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## A primary aorto-duodenal fistula associated with an inflammatory abdominal aortic aneurysm: a case report.

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# A primary aorto-duodenal fistula associated with an inflammatory abdominal aortic aneurysm: a case report.\*

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## Abstract

Primary aorto-enteric fistula (PAEF) is a serious complication of abdominal aortic aneurysm (AAA). We report a patient with PAEF associated with inflammatory AAA who underwent emergent surgery. A 52-year-old male presented with recurrent hematemesis. A computer tomography scan showed a sealed rupture of the AAA adjacent to the duodenum. At surgery, a coin-sized PAEF was noted. The aorta was replaced with a Dacron graft in situ. Histological examination revealed the characteristics of an inflammatory AAA. The postoperative course was uneventful, and there has been no evidence of infection during a follow-up period of 3 years. We discuss the etiologic and surgical considerations regarding this unusual entity.

**KEYWORDS:** primary aorto-enteric fistula, inflammatory abdominal aortic aneurysm, ruptured abdominal aortic aneurysm

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Case Report

## A Primary Aorto-duodenal Fistula Associated with an Inflammatory Abdominal Aortic Aneurysm: A Case Report

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Primary aorto-enteric fistula (PAEF) is a serious complication of abdominal aortic aneurysm (AAA). We report a patient with PAEF associated with inflammatory AAA who underwent emergent surgery. A 52-year-old male presented with recurrent hematemesis. A computer tomography scan showed a sealed rupture of the AAA adjacent to the duodenum. At surgery, a coin-sized PAEF was noted. The aorta was replaced with a Dacron graft *in situ*. Histological examination revealed the characteristics of an inflammatory AAA. The postoperative course was uneventful, and there has been no evidence of infection during a follow-up period of 3 years. We discuss the etiologic and surgical considerations regarding this unusual entity.

**Key words:** primary aorto-enteric fistula, inflammatory abdominal aortic aneurysm, ruptured abdominal aortic aneurysm

**P** rimary aorto-enteric fistula (PAEF) is a rare but serious complication of abdominal aortic aneurysm (AAA), and the outcome of surgical treatment is poor [1, 2]. The surgical results depend on preoperative condition of the patient and degree of contamination of the operative field [1, 2]. Inflammatory AAA is one of the causes of PAEF, and a few cases of PAEF caused by inflammatory AAA have been reported [3, 4]. We report here the successful treatment of a case of PAEF associated with inflammatory AAA and discuss the etiologic and surgical considerations regarding this unusual entity.

### Case Report

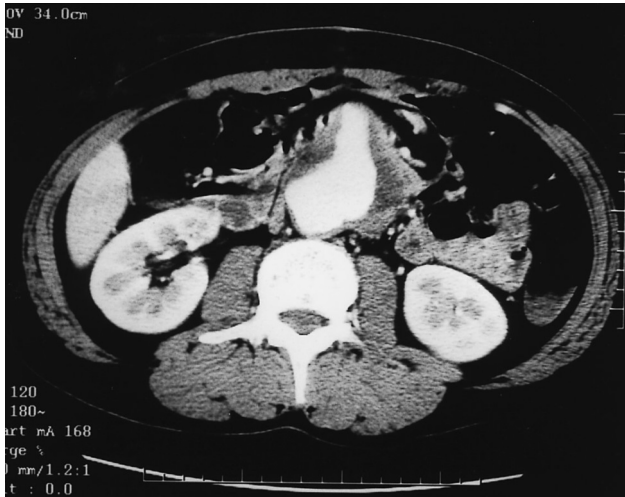
A 52-year-old male was referred to our institution for treatment of recurrent hematemesis and melena. He had had hematemesis and melena 2 weeks before admission and had visited a local hospital. Emergency exploratory laparotomy was performed due to uncontrollable hematemesis, but no obvious cause in the abdomen was found. A repeat endoscopic examination showed an extrinsic pulsating mass protruding into the internal lumen of the second portion of the duodenum. The recurrent gastrointestinal bleeding was uncontrollable, and the patient was transferred to our institution. An abdominal aortic aneurysm of 60 mm in diameter was noted on a computer tomography (CT) scan, which was adjacent to the third portion of the duodenum (Fig. 1). Abdominal aortography revealed a saccular-shaped AAA located approximately 30 mm distal of the renal arteries (Fig. 2).

At emergent surgery, the aneurysm appeared to be

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**Fig. 1** A CT scan showing the sealed rupture of the AAA adjacent to the third portion of the duodenum.

inflammatory, and the right lateral wall of the aneurysm had a communication with the third portion of the duodenum. Inspection of the internal lumen of the aorta showed a coin-sized aorto-duodenal fistula (Fig. 3). The markedly thickened arterial wall was resected to the extent that was possible. The aorta was replaced with a Dacron Y graft *in situ*. The perforated region of the duodenum was resected and was anastomosed in an end-to-end fashion. The cavity was irrigated with 10 liters of saline. The omentum was placed between the aorta and duodenum. A culture of the aneurysmal wall showed the presence of *Candida* species, and an antifungal agent was administered for 2 months. The results of histological examination showed that chronic inflammatory cells had focally infiltrated into the aneurysmal wall (Fig. 4), and that neutrophilic infiltration with abscess formation was seen around the fistula. The patient's postoperative course was uneventful, and there has been no evidence of infection during a follow-up period of 3 years.

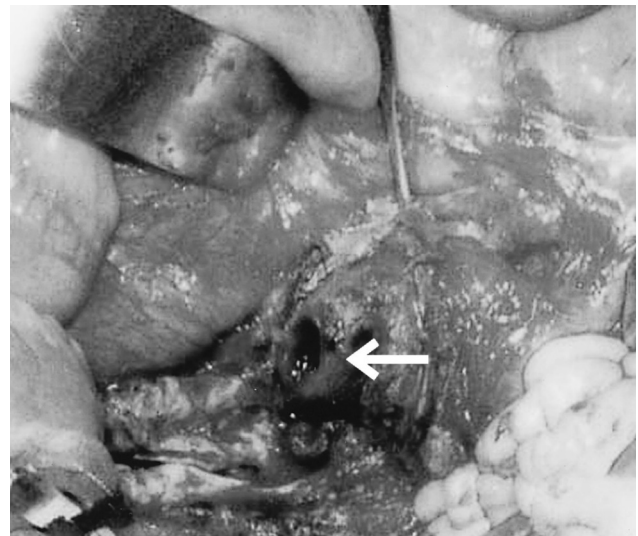
### Discussion

PAEF, direct communication between the aorta and the intestinal tract, is mainly due to an atherosclerotic aneurysm, and other causes include mycotic aneurysm, radiation trauma, and inflammatory AAA [1, 2]. Although 5% of infrarenal AAAs have the characteristics of inflammatory AAA, PAEFs resulting from inflammatory AAA are extremely rare [3, 4]. Ikeda *et*

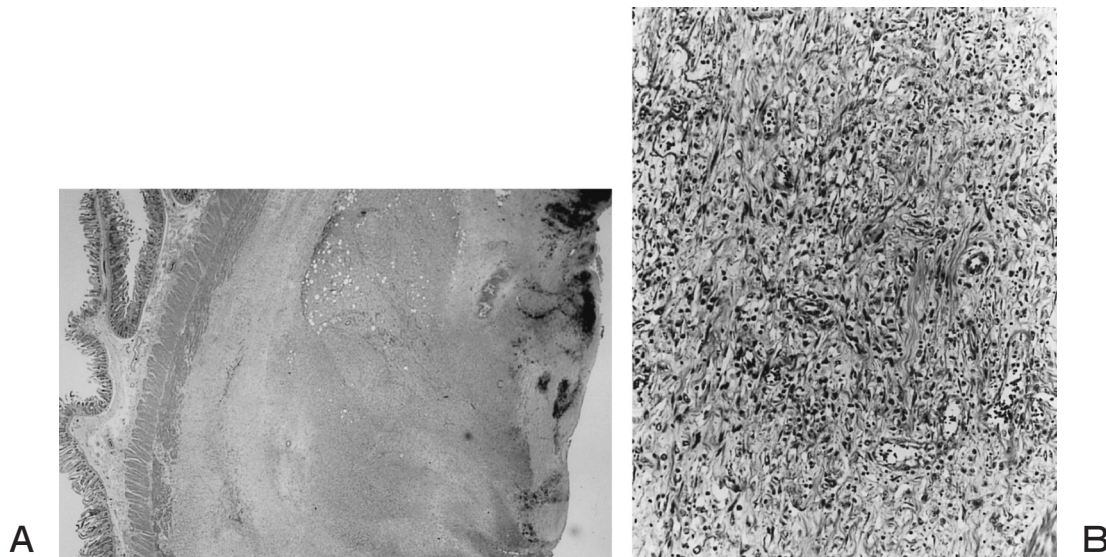


**Fig. 2** Aortography revealing a saccular-shaped AAA with no extravasation.

*al.* reported a 50-year-old woman who had PAEF associated with multiple inflammatory AAAs [3], and Mii *et al.* reported a 28-year-old man who had primary aorto-jejunal fistula associated with inflammatory AAA [4]. Together with the present case, the clinical characteristics of those patients include relatively young age and the presence of a rapidly developed saccular-shaped aneurysm. Interestingly, there were no symptoms associated with systemic inflammation such as high-grade fever,



**Fig. 3** Intraoperative finding of a coin-sized aorto-duodenal fistula. The arrow shows the fistularization.



**Fig. 4** Histological findings of the aneurysmal wall: chronic inflammatory cells infiltrated into the aneurysmal wall (A), and severe chronic inflammatory cell infiltration and localized neutrophilic infiltration with abscess formation (B).

despite the fact that all 3 patients had elevated white blood cells and C-reactive protein. The early to medium term results of those patients were encouraging, even though the overall surgical results in patients with PAEF have been poor. Such cases must receive careful follow-up, because the native aortic tissue in the anastomotic sites still has the potential of recurrent inflammation and subsequent fistulization.

It is controversial whether the chronic inflammation of the aneurysmal wall results purely from the inflammatory process, *i.e.*, a so-called “non-specific inflammatory aneurysm,” or from infection, *i.e.*, a mycotic aneurysm. In the present case, a culture of the aneurysmal wall showed the presence of *Candida* species; however, the patient had no symptoms of active infection, and there was also no evidence of endocarditis or other foci. Furthermore, bacteriological study of mycotic aneurysm [5] showed that the common organisms cultured from aneurysmal walls included *Staphylococcus aureus* and *Salmonella*, and mycotic aneurysm due to *Candida* species was extremely rare [5]. In addition, the characteristic CT findings in mycotic aneurysms, which include the sudden appearance of an aneurysm on a previously normal aorta in a febrile patient, a soft tissue mass surrounding the aneurysm, and an adjacent vertebral osteomyelitis [6], were not fully correlated with findings in present case. The clinical and pathological findings in

our case indicated that *Candida* species might not be a primary cause of inflammatory aneurysm but might be a secondary contamination through the fistula, and we concluded that it was a non-specific inflammatory aneurysm rather than a mycotic aneurysm.

Despite the presence of contamination, most surgeons have reconstructed diseased aortae with *in situ* grafts [1–4]. Extra-anatomic bypass grafting is an alternative approach for treatment of PAEF to avoid the use of a prosthetic graft in the contaminated region [7]. However, a high mortality rate has been reported in patients treated with extra-anatomic bypass, since not only does the procedure involve high risks of limb loss and aortic stump blowout, but also it is often performed in severely ill patients with gross infection in the operative field. The selection of surgical approach depends on the presence of sepsis and the degree of contamination. If a severe purulent infection is present in periaortic tissue, extra-anatomic bypass should be selected. Our patient was treated with an *in situ* graft because the contamination of the operative field was localized. Careful irrigation of the operative field is mandatory regardless of the presence or absence of gross contamination, and the proximal end of the prosthetic graft should be wrapped with the omentum to prevent recurrent fistulization, especially in cases of inflammatory aneurysm.

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