

Endovascular management of hepatic artery pseudoaneurysm hemorrhage complicating pancreaticoduodenectomy

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Gastrointestinal bleeding is a morbid complication of pancreaticoduodenectomy. Determining its etiology is often a daunting challenge in that both common and unusual mechanisms may be operative. Visceral artery pseudoaneurysms, although rare, must be considered in that minimally invasive means are available for effective therapy. Our recent experience with two cases highlights the importance for both general and vascular surgeons to be aware of the diagnostic and therapeutic role for *early* angiography and deployment of endovascular techniques to achieve a successful outcome. (J Vasc Surg 2006;43:613-7.)

Gastrointestinal bleeding is a potentially morbid complication following pancreaticoduodenectomy and requires precise identification of its source and etiology.^{1,2} Although upper endoscopy will demonstrate bleeding from enteric anastomoses and ulcers, arteriography is required to reveal vascular sources. Our experience with two recent cases emphasizes these points and illustrates the vital importance of early angiography to effect appropriate therapy and, if possible, deployment of endovascular techniques.

CASE REPORTS

Patient 1. A 61-year-old man with von Willebrand's syndrome underwent pylorus-sparing pancreaticoduodenectomy for a distal common bile duct tumor. An extraordinary finding at surgery, and presumably unrelated to the in situ carcinoma of the distal bile duct, was intense inflammation throughout the operative field that caused contraction of the gallbladder, pancreatitis, and diffuse peritonitis. Pathology confirmed an in situ carcinoma of the distal bile duct. There was no evidence for sclerosing cholangitis. The case was complicated by recurrent and persistent postoperative gastrointestinal bleeding. Computed tomography (CT) scans were nondiagnostic. Upper endoscopy suggested bleeding from the duodenojejunal anastomosis.

The patient was re-explored, and a partial gastrectomy with Billroth II reconstruction was performed. Bleeding recurred 1 week after surgery, and the patient became tachycardic and hypotensive with rapid progression to shock levels. Resuscitation included blood and blood products to replace blood loss and von Willebrand factor deficiencies. Continued bleeding prompted the use of recombinant factor VII as well. Emergency arteriography showed a pseudoaneurysm of the hepatic artery with enteric communication (Fig 1).

From the Vascular Surgery Service, Englewood Hospital and Medical Center. Competition of interest: none.

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The pseudoaneurysm was managed with a 6-mm Viabahn stent-graft (W. L. Gore & Associates, Flagstaff, Ariz) in the hepatic artery (5 mm) to cover the defect. Subsequent films showed the stent-graft had migrated distally (Fig 2). A second device was then deployed to stabilize the first and exclude the pseudoaneurysm. Thrombus was noted within the stent-grafts (Fig 3) but was lysed with 2 mg tissue-plasminogen activator (tPA). The final arteriogram revealed exclusion of the pseudoaneurysm (Fig 4). The right hepatic artery had been covered by the migrated stent-graft but was filled via collaterals from the left.

The patient rapidly regained hemodynamic stability and had an excellent subsequent recovery. There was a transient mild elevation in serum transaminases but no evidence of hepatic insufficiency or ischemia. He continues to be well after 2 years, with no evidence of tumor recurrence or hemorrhagic episodes.

Patient 2. A 67-year-old man underwent pylorus-sparing pancreaticoduodenectomy for a distal common bile duct tumor. The patient made an unremarkable recovery and was discharged home on postoperative day 10. Melena developed 6 weeks later, but a CT scan was nondiagnostic. Upper endoscopy suggested marginal ulceration. The patient was re-explored, and antrectomy with Billroth II reconstruction was performed with partial excision of the afferent limb where a right upper quadrant abscess was encountered and drained.

Thirty-six hours after the second surgery, the patient developed massive upper gastrointestinal bleeding. Emergency arteriography revealed a pseudoaneurysm of the left hepatic artery, with extravasation of contrast material into the intestinal tract (Fig 5). On this occasion, the pseudoaneurysm could not be crossed with a guidewire. Bleeding was controlled by embolization of the left hepatic artery with Gianturco coils (Cook, Bloomington, Ind) (Fig 6). The distal left hepatic artery was visualized via collaterals on delayed imaging.

The patient regained hemodynamic stability. His postprocedural course was complicated by respiratory failure, a colocolic fistula, and parenteral nutrition-induced hyperbilirubinemia. With supportive therapy, he completely recovered. The fistula spontaneously closed, and his liver function tests returned to normal levels. He remains well 21 months after the first operation.

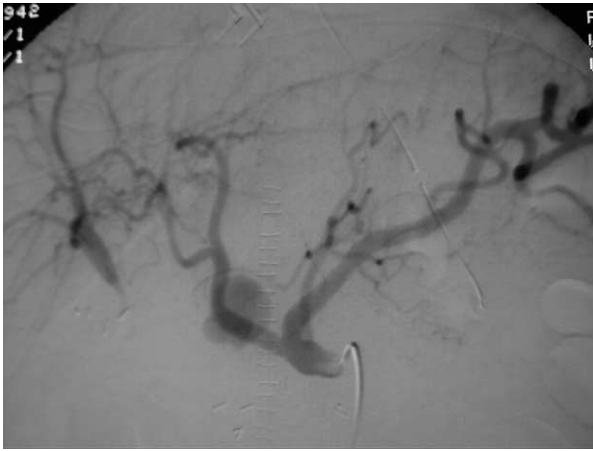


Fig 1. Hepatic artery pseudoaneurysm (patient 1) visualized with selective arteriography.



Fig 4. Patient 1: successful lysis of thrombus and exclusion of pseudoaneurysm with covered stent.

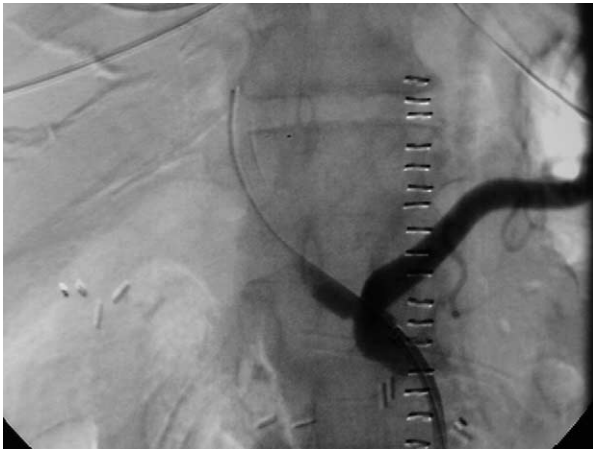


Fig 2. Migration of stent with continued visualization proximally of pseudoaneurysm.



Fig 5. Patient 2: left hepatic artery pseudoaneurysm (*black arrow*) and contrast extravasation into bowel (*white arrow*).

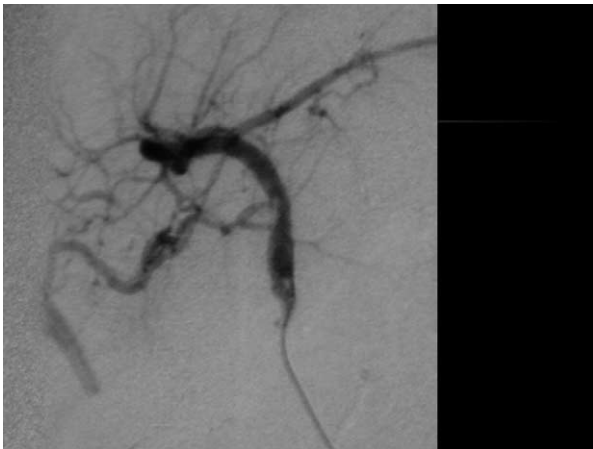


Fig 3. Thrombus formation in left hepatic arterial system occurred after the second stent-graft was placed.

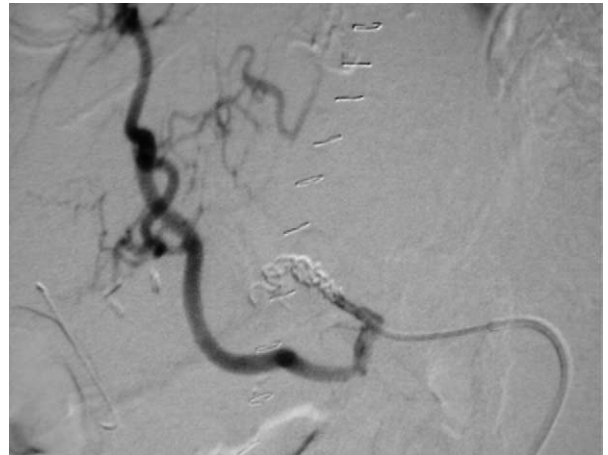


Fig 6. Arteriogram shows successful occlusion of pseudoaneurysm by coil embolization.

DISCUSSION

Complications following pancreaticoduodenectomy are common and usually consist of infection, anastomotic leak, delayed gastric emptying, and gastrointestinal bleeding. Visceral artery pseudoaneurysms are rarely encountered, however. Data from the Mayo Clinic retrospective review encompassed 18 years (January 1980 through December 1998). Of the 37 patients found with visceral artery pseudoaneurysms,³ 19 had hepatic artery pseudoaneurysms, only one of which followed pancreaticoduodenectomy. When the authors combined their personal data with their review of reports in English, 153 patients were found to have hepatic artery pseudoaneurysms following hepatic, biliary, or pancreatic procedures, only nine of which had undergone pancreaticoduodenectomy. More recently, Otah et al⁴ reported five cases of pseudoaneurysms following pancreaticoduodenectomy—two left hepatic arteries, one right hepatic artery, one gastroduodenal artery stump, and one replaced right hepatic artery. The authors also referred to six patients previously reported in the literature with post-pancreaticoduodenectomy bleeding in hepatic, celiac, or gastroduodenal arteries, or a combination.

In general, the hepatic artery is the second most frequent site of visceral pseudoaneurysms, the splenic artery being the most common.⁵ Hepatic artery pseudoaneurysms are usually iatrogenic but have also been associated with intra-abdominal inflammation, infection, or trauma. When associated with surgery, possible causes of pseudoaneurysm include direct vascular injury during dissection or retraction, clamp injuries to the vessel, or thermal injury from electrocautery close to the vessel. Surgical removal of adjacent lymph nodes, nerves, and connective tissue for malignancy renders visceral arteries more vulnerable to further trauma. Additionally, pancreatic secretions leaking from the pancreaticojejunal anastomosis may compromise the arterial wall as well as local sepsis.

Potentially lethal bleeding from pseudoaneurysms can occur early or late in the postoperative period. Teramoto et al⁶ reported a case of delayed massive hemorrhage and shock 34 days after pancreaticoduodenectomy. Angiography showed bleeding from a gastroduodenal artery stump pseudoaneurysm. The patient was successfully treated by embolization of the common hepatic artery and arterialization of the portal vein. The five cases of pseudoaneurysm following pancreaticoduodenectomy reported by Otah et al⁴ were discovered 12 to 28 days after surgery, all identified by visceral arteriography and successfully treated with coil embolization.

Choi et al¹ recently documented 22 instances of delayed hemorrhage after pancreaticoduodenectomy. Pseudoaneurysms originating from major arteries around the pancreaticojejunostomy were seen in nine of these cases. Transcatheter arterial embolization was attempted in 14 patients, four of these ultimately required laparotomy. One patient died of complications after transcatheter arterial embolization and another after massive hemorrhage. Covered stents were not used in this series.

During the pancreaticoduodenectomy of our first patient, intense inflammation was noted throughout the operative field, with pancreatitis, lymphangitis, and diffuse peritonitis. The cause of this inflammatory process was not clear. Zen et al⁷ recently emphasized the spectrum of immunoglobulin (Ig) G4-related sclerosing cholangitis and its association with biliary tumor, bile duct obstruction, infection, and pericholangitis. Our second patient was found to have an abscess in the right upper quadrant at the time of the second operation. Visceral pseudoaneurysms have been associated with inflammation.⁸ Infection and inflammation are likely factors contributing to the development of pseudoaneurysms in these patients.

In addition to the diffuse intra-abdominal inflammatory process setting the stage for potential visceral artery pseudoaneurysm, patient 1 was known to have von Willebrand's disease, the most common hereditary disorder of hemostasis. This qualitative platelet abnormality affects one in 800 to 1,000 individuals and is associated with vascular pathologic conditions. Pseudoaneurysms of the femoral, supraorbital, and superior gluteal arteries have been reported in individuals with von Willebrand's disease.⁹⁻¹¹ Garaci et al¹² presented a case report of a 37-year-old man whose only clinical manifestation was upper abdominal pain; he had not undergone surgery or abdominal trauma. There was a family history of "hemorrhagic tendencies," and subsequent blood work revealed evidence of von Willebrand's disease. Angiography clearly defined a giant hepatic artery pseudoaneurysm. This was successfully treated with transcatheter embolization.

Initially, as a result of upper endoscopy, the postoperative gastrointestinal bleeding in both of our patients was thought to be due to marginal ulceration. Both patients subsequently underwent partial gastrectomy, but bleeding recurred. Emergency arteriography revealed hepatic artery pseudoaneurysms in each case. This clearly demonstrates the value of angiography, and we presume that if angiography had been performed earlier, both patients could have been spared second operations.

Similar examples of the value of angiography were demonstrated in the five cases reported by Otah et al.⁴ Endoscopy in one of their patients showed "ulceration and clot at the gastrojejunostomy site without a visible vessel or active bleeding," but subsequent angiography revealed a 1.2-cm pseudoaneurysm of the right hepatic artery receiving retrograde flow via the left hepatic artery. After coil embolization of the left hepatic artery, the patient experienced further bleeding the next day. Repeat angiography showed recanalization of the left hepatic artery. Repeat embolization with both coils and absorbable gelatin sponge slurry was successful. A second patient with bright red blood in the stool and a decreased hematocrit (24%) underwent sigmoidoscopy that showed "normal mucosa." Endoscopy suggested "multiple gastric erosions," but a definitive visceral angiography revealed a 2-cm pseudoaneurysm of the gastroduodenal artery stump.

The traditional treatment of hepatic artery pseudoaneurysms originally consisted of open ligation or excision

and vascular reconstruction.¹³ In recent years, endovascular treatment, particularly coil embolization, has become popular.¹⁴⁻²⁰ Open surgical repair is still advocated by some groups for certain cases, for example, pseudoaneurysms after hepatic resectional procedures or after hepatic transplantation.^{21,22} Stent-grafting of hepatic pseudoaneurysms has been reported,²³⁻²⁵ but none previously, to our knowledge, following pancreaticoduodenectomy. This may be critical, in that patients undergoing hepatic artery embolization following pancreaticoduodenectomy may be at greater risk for liver infarction and failure because of decreased collateral circulation from the gastroduodenal artery and pancreaticoduodenal arcade. Transarterial embolization of the common hepatic artery resulted in fatal hepatic failure in one case.¹ Use of a covered stent, where possible, will retain prograde flow as the pseudoaneurysm is excluded from the circulation.

We elected to insert a covered stent-graft to cover the neck of the hepatic artery pseudoaneurysm in patient 1. Migration of the first stent, a complication that should be preventable, was possibly due to stent undersizing or inadequate postdeployment balloon expansion. The second complication, stent-graft thrombosis, might have been induced by the residual effects of recombinant factor VII. This complication was particularly disturbing, in that lysis needed to be instituted in a patient with a recent operation. Fortunately, there were no untoward consequences from using tPA, and the resolution of the thrombus was most gratifying. The patient is currently being maintained with aspirin. Patient 2 required coil embolization because of tortuosity of the left hepatic artery and the lesser concern for infarction in this case. Both procedures were accomplished successfully, and the patients rapidly regained hemodynamic stability.

Concern exists regarding the placement of foreign bodies such as coils and stent-grafts in the setting of infection or inflammation. Intravascular stent infection can be a devastating complication, but it is uncommon.²⁶⁻²⁸ In fact, stent-grafts have been used to repair mycotic aneurysms, infected pseudoaneurysms, and aorto-esophageal fistulae.²⁹⁻³³ The favorable outcomes reported for these cases raise provocative points that are contrary to conventional wisdom. Comparison may be made with aortoenteric fistulae following open abdominal aortic aneurysm repair; however, communication between the graft and bowel does not necessarily imply severe infection or warrant total graft removal.³⁴⁻³⁶ Both of our patients were hemodynamically unstable at the time of placement of the stent-grafts or coils and would probably not have survived an open procedure. The endovascular approach was clearly the optimal choice.

CONCLUSIONS

Gastrointestinal bleeding is a potentially devastating complication following pancreaticoduodenectomy. Although usually secondary to marginal ulceration, it may result from more uncommon etiologies such as arterial pseudoaneurysm. Inflammation or infection in the right upper quadrant raises the index of suspicion for a potential pseudoan-

eurysm. When the patient concomitantly has a hemorrhagic disorder such as von Willebrand disease, that index of suspicion is even further heightened.

Because marginal ulcers are common after pancreaticoduodenectomy, their presence should not immediately rule out other sources of bleeding. Arteriography is highly sensitive and specific in the diagnosis of massive gastrointestinal bleeding and provides immediate therapeutic opportunities. Although the ultimate etiologies of our patients' pseudoaneurysms remain uncertain, these cases emphasize the utility of angiography in identifying hepatic artery pseudoaneurysm and thus allowing rapid, optimal treatment.

As new technologies for endovascular procedures emerge, stenting for arterial pseudoaneurysms may prove to be a safe and efficacious alternative to coil embolization and surgical intervention. Use of a covered stent-graft to repair visceral artery pseudoaneurysm has proven a successful procedure and lifesaving when deployed for exsanguinating hemorrhage.

AUTHOR CONTRIBUTIONS

Conception and design:

Analysis and interpretation:

Data collection:

Writing the article:

Critical revision of the article:

Final approval of the article:

Statistical analysis:

Obtained funding:

Overall responsibility:

REFERENCES

1. Choi SH, Moon HJ, Heo JS, Joh JW, Kim YI. Delayed hemorrhage after pancreaticoduodenectomy. *J Am Coll Surg* 2004;199:186-91.
2. Santoro R, Carlini M, Carboni F, Nicolas C, Santoro E. Delayed massive arterial hemorrhage after pancreaticoduodenectomy for cancer. Management of a life-threatening complication. *Hepatogastroenterology* 2003;50:2199-204.
3. Tessier DJ, Fowl RJ, Stone WM, McKusick MA, Abbas MA, Sarr MG, et al. Iatrogenic hepatic artery pseudoaneurysms: an uncommon complication after hepatic, biliary, and pancreatic procedures. *Ann Vasc Surg* 2003;17:663-9.
4. Otah E, Cushin BJ, Rozenblit GN, Neff R, Otah KE, Cooperman AM. Visceral artery pseudoaneurysms following pancreatoduodenectomy. *Arch Surg* 2002;137:55-9.
5. Baker KS, Tisnado J, Cho SR, Beachley MC. Splanchnic artery aneurysms and pseudoaneurysms: transcatheter embolization. *Radiology* 1987;163:135-9.
6. Teramoto K, Kawamura T, Takamatsu S, Noguchi N, Arai S. A case of hepatic artery embolization and partial arterialization of the portal vein for intraperitoneal hemorrhage after a pancreaticoduodenectomy. *Hepato-Gastroenterology* 2003;50:1217-9.
7. Zen Y, Harada K, Sasaki M, Sato Y, Tsuneyama K, Haratake J, et al. IgG4-related sclerosing cholangitis with and without hepatic hepatic inflammatory pseudotumor and sclerosing pancreatitis-associated sclerosing cholangitis. *Am J Surg Path* 2004;28:1193-203.
8. Rokke O, Sondanaa K, Amundsen SR, Bjerke Larssen T, Jensen D. Successful management of eleven splanchnic artery aneurysms. *Eur J Surg* 1997;163:411-7.
9. Wautier JL, Caen JP. Angiodysplasia in acquired von Willebrand disease. *Lancet* 1976;30:973.

10. Choung KC, Wei JP. Traumatic pseudoaneurysm of the supra-orbital artery associated with von Willebrand disease: case report. *J Trauma* 1993;35:966-7.
11. De Foer B, Stockx L, Wilms G, Vermeylen J, Baert AL. Selective embolization of a superior gluteal artery pseudoaneurysm associated with von Willebrand's disease. *J Belge Radiol* 1996;79:135-6.
12. Garaci FG, Gandini R, Romagnoli A, Fasoli F, Varruciu V, Simonetti G. Hepatic artery pseudoaneurysm in von Willebrand's disease. *Eur Radiol* 2003;13:1913-5.
13. Lumsden AB, Mattar SG, Allen RL, Bacha EA. Hepatic artery aneurysms: the management of twenty-two patients. *J Surg Res* 1996;60:345-50.
14. Akatsu T, Hayashi S, Egawa T, Doi M, Nagashima A, Kitano M, et al. Hepatic artery pseudoaneurysm associated with cholecystitis that ruptured into the gallbladder. *J Gastroenterol* 2004;39:900-3.
15. Kasirajan K, Greenberg RK, Clair D, Ouriel K. Endovascular management of visceral artery aneurysm. *J Endovasc Ther* 2001;8:150-5.
16. Little AF, Lee WK. Percutaneous and endovascular embolization of ruptured hepatic artery aneurysm. *Cardiovasc Intervent Radiol* 2002;25:208-11.
17. Gabelmann A, Gorich J, Merkle EM. Endovascular treatment of visceral artery aneurysms. *J Endovasc Ther* 2002;9:38-47.
18. Finley DS, Hinojosa MW, Paya M, Imagawa DK. Hepatic artery pseudoaneurysm: a report of seven cases and a review of the literature. *Surg Today* 2005;35:543-7.
19. Reber PU, Baer HU, Patel AG, Triller J, Buchler MW. Life-threatening upper gastrointestinal tract bleeding caused by ruptured extrahepatic pseudoaneurysm after pancreatoduodenectomy. *Surgery* 1998;124:114-5.
20. Tien Y, Lee P, Yang C, Ho M, Chiu Y. Risk factors of massive bleeding related to pancreatic leak after pancreaticoduodenectomy. *J Am Coll Surg* 2005;201:554-9.
21. Maleux G, Pirenne J, Aerts R, Nevens F. Case report: hepatic artery pseudoaneurysm after liver transplantation: definitive treatment with a stent-graft after failed coiled embolization. *Br J Radiol* 2005;78:453-6.
22. Muraoka N, Uematsu H, Kinoshita K, Takeda T, Morita N, Matsunami H, et al. Covered coronary stent graft in the treatment of hepatic artery pseudoaneurysm after liver transplantation. *J Vasc Interv Radiol* 2005;16(2 Pt 1):300-2.
23. Paci E, Aantico E, Candelari R, Alborino S, Marmorale C, Landi E. Pseudoaneurysm of the common hepatic artery: treatment with a stent-graft. *Cardiovasc Intervent Radiol* 2000;23:472-4.
24. Venturini M, Angeli E, Salvioni M, Cobelli F, Trentin C, Carlucci M, et al. Hemorrhage from a right hepatic artery pseudoaneurysm: endovascular treatment with a coronary stent-graft. *J Endovasc Ther* 2002;9:221-4.
25. Bell RE, Taylor PR, Aukett M, Evans GH, Reidy JF. Successful endoluminal repair of an infected thoracic pseudoaneurysm caused by methicillin-resistant *Staphylococcus aureus*. *J Endovasc Ther* 2003;10:29-32.
26. Clarke MG, Thomas HG, Chester JF. MRSA-infected external iliac artery pseudoaneurysm treated with endovascular stenting. *Cardiovasc Intervent Radiol* 2005;8:364-6.
27. Gandini R, Pipitone V, Konda D, Pendenza G, Spinelli A, Stefanini M, et al. Endovascular Treatment of a giant superior meenteric artery pseudoaneurysm using a nitinol stent-graft. *Cardiovasc Intervent Radiol* 2005;28:102-6.
28. Kinney Ev, Kaebnick HW, Mitchell RA, Jung MT. Repair of mycotic paravisceral aneurysm with a fenestrated stent-graft. *J Endovasc Ther* 2000;7:192-7.
29. Kwon K, Choi D, Choi SH, Ko BK, Ko YG, Jang Y, et al. Percutaneous stent-graft repair of mycotic common femoral artery aneurysm. *J Endovasc Ther* 2002;9:690-3.
30. Nishibe T, Koizumi J, Kudo F, Miyazaki K, Nishibe M, Yasuda K. Successful endovascular stent-graft treatment for an aorto-esophageal fistula caused by a descending thoracic aortic aneurysm: report of a case. *Surg Today* 2004;34:529-31.
31. Qu L, Jing Z, Feng R. Endoaortic stent grafting of a giant infected hepatic-celiac pseudoaneurysm. *J Vasc Surg* 2005;42:159-62.
32. Sanada J, Matsui O, Arakawa F, Tawara M, Endo T, Ito H, et al. Endovascular stent-grafting for infected iliac artery pseudoaneurysms. *Cardiovasc Intervent Radiol* 2005;28:83-6.
33. Shiraishi S, Watarida S, Matsubayashi K, Motoishi M, Satsu T. Successful management of an aorto-esophageal fistula resulting from an aneurysm of the thoracic aorta with a covered stent. *J Cardiovasc Surg (Torino)* 2002;43:95-8.
34. Calligaro KD, Veith FJ, Yua JG, Gargiulo NJ, Dougherty MJ. Intra-abdominal aortic graft infection: complete or partial graft preservation in patients at very high risk. *J Vasc Surg* 2003;38:1199-205.
35. Hart JP, Eginton MT, Brown KR, Seabrook GR, Lewis BD, Edminston CE Jr, et al. Operative strategies in aortic graft infections: is complete graft excision always necessary? *Ann Vasc Surg* 2005;19:154-60.
36. Yoshimoto K, Shiiya N, Onidera Y, Yasuda K. Secondary aortoenteric fistulae. *J Vasc Surg* 2005;42:805.

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