Spontaneous rupture of the kidney in the patients with synchronous renal hemangioma and nephrogenic hypertension

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ABSTRACT

Most renal neoplasms in adults are epithelial in origin and mesenchymal tumors are rarely encountered. Vascular tumors and tumor-like lesions account for a very small subset. Hemangioma of the kidney is a rarely seen benign vascular neoplasm that probably arises from angioblastic cells. Its general sign is macroscopic hematuria with or without pain. Preoperative diagnosis is difficult or impossible. Previously, spontaneous rupture of the kidney caused by renal hemangioma was not reported in the English literature. In this study, two cases with a history of nephrogenic hypertension who presented with spontaneous renal rupture are presented. There wasn't any trauma history in the background of our patients. A long-standing nephrogenic hypertension was present in both patients. Patients underwent radical nephrectomy due to rupture of the renal tumor. In histopathological examination, capillary hemangioma was detected in the renal medulla in both cases. Patients didn't need antihypertensive therapy during the postoperative period.

Keywords: Nephrogenic hypertension; renal hemangioma; spontaneous rupture of the kidney.

Introduction

Hemangioma is a common benign vascular neoplasia. A substantial number of tumors encountered in the pediatric age group are hemangiomas. Although their general localization is subcutaneous tissue, they can be seen in the muscles and visceral organs. Liver is most frequent localization in one third of the patients with visceral involvement. In the kidney they more frequent as a component of angiomyolipomas, and pure hemangiomas are less frequent.^[1,2] In addition to kidney, they can be seen in the bladder, urethra in the urinary system. Though they are generally asymptomatic, the most frequent symptom is recurrent hematuria.^[2] In this study, two cases of renal hemangioma with a history of nephrogenic hypertension who presented by spontaneous renal rupture are presented. Any case presented by a similar clinical image has not been reported in the English literature.

Case presentations

Case 1

A 36-year- male patient consulted to the clinic with a deteriorated general health state and

with complaints of severe pain on the left side of his loin, nausea, and vomiting. His personal medical history revealed hypertension lasting for 16 years, and class 3rd obesity (body mass index >45 kg/m²). Systolic (SBP), and diastolic blood pressures (DBP) were detected as 220-240 mmHg, and 140-160 mmHg, respectively. On ultrasound (US) a hyperechoic mass with a homogenous appearance measuring 96x86 mm and involving upper half of the left kidney was observed. Deformation of the upper, and middle calyces, and pelvis caused by the mass lesion was detected. On intravenous urograms, functions of the right kidney were evaluated as normal, and excretion of radiopaque agent from the left kidney was not observed. On computed tomography (CT), on the left kidney a mass lesion, and subcapsular hematoma with the same dimensions were detected (Figure 1). Hemogram, and biochemical test results were as follows: hemoglobin 122 g/L, hematocrit 38%, creatinine 1.17 mg/dL, erythrocyte sedimentation rate 18 mm/hr. Based on radiological findings, the patient was diagnosed as rupture of the renal mass. The patient, and his relatives were informed about the procedure, written, and undersigned informed consent of the patient was obtained, and unilateral trans-

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Figure 1. A 36-year-old male patient. On CT, rupture of the left renal tumor, and pararenal blood clots are seen

Figure 2. A 46-year-old male patient. On CT, rupture of the left kidney tumor, and abundant pararenal blood clots, tissue edema, and fibrotic changes are seen

peritoneal left radical nephrectomy with Chevron incision was performed. During exploration, spread of hematoma into perinephric region, and the wall of the descending colon was seen. During dissection colonic serosa was inadvertently opened, and repaired with vicryl sutures. A small amount of retained fluid was seen in the peritoneal cavity. During postoperative period he did not receive any antihypertensive treatment, and SBP, and DBP were measured as 130-150 mmHg, and 80-90 mmHg, respectively. On pathological examination, mostly hemorrhagic a dark brown-coloured mass with irregular contours, extending from the upper pole of the kidney up to renal hilus measuring about 80 mm, subcapsular rupture of the mass, and subcapsular hematoma were seen. Microscopically, numerous foci of capillary vascular proliferation in association with diffuse bleeding in the renal medulla were detected. Atypia, mitotic activity, and necrosis were not encountered in endothelial cells, and perivascular connective tissue. Based on histopathological findings, the case was reported as renal capillary hemangioma. The patient was discharged on the postoperative 5th day. During 12 months of the follow-up period any complication was not seen.

At 3-month controls SBP, and DBP were determined as 130-160 mmHg, and 80-100 mmHg, respectively.

Case 2

A 46-year-old male patient consulted to our clinic with complaints of right lumbar pain, headache, and lassitude. He was suffering from hypertension which was refractory to antihypertensive treatment for 20 years. SBP, and DBP were 200-220 mmHg, and 120-140 mmHg, respectively. Fifteen days previously he felt sudden pain on the right side of his lumbar region with concomitant hypotension. It was emphasized that following bed rest, intramuscular caffeine, and cordiamine injections, his blood pressure returned to normal levels. On CT, in the middle part of the kidney a heterogenous mass with irregular contours measuring 93x65 mm, and abundant amount of blood clots in the subcapsular region were seen. Tissue edema, and fibrotic changes were detected in the pararenal region (Figure 2). In addition, 100 mL pleural effusion was detected on the right side. Since amount of the effusion was neglectible, we didn't intervene. Hemogram, and biochemical test results were as follows: hemoglobin 114 g/L; hematocrit, 31%; creatinine, 1.36 mg/dL, and erythrocyte sedimentation rate, 45 mm/hr. Glomerular filtration rate was within normal limits. Before the procedure to be performed, written, and undersigned informed consents of the patient, and his relatives were obtained. The patient underwent right radical nephrectomy because of right renal rupture. The kidney was excised together with adrenal gland, and perinephric tissues. During postoperative period, manifestations of hypertension were relieved without treatment (SBP 120-140 mmHg, and DBP 70-90 mmHg). Macroscopically, widespread bleeding in the paranephric tissue, subcapsular hematoma, and a ruptured tumor measuring 90x60 mm within the blood clots were detected. On histopathological examination hemangioma consisting of capillary vessels in the medulla was noted. Malignancy was not detected. The patient was discharged at postoperative 5th day, and any complication was not seen during 12 months of follow-up period. At 3-month-controls SBP, and DBP were determined as 120-130 mmHg, and 80-90 mmHg, respectively.

Discussion

Renal hemangiomas are localized submucosally in the renal papilla, and medulla in 90% of the cases.^[3] Rarely hemangiomas arising from cortex, and renal capsula can be seen.^[4] Mostly they are solitary, and unilateral. Only 12% of them can be multiple. In addition to sporadic lesions, renal hemangiomas can be seen as components of Sturge-Weber, Klippel-Trenaunay syndromes, and systemic angiomas.^[2] They are more prevalent in young, and middle-aged people, and they are encountered even during neonatal period.^[4] Any gender predilection has not been indicated.

Renal hemangiomas are mostly asymptomatic, and they are detected incidentally during autopsies.^[5] The most widespread clinical sign is macrohematuria.^[2,3] In some cases colicky pain can be observed. Renal rupture which was determined in our cases has not been reported in the English literature. In one study, nephrogenic hypertension associated with renal hemangioma was reported in 4 cases with end-stage renal disease, and it was emphasized that it can be a new clinicopathological comorbidity.^[6]

Juxtaglomerular cell tumor is the best known neoplasia that causes secondary hypertension.^[7] Therefore, from the clinical perspective, hemangiomas causing hypertension can be mistakenly evaluated as juxtaglomerular cell tumours. In addition, it has been reported that lesions such as intrarenal neuroblastoma, diffuse renal lymphangiomatosis, renal lymphatic malformation, multicystic nephroma induce secondary hypertension.^[8-11]

Although radiological method is not specific during the diagnostic process, as with other renal neoplasias, it is valuable as a first-step method in the detection of hemangiomas. In the intravenous urography generally signs related to renal hemangioma are not found. However submucous papillary hemangiomas can manifest themselves as calyceal deformation or filling defects.^[3] Angiography can demonstrate renal hemangioma as hypervascular, or avascular lesions or even normal anatomical structures.^[3] US which has an exceptional place in the detection of renal neoplasias, can demonstrate renal hemangiomas as lesions with isoechoic or hyperechoic content with rims without hypoechoic shadow. Blood accumulates in the center of the lesion, and characteristically manifests an anechoic image. When compared with angiomyolipoma, its echogenicity is generally less intense.^[3] In our observation in both cases, hemangiomas were observedly hyperechoic.

On CT, renal hemangioma is seen as an uncapsulated mass lesion with irregular contours protruding towards renal pelvis.^[6] Intravenous injection of radiopaque agent enhances contrast uptake. In our cases, on CT an immense mass with irregular contours whose density increased following intravenous injection of radiopaque agent, and subcapsular hematoma were observed. Preoperative diagnosis of renal hemangiomas is very difficult to make, and generally definitive diagnosis is established with postoperative histopathologic examination. On macroscopic examination renal hemangioma is visualized as an intraparenchymal lesion resembling a blood clot generally with a diameter of 0.2-3.5 cm.^[6] It rarely approaches to giant dimensions.^[12,13] Microscopically, cavernous, and capillary type angiomas are seen more frequently. In recent years, a few case with rarely seen types of hemangiomas, for instance, anastomosing subtype of hemangiomas have been reported.^[14]

Since diagnosis of renal hemangioma is very difficult, and can be made only during exploration, its treatment is generally surgical intervention. However in cases diagnosed following emergence of mild hematuria which does not effect the general status of the patient, can be kept under observance. In cases with severe clinical symptoms, nephrectomy, heminephrectomy, papillectomy or arterial embolization can be performed.^[12] In cases with macroscopic hematuria which considerably effects general health state of the patient or in cases where the possibility of renal carcinoma can not be excluded radical nephrectomy is indicated. In our cases since renal rupture, and subcapsular hematoma were seen, and renal carcinoma could not be rule out, radical nephrectomy was preferred as an appropriate treatment modality.

In conclusion, the preoperative diagnosis of renal hemangioma is impossible in many cases. Radical nephrectomy should be considered in cases with hematuria, or pararenal bleeding-as is seen in our patient-which substantially effects general status of the patient. On the other hand, possibility of chronic nephrogenic hypertension, and renal rupture caused by renal hemangioma should be also kept in mind.

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