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Summary	<p>Aortoenteric fistulas are rare (&lt;1%) but disastrous complications after open and endovascular aortic surgery. The most frequently involved anatomical sites are abdominal aorta and duodenum. Surgery is the only possible treatment and consists, in most of cases, in an axillobifemoral bypass, a very invasive procedure with a high rate of complications. In the literature, less than 10 cases of direct communication between the right iliac artery and the appendix are described. In this paper, we discuss our experience of a case of iliac appendiceal fistula. We go through clinical presentation, diagnostic path and treatment with a brief look at the literature. Our conclusion is that in some cases a less invasive surgical approach could lead to good results, but the long-term outcomes need to be studied. Iliac appendiceal fistulas are rare variations of aortoenteric fistulas with similar onset. In cases like ours, a less invasive surgical approach could lead to good results.</p>	
Keywords	<p>separated by '-' Appendix - Iliac artery - Fistula - Aortoenteric fistulas - Gastrointestinal hemorrhage</p>	

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8 **A rare but life-threatening complication of aortic surgery:  
9 iliac appendiceal fistula**  
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42 **Keywords** Appendix · Iliac artery · Fistula · Aorto-  
43 enteric fistulas · Gastrointestinal hemorrhage  
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45 **Introduction**  
46

47 An aortoenteric fistula (AEF) consists of a direct com-  
48 munication between an aortic aneurysm and the gas-  
49 trointestinal (GI) tract [1]. Primary AEF (PAEF) hap-  
50 pens in patients without a story of aortic aneurysm  
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repair. This is most frequent in patients with predis-  
posing factors such as atherosclerosis (60–80%), infec-  
tions, or mechanical stress [1]. On the other hand, sec-  
ondary aortoenteric fistula (SAEF) is a rare (<1%) [2]  
but disastrous complication after open and endovas-  
cular repair of aortic aneurysm due to an infection of  
the graft that leads to a pseudoaneurysm that pene-  
trates the bowel wall. The morbidity and mortality of  
SAEF range between 14–75% [2]. Both primary and  
secondary AEF concerns more frequently the abdom-  
inal aorta than the thoracic aorta (56% vs 44%) and  
the duodenum [1]. This is due to the anatomical po-  
sition of the third part of the duodenum, which lies  
between the superior mesenteric artery and the ab-  
dominal aorta. More rarely, conduits may involve as-  
cending, transverse, sigmoid colon or rectum. There  
are less than 10 cases of iliac appendiceal fistulas (IAF)  
[3–10, 14] in the literature. In this paper, we report our  
experience of a secondary IAF.

42 **Case report**

M.R., a 52-year-old white man, was admitted to our  
emergency room for rectal bleeding and pain to the  
lower right limb. Eleven years before this event, the  
patient had a repair of abdominal aortic stenosis  
with the interposition of an aortic-bisiliac graft after  
an unsuccessful PTA and stenting of iliac arteries.  
The only therapy he took at home is an antiplatelet  
drug (Tyclopidine). On physical examination, the  
patient was normotensive and the abdomen was  
not reactive. On rectal examination, digested blood  
was found. His laboratory values showed no sign of  
acuteness (Hgb 13.1 g/dL; WBCs 13.89 x10<sup>9</sup>/L CRP  
4.1 mg/dL). A computed tomography (CT) scan with  
contrast of the abdomen was performed. The exam  
demonstrated three pseudoaneurysms distal to the  
previous vascular anastomosis, with diameters of

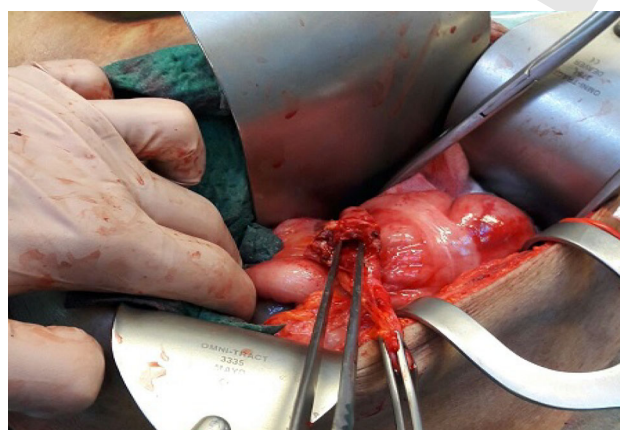


**Fig. 1** A computed tomography scan with contrast showing the pseudoaneurysm in right iliac artery enhanced in arterial phase



**Fig. 2** Three-dimensional reconstruction of the iliac vessels: A primitive iliac artery stump, B graft in right iliac artery with the pseudoaneurysm, C left iliac artery with pseudoaneurysms

42.5 mm on the right, 15 mm and 21 mm on the left, respectively. There were no signs of extravasation of contrast or any communication with bowel. Because of the stable situation, the initial approach was conservative. After about 24 h of observation his blood tests demonstrate anemia (Hgb 8.9 g/dL) with no other concerning laboratory or clinical signs. After about 30 h, the patient experienced repeated episodes of severe rectal bleeding. He underwent gastroscopy and colonoscopy, but no source of active bleeding was found. The only findings were clots in the sigmoid colon and blood painted intestine walls until the cecum. At 36 h, his condition abruptly changed and the patient went into hemorrhagic shock. Because of the



**Fig. 3** Situs of appendiceal fistulization

severe scenario, the patient was transported to operating room (OR) for an urgent exploratory celiotomy. No hemoperitoneum was found nor an active source of bleeding was evident. During surgery, the patient became dangerously hypotensive. A pseudoaneurysm of the right iliac artery that was in direct communication with the appendix was found. Clamping of the artery was necessary to keep tension up. The decision was made to cut open the pseudoaneurysm capsule and to partially remove the right limb of the Dacron graft that was severely damaged because of inflammation. A first attempt to connect the previous graft to the distal external iliac artery was made, but the inflamed tissues were not safe enough to perform an anastomosis. So, after right inguinal incision, the femoral artery was isolated and an iliac–femoral bypass was performed with the interposition of a new 8 mm Dacron graft. The peritoneum was closed above the new graft as protection. Immediate postoperative progress was in ICU. He was transferred in our department on postoperative day 3. During recovery, he had no clinical or surgical complications. His blood pressure was poorly controlled so antihypertensive therapy was initiated. He was administered a broad-spectrum intravenous antibiotic therapy (piperacillin/tazobactam; teicoplanin) until postoperative day 10 when he was discharged. Recommendations of continuing oral antibiotic therapy for more 4 weeks (sulfamethoxazole/trimethoprim) were given. Three months after surgery the morphology of the distal right iliac anastomosis, the inner blood flow, the distal vascularization of the right lower limb and CRP blood levels were all in normal range.

### Discussion

In patients with rectal bleeding and a history of previous aortic graft, a SAEF should always be suspected until proven wrong [6]. The appendix is a site of fistulization with an incidence of 2.4–4% [1]. The typical clinical presentation of IAF is not different from other SAEF, that is with repeated episodes of severe

119 rectal bleeding that could lead to hemorrhagic shock  
 120 if not stopped [3–5]. Some cases of onset with me-  
 121 lena, abdominal and right limb pain and anemia are  
 122 described [7–9]. Diagnosis is challenging and could  
 123 be reached with the aid of CT scan and endoscopy in  
 124 stable patients [9]. The CT findings related to SAEF in-  
 125 clude perigraft fluid, perigraft soft tissue attenuation,  
 126 ectopic gas, pseudoaneurysm and focal bowel wall  
 127 thickening [11]. In our case, the CT showed a pseu-  
 128 doaneurysm, but there was no evidence of commu-  
 129 nication with the bowel. Tagged red blood cell study  
 130 has been useful in one case [8]. Gadolinium mag-  
 131 netic resonance imaging was used in another case but  
 132 the result was not determining [12]. Laparotomy may  
 133 be negative in almost 50% of patients bleeding after  
 134 aortic surgery [13]. Though, it is a mandatory step  
 135 when SAEF is suspected because of the inevitably fatal  
 136 outcome if surgical treatment is not promptly in-  
 137 stituted. In the literature, different treatments of IAF  
 138 are described. Per some authors, complete graft re-  
 139 moval and performing of an axillobifemoral bypass is  
 140 mandatory [6]. This is a very traumatic surgery espe-  
 141 cially in patients who are already compromised and  
 142 the risk of aortic stump disruption is 20% [9]. For  
 143 other authors, partial graft removal associated with  
 144 radical debridement of perigraft infected tissues is suf-  
 145 ficient, especially when the infection is remote from  
 146 the aortic stump. Chiche et al. presented a case of par-  
 147 tial graft removal with in situ rifampin-bonded graft  
 148 reconstruction [9]. Their patient follow-up was nega-  
 149 tive for recurrent infections after 17 months. Another  
 150 strategy supported by some authors [14] could be an  
 151 endovascular approach. The main problem about this  
 152 technique is the risk of infection from inserting pros-  
 153 thetic material into an infected field. In the paper  
 154 by Danneels et al., 70% of all patients studied had  
 155 a recurrent infection or new AEF within 1 year after  
 156 an endovascular approach [15]. Their conclusion  
 157 was that, especially in high-risk patients, endovascular  
 158 treatment of AEF should be a short bridge to surgery.

## 160 Conclusion

162 SAEF must be suspected in patients with rectal bleed-  
 163 ing and a history of aortic surgery. IAF is a rare varia-  
 164 tion of this complication with similar clinical presen-  
 165 tation and diagnosis. Treatment could be less inva-  
 166 sive than the classical axillobifemoral bypass. A par-  
 167 tial graft excision, debridement of perigraft infected  
 168 tissues and reconstruction with a new graft could be

sufficient. The long-term outcomes need to be stud-  
 169 170 171 172 173 174 175 176 177

**Conflict of interest** A. Bondurri and L. Zampino declare that they have no competing interests.

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**1** page 1, column A

Schlüsselwörter/Keywords: Since both the title and keywords are scanned by search engines we aim to avoid duplicating terms to improve the “hit rate”. Five keywords should be chosen that are listed in the MeSH browser. Please try to apply changes accordingly.

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All acronyms/abbreviations should be written out at first usage in the text. Please attend to all acronyms whose explanations are still missing. Here: PTA.

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Do you mean 'ticlopidine' instead of Tyclopidine?

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**6** page 3, column B

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