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CASE REPORT

Idiopathic unilateral adrenal hemorrhage in an elderly patient

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Abstract We report the case of an 85-year-old woman who has been undergoing treatment for hypertension but has not received anticoagulation therapy. The patient was admitted to our hospital for the evaluation of a right adrenal tumor (size, 10×9 cm^2). Preoperative contrast-enhanced computed tomography and magnetic resonance imaging findings were indicative of adrenal hemorrhage (AH). Laboratory data revealed mild anemia but no adrenal dysfunction. The final pathological diagnosis was simply idiopathic adrenal hematoma. There is no case report of exactly idiopathic AH over 80 years old. We report an unusual case of idiopathic unilateral adrenal hematoma in an elderly patient. It is important to distinguish this benign lesion from a neoplasm and to consider idiopathic AH in an adrenal tumor during differential diagnosis in elderly patients who have not received anticoagulation therapy or suffered from trauma.

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Introduction

Adrenal hemorrhage (AH) is an uncommon condition, and its diagnosis is usually made at autopsy because the adrenal gland is located deep within the abdomen and is well cushioned by surrounding soft tissue structures [1]. AH is associated with meningococcal septicemia and disseminated intravascular coagulation (Waterhouse–Friderichsen syndrome), but trauma, hypotension, tumors, complications of pregnancy, surgical stress, or anticoagulation therapy may also cause AH [1]. The incidence of AH in children is seven times greater than that in adults [2–5]. AH in neonates and infants is known to manifest typically as an incidental abdominal mass, vomiting, anemia, jaundice, or scrotal swelling [6]. In adults, AH is mainly caused by trauma, surgical stress, or anticoagulation therapy; however, spontaneous or idiopathic AH is extremely rare in adults [1–5]. There is no case report of exactly idiopathic AH over 80 years old.

In the present study, we report the case of an elderly patient with spontaneous unilateral AH.

Case report

An 85-year-old woman was admitted to our hospital for the evaluation of a right adrenal mass. She had no history of abdominal trauma. The patient was diagnosed with mild hypertension at the age of 45 years and was treated with drugs but had never received anticoagulation therapy. She did not have a history of any specific systemic disease. The

patient complained of chronic abdominal pain for 4 months, and experienced general fatigue and palpitations. She was admitted to another hospital for examinations; abdominal computed tomography (CT) was performed, which revealed a right adrenal tumor. On admission, the results of her physical examination were normal and were as follows: height, 152 cm; weight, 56 kg; and blood pressure, 120/70 mm Hg. The heart rate reflected a normal sinus rhythm.

Laboratory data showed mild anemia (hemoglobin, 11.4 g/dl) but no hepatic, renal, or adrenal dysfunction {ACTH; 19.8 pg/ml, serum cortisol; 11.1 μg/dl at 3 p.m., urine free cortisol 32.3 μg/day (11.2–80.3), urine vanillylmandelic acid, 3.4 mg/day (1.3–5.1), urine adrenaline 14.2 μg/day (3.0–15.0), urine noradrenalin 110 μg/day (26–121), urine metanephrine 0.1 μg/day (0.05–0.23), urine normetanephrine 0.18 μg/day (0.07–0.26)}. The tumor markers {DHEA-S 1020 ng/ml (70–1770), CEA 2 ng/ml (≤5), CA19–9 5 U/ml (≤37)} levels in the patient's serum were within the normal range. Her endocrinologial examinations showed an Aldosterone:Renin ratio (ARR) 5 (≤30) and overnight 1 mg dexamethasone suppression test (DST): cortisol 0.1 μg/dl. We have concluded the tumor as non-functioning tumor.

Abdominal ultrasound revealed a giant echogenic mass adjacent to the stomach, transverse colon, and right kidney. The tumor was 10 × 9 cm^2 in size, well circumscribed, encapsulated, and mainly solid. Contrast-enhanced CT revealed a large adrenal tumor with low attenuation, and mild enhancement of the capsule around the tumor (Fig. 1). No liver metastasis or lymph node swelling was detected. Magnetic resonance imaging (MRI) showed low signal intensity of the tumor. On T1-weighted images, the signal intensity of the tumor was hypointense with some irregular areas of high signal intensity. T2-weighted images showed heterogenous low signal intensity of the tumor (Fig. 2a, b). Gadolinium-enhanced MRI revealed no lesion within the tumor and no findings suggestive of a neoplasm, such as hypervascularity, apart from the faint enhancement at the rim of the tumor (Fig. 2c). A whole-body scan performed using iodinated metaiodobenzylguanidine ([^{131}I]-MIBG) did not show sufficient ^{131}I accumulation in the adrenal tumor to indicate a pheochromocytoma. [^{18}F]-Fluorodeoxyglucose-positron emission tomography (FDG-PET) revealed no significant uptake of the radiotracer in the right adrenal tumor; therefore, it was clinically diagnosed as an adrenal hematoma. However, we could not rule out the possibility of a malignancy.

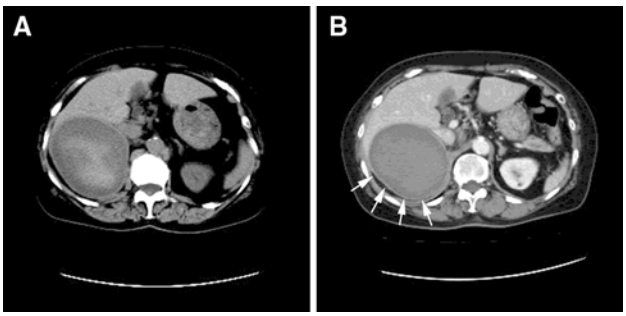

Fig. 1 CT scan of the abdomen. **a** CT scan demonstrates a large adrenal tumor with heterogeneous attenuation in right adrenal gland. **b** On contrast-enhanced CT, a right adrenal tumor has low attenuation with mild thin peripheral rim of enhancement (*arrow*)

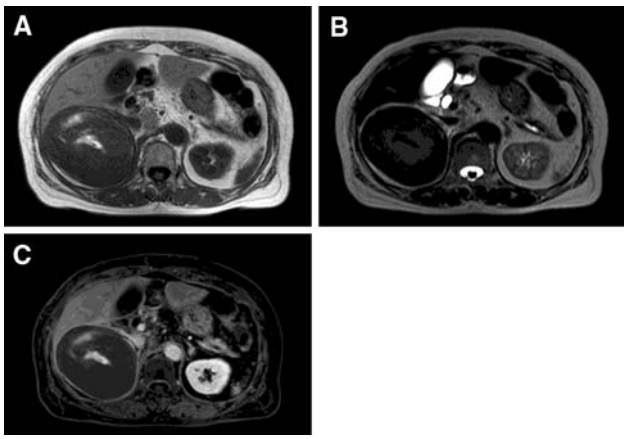

Fig. 2 MRI of the abdomen. **a** T1-weighted image shows hypointense right adrenal tumor with some irregular areas of high signal intensity. **b** On T2-weighted image, the signal intensity of the right tumor is heterogeneously low. **c** Gadolinium-enhanced T1-weighted fat saturation image demonstrates no significant enhancement except for faint enhancement at the rim of the tumor

The excised specimen contained a tumor which was 10 × 8 × 8 cm^3 in size, encapsulated, and partially connected to the adrenal gland. The tumor consisted of a large capsule and an old hematoma occupied the cavity (Fig. 3). On microscopic observation, it was found that the tumor was encapsulated by a thin fibrous tissue and that it involved the normal adrenal gland beneath the capsule. The tumor consisted of fibrinoid exudate and an old hematoma with peripheral organization (Fig. 4). No neoplastic lesion such as an adenoma, carcinoma, pheochromocytoma, or hemangioma was detected on histologic examination. Therefore, the final pathological diagnosis was simply idiopathic adrenal hematoma. In summary, we have reported a case of idiopathic AH in an elderly woman.

Discussion

Spontaneous or idiopathic AH is extremely rare in adults; there is no case report on AH in patients aged >80 years. AH can occur spontaneously in elderly patients because of anticoagulation therapy. Swift et al. [7] have reported that pheochromocytomas and a variety of primary adrenal tumors and cysts have been reported in association with

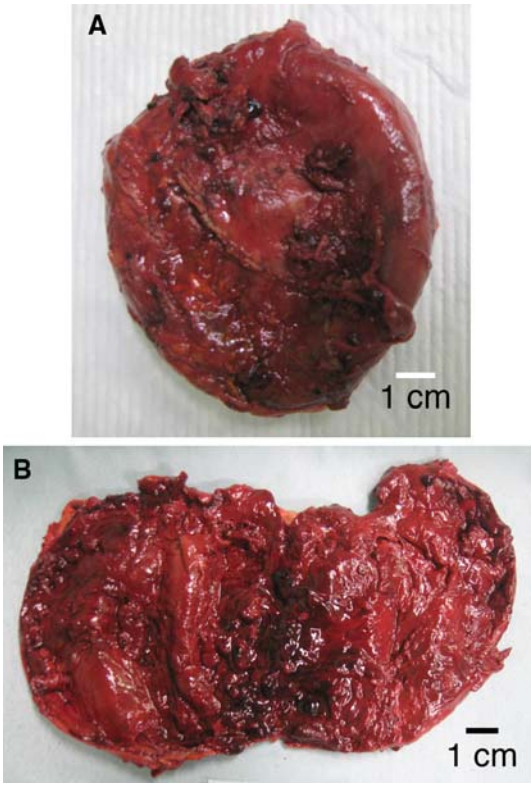

Fig. 3 Macroscopic findings of the excised tumor. The tumor consists of a large capsule which is 10 × 8 × 8 cm^3 in size. **a** and **b** are outside and cut section of the tumor, respectively, after the aspiration of the liquid occupied in the capsule

unilateral AH. In this case, an 85-year-old woman suffered from spontaneous or idiopathic AH although she did not receive anticoagulation therapy or have any adrenal tumors. Vella et al. [1] have summarized 141 cases of AH in patients aged >15 years during 25 years of experience at the Mayo Clinic. They classified AH into the following seven categories: incidentaloma (28 cases), spontaneous AH (16 cases), AH associated with antiphospholipid- and heparin-associated thrombocytopenia (20 cases), postoperative AH (14 cases), AH associated with anticoagulation therapy (3 cases), AH associated with trauma (4 cases), and AH associated with severe stress or sepsis (56 cases). According to their criteria, spontaneous AH is presented as acute, spontaneous hemorrhage of the adrenal gland with acute abdominal pain in the absence of prior trauma or anticoagulation therapy. In contrast, an incidentaloma is presented as an apparent nonfunctional adrenal mass detected by imaging studies performed for other reasons. Although Moore and Biggs [5] initially reported a case of AH manifesting as an asymptomatic adrenal mass, the mass can now be categorized as an incidentaloma.

Hoeffel et al. insisted that MRI was more useful than CT for the diagnosis of spontaneous unilateral AH, the most important finding being the high intensity on T1-weighted images, especially when the high signal intensity is mainly located peripherally. Furthermore, heterogeneously low signals on T2-weighted images were characteristic of an AH [3]. They also suggested that gadolinium-enhanced MRI may be helpful in distinguishing a collection of fluid, such as blood, from a neoplasm, such as a pheochromocytoma or carcinoma. However, the hemorrhagic stages in the eight cases of spontaneous unilateral AH reported by Hoeffel et al. [3] varied from subacute to chronic organizing hematoma. Therefore, although MRI may be useful for revealing tumor contents, making a precise diagnosis of AH is not easy when the entity and natural course of the disease are unknown. In a recently reported case, increased F-18 FDG uptake was observed in both adrenal glands in a 61-year-old woman who developed atraumatic bilateral AHs while on anticoagulation therapy [8]. However, in our case, the tumor showed no F-18 FDG uptake. More cases are needed to demonstrate the usefulness of FDG-PET in the diagnosis of AH.

Majority of the patients with adrenal bleeding do not show any signs of adrenal insufficiency. The most common symptoms are hypotension, confusion, lethargy, nausea, vomiting, tachycardia, and fever. More specific symptoms such as central abdominal pain radiating to the flank are experienced by 45% of the patients [9]. These symptoms together with anticoagulation therapy in an elderly or very young patient should raise clinical suspicion. In these cases, CT is an essential diagnostic tool [10]. Acute adrenal insufficiency (Addisonian crisis) may occur when the

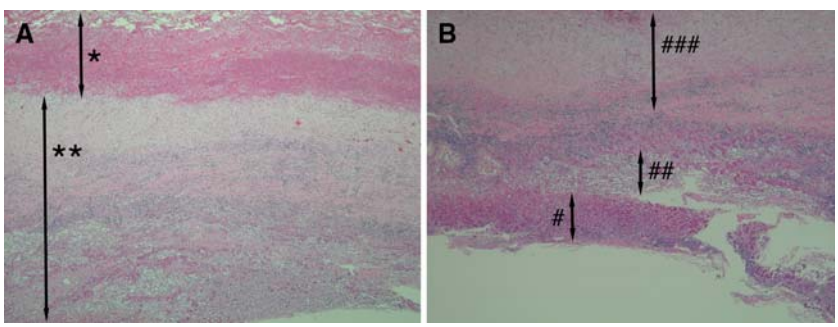

Fig. 4 Histology of the right adrenal tumor. **a** The tumor consists of an old hematoma (*) with fibrinoid exudate and organization (**) (100×). **b** The tumor involved the normal adrenal (#, adrenal cortex and ##, medulla of adrenal gland) beneath the capsule (###) (100×)

bleeding is bilateral. Such anticoagulant-related adrenal insufficiency may be life-threatening but can easily be treated with steroids [11].

Several cases of adrenal hematoma have been reported in elderly patients who were receiving anticoagulation therapy [1]. Steensrud et al. [12] pointed out the clinical suspicion of AH in elderly patients who receive anticoagulation therapy. In this case, an AH was detected in an 85-year-old woman who had not suffered trauma or received anticoagulation therapy and was not at any risk of AH. Hence, it is difficult to diagnose an idiopathic AH if the entity is unsuspected, such as in our case.

In conclusion, we report the first case of an elderly patient who suffered from idiopathic AH without any endocrinological disturbances. Our findings suggest that idiopathic AH in an adrenal tumor should be considered during differential diagnosis in elderly patients who have not received anticoagulation therapy or suffered trauma.

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