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Duodenal fistula associated with a peri-appendiceal abscess: A case report $^{\diamond}$



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ABSTRACT

INTRODUCTION: Retroperitoneal abscess is an unusual presentation of perforated appendicitis. A fistula between the duodenum and an abscess resulting from appendicitis has not been previously reported. PRESENTATION OF CASE: A 53-year-old Japanese man with a past medical history of hypertension and iron deficiency anemia presented with a 10-day history of fever and right lower abdominal pain, and was diagnosed with a retroperitoneal abscess secondary to perforated appendicitis. He was then treated with piperacillin and tazobactam after undergoing ultrasound-guided drainage, after which his overall condition improved. Due to iron deficiency anemia, we performed further evaluation for gastrointestinal bleeding and esophagogastroduodenoscopy showed an elevated lesion with granulomatous tissue in the duodenum, without an associated ulcer. At 10 days after abscess drainage, duodenography with contrast showed continuity between the abscess cavity and the duodenum. At 74 days after drainage, we performed laparoscopic appendectomy. Pathological examination showed granulomatous tissue inside the appendix with an inflammatory background and fecaliths infiltrated by macrophages.

DISCUSSION: Perforated appendicitis has various presentations and many unusual fistulae have been reported, however, a fistula between a peri-appendiceal abscess and the duodenum has not yet been reported. A retroperitoneal abscess around the duodenum and appendix should be checked to differentiate it from Valentino's syndrome.

CONCLUSION: We present the rare complication of a duodenal fistula during the treatment of perforated appendicitis. The possibility of fistula formation should be considered in patients with complicated appendicitis.

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1. Introduction

Acute appendicitis is a common disease in all age groups, and typically presents with right lower abdominal pain. However, atypical presentations are well known and may be associated with unusual manifestations, leading to diagnostic delays with an increase in morbidity and mortality.^{1,2} Retroperitoneal abscess is one of the unusual presentations of perforated appendicitis.¹ The optimal management of this condition depends on the patients' clinical condition. If abscess formation is seen on imaging studies,

CT- or ultrasound-guided drainage is often performed as the initial treatment.² Various types of fistulae associated with the appendix have been reported,³ but there are no previous reports of a non-malignant connection between an abscess and the duodenum. We herein present a patient with a duodenal fistula that developed during the treatment of perforated appendicitis presenting with a retroperitoneal abscess.

2. Case presentation

A 53-year-old Japanese man presented with a 10-day history of lower abdominal pain. He reported no diarrhea, weight loss or back pain. He had a past medical history of hypertension and iron deficiency anemia, and his only medication was for iron deficiency anemia. His weight was 81.3 kg and BMI 28.3 kg/m². The remainder of the physical examination revealed a fever of 38.3 °C and right lower abdominal tenderness on palpation. Laboratory data revealed a mild leukocytosis (white blood cell count of $10,400/\text{mm}^3$) and iron deficiency anemia. Computed tomography of the abdomen showed a $7\,\text{cm} \times 5\,\text{cm} \times 8\,\text{cm}$ retroperitoneal

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Fig. 1. Abdominal computed tomography showed a $7 \text{ cm} \times 5 \text{ cm} \times 8 \text{ cm}$ retroperitoneal abscess with high density lesions and compression of adjacent organs including the duodenum and ascending colon.

abscess with high-density lesions and compression of adjacent organs including the duodenum and ascending colon (Fig. 1). This was felt to be consistent with perforated appendicitis, and treatment was begun with intravenous piperacillin and tazobactam. Ultrasound-guided drainage of the abscess was also performed.

After starting treatment, his condition generally improved, and workup regarding the iron deficiency anemia was then undertaken. Colonoscopy showed no abnormalities, but esophagogastroduodenoscopy showed an elevated lesion with granulomatous tissue in the duodenum without ulcer formation. Ten days after drainage was performed, duodenography showed granulomatous tissue with flow of contrast demonstrating a connection between the abscess and the duodenum (Fig. 2). The drain was removed 38 days after placement. At 74 days after drainage, the patient underwent laparoscopic appendectomy. Pathological examination showed granulomatous tissue inside the appendix with an inflammatory background and fecaliths surrounded by macrophages. Repeat EGD showed no abnormality at 90 days after drainage and the iron deficiency anemia resolved spontaneously.

3. Discussion

Perforated appendicitis has many presentations, including an abdominal mass, abscess, peritonitis or, rarely, fistulae. 1,2 Many unusual fistulae have been reported including cutaneous, umbilical, colon, kidney, bladder and aorta but a fistula between a periappendiceal abscess and the duodenum has not yet been reported. 3 Management of fistulae, especially to the duodenum, are challenging because approximately 10 L of fluid from the stomach, bile duct, and pancreas pass through the duodenum each day. A retroperitoneal abscess around the duodenum and appendix should be checked to differentiate it from Valentino's syndrome (a perforated duodenal ulcer presenting as appendicitis). 4

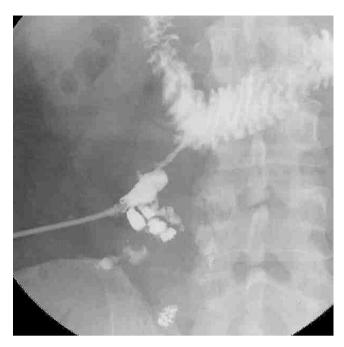


Fig. 2. Duodenography demonstrated continuity between the peri-appendiceal abscess and the duodenum.

In the present case, EGD showed no ulcer formation but did demonstrate granulomatous tissue. Computed tomography also showed that the abscess compressed the adjacent duodenum. In addition, the specimen showed the appendix with inflammatory background and granulomatous tissues, compatible with perforated appendicitis. We usually manage the fistula by administering total parenteral nutrition, and withholding oral intake. However, in this patient an influx from the duodenum into fistula was not seen on the fistulogram and oral intake was continued. Fortunately, this patient had no complaints of abdominal pain, gradually improved and the amount of drainage did not change with oral intake. The fistulogram showed a connection but the tract was small. Computed tomography showed the appendix, no sign of inflammation and no fistulae. We removed the drainage tube on day 38, and the patient fully recovered.

The role of interval appendectomy in this situation is not fully defined.² In consideration of his age and the remaining high-density lesions, we performed an appendectomy. The specimen showed granulomatous tissue inside the appendix with an inflammatory background, compatible with appendicitis

4. Conclusion

In summary, we present here the first case of perforated appendicitis associated with a duodenal fistula. The possibility of fistula formation in patients with complicated appendicitis must be considered.

Conflicts of interest

The authors report that there are no conflicts of interest.

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Ethical approval

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

Author contributions

Kenji Okumura undertook the gathering of information for this case and was a major contributor in writing the manuscript. Tadao Kubota and Alan T. Lefor conceived the manuscript and were a

major contributor to the manuscript. All authors read and approved the final manuscript.

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