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An unusual cause of hematuria; primary epiploic appendagitis



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ABSTRACT

INTRODUCTION: Primary epiploic appendagitis (PEA) is self limiting inflammatory disease of colonic epiploic appendices.

PRESENTATION OF CASE: Herein, a 40 years old patient describing abdomino-inguinal pain with clotty hematuria having PEA was presented. At first, the patient was thought to have a primary bladder pathology, but after a metociolus examination, he found to have PEA and managed by conservative measures. **DISCUSSION:** Although PEA does not require surgical intervention, it may mimic other acute abdominal disorders which can be difficult to differentiate. Appendices overlying the sigmoid colon and cecum are more prone to be affected as they are more elongated and wider in size. The patient is usually admitted due to sudden onset of abdominal pain accompanied with fever, abdominal tenderness and leucocytosis. **CONCLUSION:** The present case demonstrated that PEA located close to the lower urinary tract especially urinary bladder might present with urinary symptoms such as hematuria, dysuria, pollakuria and inguinal pain.

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1. Introduction

Epiploic appendices are tiny pouches of fat protruding from the serosa of colon distributed from cecum to rectosigmoid.¹ They are usually arranged in two longitudinal rows along the tinea libera and omentalis supplied by one or two arterioles from the vasa recta of the colon, from cecum till to the distal sigmoid colon, and drained by a single venule. PEA is the acute inflammation of these tiny structures. It is usually caused by torsion, but the exact pathophysiology remains unclear. It is supposed that spontaneous venous thrombosis or torsion followed by hemorrhagic infarction, fatty necrosis, inflammatory reaction and subsequent peritoneal irritation cause the symptoms.² Lastly, the vein which is longer than the artery by virtue of its tortuous course, which makes the pedicle predisposed to twisting.

Given that PEA is a benign and self-limited condition, its recognition is important to clinicians to avoid unnecessary hospitalization, antibiotic therapy, surgical interventions, and overuse of medical

resources.^{4,8} PEA cases are infrequent and may often be missed even after imaging studies.⁶ So far no hematuria in PEA has been reported in the literature. A patient with gross hematuria due to PEA was presented in this report with a short review of the literature.

2. Case

A 40 years old patient weighing 103 kg and 1.77 m tall describing abdomino-inguinal pain with hematuria was referred to urology out patient clinic. The history revealed temporary constipation and gastric complaints without abdominal pain. He had no history of previous surgery. Patient's temperature was 36.7 °C, blood pressure was 141/96 mm/Hg and pulse rate was 75 beats per minute. On abdominal examination he had guarding and mild tenderness in the right iliac fossa. The complete blood count and biochemistry were unremarkable. Urine analysis showed abundant hematuria. Urinary US documented a 13 mm diameter hyperechoic lesion in the bladder wall. Prostate gland had 20 cc volume with regular contours (Fig. 1) A contrast CT detailed mentioned lesion as having peripheral rim-like calcification with irregular mild contrast uptake. The center of the lesion was hypodense. It was located between the

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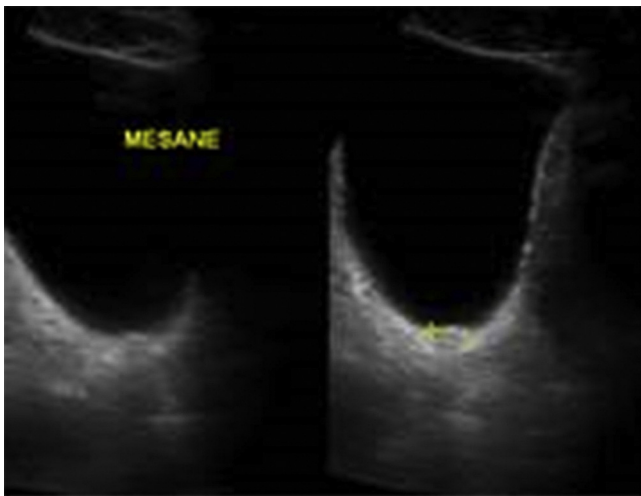


Fig. 1. Hyperechoic 13 mm lesion at the posterior bladder wall not displaced with movement.

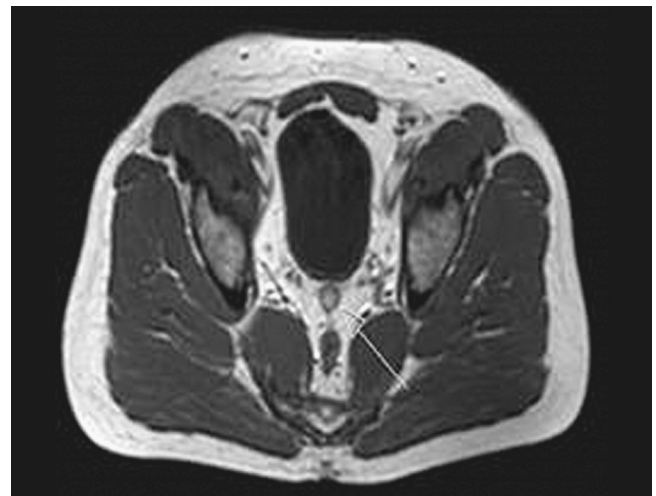


Fig. 3. Axial T1 weighted MR images revealed that central of the lesion has similar intensity to the fatty tissue correlated with epiploic appendagitis.



Fig. 2. (a) A slightly calcified 14 mm lesion between the bladder and rectosigmoid junction. (b) The cystic lesion without penetration of contrast medium from the bladder (cystographic lateral view).

urinary bladder and adjacent rectum causing an indentation on the bladder wall (Fig. 2a and b).

The patient underwent cystoscopy with a suspicion of primary bladder pathology. However, nothing was found but a hyperemic area at the junction of the posterior wall and bladder base (Fig. 3).

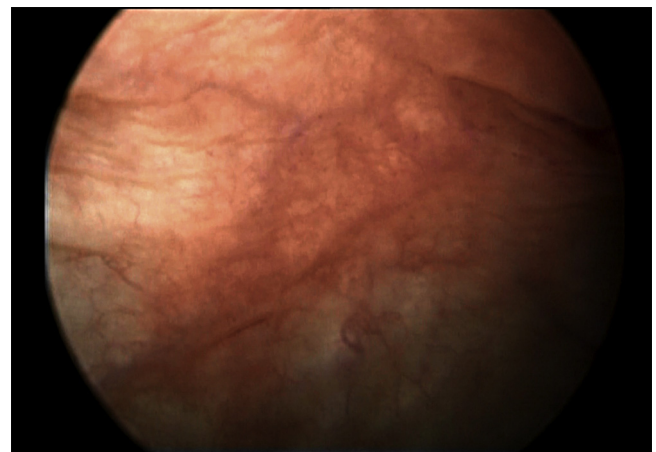


Fig. 4. Image cystoscopy; on the rear wall of the bladder mucosa hyperemic area.

Nevertheless, a punch biopsy of the area was performed. A transrectal biopsy of the aforementioned lesion was also tried but the lesion was not reached and the attempt was failed. The histopathological examination of the bladder biopsy demonstrated chronic cystitis with vascular ectasia, edema and mononuclear inflammatory infiltrate. Afterwards, a pelvic MR has been taken. Axial T1 weighted MR images revealed that center of the lesion has similar intensity with fatty tissue correlating with epiploic appendagitis (Fig. 4). The final diagnosis of PEA was made after retrospective analysis of the patient's all imaging modalities and clinical findings. He responded well to antibiotic treatment and discharged uneventfully in a week. During the follow-up period of 6 months the patient was not experienced any problem related with this disease.

3. Discussion

PEA affects individuals at 2nd to 5th decades of life, with equal distribution between men and women.⁹ Patients may present with localized abdominal pain of variable intensity and duration, rebound tenderness, abdominal mass and mild fever. Nausea, vomiting and loss of appetite are the other less frequently seen symptoms. The pain may be exacerbated by coughing, deep

breathing or stretching because the infarcted appendice is adherent to the parietal peritoneum. The site of pain may vary depending on the location of the appendice involved. Thus the disease can be often mistaken for either diverticulitis or appendicitis.¹¹ The commonest site of epiploic appendagitis is in the sigmoid colon (57%) followed by the cecum (26%).^{2,3}

PEA is a rare cause of localized abdominal pain in otherwise healthy patients. The only clinical feature of PEA can be focal abdominal pain and tenderness, without pathognomonic laboratory findings. Historically, the diagnosis of PEA had been made at diagnostic laparotomy, performed for presumed appendicitis or diverticulitis with complications.^{4,5} Urological signs and symptoms have not been reported yet in PEA cases although it is well known that gastrointestinal diseases such as acute appendicitis may occur with gross hematuria or manifestations mimicking various disorders of the genitourinary tract. Furthermore one should not forget that up to one third of appendicitis cases occur with “urologic” symptoms. The proximity of intestinal structures to the genitourinary tract, mainly the bladder, appears to determine the associated symptoms and signs. To our knowledge, our case is the first one in English literature that urological symptoms associated with PEA has been mentioned.

In PEA signs and symptoms are self limiting and rarely last more than 1 week.^{6,10} Chronic torsion of an epiploic appendice has been reported to be associated with volvulus of a bowel segment with strangulated bowel obstruction.¹² Four deaths relating to the disease have also been reported in literature.¹³ Interestingly, a significant number of patients with disease of epiploic appendices were found to have disorders of fat metabolism in a series.¹⁴

With advancements in radiologic techniques, such as US, CT, or MR, PEA can be distinguished preoperatively due to its characteristic radiologic findings, and it is already being diagnosed increasingly.^{6,7} In CT examinations, the lesions are generally fatty masses which are connected to the serosal surface of the colon and have slightly higher attenuation than peritoneal fat. Most masses have periappendiceal fat stranding, and a few may have a central dot of high attenuation, possibly caused by a thrombosed vessel in the epiploic appendice or by the opposing surfaces of two adjacent appendices.^{5,17,18} In our case, the diagnosis was reached after the elimination of differential diagnosis with several unnecessary invasive measures, and at the end, relied on careful evaluation of the CT and MR findings.

A CT scan was claimed to be pivotal in formulating the principles of the PEA treatment.^{15,16} Following an equivocal CT imaging, a laparoscopic exploration of peritoneal cavity will establish the correct diagnosis and the treatment can be provided during the same procedure.¹⁹ The operation reveals several different findings such as a phlegmon, a gaseous epiploic abscess, an infarcted epiploic appendice or a colonic mass depending on the amount of torsion and/or inflammation.^{20,21} However pathological confirmation of the disease is uncommon and, as most PEA cases are self-limiting, conservative treatment with anti-inflammatory drugs and a moderate to severe pain medication as needed is usually thought to be sufficient.²² As in the present case, our patient's signs and symptoms relieved by simple conservative measures. On the other hand, some authors²³ still recommend laparoscopic surgery to excise the inflamed appendage in most cases in order to prevent recurrence.

In conclusion, the present case demonstrates that PAE located close to the lower urinary tract, especially urinary bladder might present with urinary symptoms such as hematuria, dysuria, polakuria and inguinal pain, and this rare disease mimicking acute abdomen can be diagnosed radiologically and managed conservatively. Dispersion of this information may be useful for physicians regarding the accurate therapeutic approach to adopt, thus avoiding improper procedures and superfluous expenditure in caring for this disease.

Conflict of interest

None.

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None.

Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying image. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contributions

All authors were involved in drafting the article or revising it critically for important intellectual content and all authors approved the final version for publication. Dr. Basri had full access to all of the data in the study and takes responsibility for the integrity of the data and accuracy of the data analysis. Study conception and design: Basri Cakiroglu, Orhun Sinanoglu, Tuncay Tas. Acquisition of data: Akif Nuri Dogan, İlker Abci, Suleyman H. Aksoy. Analysis and interpretation of data: Basri Cakiroglu, Yilmaz Bilsel, Orhun Sinanoglu, Akif Nuri Dogan, İlker Abci.

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