The stress-induced adrenal hemorrhage is hypothesized to be an exacerbation of the physiological combination of increased arterial blood flow to the adrenal gland and slow drainage by relatively few venules and a single adrenal vein, leading to intraglandular vascular congestion and possible subsequent hemorrhage [2,3]. This dramatic event may manifest because of several predisposing factors, among which stress caused by surgery, severe illness, overwhelming sepsis, burns, hemorrhagic diatheses (e.g., anticoagulant use, thrombocytopenia), thromboembolic disease (including antiphospholipid antibody syndrome) are the main nontraumatic ones [2–4,7–9].

Because of nonspecific clinical and laboratory findings, adrenal hemorrhage is rarely suspected. The most frequently described signs are abdominal pain, vomit, fever, weakness, severe hypotension and altered conscious state, which often overlap with other concurrent severe illness making difficult the diagnosis [3–6]. Fever is the most common physical sign occurring in about 70% of cases [2]. Hypotension is not often seen before the development of dramatic hypotension and shock [2,3].



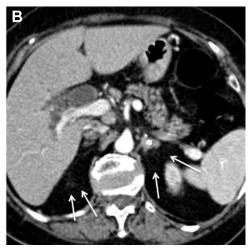


Fig. 1 — Computed tomography, axial image, portal phase showed bilateral (A: right; B: left) mild adrenal enlargement (stars) with surrounding fat stranding (arrows).

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