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Case Report

Spontaneous Left Hypochondrium Pain Revealing a Unilateral Adrenal Ischemia: A Case Report

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Abstract

Adrenal glands can be the site of rare hemorrhagic or ischemic phenomena causing abdominal pain. The diagnosis is made with an abdominal contrast enhanced CT scan. Unilateral adrenal ischemia is not usually responsible for Addison's disease, as the contralateral gland compensates for the hormonal secretion deficit. The etiological assessment of adrenal ischemia should include a look for lodging clot and thrombophilia. Treatment is based on analgesics administration and anticoagulation at a therapeutic dose. The anticoagulation duration must be adapted according to the etiology.

Keywords: Adrenal gland; Ischémia; Thrombophilia; Embolism; Steroid withdrawal

Introduction

Adrenal glands can be the site of rare hemorrhagic or ischemic phenomena causing abdominal pain rather localized in the hypochondrium. The diagnosis is made with an abdominal contrast enhanced CT scan. These Adrenal glands, because of their vascular network anatomy, are vulnerable to hemorrhagic accident. Adrenal hemorrhagic accidents are favored by sepsis, anti-phospholipid syndrome or localized cancer. Adrenal ischemia not secondary to hemorrhagic accident is extremely rare, some case are reported in pregnant women. We describe a spontaneous unilateral adrenal ischemia found in a patient with abdominal pain.

Case

A 60-year-old patient presented to the emergency department with abdominal pain mainly localized in the left hypochondrium. The pain started suddenly a day ago and was resistant to common analgesics such as acetaminophen and NAIDS. The patient's medical history was marked by substituted hypothyroidism and appendectomy in childhood. The admission laboratory test reported an anormal leukocytosis with 24,910 white blood cells / µL (normal <9500 el / µL) and thrombocythemia with 551,000 platelets / µL (normal <400,000 el / µL). An enhanced abdominal CT scan was performed (Figure 1). It showed a defect in the enhancement of the left adrenal gland associated with an infiltration of the neighboring fat. The patient had been admitted to the hospital. During hospitalization, the cardiological assessement including a trans-oesophageal cardiac ultrasound and a rhythmic holter to look for an embologenic focus was performed and didn't highlight any abnormality. Because of the abnormal blood count objectified on admission, a haematological assessment by bone marrow puncture was performed and showed a mixed myelodysplastic and myeloproliferative syndrome. An anticoagulant treatment for a period of 3 months was started and associated with a cytoreductive treatment with hydroxycarbamide allowing a normalization of the blood count after 30 days. Endocrine function was checked by measuring the morning cortisol 48 hours after admission, this was measured at 8.69 µg / dL (normal: 4.46 - 22.7 µg / dl).

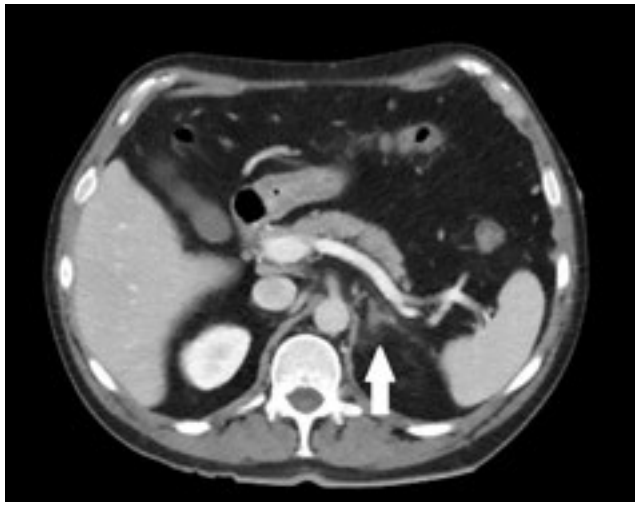

Figure 1: Abdominal enhanced CT scan showing a left adrenal ischemia.

Discussion

The adrenals glands are two triangular asymmetric glands located in the retro peritoneum, at the level of the kidney upper pole. They weigh on average four to six grams. They have an essential endocrine role; the mineralocorticoids, glucocorticoids and sex steroids synthesis in the cortex area and the catecholamine production in the medulla. These glands have a rich arterial vascularization. The blood is supplied from the upper adrenal arteries (originating from the inferior phrenic artery), the middle adrenal arteries (originating from the abdominal aorta) and the lower adrenal arteries (originating from the renal artery). Venous return takes place only through the adrenal vein. The left adrenal vein empties into the left renal vein while the right adrenal vein empties directly into the inferior vena cava. This unique venous return exposes the adrenal gland to a vulnerability to hemorrhages, for example in a situation of vasoconstriction such as sepsis or thrombosis of the adrenal vein. These adrenal hemorrhages can even be bilateral, they are then called Waterhouse-Friderichsen Syndrome. Risk factors for adrenal haemorrhage are thrombocytopenia, anticoagulant use and sepsis [1]. Patients with anti-phospholipid syndrome are particularly at risk for this type of bleeding.

Isolated ischemic phenomena are rarer. Cases of unilateral adrenal ischemia secondary to venous thrombosis have been described in the third trimester of pregnancy [2,3]. These patients present sudden unilateral hypochondrium pain, similar to the pain described in urolithiasis pathologies. The diagnosis mean is an enhanced abdominal CT scan or magnetic resonance. The radiological features of adrenal ischemia are a larger hypodense gland, without significant enhancement after injection of contrast agent. A weak enhancement of the capsule can be observed and bears the name of capsular sign. Venous or arterial thrombosis is sometimes also visible [4].

The aetiological assessment is focused mainly on the look for lodging clot (namely a trans-esophageal and rhythmic holter ultrasound) in the event of arterial thrombosis and coagulation abnormality (in particular Leiden factor V mutation) in the event of venous thrombosis

Endocrineically, adrenal insufficiency is unusual after the loss of one of the two glands. The contralateral gland has the ability to compensate for endocrine function after unilateral adrenalectomy, for example [5]. The basal cortisol level remains unchanged after this type of surgery although the level of ACTH increases and the response to ACTH administration decreases [6]. Steroid replacement therapy is therefore only indicated in the case of loss of the two adrenal glands or unilateral adrenalectomy associated with Cushing's syndrome [7]. As a precaution, measuring a basal cortisol level forty-eight hours after the ischemic event is recommended especially in the presence of adrenal insufficiency characteristic symptoms (Addison's disease) such as asthenia, hypotension, anorexia, abdominal pain and confusion.

Treatment is based on analgesics administration and initiation of anticoagulation at a therapeutic dose, pending the results of the aetiological assessment.

Conclusion

Unexplained pain in the hypochondrium can be caused by rare condition such as unilateral adrenal ischemia. The diagnosis is made by performing an enhanced CT scan. The assessment should include a look for lodging clot and thrombophilia. Steroid supplementation is not necessary in unilateral disease. Treatment is based on the administration of analgesics and anticoagulants.

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