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Case Report / Приказ болесника

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**A rare case of spontaneous rupture of renal artery pseudoaneurysm in a
previously hypertensive patient**

Редак случај спонтане руптуре псеудоанеуризме бубрежне артерије
код претходно хипертензивног болесника

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A rare case of spontaneous rupture of renal artery pseudoaneurysm in a previously hypertensive patient

Редак случај спонтане руптуре псеудоанеуризме бубрежне артерије код претходно хипертензивног болесника

SUMMARY

Introduction The renal artery and segmental renal artery pseudoaneurysm is a rare and usually asymptomatic vascular lesion which in most of the cases thrombose spontaneously, but at same time it can be a source of life-threatening hemorrhage and shock. Today, these pseudoaneurysms are discovered with increasing frequency due to unrelated abdominal imaging or on screening work-ups for hypertension, as well as widespread use of angiography. Typically, they are seen in patients after trauma, inflammation, or renal surgery or biopsy.

Case outline In our case, a 52-year-old male patient with no prior history of surgery, significant abdominal trauma and systemic disease, presented with left flank pain and signs of hypovolemic shock that manifested before the admission in the surgical emergency room. The CT scan promptly demonstrated rupture of large retroperitoneal hematoma with the massive intraperitoneal hemorrhage. The angiography confirmed the rupture of the renal artery pseudoaneurysm. The patient had the urgent operation. A life-saving nephrectomy was performed while intraperitoneal hemorrhage and retroperitoneal hematoma was evacuated. The fourteen days after surgery the patient was discharged fully recovered, with normal diuresis and serum levels of creatinine and urea within referential values. During the period of hospitalization, he was diagnosed and treated hypertension.

Conclusion Rupture of pseudoaneurysms with the following hemorrhage into the intraperitoneal cavity and retroperitoneum is a life-threatening condition, as proven with this case in which hypovolemic shock manifested before the admission. We would like to highlight the importance of high blood-pressure control and the importance of regular check-ups.

Keywords: renal artery pseudoaneurysm; rupture; life-threatening procedure

САЖЕТАК

Увод Псеудоанеуризма бубрежне артерије или њених сегментних грана је ретка и обично асимптоматска васкуларна лезија, која у већини случајева спонтано тромбозира. Истовремено, она може руптурирати и довести до животно угрожавајућег крварења и стања хиповолемичног шока. Данас се ове псеудоанеуризме откривају задесно у склопу широко распрострањене СТ ангиографске дијагностике неког другог абдоминалног патолошког стања или током скрининга на хипертензију. Типично се примећују код пацијената након трауме, упалних процеса или бубрежне операције или биопсије.

Приказ болесника У нашем случају, 52-годишњи болесник мушког пола без претходне историје хирургије, значајне трауме стомака и системске болести, примљен је као хитан случај са јаким боловима у левој половини трбуха и знаковима хиповолемичног шока. Хитна комјутеризована томографија је показала руптуру великог ретроперитонеалног хематома са масивним интраперитонеалним крварењем. СТ ангиографија је потврдила руптуру псеудоанеуризме бубрежне артерије која је дисецирала према доњем полу левог бубрега. Болесник је хитно оперисан, када је урађена лева нефректомija уз евакуацију велике количине слободне свеже крви и коагулума интраперитонеално, као и ретроперитонеалног хематома. Четрнаест дана након операције болесник је отпуштен потпуно опорављен, са нормалном диурезом и нивоом креатинина и урее у серуму унутар референтних вредности. Током периода хоспитализације дијагностикована му је хипертензија, која је медикаментозно искоригована.

Закључак Руптура реналне псеудоанеуризме са крварењем у интраперитонеалну дупљу и ретроперитонеум је животно угрожавајуће стање, што је показано у нашем случају, у којем се хиповолемички шок манифестовао приликом пријема. Посебно истичемо важност контроле високог крвног притиска и медикаментозног третмана оваквих пацијената.

Кључне речи: псеудоанеуризма бубрежне артерије; руптура; крварење

INTRODUCTION

Aneurysmal vascular lesions represent anomalous dilatations of the blood vessel lumen. Following the pathological condition of its wall, these lesions are classified into true aneurysms and pseudoaneurysms. A true aneurysm signifies a circumscribed dilatation of an artery with the preservation of all three wall layers - intima, media and adventitia. In 90% of cases, they are extraparenchymal. [1, 2] In contrast, a pseudoaneurysm arises from the disruption of arterial wall continuity and represents a perfused hematoma contained solely by the adventitia and perivascular tissues. These tissues usually wield sufficient compressive force to decrease the bleeding from the site of the lesion, allowing reactive fibrosis to occur and encapsulate the hemorrhage. [3–6] It may involve both the extraparenchymal or intraparenchymal renal artery as well as its branches. [7, 8]

While it is a rare and usually asymptomatic vascular lesion which in most of the cases thrombose spontaneously, renal artery pseudoaneurysm can be a source of life-threatening hemorrhage and shock. [9,10] Spontaneous pseudoaneurysm of segmental renal artery is also a rare entity. Today, these pseudoaneurysms are discovered with increasing frequency due to unrelated abdominal imaging or on screening work-ups for hypertension, as well as widespread use of angiography. [11] Typically, they are seen in patients after trauma, inflammation, or renal surgery or biopsy [12, 13].

Here, we report a rare case of rupture of spontaneous pseudoaneurysm from a segmental renal artery branch, presenting itself with extensive intra and retroperitoneal hemorrhage in a patient with previously uncontrolled high blood pressure.

CASE REPORT

A 52-year-old male patient was admitted to surgical emergency room of our Institution in a serious condition, somnolent to soporous, hemodynamically unstable with extreme hypotension 60/40mmHg, tachycardic with heart rate around 160bpm, tachypneic with breath rate 18/min, pale and drenched in sweat. The abdomen was diffusely tender, guarded and distended, suggestive of an acute surgical condition. Heteroanamnestic data obtained from the

ambulance physician suggested that the patient had gross hematuria immediately after a sudden onset of severe left flank pain during an intense physical activity. Further data were not available. A Foley catheter was inserted and around 500ml of bloody urine was obtained, along with small blood clots. Due to the high suspicion of internal bleeding, the patient was sent for an emergency CT scan, which was performed without and after intravenous injection of contrast medium. Meantime, blood analysis results revealed hemoglobin at 7.2 g/dl (14-17.5), creatinine at 1.5 mg/dl (0.7-1.2) and urea at 49.8 mg/dl (12.8-42.8). CT finding in the arterial phase indicated tortuous and up to 18.8mm dilated left renal artery with active contrast extravasation into the left retroperitoneal space from its inferior segmental branch and the intraperitoneally large amount of free fluid of blood consistency. (Figure 1).

The CT scan also demonstrated rupture of large retroperitoneal hematoma with the massive intraperitoneal haemorrhage. Left kidney was pushed cranially towards the spleen by a massive hematoma that occupied the entire left retroperitoneal space, measuring almost 220mm in length, and propagated perirenally, pararenally and partially to the contralateral side (Figure 2).

A giant cyst of the lower kidney pole with diameter up to 80mm was also reported. The pyelon, left ureter and urin bladder were completely filled with blood. Active bleeding was detected at the time of the angiogram (Figure 3).

The patient was rushed to the operating room, where a life-saving nephrectomy was performed while intraperitoneal haemorrhage and retroperitoneal hematoma was evacuated. The estimated blood loss was around 2500ml. 9 units of blood and 10 units of blood plasma, cryoprecipitate and platelets were administered intraoperatively, each. Later, examination of the removed kidney verified the destruction of its lower pole, while the rest was not pathologically altered (Figure 4).

The postoperative course was uneventful and after fourteen days the patient was discharged fully recovered, with normal diuresis and serum levels of creatinine and urea within referential values. During the period of hospitalization, he was diagnosed with hypertension.

Written consent to publish all shown material was obtained from the patient.

DISCUSSION

Renal artery aneurysms including pseudoaneurysms, represent localized anomalous dilations of the renal artery or its branches. Once thought rare, today are discovered with increasing frequency due to unrelated abdominal imaging or on screening work-ups for hypertension, as well as widespread use of angiography. Overall incidence ranges between 0.01% and 1%, and increases up to 2.5% in hypertensive patients, and can be as high as 39% in patients with hypertension unresponsive to therapy. The average age at diagnosis is between 40 and 60 years. They occur more frequently in men, and are primarily located on the right side. They can be congenital or acquired. Congenital aneurysms are associated with autosomal dominant disorders, such as polycystic disease. Acquired etiologies include long-standing and untreated hypertension, atherosclerosis, trauma, inflammation, renal surgical manipulation (open, laparoscopic or endovascular), malignancy, irradiation, coagulopathy etc. [10–14]. Clear pathophysiology is yet uncertain, but their development is reported to be related to atherosclerosis and fibromuscular dysplasia in 60% of the cases, and to renal arterial hypertension in 25% of the cases [15, 16].

Renal artery pseudoaneurysm (RAP) arises from the disruption of renal artery wall continuity. At first, a combination of hypotension, coagulation and sufficient compressive force exerted by the surrounding tissue, such as adventitia, renal parenchyma and Gerota's fascia, results in decrease and cessation of bleeding. Later, dissolution of the blood clot results in restoration of a normal blood flow and communication between the intravascular and extravascular space, leading to formation of a pseudoaneurysm. In time, pseudoaneurysm can grow in size and eventually become unstable, susceptible to rupture [17].

Signs and symptoms may include hematuria, anemia, flank pain or abdominal tenderness, pulsatile abdominal mass and shock. They may develop immediately after the lesion occurred or may be delayed, as reported by several studies [17, 18]. Hematuria is the most common symptom which results from the erosion into the adjacent renal collecting system [18]. However, patients with RAP may present with nonspecific symptoms, may be completely asymptomatic, or may not have any medical history related to RAP, thus making diagnosis challenging due to potential lack of suspicion from physicians.

Bearing in mind all of the above, and in the case when patient presents with one or several complications, physicians need to be aware of them and to act quickly, given a high mortality rate in case of rupture, which is up to 80% [17, 18, 19].

Diagnosis of RAP is primary radiologic, either if discovered incidentally or due to suspicion in the presence of a complication. Doppler ultrasound may indicate the existence of aneurysm or pseudoaneurysm, as well as the active hemorrhage and existing hematoma. Contrast CT scan can confirm the aneurysmal dilatation and define its anatomic details, and can also demonstrate active extravasations of contrast in case of bleeding and quantify the retroperitoneal hematoma. Angiography is the imaging modality of choice [20].

In our case, a male patient with no prior history of surgery, significant abdominal trauma and systemic disease, presented with left flank pain and signs of hypovolemic shock that manifested before the admission in the surgical emergency room. The CT scan promptly demonstrated rupture of large retroperitoneal hematoma with the massive intraperitoneal haemorrhage. The angiography confirmed the rupture of the renal artery pseudoaneurysm.

Methods of managing RAP are also a challenging issue. A few modalities have been exploited so far. Depending on the patient's clinical condition RAP can be treated by nephrectomy, open vascular surgery, endovascular treatment or angiographic embolization [21, 22, 23]. The urgent surgical indications include overt ruptures, existing renal damage, expansion of the aneurysm and renovascular hypertension. It is suggested that angiographic embolization is the procedure of choice for RAP management due to its minimally invasive and selective nature along with maximal preservation of renal parenchyma, surgery remains a very important treatment in case of a RAP, particularly in the presence of hypovolemic shock. [24, 25]. In our case, a left nephrectomy was performed due to the urgency of the entire procedure, large blood loss and a large destroyed area of the renal parenchyma. Prior to performing the nephrectomy, we had to evacuate large retroperitoneal and intraperitoneal hematomas with vascular control of the abdominal aorta. In this situation, it was out of the question to do some minor invasive procedures, so left nephrectomy was the surgery of choice in our case.

Even though renal artery pseudoaneurysm is a rare entity, especially in the case of the above, physicians may come across it during their work. Rupture of pseudoaneurysms with the

following hemorrhage into the intraperitoneal cavity and retroperitoneum is a life-threatening condition, as proven with this case in which hypovolemic shock manifested before the admission. Therefore, we would like to highlight the importance of high blood-pressure control and the importance of regular check-ups.

Conflict of interest: None declared.

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Figure 1. Large retroperitoneal hematoma on the left (axial CT section) involving both the perirenal and pararenal space; aneurysmal dilatation of the left renal artery up to 18.8 mm in diameter



Figure 2. Large hematoma of the retroperitoneum on the left (coronal CT section) with the largest intercommissural diameter up to 213 mm



Figure 3. Virtual reality 3D imaging angiographic view of the aorta; there is a large fusiform aneurysmal dilatation of the terminal part of the left renal artery (after separation of a separate branch for the lower part of the kidney) with rupture of the lower branch of one of the middle interlobar arteries with arrow marking the site of contrast extravasation into a hematoma around the left kidney



Figure 4. The destruction of the lower pole of the left kidney, as a source of massive bleeding