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A Rare Cause of Retroperitoneal Hemorrhage: Spontaneous Rupture of a Giant Adrenal Myelolipoma

Retroperitoneal Kanamanın Nadir Bir Nedeni: Dev Adrenal Myelolipomun Spontan Rüptürü

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Abstract

Adrenal myelolipoma (AM), with its unique histological appearance, is a rarely encountered benign tumor. It is mostly asymptomatic and is incidentally detected. Rarely, in some large masses, retroperitoneal hemorrhage, which could cause life-threatening shock due to spontaneous rupture of AM may develop. In this case report, we presented a 60-year-old male patient who was admitted with abdominal pain and rapid decline in hemoglobin values. Computed tomography (CT) showed bleeding into the retroperitoneal space secondary to the rupture of myelolipoma measuring 14-cm in diameter in the right adrenal gland. Emergency exploratory laparotomy was performed, and the mass was excised. The possibility of rupture and bleeding of large adrenal myelolipomas should be taken into consideration and accordingly, follow-up of asymptomatic patients should be done carefully.

Keywords: adrenal myelolipoma, retroperitoneal hemorrhage, spontaneous rupture

Öz

Adrenal miyelolipom, kendine özgü histolojik görünüm ile karakterize nadir görülen iyi huylu bir tümördür. Çoğunlukla asemptomatiktir ve insidental olarak saptanır. Nadiren bazı büyük boyutlu kitlelerde spontan rüptüre bağlı yaşamı tehdit eden şok tablosuna yol açan retroperitoneal kanama gelişebilir. Biz, karın ağrısı şikayetiyle gelen ve hemoglobin değerlerinde hızlı bir düşüş izlenen 60 yaşındaki erkek hastayı sunduk. Bilgisayarlı tomografide (BT) sağ adrenal bezde izlenen 14 cm lik miyelolipomun rüptüre olmasına sekonder retroperitoneal alan içerisine kanadığı görüldü. Acil eksploratif laparotomi yapıldı ve kitle eksize edildi. Olgumuzda da görüldüğü gibi büyük boyutlu adrenal myelolipomların spontan rüptüre bağlı kanama ihtimali göz önünde bulundurulmalı ve buna bağlı olarak asemptomatik hastaların takibi dikkatli bir şekilde yapılmalıdır.

Anahtar kelimeler: adrenal myelolipom, retroperitoneal kanama, spontan rüptür

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Introduction

Although spontaneous retroperitoneal hematomas have a low incidence, the morbidity and mortality of the condition is high. Acute abdominal pain is a common presenting complaint in patients visiting the Emergency Department (ED). Spontaneous retroperitoneal hematoma is mostly of renal origin (rupture of parenchymal lesions such as angiomyolipomas, cysts and kidney carcinomas) [1]. Acute adrenal hemorrhage is rare but may have devastating outcomes. Main causes of acute adrenal bleedings are trauma and ruptured neoplasms. Adrenal myelolipoma is a benign tumor that contains mature fat cells and relatively young hematopoietic elements. Rarely, spontaneous rupture leading to retroperitoneal hemorrhage develops in some large lesions.

Herein, we report a patient with a giant adrenal myelolipoma that presented with retroperitoneal hemorrhage and underwent emergency surgery.

Case

A 60-year-old male patient was admitted to the ED of our hospital with a complaint of acute abdominal pain with sudden onset, and rapid progression. Physical examination of the patient revealed mild tenderness on the right upper quadrant of the abdomen. No abdominal rigidity or rebound were noted during palpation. His blood pressure was 104/62 mmHg, and the heart rate was 96 bpm. He had flatulence and defecation within the last twelve hours. No history of trauma was described. The patient had a history of coronary artery bypass surgery 3 years ago and was using clopidogrel. At admission, baseline hemoglobin (9.2 g/dl), white blood cell count (11400/mm^3) and the creatinine (2.2 mg/dl) values were as indicated, while 3 days previously his hemoglobin value was 15 g/dl. In the abdominal CT performed in the ED, a solid mass with a size of 14 cm in diameter in the right adrenal gland, with areas of fat density and hemorrhagic foci was observed (**Figure 1**). The decision of emergency exploratory laparotomy was made. During the operation, a hematoma occupying the retroperitoneal space was observed. Total blood loss was 3000 ml. Fragmented adrenal tissue required the excision of the adrenal gland (**Figure 2**). After the operation, the patient's hemoglobin level was stabilized, and the patient was discharged on the postoperative 4th day in a stable health state. No blood transfusions were needed during the perioperative and postoperative follow-up of the patient. Histopathological assessment of the biopsy specimen revealed the presence of the myelolipomatous metaplasia, with myeloid cells and adipocytes in the lesion (**Figure 3**).

Discussion

Adrenal myelolipoma is a benign tumor containing hematopoietic and fat cells that are usually non-functional in hormonal evaluation. They are frequently detected incidentally and are the second most common cause of adrenal incidentalomas, preceded by adrenal adenomas (6-16%) [2]. They are usually diagnosed after the fifth decade of life. They are observed equally in men and women. They can be present in one or both adrenal glands. Myelolipomas may also be seen in extraadrenal

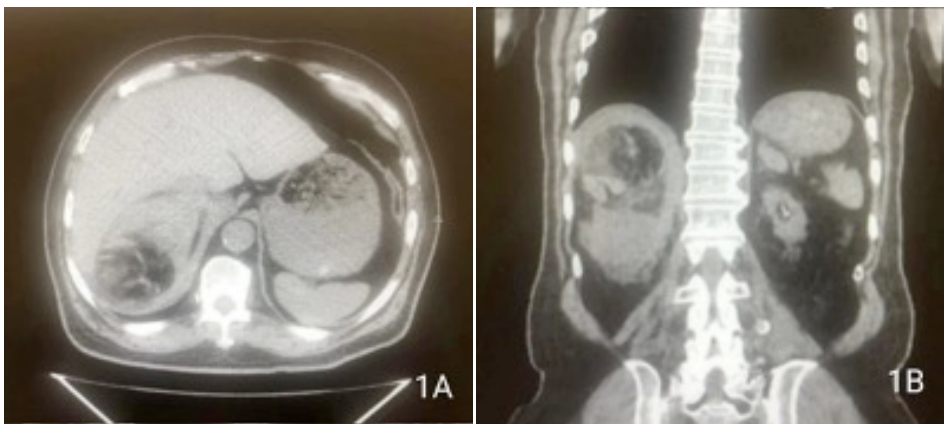

Figure 1. 1AB: Computed tomography images of the 14 cm right adrenal mass and the retroperitoneal hemorrhage

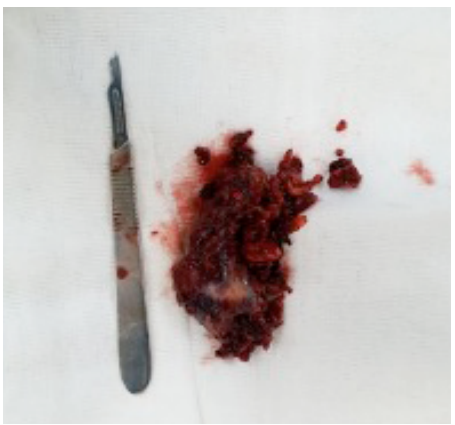

Figure 2. Macroscopic view of the excised right adrenal gland

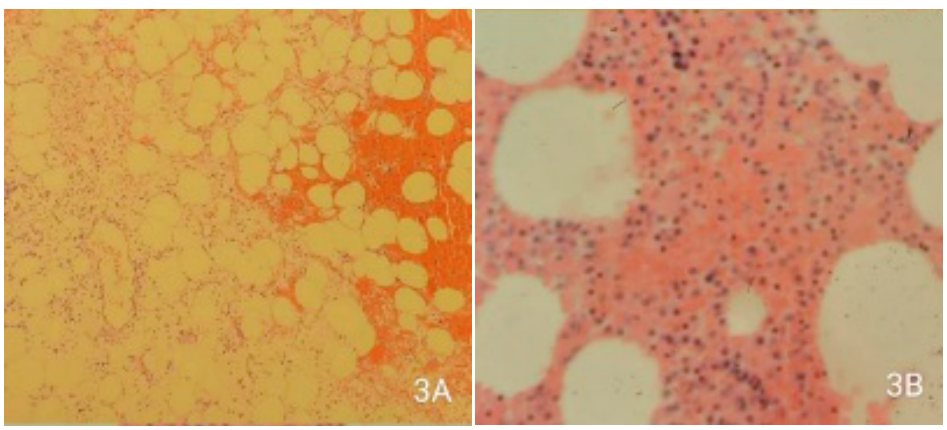

Figure 3. AB: Microscopic examination of the surgical specimen, showing adipocytes and myeloid cells

localizations such as thoracic, pelvic, retroperitoneal, renal, and hepatic regions.

Myelolipomas are usually asymptomatic. However, in some patients, symptoms such as abdominal pain, hypochondriac pain, abdominal mass, back pain, fever, weight loss, shortness of breath and endocrine disorders may be seen [2]. These patients usually present with sudden onset of severe abdominal pain, nausea, and vomiting. Unless promptly intervened, the condition progresses to life-threatening shock due to hypotension.

In diagnosis of AM, imaging methods such as ultrasonography, CT, magnetic resonance imaging (MRI) are used [3]. The most sensitive diagnostic imaging method is CT. Lesions are seen on CT as contrast-enhancing, hypodense, well-circumscribed, heterogeneous masses with an attenuation value ranging from −20 to −120 HU depending on their myeloid and adipose tissue content [4]. However, it may be difficult to distinguish the lesions from the surrounding retroperitoneal adipose tissue due to the abundant fat content of some masses.

The clinical condition of the patient and the size of the lesion should be taken into consideration in the management of AMs. Asymptomatic lesions smaller than 10 cm in diameter should be followed up with imaging methods for 1 or 2 years. If symptoms occur, surgery is recommended. Asymptomatic masses bigger than 10 cm in diameter should be surgically excised due to the possibility of life-threatening retroperitoneal bleeding in case of spontaneous rupture, as in the case presented [5]. In cases with smaller mass lesions, minimal invasive or endoscopic procedures can be applied. Transabdominal, lumbar, subcostal, or posterior approach may be preferred for surgical excision [2]. After adrenalectomy, myelolipoma may also develop in the contralateral adrenal gland [5]. Therefore, patients should be followed up regularly in the postoperative period.

Conclusion

Spontaneous retroperitoneal hemorrhage is a considerably rarely seen life-threatening condition. Case series and retrospective cohort studies account for most of the literature on the subject. Spontaneous retroperitoneal hematoma is most common in the elderly patients, who are receiving anticoagulation treatment, or patients with underlying coagulopathy [6]. Similar to the literature, our patient was 60 years old and was receiving anticoagulant therapy. This uncommon adrenal gland neoplasm should be considered in patients presenting to ED with unexplained abdominal pain and receiving anticoagulant treatment while keeping its unique presentation in mind.

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