An unusual cause of hematuria: primary epiploic appendagitis

Basti Cakiroglu a,*, Orhun Sinanoglu b, Ilker Abci c, Tuncay Tas d, Akif Nuri Dogan e, Suleyman Hilmi Åksoy f, Yilmaz Bilse c

a Department of Urology, Hisar Intercontinental Hospital, Saray Mahallesi Site Yolu Caddesi No. 7, Umranıye, 34768 Umranıye, İstanbul, Turkey
b Department of Urology, Maltepe University Medical School, 34000 Maltepe, Istanbul, Turkey
c Department of General Surgery, Hisar Intercontinental Hospital, Saray Mahallesi Site Yolu Caddesi No. 7, Umranıye, 34768 Umranıye, Istanbul, Turkey
d Department of Urology, Taksim Training and Research Hospital, Sarıçevher Cadd., 34200 Istanbul, Turkey
e Department of Internal Medicine, Hisar Intercontinental Hospital, Saray Mahallesi Site Yolu Caddesi No. 7, Umranıye, 34768 Umranıye, İstanbul, Turkey
f Department of Radiology, Hisar Intercontinental Hospital, Saray Mahallesi Site Yolu Caddesi No. 7, Umranıye, 34768 Umranıye, İstanbul, Turkey

A R T I C L E   I N F O

Article history:
Received 12 April 2014
Received in revised form 2 July 2014
Accepted 7 September 2014
Available online 28 October 2014

Keywords:
Appendagitis
Epiploic appendice
Hematuria
Dysuria
Diverticulitis

A B S T R A C T

INTRODUCTION: Primary epiploic appendagitis (PEA) is a 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 meticulous 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.

© 2014 The Authors. Published by Elsevier Ltd. on behalf of Surgical Associates Ltd. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/3.0/).

1. Introduction

Epiploic appendices are tiny pouches of fat protruding from the serosa of colon distributed from cecum to rectosigmoid. They are usually arranged in two longitudinal rows along the tinea libera and omentalis supplied by one or two arteries from the vasa recta of the colon, from cecum till to the distal sigmoid colon, and drained by 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. 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. PEA cases are infrequent and may often be missed even after imaging studies. 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

* Corresponding author. Tel.: +90 216 5241300; fax: +90 216 5241223.
E-mail address: drcarsi@gmail.com (B. Cakiroglu).
Fig. 1. Hyperechoic 13 mm lesion at the posterior bladder wall not displaced with movement.

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).

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. 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. 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 appendix is adherent to the parietal peritoneum. The site of pain may vary depending on the location of the appendix involved. Thus the disease can be often mistaken for either diverticulitis or appendicitis. The commonest site of epiploic appendagitis is in the sigmoid colon (57%) followed by the cecum (20%).

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. 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, namely 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. Chronic torsion of an epiploic appendix has been reported to be associated with volvulus of a bowel segment with strangulated bowel obstruction. Four deaths relating to the disease have also been reported in literature. Interestingly, a significant number of patients with disease of epiploic appendices were found to have disorders of fat metabolism in a series.

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. 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 peripendical fat stranding, and a few may have a central dot of high attenuation, possibly caused by a thrombosed vessel in the epiploic appendix or by the opposing surfaces of two adjacent appendices. 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. 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. The operation reveals several different findings such as a phlegmon, a gaseous epiploic abscess, an infarcted epiploic appendix or a colonic mass depending on the amount of torsion and/or inflammation. 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. As in the present case, our patient’s signs and symptoms relieved by simple conservative measures. On the other hand, some authors recommend laparoscopic surgery to excise the inflamed appendix 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, pollakuria 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.

Funding
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.

References
1. Ross JA. Vascular loops in the appendices epiploicae; their anatomy and surgical significance, with a review of the surgical pathology of appendices epiploicae. Br J Surg 1950;37(April 148):464–6.
2. Golash V, Wilton PD. Early laparoscopy as a routine procedure in the management of acute abdominal pain: a review of 1320 patients. Surg Endosc 2005;19:882–5.
3. Thomas J, Rosato F, Patterson L. Epiploic appendagitis. Surg Gynecol Obstet 1974;13823–5.
4. Rao PM, Rhea JT, Wittenberg J, Warshaw AL. Misdiagnosis of primary epiploic appendagitis. Ann J Surg 1998;176:81–5.
5. Rao PM, Wittenberg J, Lawresson J. Primary epiploic appendagitis: evolutionary changes in CT appearance. Radiology 1997;204:713–7.
6. Rioux M, Langis P. Primary epiploic appendagitis: clinical, US, and CT findings in 14 cases. Radiology 1994;191:523–6.
7. Choi YU, Choi PW, Park YH, Kim JL, Heo TG, Park JH et al. Clinical characteristics of primary epiploic appendagitis. J Kor Soc Coloproctol 2011;27:114–21.
8. Legome EL, Belton AL, Murray RE, Rao PM, Novelline RA. Epiploic appendagitis: the emergency department presentation. J Emerg Med 2002;22:9–13.
9. Kantarcı M, Duran C, Sivranci M. Images of interest. Gastrointestinal: epiploic appendagitis. J Gastroenterol Hepatol 2005;20:482.
10. Molè F, Ropellì T, Martinez MJ, Morote V, Roselló-Sastre E. Primary epiploic appendagitis: US and CT findings. Eur Radiol 1998;8(3):435–8.
11. Pereira JM, Sirlin CB, Pinto PS, Jeffrey RB, Stella DL, Casola G. Disproportionate fat stranding: a helpful CT sign in patients with acute abdominal pain. Radiographics 2004;24:703–15.
12. Osdachy A, Shapiro-Feingold M, Zissin R. Strangulated small bowel obstruction related to chronic torsion of an epiploic appendix: CT findings. Br J Radiol 2001;74:1062–4.
13. Shamblin JR, Payne CL, Soilean MK. Infarction of an epiploic appendix. South Med J 1986;79(March (3)):374–5.
14. Effendiev ShM, Volkov OV, Kurbanov MA, Volkov MO, Dzhahalov DM. Diseases of the fatty appendices of the colon. Khiruruga (Mosk) 2003;10:64–6.
15. Sandrasegaran K, Maglente DD, Rajesh A, Aikins FM, Primary epiploic appendagitis: CT diagnosis. Emerg Radiol 2004;11(August (1)):9–14 [Epub 2004 July 6].
16. Singh AK, Gervais DA, Hahn PF, Rhea J, Mueller PR. CT appearance of acute appendagitis. AJR Am J Roentgenol 2004;183(November (5)):1303–7.
17. Oliphant UJ, Rosenthal A. Hematuria: an unusual presentation for mucocele of the appendix. Case report and review of the literature. J Mosk 1999;3(January–March (1)):71–4.
18. Ng KC, Tan CK, Lai SW, Chen DR, Chen WK. Mucocele of the appendix with hematuria. Yale J Biol Med. 2001;74(January–February (1)):9–12.
19. Vázquez-Frias JA, Castaneda P, Valencia S, Cueto J. Laparoscopic diagnosis and treatment of an acute epiploic appendagitis with torsion and necrosis causing acute abdomen. JSLS 2000;4(July-September (3)):247–50.
20. Legome EL, Bolton AL, Murray RE, Rao PM, Novelline RA. Epiploic appendagitis: the emergency department presentation. J Emerg Med 2002;22(January (1)):9–13.
21. Hiller N, Berelowitz D, Hadas-Halpern I. Primary epiploic appendagitis: clinical and radiological manifestations. Isr Med Assoc J 2000;2(December (12)):896–8.
22. Boardman J, Kaplan KJ, Hollcraft C, Bui-Mansfield LT. Torsion of the epiploic appendage. AJR Am J Roentgenol 2003;180(March (3)):748.
23. Sand M, Gelos M, Bechera FG, Sand D, Wiese TH, Steinstraesser L, Mann B. Epiploic appendagitis – clinical characteristics of an uncommon surgical diagnosis. BMC Surg 2007;July (7):11.

Open Access
This article is published Open Access at sciencedirect.com. It is distributed under the IJSCR Supplemental terms and conditions, which permits unrestricted non commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.