Radiology Case Reports 18 (2023) 169–172

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Case Report

Necrotizing fasciitis of the thigh due to a secondary aortoduodenal fistula ☆

Fahd Khefacha, PhD[a,], Aouini Fatma, PhD[a], Amel Changal, PhD[b], Raja Taieb, PhD[a], Chaima Chihaoui, PhD[a], Haifa Jenni, PhD[a], Achraf Saaidi, PhD[a], Mohamed Bechir Khalifa, PhD[b], Nabil Ben Romdhane, PhD[a]*

[a] *Department of Peripheral Vascular Surgery, Military Hospital Of Tunis, Tunisia*
[b] *Department of General Surgery, Military Hospital Of Tunis, Tunisia*

ARTICLE INFO

Article history:
Received 15 September 2022
Revised 20 September 2022
Accepted 25 September 2022

Keywords:
Blood vessel prosthesis
Necrotizing fasciitis
Retroperitoneal space
X-ray computed tomography
Duodenum

ABSTRACT

Background: Secondary aortoenteric fistula is an iatrogenic complication after aortic reconstructive surgery presenting with gastrointestinal bleeding and/or infectious symptoms. Infrequently, it may manifest with nonspecific and atypical clinical signs. We present a case of necrotizing fasciitis of the thigh complicating secondary aortoduodenal fistula, diagnosed with CT-scan. *Case presentation:* A 67-year-old man with a history of an open aortic-bifemoral bypass 6 years ago was admitted for a progressively swollen and painful right thigh for the last month. Through laboratory and morphological (CT-scan) investigations, a secondary aortoduodenal fistula associated with necrotizing fasciitis of the right thigh was discovered. After general supportive care and empiric antibiotherapy, the patient underwent a prosthetic explantation, a resection of the perforated bowel with end-to-end anastomosis, and extensive debridement of the necrotic tissue of the thigh. No revascularization has been attempted. The patient died the next day of multiple organ failure. *Conclusion:* Secondary aortoenteric fistula is rare but with a poor prognosis. Clinical presentation is not always typical. A high index of suspicion is the most important factor for improving outcomes. There is not a consensus about optimal management. Axillo-bifemoral revascularization and subsequent graft removal seem to be the best therapeutic option.

Introduction

Secondary aortoenteric fistula (SAEF) is a rare but serious iatrogenic complication after aortic reconstructive surgery. The most common presenting features are upper gastrointestinal bleeding and infectious symptoms. Infrequently, it may manifest with nonspecific and atypical clinical signs.

We present herein a case of a secondary aortoduodenal fistula presenting as necrotizing fasciitis of the thigh. Consent for the publication was obtained from the family.

Abbreviations: SAEF, Secondary aorto-enteric fistula.

☆ Competing Interests: All authors declare no conflict of interest.

* Corresponding author.
E-mail address: fahdkh90@gmail.com (F. Khefacha).

https://doi.org/10.1016/j.radcr.2022.09.086

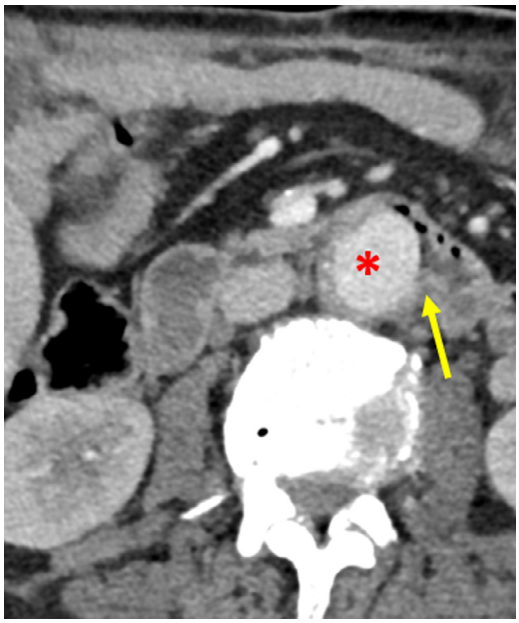

Figure 1 – CT-scan with intravenous injection of contrast medium showing the migration of the aortic prosthesis (asterisk) into the duodenum with breach into the posterior duodenal wall (arrow).

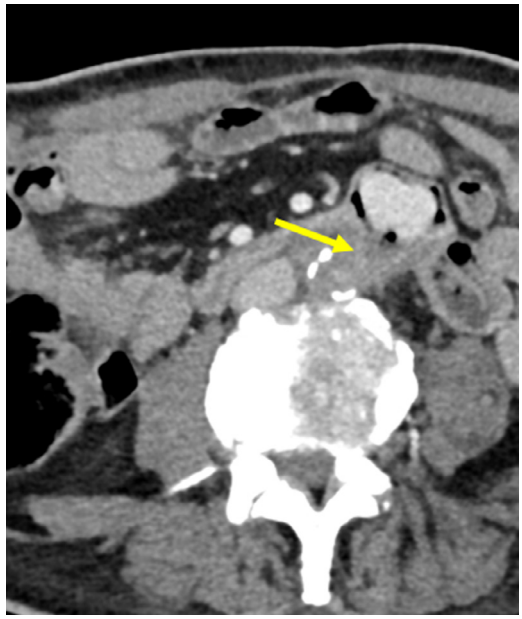

Figure 2 – CT-scan with intravenous injection of contrast medium showing the fluid and gas (yellow arrow) surrounding the aortic prosthesis.

Case presentation

A 67-year-old man presented with a progressively swollen and painful right thigh for the last month. His past medical history included cigarette smoking, hypertension, coronary artery angioplasty, and an open aortic-bifemoral bypass (Dacron) 6 years ago because of Leriche syndrome. He also reported intermittent subjective fever, low back pain, and weight loss but denied abdominal pain, nausea, vomiting, or melena. On admission, he exhibited signs of hypothermia of 36.5°C, chills, and he was unable to walk. The physical findings revealed tenderness and swelling of the right thigh without erythema or crepitus. His vital signs were: a blood pressure of 130/58 mm Hg, a heart rate of 140 bpm, and a respiratory rate of 20/min. Lower extremity pulses were present and the abdomen was soft. Blood test evaluation showed an elevated white blood cell count of 40,000/mm^3 (normal ranges [4000-10,000/mm^3]), an elevated C-reactive protein of 320 mg/L (normal range ≤5 mg/L), a hemoglobin level of 7.2 g/dl (normal ranges [12-16/g/dL]), creatinine of 53 μmol/L (normal ranges [44-80 μmol/L]), creatinine clearance of 143.1 mL/min, and urea of 7.2 g/L (normal range ≤8,3 mmol/L). Given the presence of the aortic prosthesis, a secondary fistula was suspected and the patient underwent a CT-scan with intravenous contrast which demonstrated an aortic prosthesis migrated into the 4th duodenum associated with an infiltration of the retroperitoneal fat (Fig. 1). It was surrounded by fluid and gas (Fig. 2), with downward extension into the thigh around the femoral artery and involvement of the adjacent muscles (Fig. 3). Moreover, the CT-scan showed a 240 × 110 mm fluid and gaseous collection between the anteromedial and posterior compartments of the right thigh (Fig. 4). The diagnosis of a secondary

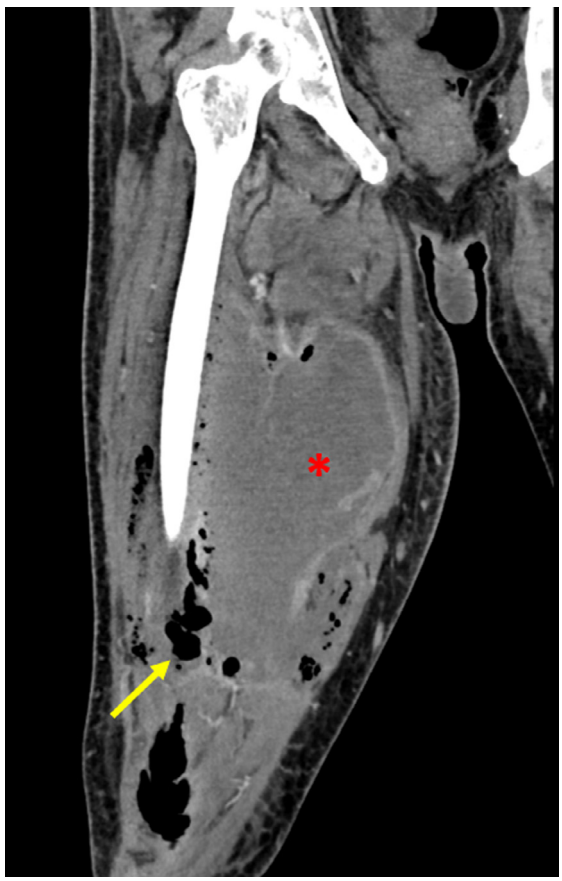

Figure 3 – CT-scan with intravenous injection of contrast medium showing downward extension of fluid (asterisk) and gas (arrow) into the thigh.

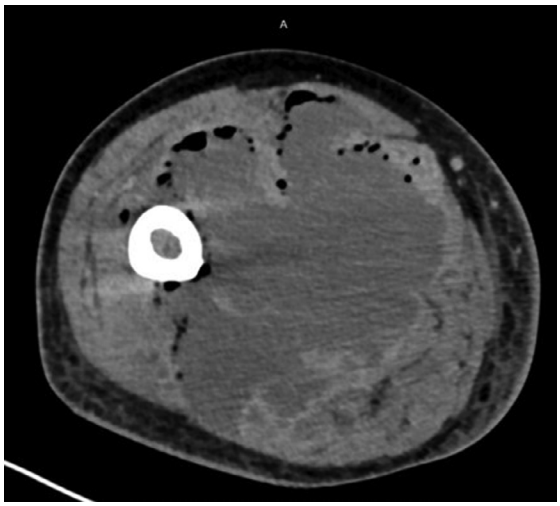

Fig. 4 – CT-scan with intravenous injection of contrast medium showing the fluid and gas collection within the deep layers of the thigh.

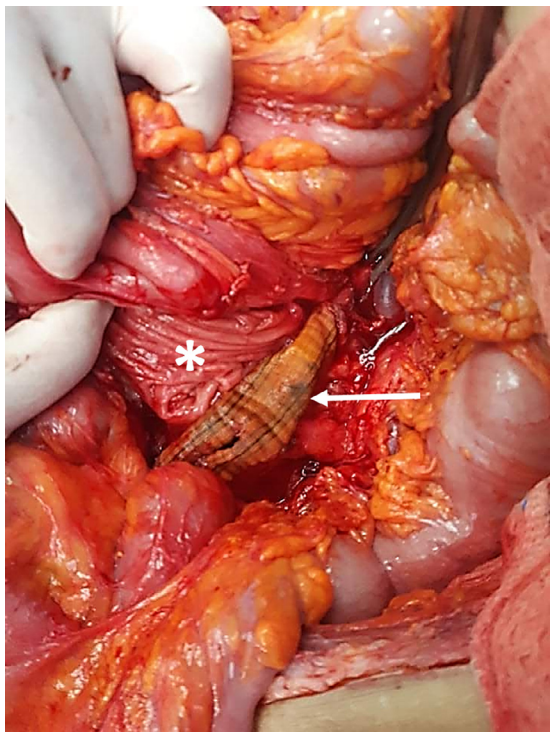

Fig. 5 – Biliary contaminated aortic prosthesis (arrow) perforating the Treitz angle. The asterisk showed the duodenal mucosa.

aortoduodenal fistula with direct spread to the right thigh was considered. The patient was started empirically antibiotherapy with Imipenem + Cilastatin (3 g/day), Teicoplanin (800 mg/day), and Amikacin (1.5 g/day) and general supportive treatment. He had a blood transfusion and underwent an urgent laparotomy. Intraoperative exploration found an infected prosthesis migrated into the angle of Treitz with large ulceration on its posterior wall (Figs. 5 and 6). There was no direct communication between the aorta lumen and the bowel

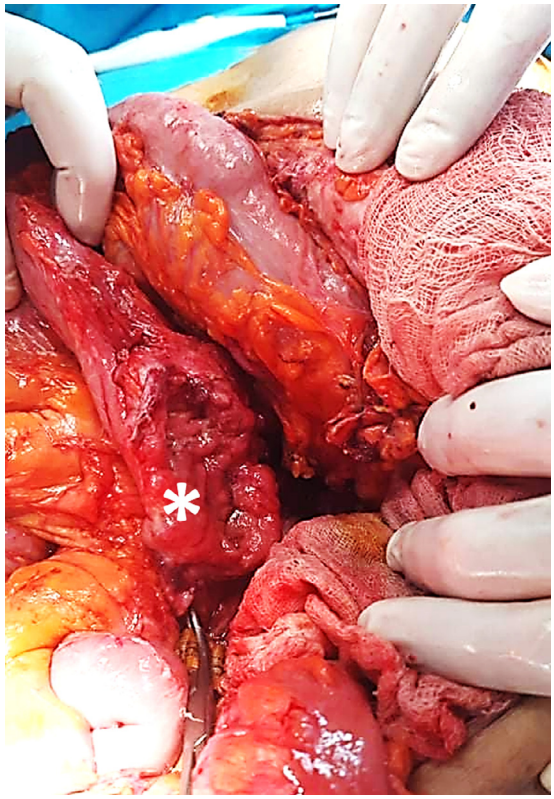

Fig. 6 – Intraoperative aspect of the duodenal perforation (asterisk).

lumen. A prosthetic explantation, a resection of perforated bowel with end-to-end anastomosis between the 3rd duodenum and the first jejunal loop, and an extensive debridement of the necrotic tissue of the thigh were performed (Fig. 7). Because of local and general conditions, axillo-bifemoral revascularization was not possible. We decided to not proceed any further. Corrugated rubber drains were kept in the abdomen behind the anastomosis and at the level of the thigh. The patient died the next day due to multiple organ failure. The culture of the abscess fluid of the right thigh grew *Escherichia Coli*, and the culture of the prosthetic graft grew *Escherichia Coli*, *Klebsiella Pneumoniae*, and *Enterococcus Faecalis*.

Discussion

SAEF is a complication of aortic reconstructive surgery even in the absence of aortic stent-graft placement [1]. The first case of SAEF was described in 1953 [1]. It is a rare event with an incidence within the vicinity of 1% [1,2] but with a high mortality rate of 57% [2]. Its pathogenesis is multiple and includes the extension of infection from an infected graft to a suture line, bowel wall trauma and/or ischemia during emergency graft insertion, mechanical injury, bowel lumen invasion due to pseudo aneurysms or paragraft abscesses, which may be facilitated by prosthesis-induced inflammation [3]. Continuous pulsating pressure of the graft on the bowel wall, especially among patients with uncontrolled hypertension, may be involved too [4]. The entire gastrointestinal tract may be

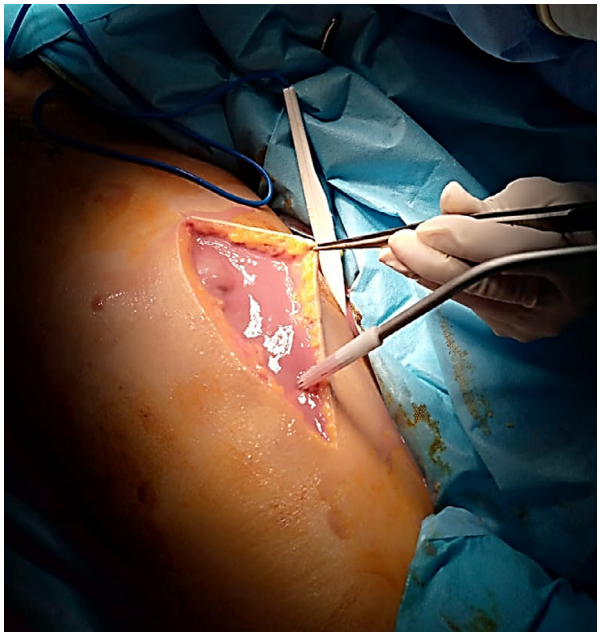

Fig. 7 – Intraoperative aspect of the pus into the thigh.

involved. However, the duodenum is the most affected site for anatomic reasons [1,2]. Bergqvist and Björck reported the possibility of several aortoenteric fistulae in the same patient [2].

The most common signs are infectious signs or gastrointestinal bleeding [2]. The latter may vary from herald bleeding to a state of severe hemorrhagic shock [5]. Our patient denied gastrointestinal bleeding but was anemic on admission. This may be explained by so-called herald bleeding, which is present in half of the cases [2] and can be explained by mucosal bleeding or a clot occlusion of a small fistula [3]. Atypical and nonspecific features may be encountered, such as abdominal or groin mass, visceral or muscle abscesses, limb ischemia, septic arthritis, or weight loss [3,5]. To the best of our knowledge, this is the first case of a SAEF associated with necrotizing fasciitis of the thigh from a direct spread.

Because of these diverse and potentially misleading features, the diagnosis is usually delayed [2,3,5]. A review of 194 papers showed that the diagnosis delay varies from hours to 3 years, with the median being 4 days [2]. In our case, the diagnosis was delayed for 1 month before it developed into necrotizing fasciitis of the thigh.

The main diagnostic tools are CT-scan with intravenous contrast and endoscopy and depend on the hemodynamic state [3]. CT-scan is the test of choice because of its availability, short acquisition time, and high resolution [1]. Pathognomonic signs are ectopic gas, focal bowel wall thickening, breach of the aortic wall, and extravasation of contrast material into the bowel lumen [1]. Endoscopy can demonstrate the fistula and exclude alternative causes of bleeding [3,6]. The confirmation of the diagnosis is therefore made by surgical exploration [3]. However, the most important tool to achieve the right diagnosis is clinical suspicion [2–6]. Bergqvist and Björck reported a few negative laparotomies because the possibility of SAEF was not considered [2].

Yet, there are no established guidelines for the repair of the SAEF [5]. Several treatment modalities are described in the literature [2]. Open aortic repair seems better than endovascular stent placement [1]. Axillo-bifemoral revascularization and subsequent graft removal seem to have the best outcomes. Our patient did not have revascularization. In the literature, 20% of patients did not have a reconstruction [2]. Endovascular management in high-risk patients may lower perioperative morbi-mortality rates [1] but does not seem optimal as a final solution because the bowel defect is not adequately dealt with [2]. It can be proposed as a bridging procedure [2]. Bowel repair consists of primary closure, resection with anastomosis, or resection with diversion [1]. Because of the instability of the patient, we considered the simplest solution.

Conclusion

Secondary aortoenteric fistula is a rare but serious event with a poor prognosis. Clinical presentation is not always typical, and a high index of suspicion is the most important factor to improving outcomes. There is not a consensus about optimal management. Axillo-bifemoral revascularization and subsequent graft removal seem to be the best therapeutic options.

Patient consent

A written consent was obtained from the next kin of the patient.

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