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Summary

Aortoenteric fistulas are rare (<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.

Keywords separated by '-'

Appendix - Iliac artery - Fistula - Aortoenteric fistulas - Gastrointestinal hemorrhage

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A rare but life-threatening complication of aortic surgery: iliac appendiceal fistula

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Summary Aortoenteric fistulas are rare (<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.

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Introduction

An aortoenteric fistula (AEF) consists of a direct communication between an aortic aneurysm and the gastrointestinal (GI) tract [1]. Primary AEF (PAEF) happens in patients without a story of aortic aneurysm repair. This is most frequent in patients with predisposing factors such as atherosclerosis (60–80%), infections, or mechanical stress [1]. On the other hand, secondary aortoenteric fistula (SAEF) is a rare (<1%) [2] but disastrous complication after open and endovascular repair of aortic aneurysm due to an infection of the graft that leads to a pseudoaneurysm that penetrates the bowel wall. The morbidity and mortality of SAEF range between 14–75% [2]. Both primary and secondary AEF concerns more frequently the abdominal aorta than the thoracic aorta (56% vs 44%) and the duodenum [1]. This is due to the anatomical position of the third part of the duodenum, which lies between the superior mesenteric artery and the abdominal aorta. More rarely, conduits may involve ascending, 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.

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 x10^9/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

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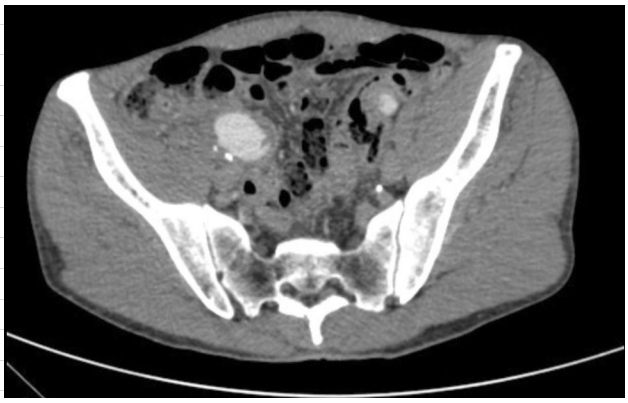

Fig. 1 A computed tomography scan with contrast showing the pseudoaneurysm in right iliac artery enhanced in arterial phase

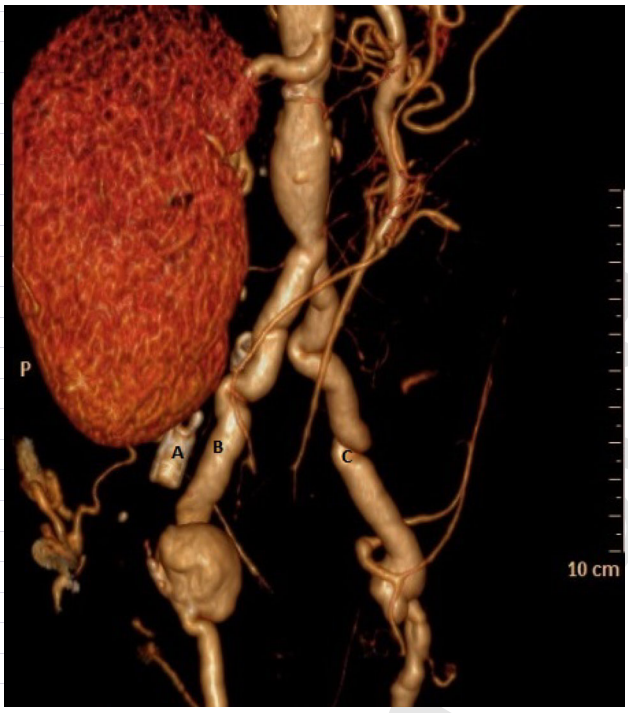

Fig. 2 Three-dimensional reconstruction of the iliac vessels: *A* primitive iliac artery stump, *B* graft in right iliac artery with the pseudoaneurysm, C left iliac artery with pseudoaneurysms

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 anemization (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

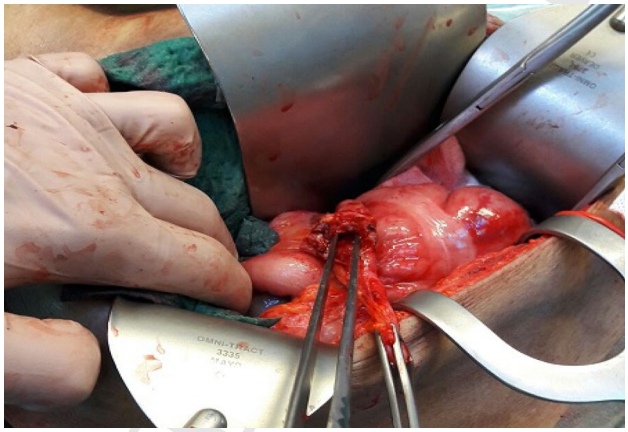

Fig. 3 Situs of appendiceal fistulization

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

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rectal bleeding that could lead to hemorrhagic shock if not stopped [3–5]. Some cases of onset with melena, abdominal and right limb pain and anemia are described [7–9]. Diagnosis is challenging and could be reached with the aid of CT scan and endoscopy in stable patients [9]. The CT findings related to SAEF include perigraft fluid, perigraft soft tissue attenuation, ectopic gas, pseudoaneurysm and focal bowel wall thickening [11]. In our case, the CT showed a pseudoaneurysm, but there was no evidence of communication with the bowel. Tagged red blood cell study has been useful in one case [8]. Gadolinium magnetic resonance imaging was used in another case but the result was not determining [12]. Laparotomy may be negative in almost 50% of patients bleeding after aortic surgery [13]. Though, it is a mandatory step when SAEF is suspected because of the inevitably fatal outcome if surgical treatment is not promptly instituted. In the literature, different treatments of IAF are described. Per some authors, complete graft removal and performing of an axillobifemoral bypass is mandatory [6]. This is a very traumatic surgery especially in patients who are already compromised and the risk of aortic stump disruption is 20% [9]. For other authors, partial graft removal associated with radical debridement of perigraft infected tissues is sufficient, especially when the infection is remote from the aortic stump. Chiche et al. presented a case of partial graft removal with in situ rifampin-bonded graft reconstruction [9]. Their patient follow-up was negative for recurrent infections after 17 months. Another strategy supported by some authors [14] could be an endovascular approach. The main problem about this technique is the risk of infection from inserting prosthetic material into an infected field. In the paper by Danneels et al., 70% of all patients studied had a recurrent infection or new AEF within 1 year after an endovascular approach [15]. Their conclusion was that, especially in high-risk patients, endovascular treatment of AEF should be a short bridge to surgery.

Conclusion

SAEF must be suspected in patients with rectal bleeding and a history of aortic surgery. IAF is a rare variation of this complication with similar clinical presentation and diagnosis. Treatment could be less invasive than the classical axillobifemoral bypass. A partial graft excision, debridement of perigraft infected tissues and reconstruction with a new graft could be sufficient. The long-term outcomes need to be studied.

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

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