

Case Report

Spontaneous Unilateral Adrenal Hemorrhage

Deniz Bolat^{1*}, Volkan Sen¹, Tansu Degirmenci¹, Ozgu Aydogdu¹, Ibrahim Halil Bozkurt¹, Tarik Yonguc¹ and Mehmet Bilgehan Yuksel²

¹Department of Urology, Bozyaka Training and Research Hospital, Turkey

²Department of Urology, Celal Bayar University, Turkey

*Corresponding author

Deniz Bolat, Department of Urology, Izmir Bozyaka Training and Research Hospital, Izmir, Turkey, Tel: 905056383010; Email: drbolat@hotmail.com

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Keywords

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Abstract

Spontaneous adrenal hemorrhage is a rare and potentially life-threatening condition. Preoperative diagnosis is extremely complicated because of the formation of hematoma. Current treatment modalities range from supportive management to remobilization or immediate laparotomy. Herein, we present a huge idiopathic unilateral adrenal hemorrhage which was not associated with any trauma, previous surgery or coagulative disorders and without any underlying pathology such as a cyst, myelolipoma or tumor.

ABBREVIATIONS

AH: Adrenal Hemorrhage; CT: Computed Tomography; MRI: Magnetic Resonance Imaging; ACTH: Adrenocorticotropic Hormone; DHEASO4: Dehydroepiandrosterone-sulphate

INTRODUCTION

Adrenal hemorrhage (AH) is an extremely rare condition. The presentation of AH range from clinically insignificant signs to acute adrenal crisis and death. Associated risk factors with AH are based on case reports [1].

Idiopathic, unilateral AH is a rare entity that may have an acute presentation (e.g., idiopathic adrenal rupture) or may present as an asymptomatic adrenal mass [2,3]. Herein, we present an idiopathic unilateral AH without any demonstrable etiology and any underlying pathology such as a cyst, myelolipoma or tumor.

CASE PRESENTATION

A 57-year-old woman admitted to our hospital emergency service with acute right flank and right upper quadrant pain with nausea and vomiting for the last 8 hours in September 2014. She had a history of insulin-dependent diabetes mellitus. There was no history of a recent trauma or an operation. On admission, she had a temperature of 37.6 °C, her blood pressure was 100/50 mmHg and pulse rate was 120/minute. Abdominal examination revealed no defense or rebound. However, she had costovertebral angle tenderness on the right side. The laboratory test results showed hemoglobin 8, 7 g/dL and white blood cell count of 18,200/μL, platelet count of 360,000/μL. As well as, activated partial thromboplastin time (APTT) was 26, 9 sec and prothrombin time was 10, 6 sec. Biochemical analysis revealed

serum sodium 139 mmol/L, potassium 4, 2 mmol/L, calcium 9 mg/dl.

In hormonal work-up, serum cortisol level was 17, 9 μg/dl, Adrenocorticotropic hormone (ACTH) was 21, 4 pg/ml, dehydroepiandrosterone-sulphate (DHEASO4) was 60, 3μg/dl. As well as, metanephrine, normetanephrine and vanillylmandelic acid levels in 24 hour urine sample were normal.

Abdominal contrast-enhanced computed tomography (CT) showed 95x85 mm well-circumscribed, some low-densities areas included and milimetric lipid values demonstrated, heterogeneous lesion on the upper pole of the right kidney. CT scan result was suspicious for complicated right adrenal myelolipoma (Figure 1 a,b). After the hospitalization of the patient, her vital signs were stabilized, fluid replacement and erythrocyte transfusion were started and ampicillin intravenous antibiotic prophylaxis was initiated. Serial vital signs and hemoglobin levels were followed and hematoma size was measured once a day by ultrasound. On the 2nd day, abdominal magnetic resonance imaging (MRI) was performed. MRI demonstrated 90x87 mm sized heterogeneous solid mass on the right adrenal gland localization. In dynamic studies, no enhancement was detected in the mass. There were no lipid intensities, so that myelolipoma was ruled out. However, malignancy could not be excluded. Because of the suspicion of malignancy and decline in hemoglobin levels on the 3rd day, we decided to explore the patient (Figure 1 c,d). Right adrenalectomy was performed. During the operation, 9 cm organized hematoma was observed within the right adrenal gland which attached to the upper pole of the kidney, liver, inferior vena cava with right renal vein. Lesion was carefully dissected from the surrounding tissues and hematoma was aspirated. Right adrenal gland and the hematoma was surgically removed (Figure 2 a,b). The

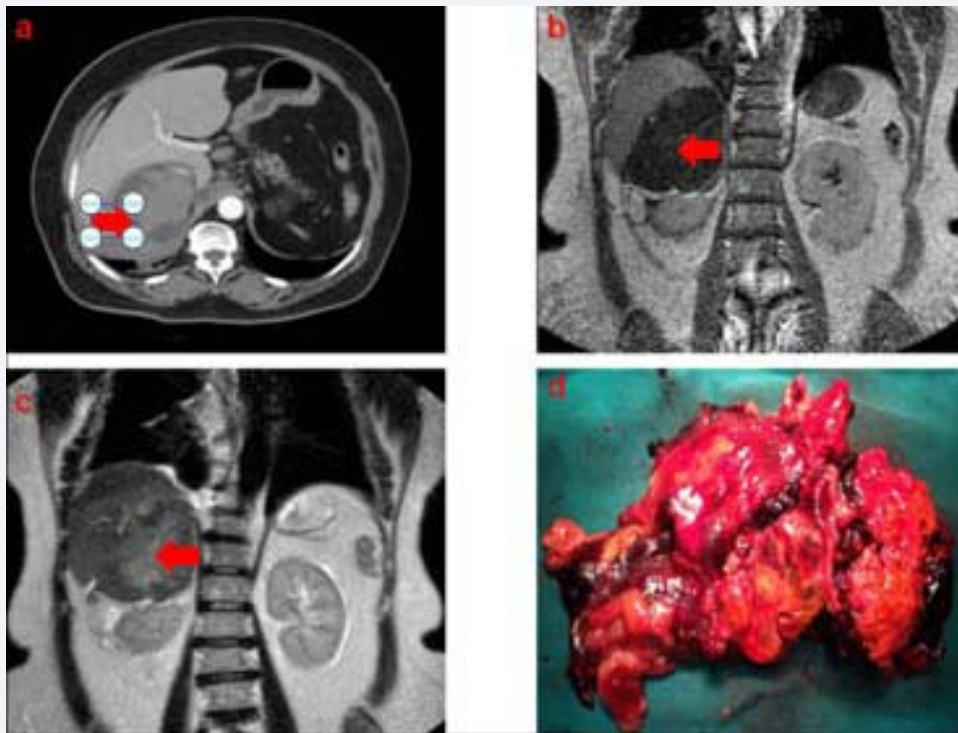


Figure 1 (a-d): Preoperative contrast-enhanced computed tomography showed 95x85 mm hematoma on the upper pole of the right kidney (a). Preoperative magnetic resonance imaging demonstrated 90x87 mm sized heterogeneous solid mass on the right adrenal gland localization. On T1-weighted imaging the lesion showed low intensity and it was hyperintense on T2-weighted imaging (b and c). Intraoperatively, 9 cm organized hematoma was observed within the right adrenal gland. Lesion was carefully dissected from the surrounding tissues and hematomas were aspirated. Hematoma with the right adrenal gland was surgically removed (d).

histopathology report revealed only hematoma and adrenal tissue without any tumor cells.

DISCUSSION

AH has been reported in 0.3%-1.8% of undetected cases in autopsy studies, although extensive bilateral AH may be present in 15% of individuals who die of shock [4]. The incidence of spontaneous AH has been reported from 0.14% to 1.1% and it usually involves the right gland [5]. The Pathophysiology that leading to AH is unclear; in non-traumatic cases available evidence has implicated ACTH, adrenal vein spasm and thrombosis and normally limited venous drainage of adrenal in the pathogenesis of the condition [4]. According to 25 year experience of Vella *et al.*, AH was associated with incidentaloma, spontaneous rupture, antiphospholipid and heparin-associated thrombocytopenia, postoperative conditions, anticoagulation therapy trauma, sepsis (especially meningococemia) or severe stress [6]. In this case, there was no history of infection, trauma, anticoagulant usage, or evidence of underlying adrenal tumor. Considering the etiological factors, our case is an example of idiopathic/spontaneous AH.

Our patient was presented with right flank and abdominal pain, vomiting, nausea with mild hypotension, sub febrile fever, tachycardia and low hemoglobin levels. All these findings were meaningful for the hemorrhage from anywhere, and further investigation was mandatory in order to confirm the diagnosis. Therefore, our patient was scanned by contrast-enhanced CT. AH is usually first diagnosed via CT or MRI. AH on CT scan is

characterized by a round or oval mass in the location of adrenal gland. The attenuation value of an AH depends on the age of the bleed [7,8]. MRI shows the adrenal hematoma more accurately than the other imaging tools, with high signal intensity on T1-weighted images. MRI also may differentiate sub acute and chronic hemorrhages [7-9]. Management of AH includes operative and nonoperative procedures. It depends on the presence of malignancy, viability of residual adrenal tissue, status of contralateral adrenal gland, patient hemodynamic and extension of the injury in traumatic AH.

In hemodynamically stable patients, conservative treatment can be considered. In these cases, intravenous hydration, ant biotherapy prophylaxis, measurement of serial blood counts, and administration of blood transfusion if necessary is essential [7].

In case of persistent bleeding, angioembolization of an AH may be an option [8,10]. Because of availability difficulties of interventional radiology capabilities, urgent laparotomy is required for unstable patients. If timing did not let a hormonal evaluation, for the possibility of pheochromocytoma and acute adrenal crisis, preoperative preventive measures should be taking [7]. In this case; we applied conservative treatment to the patient. Although upon conservative therapy the clinical condition improved and no progression was observed in the size of hematoma on control ultrasound scans, there was a suspected malignancy on her MRI imaging. For this reason, we explored the patient and performed right adrenalectomy. Her pathology result revealed hematoma and adrenal tissue without any tumor cells.

In conclusion, in patients presenting with hemorrhagic shock and abdominal pain, adrenal hemorrhage should be kept in mind as a differential diagnosis. If the endocrinologic work-up or imaging modalities have a suspicion for the adrenal malignancy, histopathological examination is necessary to determine the nature of these lesions. Management of AH depends on the extent of injury, the viability of residual adrenal tissue, the status of the contra lateral adrenal gland, and stability of the patient. If there is no indication for abdominal exploration, non-operative management which includes supportive care, measurement of serial hematocrits, and administration of blood transfusion as needed, is recommended. Otherwise angioembolization of the adrenal gland or laparotomy should be considered.

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