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Endovascularly treated superficial femoral artery aneurysm rupture secondary to

_Campylobacter fetus_ bacteremia: A case report

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ABSTRACT:

Degenerative aneurysms of the superficial femoral artery (SFA) are relatively rare and often recognized when they become symptomatic such as rupture. Infected SFA aneurysms are much rarer, especially those caused by *Campylobacter fetus* bacteremia. We report a case of a 67-year-old woman referred to our hospital owing to the presence of a painful reddish swelling on her left thigh. A huge SFA aneurysm rupture was diagnosed, and endovascular treatment with covered stent was performed. *C. fetus* was detected in the blood culture thereafter, and antibacterial therapy was successfully performed without any additional surgical interventions. She remained well without any evidence of indolent infection 19 months after the endovascular treatment. The endovascular approach with appropriate prolonged antibacterial therapy would be a feasible alternative for managing selected infected aneurysm cases.
INTRODUCTION

Degenerative true aneurysms of the superficial femoral artery (SFA) are relatively rare and usually not diagnosed until they reach a considerable diameter or become symptomatic. Aneurysm rupture is one of the most common symptoms. We present the case of a 67-year-old woman referred to our hospital with the diagnosis of left thigh cellulitis, which turned out to be an infected SFA aneurysm rupture due to Campylobacter fetus bacteremia.

Written informed consent was obtained from the patient for the publication of this report and any accompanying images.

CASES REPORT

A 67-year-old woman was referred to our hospital due to the presence of painful reddish swelling on her left thigh with the diagnosis of cellulitis. Eleven days prior to the admission, she had low-grade fever and diarrhea. High fever up to 38.8 °C had persisted intermittently thereafter and she had severe pain on her left thigh 3 days...
prior to the admission. Her medical history included hypertension, hyperlipidemia, and diabetes mellitus, and she is a current smoker.

On the day of admission, her temperature was 38.0°C, and the laboratory findings were as follows: white blood cell count: 15,900/mm³, hemoglobin: 11.6 g/dL, C-reactive protein: 20.57 mg/dL, procalcitonin: 0.28 ng/ml, and HbA1c: 7.3%. No bacteria were detected in the blood culture. A physical examination confirmed the presence of a subcutaneous hematoma throughout her left thigh (Fig. 1). A contrast-enhanced computed tomography (CT) scan revealed a ruptured SFA aneurysm with a mural thrombus of 58 mm in diameter and an extended hematoma (Fig. 2).

Contralateral SFA was within normal limits, and no abdominal aortic aneurysm was observed.

We proceeded to aneurysm exclusion with a self-expandable covered stent (GORE®, Viabahn®) (Fig. 3A, B). Intravenous antibiotics were administered with sulbactam sodium/ampicillin sodium (SBT/ABPC) 3 g 4 times daily (QID) until postoperative day 6. Her postoperative course was uneventful, inflammation markers were improved, and she had been afebrile. She was discharged on postoperative day 8.
On postoperative day 17, she was readmitted to our hospital owing to high fever (temperature up to 39.0°C), and a contrast-enhanced CT scan revealed the severe inflammation around the covered stent (Fig. 4). Empiric treatment was initiated with intravenous cefazolin (CEZ) 1 g 3 times daily. However, *C. fetus* was detected in the blood culture, which is resistant to CEZ. Therefore, we immediately switched to intravenous ABPC 2 g QID for 2 weeks. The level of inflammatory markers decreased without any invasive management. A follow-up contrast-enhanced CT scan revealed significantly improved inflammation (Fig. 5), and antibiotics were switched to chronic oral amoxicillin for 1 year. She remained well without any evidence of indolent infection 19 months after the endovascular treatment.

**DISCUSSION**

The incidence of isolated degenerative femoral artery aneurysms is reported to be approximately 0.005% (1). They represent only 3% of all peripheral aneurysms, and 80% of them have been reported to be in the common femoral artery, 15% in the SFA, and 5% in the deep femoral artery (2-5). Most of SFA aneurysms are located in the
middle-distal third of the SFA. Therefore, SFA aneurysms are less likely to be palpable and recognized, and the majority of them are not detected until they reach a considerable diameter or patients exhibit any symptoms (3, 6). Moreover, an SFA aneurysm is usually caused by iatrogenic or other traumas, or conditions related to inflammatory and infectious processes, or connective tissue diseases (7, 8). Representative rheumatologic diseases related to vasculitis or aneurysms include Bechel and nodular polyarteritis; however, our case did not have stomatitis aphthosa, genital ulcers, and any skin diseases. She also had no signs of other connective tissue disorders or vasculitis. In addition, the rarity of an SFA aneurysm without any traumas or symptoms, such as rupture, acute ischemia, or limb edema due to compression on the deep vein, makes the diagnosis much challenging.

Since infected aneurysms were first described by Osler (9), they have been reported to constitute up to 3% of aneurysms, and most of them are the abdominal aorta and femoral artery aneurysms (10, 11). Immune status is considered to be an important factor in the formation of an infected aneurysm (12). Atherosclerosis allows bacterial colonization in the arterial intima, resulting in a local immune response and the activation
of T-cells, which release cytokines that increase protease activity (10, 13). The reduced structural integrity of the artery, due to breakdown of elastin and collagen, has been considered to result in the formation of an infected aneurysm (10, 13).

It is crucial to obtain a microbiological diagnosis for managing a critical course of an infected aneurysm. *Staphylococcus aureus* is a common causative pathogen, and enteric organisms such as *Salmonella* species, *Escherichia coli*, and *Pseudomonas aeruginosa* have also been isolated (14-19).

As of now, more than 15 different *Campylobacter* species have been identified (20). *C. fetus*, comprising two species, has been recognized as a major species causing extraintestinal lesions (20) and bacteremia, particularly in elderly and immunocompromised patients (21, 22). The most common clinical symptoms are diarrhea and cellulitis; however, endovascular infection is serious and relatively frequent compared to infection caused by other *Campylobacter* species (21, 22). *C. fetus* has been reported to have a tropism for vascular endothelium or arterial destructive potential particularly where preexisting damage is preexisting (23). Farm animals are considered to be the major reservoir of *Campylobacter* species, and the major cause of bacteria-
related food poisoning and foodborne gastrointestinal infections. Our patient had no
contact with any animals and had not eaten any suspicious food, except for a properly
heated chicken dish before the onset of diarrhea. She also did not have any signs of
peripheral artery disease or aneurysms, and was not even under an immunocompromised
status, except for diabetes. The reason behind the bacterial colonization occurred in the
SFA still remains unclear.

Currently, complete resection of the infected aneurysm with adjacent tissues
and autologous reconstruction is the gold standard to manage infected aneurysms. The
prosthetic graft was considered to be avoided or resected if used. However, even if
aggressive debridement was performed, reinfection could occur at a relatively high rate
(24). Recently, the strategy of in situ preservation of the infected graft has also been
reported. (25-27). Endovascular aneurysm repair (EVAR) has been introduced as an
alternative for infected abdominal aortic aneurysms (IAAAs); this repair is particularly
essential in the case of severely ill or elderly patients. Endovascular treatment was
considered to be a temporary treatment followed by definitive open repair, which was
therefore called a “bridge to open surgery.” However a European multicenter trial showed
the durability of EVAR by assessing late infection-related complications and long-term survival. The study concluded that EVAR can be a durable treatment option for most patients (15). Additionally, another recent study showed acceptable long-term outcomes of EVAR for IAAAs (16). Appropriate antibacterial therapy with suitable duration would be the most definitive factor for this treatment.

* C. fetus is typically very sensitive to ampicillin, third-generation cephalosporins, aminoglycosides, imipenem, and meropenem (21, 28, 29); therefore, our antibiotics alone strategy was acceptable even after the second admission with the presence of a prosthetic device. As for the duration of postoperative antibacterial therapy for infected aneurysms to prevent indolent infection, although some authors recommended a minimum of 1–2 months, recent reports have claimed that 6 months or even a lifelong use may be required (15, 30, 31). We consider that prolonged antibacterial therapy is necessary, and oral amoxicillin was administered for 1 year. If *S. aureus* or *Candida species* was the causative pathogen, the lifelong treatment with antibiotics may be required. The duration should be considered depending on the pathogens, and continuous follow-up should be done even after discontinuing the antibacterial therapy.
Arterial destruction is often observed in the presence of arterial infection, especially in the presence of the prosthetic devices (32, 33), and even without the presence of devices (34, 35). Biofilm constructed around the prosthetic devices may inactivate antibiotics or would prevent antibiotics from penetrating, making infection control difficult (36). However, depending on the bacteria or the materials of the prosthetic device, antibiotics alone strategy could be a feasible option in selected cases (36-39).

We should have considered the potential indolent infection at the time of initial treatment from several clinical signs, and should have used a prolonged antibiotic therapy. The diarrhea 2 weeks before the first admission should have made us consider *C. fetus* infection, which was resistant to CEZ. No bacteria were detected in the first blood culture; therefore, deciding the appropriate antibiotics and their dosage and duration was difficult. However, even though an infection was apparent in the first place, endovascular treatment with appropriate antibacterial therapy could be a feasible alternative for managing infected aneurysms.

We consider that an emergent case, such as ours, could be treated endovascularly, and even in an elective case, it could be tried unless the bacteria is multi-
drug resistant. However, clinicians should be prepared to perform appropriate drainage or surgical intervention in case the infection becomes uncontrollable. If a multi-drug resistant bacterial infection was apparent in the first place, surgical repair should be the first choice.

Another problem is that the use of a covered stent use for SFA or popliteal artery aneurysms is not covered by the health insurance in Japan. Analysis of further cases will be needed to ensure that kind of treatment is effective and feasible.

The findings from our case highlight the rarity of a *C. fetus*-infected aneurysm and the efficacy of endovascular treatment with appropriate antibacterial therapy for infected aneurysms.

**CONCLUSION**

We observed a rare case of *C. fetus*-infected SFA aneurysm rupture. Endovascular treatment with prolonged antibacterial treatment might be a feasible alternative to treat selected infected aneurysms.
We all have no conflict of interest to declare.
Fig. 1 A huge hematoma on the patient’s left thigh.

Fig. 2 Contrast-enhanced computed tomography scan shows a ruptured superficial femoral artery (SFA) aneurysm with a mural thrombus of 58 mm in diameter and an extended hematoma on the left thigh. Contralateral SFA was within normal limits.

Fig. 3A The initial angiogram shows a ruptured superficial femoral artery aneurysm.

Fig. 3B The final angiogram shows a complete exclusion of the aneurysm.

Fig. 4 Contrast-enhanced computed tomography scan shows severe edema and inflammation around the covered stent.

Fig. 5 Contrast-enhanced computed tomography scan shows significant improvement in inflammation.
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