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CASE REPORT

Secondary aortoduodenal fistula identified by ultrasonography

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Abstract A 73-year-old man underwent coronary artery bypass grafting, abdominal aortic aneurysm resection, and prosthetic implantation as a single procedure in 2002. His progress was favorable until April 2008, when he was admitted to our hospital with melena. B-mode ultrasonography revealed a 5-mm defect in the abdominal aorta at the graft anastomosis, and an umbilicated lesion was seen projecting between the posterior wall of the third part of the duodenum and the abdominal aorta. A color signal was noticed at this site on color Doppler ultrasonography, leading to the diagnosis of a secondary aortoduodenal fistula (ADF). We resected the inflammatory mass comprising the graft and the third part of the duodenum, and performed prosthetic re-implantation, omentopexy, and duodenojejunostomy. We could not find any previous reports of successful identification of secondary ADF using ultrasonography. When a patient with gastrointestinal hemorrhage following reconstructive aortic surgery is encountered in the emergency department, ultrasonography may be considered to be a useful modality in the diagnosis of secondary ADF.

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Introduction

The prevalence of vascular diseases has recently increased with aging of the population, changes in dietary habits, and an increased prevalence of diabetes mellitus and hyperlipidemia. Aortic grafting is often performed in patients with these risk factors. Aortoduodenal fistula (ADF), a condition in which a direct connection develops between the abdominal aorta and the intestinal lumen, is a rare long-term postoperative complication of abdominal aortic aneurysm (AAA) surgery. Not only is early detection difficult but treatment is also often problematic due to associated graft infection and massive bleeding, leading to a high mortality rate [1]. In this paper, we describe a case of secondary ADF in which abdominal ultrasonography was useful in making the diagnosis, and we present a review of the literature.

Case report

A 73-year-old man with a history of myocardial infarction and AAA underwent coronary artery bypass grafting (CABG), AAA resection, and prosthetic implantation as a single procedure in 2002. His progress was favorable until April 2008, when he developed melena and was admitted to this hospital on the following day with hematemesis and fever (39.0°C). Examination on admission revealed mild hypotension (90/55 mmHg), tachycardia (102 bpm), and mild periumbilical tenderness. Laboratory investigations demonstrated an inflammatory response, with white blood

cell count (WBC) 13.4 × 10^3/μL, erythrocyte sedimentation rate (ESR) 91 mm/h, and C-reactive protein (CRP) 11.4 mg/dL. There was mild anemia with red blood cell count (RBC) 286 × 10^4/μL, hemoglobin (Hb) 9.5 g/dL, and hematocrit value (Ht) 26.6%. Examination as far as the second part of the duodenum by esophagogastroduodenoscopy (EGD) failed to reveal an obvious bleeding source. Contrast-enhanced abdominal computed tomography (CT) scanning revealed a para-aortic area of weak contrast enhancement in the late phase at the level of the graft anastomosis, with an indistinct border with the duodenum, raising suspicion of a fistula between the abdominal aorta and the duodenum associated with infection (Fig. 1). Abdominal ultrasonography (Logic 9, GE Healthcare, Milwaukee, WI, USA) demonstrated a 5-mm defect in the vascular wall on the ventral side of the abdominal aorta at the graft anastomosis, and an umbilicated anechoic lesion, continuous with the aorta. A heterogeneous hypoechoic lesion and calcification were seen projecting between the aorta and the posterior wall of the third part of the duodenum. The duodenal wall adjacent to this lesion was thickened, suggesting bleeding from ADF (Fig. 2a, b). A color signal was seen at this site on color Doppler ultrasonography, confirming the diagnosis of a secondary ADF (Fig. 2c, d). Angiography revealed a pseudoaneurysm (Fig. 3a). Initially, we performed a percutaneous endovascular aneurysm repair (EVAR) to prevent further hemorrhage (Fig. 3b). We then commenced antimicrobial therapy to reduce inflammation of the graft, checked by CT scanning that the stent was fully occluding the fistula, and confirmed disappearance of the hot spot with scintigraphy. We resected the inflammatory mass comprising the infected graft and the third part of the duodenum, and performed prosthetic re-implantation, omentopexy, and duodenojejunostomy (Fig. 4a, b). Elastica van Gieson (EVG) stain showed the aortoduodenal fistula and revealed fibrosis and inflammatory cell invasion of the aortic wall. Development of the duodenal mucous membrane had progressed along the vascular wall around the fistula (Fig. 4c). The patient's postoperative course was uneventful, and abdominal CT findings were also favorable (Fig. 5). He was discharged after a 45-day hospital stay, and follow-up continued on an outpatient basis.

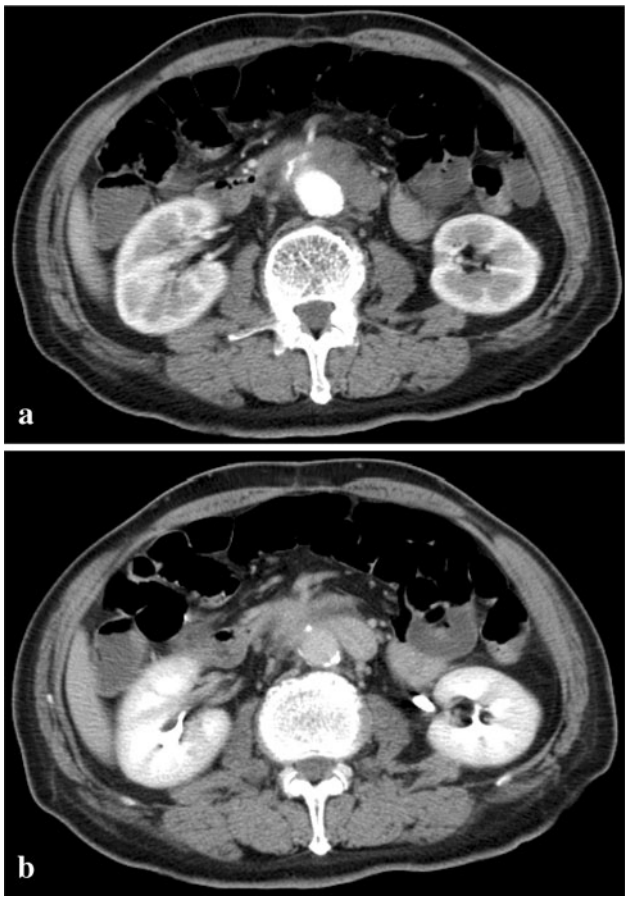

Fig. 1 Contrast-enhanced abdominal CT scans on admission. **a** Early arterial phase. **b** Late perfusion phase. Contrast-enhanced abdominal CT scanning revealed a para-aortic area of weak late-phase contrast uptake at the level of the graft anastomosis, with an indistinct border with the duodenum, raising the suspicion of a fistula between the abdominal aorta and the duodenum associated with infection

Discussion

ADF can be primary, where the aorta perforates the duodenum, e.g., in association with AAA, or secondary, following AAA repair with prosthetic implantation. ADF is a rare condition, but it is often severe and life-threatening. As prosthetic implantation is now currently considered a common procedure for AAA, secondary ADF should be recognized as a serious long-term postoperative complication. The incidence is less than 1%, and the interval between the initial procedure and development of secondary ADF is generally between 32 and 90 months, with a high mortality rate [2]. According to the different pathogenic mechanisms, secondary ADF is classified into two subtypes; namely, true aorto-enteric fistula (AEF) and paraprosthetic enteric fistula [3]. In the former, a pseudoaneurysm forms at the graft anastomosis site, then perforates the alimentary tract, often presenting with gastrointestinal hemorrhage. In the latter, perforation first develops in the bowel wall adjacent to the aortic prosthesis, and infection then spreads along the prosthesis, resulting in dehiscence of the prosthesis. This form often presents with pyrexia of unknown origin (PUO), abdominal pain, or other signs of sepsis. In either case,

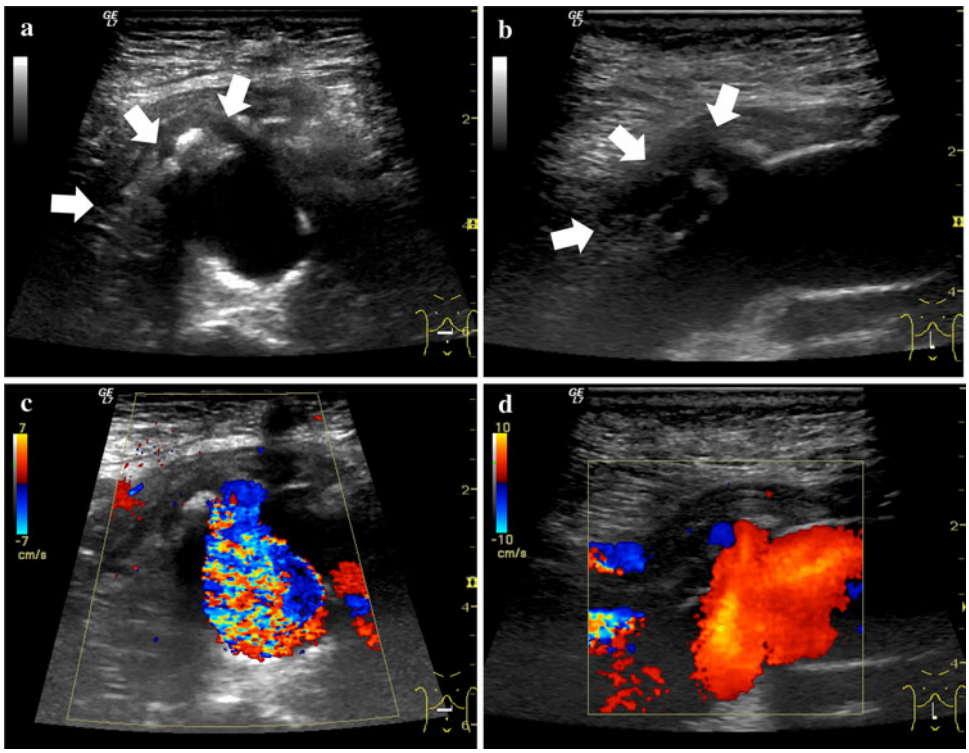

Fig. 2 Abdominal ultrasonograms on admission. **a** Horizontal scan of B-mode ultrasonography. An anechoic lesion (*arrows*) was detected between the duodenum and aorta. The duodenal wall adjacent to this lesion was thickened, suggesting bleeding from ADF. **b** Vertical scan of B-mode ultrasonography. Abdominal B-mode ultrasonography demonstrated a 5-mm defect in the vascular wall on the ventral side of the abdominal aorta at the graft anastomosis, and an umbilicated anechoic lesion (*arrows*), continuous with the aorta, was seen projecting between the aorta and the posterior wall of the third part of the duodenum. **c** Horizontal scan of color Doppler ultrasonography. A color signal was seen corresponding to the anechoic lesion between the duodenum and aorta. **d** Vertical scan of color Doppler ultrasonography. A color signal was seen at this site on color Doppler ultrasonography, confirming the diagnosis of a secondary ADF

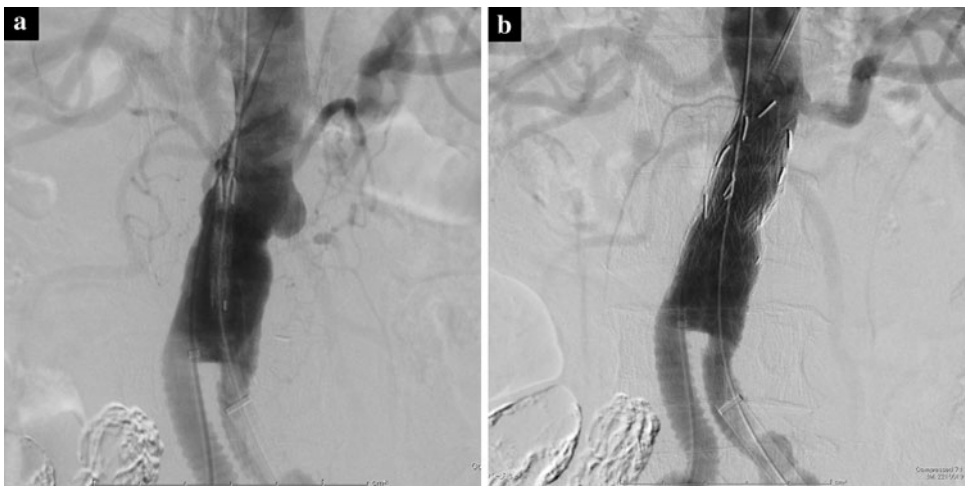

Fig. 3 Abdominal aortic angiograms. **a** Angiogram before the operation demonstrating a pseudoaneurysm. **b** Angiogram after the operation. A percutaneous endovascular aneurysm repair (EVAR) was performed to prevent further hemorrhage

when the fistula is small, bleeding is readily stopped by thrombus formation, and so the patient often has repeated transient small gastrointestinal hemorrhages, known as 'herald bleeds', followed by a massive fatal hemorrhage associated with enlargement of the fistula [4]. To save patients with secondary ADF, we must diagnose and treat this condition before this fatal hemorrhage occurs.

The accurate diagnosis of secondary ADF requires selection of investigation modalities appropriate to the clinical presentation. For patients presenting with

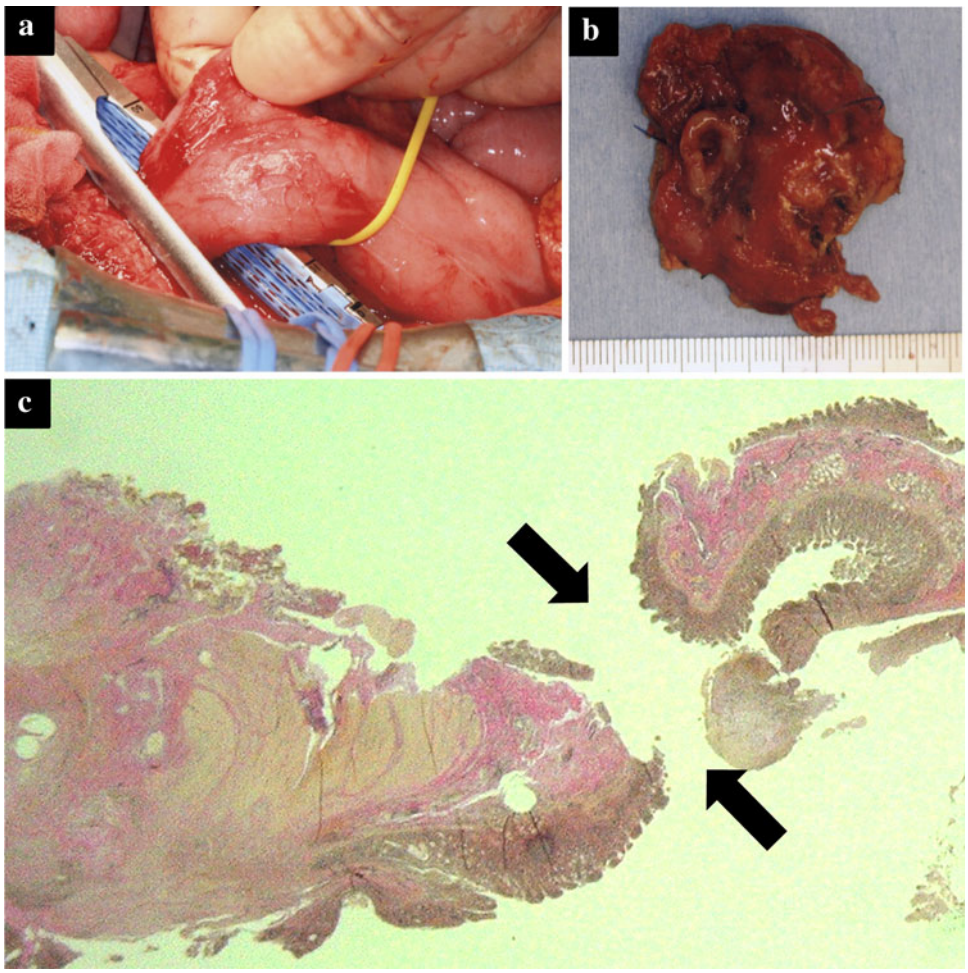

Fig. 4 **a** The third part of the duodenum and aorta. **b** The resected inflammatory mass comprising the infected graft and the third part of the duodenum. **c** Elastica van Gieson (EVG) stain (×10). The inflammatory mass was resected. Prosthetic re-implantation, omentopexy, and duodenojejunostomy were performed. The EVG stain showed the aortoduodenal fistula (*arrows*) and revealed fibrosis and inflammatory cell invasion of the aortic wall. Development of the mucous membrane of the duodenum had progressed along the vascular wall around the fistula

gastrointestinal hemorrhage, endoscopy should be performed first [5]. However, the site of the fistula is often the third part of the duodenum or even more distal. Consequently, with EGD we are often unable to pass the scope far enough to identify the bleeding point [6]. Furthermore, the procedure often raises the patient's blood pressure or dislodges the clot, disrupting hemostasis and causing a massive hemorrhage [3]. Abdominal CT scanning is non-invasive and can be performed repeatedly, and its sensitivity and specificity are both high for detection of serious complications following AAA repair. Accordingly, as with EGD, CT scanning is an important diagnostic modality for ADF [7]. However, in some cases no diagnosis can be made using CT, and gallium scintigraphy or angiography is needed to make the diagnosis [8]. In our case, CT and EGD failed to yield a diagnosis. In contrast, abdominal ultrasonography was able to continuously provide a color Doppler image in real time as an umbilicated anechoic area between the aorta and the posterior wall of the third part of the duodenum, enabling a diagnosis of bleeding due to the secondary ADF.

Recent advances in diagnostic ultrasound systems and progress in diagnostic methods have led to more widespread use of ultrasonography in investigations of the gastrointestinal tract [9, 10]. However, our PubMed search revealed no reports of ADF detected using ultrasonography. When a patient with gastrointestinal hemorrhage following reconstructive aorto-iliac surgery is encountered in the emergency department, the possibility of ADF should be considered. We believe that non-invasive abdominal ultrasonography may be useful in the diagnosis of secondary ADF.

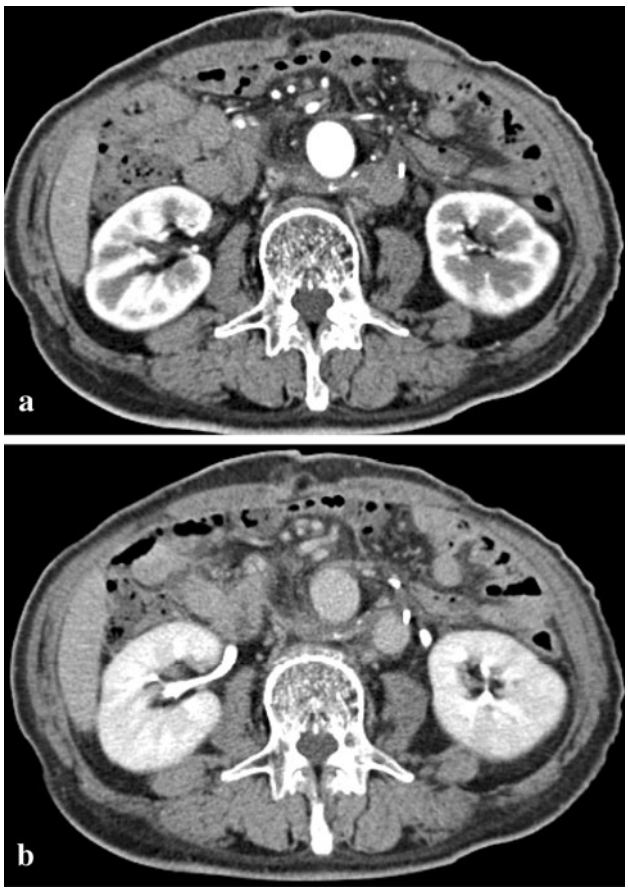

Fig. 5 Contrast-enhanced abdominal CT scans on admission. **a** Early arterial phase. **b** Late perfusion phase. The abdominal CT findings were also favorable

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