Clinical Case Report

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Primary epiploic appendagitis as an unusual cause of acute abdominal pain in a middle-aged male

A case report

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Abstract

Rationale: Primary epiploic appendagitis (PEA) is a rare cause of acute abdomen caused by spontaneous torsion or venous thrombosis of epiploic appendices, it commonly manifests with acute lower quadrant pain, thus may mimic acute diverticulitis, appendicitis, or mesenteric infarction.

Patient concerns: In this case report, we report a 44 years old man who presented with persistent sharp pain in the left lower quadrant abdomen, Laboratory tests were mostly normal, contrast enhanced computed tomography (CECT) revealed a slightly high density shadow with fat foci in the middle was presented around the local descending colon, accompanied by the adjacent peritoneal thickening.

Diagnoses: He was diagnosed with PEA as confirmed by an abdominal contrast enhanced computed tomography (CECT) scan.

Interventions: He was followed up in the clinic without any dietary restrictions, antibiotic or analgesic drugs use.

Outcomes: The abdominal pain gradually subsided a week later, and there were no recurrence of the symptoms during follow-up.

Lessons: In our case, the diagnosis of PEA using CECT allows the patient to avoid surgery and other invasive treatment.

Abbreviations: CDUS = Color Doppler ultrasound, CECT = contrast enhanced computed tomography, CEUS = contrast-enhanced ultrasound, hs-CRP = high-sensitivity C-reactive protein, PEA = primary epiploic appendagitis.

Keywords: acute abdominal pain, Color Doppler ultrasound, contrast enhanced computed tomography, primary epiploic appendagitis

1. Introduction

Epiploic appendices also known as appendices epiploicae, epiploic appendages, appendix epiploica, or omental appendices, are small, mobile pouches of the peritoneum filled with fat and situated parallel to the outer surface of the tenia coli.[1] There are about 50 to 100 epiploic appendices in the adult, extending from the cecum to the sigmoid colon. Each epiploic appendice is supplied by 1 or 2 terminal arteries of the marginal artery of the colon and is drained by a single vein[2,3]; Epiploic appendices are pedunculated and have larger dimensions. Based on these special structures, epiploic appendices are particularly prone to torsion and susceptible to ischemic or hemorrhagic infarction, spontaneous venous thrombosis, and inflammatory reaction.[4,5] This pathological process is known as primary epiploic appendagitis (PEA).

PEA is a rare cause of acute abdomen and lacks of relevant data on the morbidity. Patients with PEA often present with acute onset of pain in the left or right lower quadrant abdomen, thus it can mimic and be mistaken for acute diverticulitis, appendicitis, hernia, and cholecystitis.[5–7] Part of PEA patients are diagnosed during abdominal exploration misdiagnosing of acute appendicitis.[8] Here we report 1 case presented with left lower abdominal quadrant pain which was diagnosed as PEA, and finally avoided surgery and other invasive treatment.

2. Consent

This study was approved by the Ethics Committee of Tongji Hospital, Tongji Medical College, Huazhong University of Science and Technology. Informed written consent was obtained from the patient for publication of this case report and accompanying images.

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3. Case description

A 44 years old man presented to our internal medicine department with persistent sharp pain in the left lower quadrant abdomen that had started 3 days before his presentation. He is a building worker with no significant medical or surgical history. He described his pain as sharp, constant, localized, and exacerbated by sudden movements, the pain was not found to be related with eating and defecation. He denied trauma to the abdomen, fever, chills, nausea, vomiting, dysuria, hematuria, diarrhea, and astriction. Physical examination of the patient was performed, and abdominal examination revealed a non-distended abdomen with tenderness in the left lower quadrant. There was rebound tenderness while no guarding, pulsatile, or palpable mass, or costovertebral angle tenderness. Physical examination was otherwise unremarkable. Laboratory investigation showed total leukocyte count 8100/mm^3, with 71% polymorphonuclear leukocytes; hemoglobin, 14.7 g/dl; platelet count, 1.42×10^5/mm^3; high-sensitivity C-reactive protein(hs-CRP), 12 mg/L; liver function tests, kidney function tests, serum electrolytes, serum lipid, and urinalysis were all within the normal range. He underwent a contrast enhanced computed tomography (CECT) abdomen which revealed a slightly high density shadow with fat foci in the middle was presented around the local descending colon (Fig. 1), accompanied by the adjacent peritoneal thickening (Fig. 2). The CECT suggested PEA. In view of the clinical presentation, laboratory data and the radiological findings, the patient was diagnosed as having PEA. The patient was placed on observation without antibiotic or analgesic drugs. The abdominal pain was gradually remitted a week later.

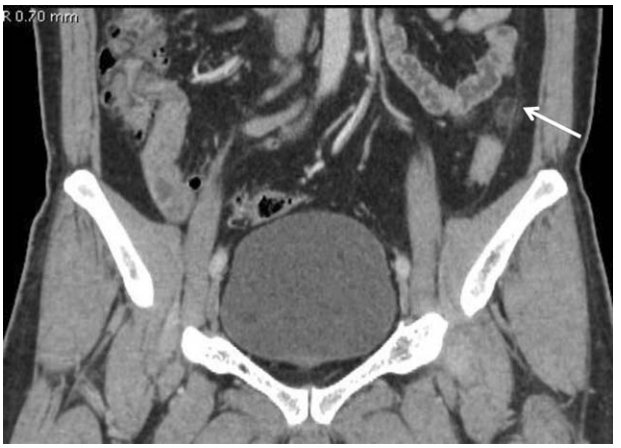

Figure 2. Coronal CT image of the patient. The arrow points to the PEA.

4. Discussion

Epiploic appendagitis is a rare cause of the acute abdomen and first described by Lynn et al in 1956,[9] it can be classified as primary (PEA) and secondary epiploic appendages.[10] PEA is mainly caused by epiploic appendage torsion or spontaneous thrombosis of the epiploic appendage central draining vein resulting in vascular occlusion and focal inflammation[4,5]; while the secondary epiploic appendages are usually generated by torsions secondary to another process, such as diverticulitis, appendicitis, pancreatitis, Crohn ileitis, or surgical adhesions.[11]

PEA can occur at any age, with a peak incidence in the fourth to fifth decades, and is more common in men.[6] Obesity is considered as a risk factor of PEA, and it is reported occurs more common in obese people,[11] especially in those experienced weight loss in a short period of time and who had done strenuous exercise,[7] however, our case and some other reports do not belong to this situation.[6]

PEA has a nonspecific clinical features, while it usually presents as an abrupt onset of focal abdominal pain in the left or right lower abdominal quadrant, it is non-migratory, continuous and intense, and may be worsens by cough or movement.[12] Chen et al reviewed 21 PEA patients diagnosed by computed tomography (CT),[7] they found that abdominal pain was the leading symptom, 17 patients suffered the pain in the left lower quadrant (81.0%), and 2 patients was in the left middle abdomen (9.5%) and right lower quadrant (9.5%), respectively. PEA often affects patients with good overall condition, their appetite and bowel function are usually unchanged; most patients do not have any associated symptoms such as fever, nausea, vomiting,

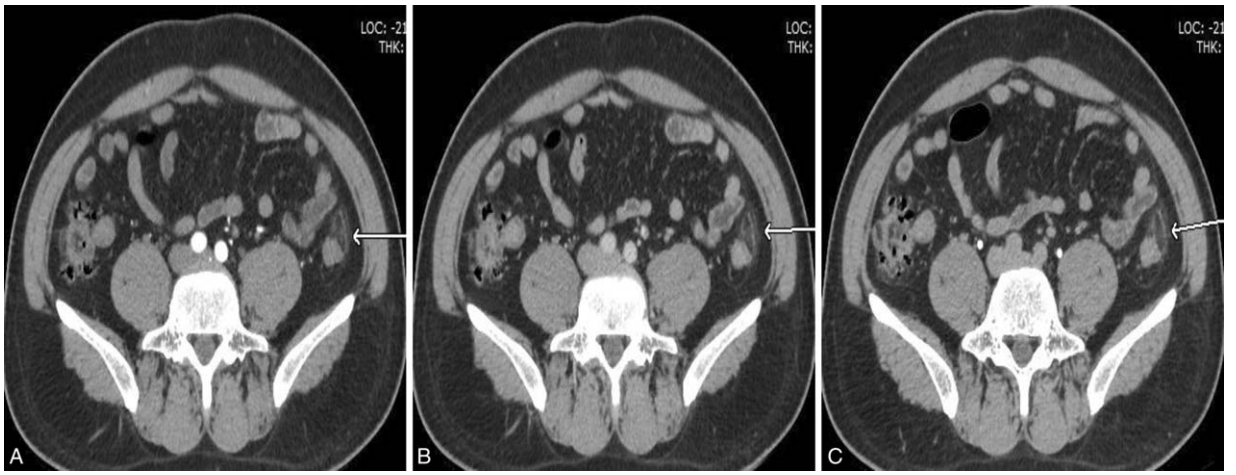

Figure 1. Axial CT images of the patient. A slightly high density shadow with fat foci in the middle beside the local descending colon, accompanied by the adjacent peritoneal thickening. A, B, C stands for the arterial phase, venous phase, and delayed phase, respectively. The arrow points to the PEA.

diarrhea, or constipation. Choi et al retrospective analyzed 31 PEA patients,[12] and found that abdominal pain exited in all patients, other symptoms and their occurrence rate were anorexia (9.7%), nausea (12.9%), vomiting (3.2%), diarrhea(6.5%), and constipation (12.9%).

Localized abdominal tenderness and abdominal guarding are usually presented on physical examination. Rebound pain is elicited in 25% of cases and an inflmmatory resistant mass is identified in 10% to 30% patients as reported.[12,13] PEA has not any pathognomonic laboratory finding. The abnormal laboratory results may include slightly elevated leukocytes and serum C-reactive protein in a few cases. Other routine laboratory parameters, such as erythrocyte, liver, and renal function are usually within normal ranges.[5,12]

As the lack of nonspecific presentation and distinctive clinical features, the diagnosis of PEA can be challenging. The right lower PEA is often confused with acute appendicitis; while left sided PEA is often misdiagnosed as sigmoid diverticulitis in clinic. Therefore, diagnosis of PEA was often the result of an unexpected finding during an exploratory laparotomy in the past.[14] Today, however, this condition is usually accurately diagnosed by Color Doppler ultrasound (CDUS) or CT pre-operatively. CDUS examination sometimes shows nonspecific images such as an oval noncompressible hyperechoic mass with a subtle hypoechoic rim with no color Doppler flow under the site of maximum tenderness. And this mass is adherent to the anterior abdominal wall and would not move during the patient's deep breathing.[10,15,16] However, establishing the accurate diagnosis of PEA would be very difficult without prior experience and knowledge of the typical imaging findings about this rare disease. More recently, contrast-enhanced ultrasound (CEUS) further described the characteristic features of PEA as a thick rim of enhancement and a hypoechoic central area of the mass.[17,18] Therefore, CEUS might be helpful to further confirm the diagnosis of PEA in some equivocal cases. Although CDUS has the advantage of correlating the position of the lesion with the location of maximum tenderness, some researchers propose that when CDUS detects an image compatible with PEA, CT should be performed to confirm the fatty nature of this mass.[19] PEA has a predictable appearance in terms of location, size, and density. In a study to describe the CT findings in 50 patients with PEA by Singh et al,[4] the location of the colon lesion in decreasing order of frequency was the sigmoid colon (62%), descending colon (18%), cecum (12%), and ascending colon (8%); in most cases(86%), the size of the fatty central core was between 1.5 and 3.5 cm; morphologically, a fatty central core contiguous the colon wall with inflammatory changes surrounded and a base narrower than the equator was showed on CT images in all cases. The fatty attenuation centers were overt in all patients on soft-tissue window, and a central high-density dot, irregular or linear focus, was presented in more than half the cases; most cases had normal colon wall thickness, and only a small part were thickened (4%).[4] Another study found that in 21.4% cases of PEA, the mass on CT images had a lobulated appearance.[20] Therefore, the lobulated appearance of the inflamed epiploic appendages may aid in differentiating PEA from other diseases, such as omental torsion, and increase the accuracy of diagnosis.[20] Magnetic resonance imaging (MRI) is not a routine diagnostic method for PEA in most hospitals, but MRI has advantage on high tissue resolution compared to CT, and it lacks ionizing radiations, so it should be preferred in the diagnosis of PEA, particularly in pediatric and obstetric patients.[21,22]

PEA has been known as a self-limiting disease with a benign course now. However, in the past, the regular therapy for PEA was surgical excision because most cases were discovered during exploratory operation. Some researchers have reported that pain recurred or persisted until the epiploic appendagitis was removed in a part of their cases, so they strongly recommended excision of PEA when it was diagnosed.[14] Later, with the further understanding of this disease, more and more researchers proposed nonsurgical management of PEA. With conservative management, the symptoms gradually alleviate between 1 weeks and 4 weeks.[12] In a retrospective study of 12 PEA, Ozdemir et al found that 7 patients treated with conservative therapy overcame the symptoms within approximately 3 weeks, and no recurrence was observed during the mean follow-up of 7 weeks.[19] Oral anti-inflammatory medication is usually recommended for 4 to 7 days if abdominal pain is unbearable, and antibiotics are not often indicated.[11] Some small-scale studies suggest that the conservative management is successful for 97% to 100% of cases, and therefore, it is suggested to be performed on an outpatient basis.[10] Antibiotics and surgical treatments (laparotomy or laparoscopic) are rarely warranted, however, surgical intervention can be used for patients whose symptoms fail to improve with conservative management or for complications that cannot be managed nonoperatively, such as inflammation induced adhesions and ileus, intussusceptions, abscess formation, intestinal occlusions, or doubtful cases.[6,19,23]

In conclusion, PEA is a very rare but self-limiting disease, and the symptoms and signs are non-specific, which often results in clinical misdiagnosis; with the improvement of radiological technology, more and more cases have been diagnosed and treated. However, radiologists and clinicians should bear in mind PEA as a cause of the acute abdomen to prevent unnecessary hospitalization and surgery.

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