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A Rare Cause of Haematuria: Angiomyolipoma

Nadir Bir Hematüri Sebebi: Anjiyomiyolipom

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Abstract

Haematuria, basically means seeing red blood cells in urine, is one of the causes of emergency service admissions and it has a wide range of etiology from urinary tract infection to malignancy. Renal angiomyolipoma, which is common in tuberous sclerosis patients, is one of these etiologies. These masses often manifest themselves with haematuria and can cause significant morbidity. In this paper, we present the case of a twenty-four-year-old female patient who complained of haematuria and left flank pain. She had been followed for tuberous sclerosis for eleven years and we diagnosed her as postpartum spontaneous renal angiomyolipoma rupture accurately, after performing abdominal ultrasonography and computed tomography. Then she recovered fully and discharged after stopping bleeding by transarterial embolization method.

Keywords: Haematuria; Angiomyolipoma; Pregnancy; Emergency.

Öz

En basit tanımıyla idrarda kan hücresi görülmesi demek olan hematüri, basit idrar yolu enfeksiyonundan maligniteye kadar çok geniş etiyolojisi olan bir acil servis başvuru nedenidir. Özellikle, tüberoz skleroz tanılı hastalarda görülen renal anjiyomiyolipom ise bu sebeplerden biridir. Çoğunlukla hematüri ile ortaya çıkan bu kitleler önemli derecede morbiditeye sebep olabilirler. Çalışmamızda acil servisimize hematüri ve sol yan ağrısı şikayetleriyle başvuran ve aynı zamanda on bir yıldır tüberoz skleroz tanısı ile takip edilen yirmi dört yaşındaki bayan hastada, postpartum dönemde görülen spontan renal anjiyomiyolipom rüptürü vakası sunulmuştur. Hastaya ultrasonografi ve bilgisayarlı tomografi vasıtasıyla hızlıca doğru tanı konuldu. Sonrasında ise transarteryel embolizasyon işlemi ile kanama durduralarak hasta başarıyla tedavi edildi.

Anahtar Kelimeler: Hematüri; Anjiyomiyolipom; Gebelik, Acil.

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INTRODUCTION

Hematuria refers to the presence of RBCs in urine detected through microscopic examination. The most common causes for hematuria are urinary tract infections, kidney stones, traumas, nephropathies, prostate cancer, renal cell cancers, bladder induced cancers, benign prostatic hypertrophy, angiomyolipoma (AML), oncocytoma, and surgical complications (1). As can be seen, hematuria has a wide etiology from urinary tract infections to cancer (2).

In this study, we present the case of a tuberous sclerosis (TS) patient who was admitted to our emergency department with hematuria and was diagnosed with renal AML rupture. What makes our patient an extraordinary case is the manifestation of hematuria after birth, which is a rare entity in the literature.

CASE REPORT

A twenty-four-year-old female patient presented in our emergency department with hematuria that had started five hours ago and left flank pain. With no history of trauma or use of anticoagulants or similar drugs, the patient related that the complaints began spontaneously and suddenly. The patient had been followed for TS for 11 years and had given birth by vaginal delivery three months ago. On physical examination, the patient's general condition was moderate with fever at 36,5^{0}C, blood pressure around 90/70mmHg, and heart rate at 95/min. The abdominal examination revealed costovertebral angle tenderness on the left with no rebound or defense. The initial laboratory tests at the emergency room were as follows: haemoglobin: 8,3 g/dL; hematocrit: 25,1%; international normalised ratio (INR): 1,2; creatinine: 1,13 mg/dL; urea 21 mg/dl; and with densely present erythrocytes in the urinalysis. The other laboratory tests were normal. The abdominal ultrasonography (USG) performed in the emergency department showed hyperechoic masses in both kidneys. The computed tomography (CT) revealed many cortical lesions as dense as fat with vague limits in both kidneys (angiomyolipomas). We also observed bleeding in the pelvicaliceal system of the left kidney (Figure 1). With the results of the existing imaging modalities and clinical picture, we diagnosed the patient with TS-related renal AML rupture.

We first monitored the patient for hemodynamics in the emergency room. We followed the course of hematuria after inserting a urinary catheter and started erythrocyte suspension (ES). The patient was referred to the urology department where she was admitted to the urology service for hospital stay. Although she was given ES in the urology service, the haemoglobin values continued to fall. With no planned surgical intervention, the patient was referred to the radiology department. In the bilateral renal angiography performed on the same day by the radiology department, we identified a bleeding locus in the left kidney. This was stopped by administrating transarterial embolization (TAE). In time, the patient was followed in the urology department with decreasing hematuria and improving haemoglobin values. After a five-day-follow-up in the hospital and due to the improvement of the patient's general condition, the patient was discharged and recommended to take AML tests at short intervals.

DISCUSSION

AMLs are benign tumour formations composed of smooth muscles, blood vessels, and fat tissues. Its prevalence is approximately 0.3% while its incidence rate among renal masses is 3% (3). AML has two types; the first and most common type is the isolated AML with an incidence rate of 80% while the other is the TS-related AML. Isolated AML is generally asymptomatic, large, and solitary and occurs later in life. TS-related AML is bilateral, small, and multiple; it is seen at earlier ages and surfaces with bleeding (4). Affected patients often present with hematuria and flank pain while 20% of cases may develop retroperitoneal haemorrhage that might lead to hemorrhagic shock. It is often thought that those greater than 4 cm in size have serious risk of bleeding. Another complication of AML is abdominal pain and blocked urine flow caused by the mass (5).

AML is usually incidentally detected in CT or magnetic resonance imaging (MRI) taken during other investigations (6). The USG view shows dense haemorrhage and hyperechoic masses. Since it is capable of showing the fat tissue within the mass, CT is the gold standard for diagnosis. If CT is contraindicated and the lesions are small and mixed, MRI can be helpful (4). In our case, the initial abdominal ultrasonography showed macroscopic hematuria with renal masses that could not be fully evaluated; yet, the following CT enabled us to diagnose the patient with bilateral and multiple AML. Besides, the fact that the patient had been followed for TS for 11 years supported our diagnosis. Because, the incidence of sporadical renal AML is 1-2% whereas this rate is around 50-75% in TS patients (6).

The increase in intra-abdominal pressure and renal blood flow during pregnancy increases the possibility of spontaneous rupture of renal AML development (7). In addition, oestrogen and progesterone receptors that are known to be present in the muscle tissues at high rates also cause AML's growth and rupture with the increasing stimulating effect of these hormones during pregnancy (3). In our study, our patient had a history of vaginal delivery 3 months ago. Despite our detailed questioning, we failed to

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determine any related history of trauma and drug use and we concluded that the cause of spontaneous AML rupture was the hormonal and physiological changes during the pregnancy and postpartum periods. The literature survey conducted in 2012 showed 26 AML rupture cases during pregnancy while there was only one AML rupture case in the postpartum period (7). Therefore, having developed after the delivery, our patient is a rare case.

Management of AML patients have been much discussed in the literature. The asymptomatic AMLs smaller than 4 cm should be periodically checked and followed every 6 months with ultrasound and CT. It is indicated that bilateral and multiple AMLs with a potential to increase in size over time when they are accompanied by TS should be monitored more closely. But the symptomising bilateral lesions must be intervened with arterial embolization or partial nephrectomy. Patients with retroperitoneal bleeding and deteriorating hemodynamics may need radical nephrectomy (3, 6). In our case, we were able to stop the bleeding in the left kidney by applying a successful TAE process.

CONCLUSION

Consequently, practitioners should keep in mind AML as a rare cause of hematuria in patients presenting at the emergency department. Such patients should be monitored closely and receive a swift diagnosis through abdominal CT. In addition, the TS patients with renal AML should be followed at frequent intervals due to the risk of growth during pregnancy.

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