

[CASE REPORT]

An Infected Massive Persistent Sciatic Artery Aneurysm Treated by an Aneurysmal Incision and Drainage after Angiographic Embolization

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Abstract:

A man in his 80s undergoing chronic hemodialysis presented with a high fever. A 10-cm soft mass was palpable in his right buttock. Abdominal computed tomography and angiography showed an incomplete-type unilateral persistent sciatic artery aneurysm (PSAA) with gas patterns and a blood flow through the aneurysm. Incision drainage was performed after arterial embolization. Gram staining of the hematoma showed gram-positive cocci that had formed chains, thus leading to a diagnosis of an infected PSAA. Drainage by incision after arterial embolization was used as the therapeutic method of choice for a massive infected PSAA with a sustained blood flow in order to prevent sciatic nerve injury and bleeding associated with PSAA resection.

Key words: infected persistent sciatic artery aneurysm, incomplete-type, sustained blood flow, incision drainage, arterial embolization

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Introduction

A persistent sciatic artery (PSA), which is characterized by a failure to regress with femoral artery development in the embryonic phase, is very rare, with an incidence of only 0.025-0.04% (1). PSA is divided into two types: complete-type PSA (70-80%), which is connected to the popliteal artery, and incomplete-type PSA (20-30%), which is not connected to the popliteal artery. With incomplete-type PSA, the superficial femoral artery (SFA) is connected to the popliteal artery and it also supplies its blood flow (2). While PSA may be observed from 6 months to 89 years of age (1), the average age associated with occurrence of certain complications, including lower limb ischemia (31-63%) and aneurysm formation (45-50%), is 57 years of age (3, 4). PSA is prone to develop an aneurysm due to either an inherent collagen defect or hypoplasia of the connective tissue in the primitive arterial wall (2, 5). Repeated compression can subsequently result in the development of a PSA aneurysm

(PSAA) (6, 7). In addition to the pain caused by the aneurysm itself or neuralgia due to compression of the sciatic nerve by the aneurysm, PSAA can also cause ischemic manifestations due to thrombotic occlusion of the lower limb arteries (2-4, 8), thus resulting in the amputation of the lower limb in 18% of such patients (9). Furthermore, a previous report has focused on a few cases of a ruptured PSAA with a maximum diameter of ≥ 5 cm (4). PSAA complicated with a bacterial infection (infected PSAA), which is extremely rare and has only been reported in a few case reports (8, 10-13), tends to grow rapidly and rupture more easily due to the fragility of the aneurysm wall caused by bacterial inflammation. Such cases therefore require rapid intervention (14).

We herein report a case of unilateral massive infected PSAA with a maximum diameter of 10 cm in an elderly patient undergoing chronic hemodialysis due to terminal-stage chronic kidney disease (CKD) caused by type 2 diabetes mellitus (DM). The patient's condition improved after drainage of the infected hematoma by aneurysmal incision after

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transcatheter arterial embolization (TAE) with the subsequent long-term administration of antibiotics, which was performed to prevent severe bleeding, instead of performing highly invasive surgery to completely remove the infected aneurysm and surrounding tissue which would thus have required revascularization.

Case Report

A man in his 80s undergoing chronic hemodialysis, owing to terminal-stage CKD caused by type 2 DM, presented with a fever of $>38^{\circ}\text{C}$ 8 days which had initially been observed 8 days previously. Since then, he had tried to avoid touching his right buttock against a chair or against the floor when sitting. He had not undergone any invasive dental care within several months before admission. Blood tests performed at his previous hospital 5 days before admission showed a white blood cell (WBC) count of $4.9 \times 10^3/\mu\text{L}$ and a C-reactive protein concentration of 8.9 mg/dL. Abdominal computed tomography (CT) without contrast enhancement showed a mass lesion with a calcified lesions in the wall in the right buttock, whose maximum diameter was 8 cm, without revealing the definite origin of fever (Fig. 1a). Ceftriaxone (CTRX) (1 g/day) had been administered 4 days prior to presentation without any improvement, so he was therefore transferred to our hospital. On admission to our hospital, the patient's Glasgow Coma Scale classification was E4V4M6, body temperature was 37.3°C , pulse rate was 71 beats/min, blood pressure was 140/66 mmHg, respiratory rate was 19 breaths/min, and oxygen saturation was 100% in room air. Upon physical examination, an apical systolic murmur was auscultated, and a soft, non-pulsatile mass lesion with a diameter of approximately 10 cm with mild tenderness was palpable in the right buttock. No signs of conjunctival petechial hemorrhaging, Osler nodes, Janeway lesions, dental caries, or periodontitis were detected. Laboratory findings (Table) showed a WBC count of $6.0 \times 10^3/\mu\text{L}$ (neutrophils: 83.5%), a C-reactive protein concentration of 9.4 mg/dL, a D-dimer concentration of 9.2 $\mu\text{g}/\text{mL}$, and a rheumatoid factor concentration of 106 IU/mL. Abdominal CT without contrast enhancement on admission showed the same mass lesion in the right buttock. The densities of the inside of the mass were heterogenous with some gas patterns. The maximum diameter of the mass lesion had increased to 10 cm, which clearly showed a tendency of the aneurysm to increase in size (Fig. 1b). Subsequently contrast enhancement revealed the mass to be a massive PSAA. The early phase of contrast enhancement showed the presence of slightly and irregularly enhanced lesions at the deepest part in the mass (Fig. 1c). In addition, a delayed phase examination revealed definite enhancement of the wall (Fig. 1d). The tendency of rapid growth of its size, the presence of gas, and clear enhancement of the wall of the aneurysm highly suggested a diagnosis of an infected PSAA. We subsequently performed an angiographic examination, which revealed a sustained blood flow through the PSAA (Fig. 2a).

The PSAA was classified as an incomplete-type PSAA due to a lack of a direct connection with the popliteal artery, with the right SFA being apparently connected to the popliteal artery (Fig. 2b). Thus, TAE was performed to stop the blood flow inside the PSAA to prevent serious bleeding without any adverse outcomes, such as necrosis of the gluteal muscle and lower-limb ischemia. Thereafter, the administration of CTRX (2 g/day) and metronidazole (750 mg/day) was started after blood culture specimens were obtained. On the 5th hospital day, we performed drainage of the right infected gluteal hematoma by aneurysmal incision, revealing a mixture of large amounts of old and fresh hematoma (Fig. 3a, b). We avoided highly invasive surgery to remove the infected PSAA with surrounding tissues, since such procedures would have required vascular reconstruction, considering the possibility of serious complications, such as massive bleeding or sciatic nerve injury (11). Instead, we tried to drain the infected aneurysm as far as possible through the incision. Although cultures of both the hematoma and blood were negative, probably due to previously administered antibiotics, gram staining of the hematoma showed gram-positive cocci that had formed chains with a large number of neutrophils (Fig. 4). As a result, we made a definite diagnosis of unilateral infected PSAA. Transthoracic echocardiography (TTE) on the 3rd hospital day and transesophageal echocardiography (TEE) on the 17th hospital day revealed only trivial mitral regurgitation and mild tricuspid regurgitation without any heart valve vegetations. Abdominal CT with contrast enhancement on the 11th, 18th, and 36th hospital days showed a consecutive decrease in the size of the hematoma. Antimicrobial agents were changed to intravenous ampicillin/sulbactam (3 g/day) on the 10th hospital day, followed by oral amoxicillin/clavulanate on the 38th hospital day. Because the patient's general condition thereafter improved, he was transferred to another hospital for further recuperation on the 44th hospital day. We prescribed a prolonged course of oral antibiotics (at least 3 months), with the duration of the treatment modified in accordance with imaging findings. After 3 months of antibiotic therapy, follow-up abdominal CT confirmed the disappearance of fluid accumulation within the infected-PSAA, and thus the antibiotic therapy was stopped; there was no exacerbation detected after antibiotic discontinuation.

Discussion

PSA is a very rare condition with an incidence of 0.025-0.04% (1). Lower-limb ischemia due to thrombotic occlusion of the feeding arteries (31-63%) and PSAA formation (45-50%), especially a ruptured PSAA with a maximum diameter of ≥ 5 cm, have been previously reported to have significant complications (4, 8, 9). PSA is susceptible to aneurysm formation due to either an inherent collagen defect or hypoplasia of the connective tissue in the primitive arterial wall (2, 5). Repeated compression can subsequently result in PSAA. In the present case, the PSAA likely developed be-

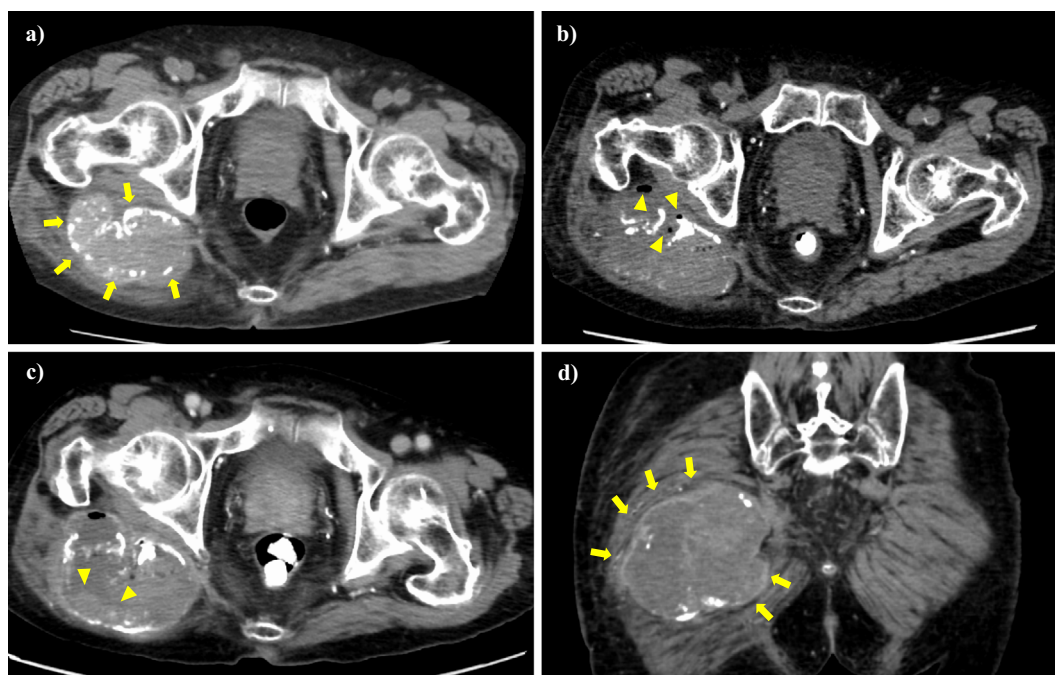


Figure 1. The findings of abdominal computed tomography. (a) Axial imaging without contrast enhancement 7 days before admission: Abdominal computed tomography (CT) without contrast enhancement taken at the previous hospital shows a mass lesion with a maximum diameter of 8 cm with calcified lesions in the wall (arrows). (b) Axial imaging without contrast enhancement on admission: The maximum diameter of the mass lesion has increased to 10 cm, showing a tendency to increase in size. The densities of the inside of the mass are heterogeneous with some gas patterns (arrowheads). (c) Axial imaging with an early phase of contrast enhancement on admission: The early phase of contrast enhancement shows a slight and irregular enhancement of the most dorsal part in the aneurysm (arrowheads) without any extravasation. (d) Coronal imaging with delayed phase of contrast enhancement on admission: The delayed phase of the abdominal CT with contrast enhancement shows enhancement of the wall of the aneurysm (arrows).

Table. Laboratory Data on Admission.

Complete blood count		Biochemistry	
White blood cell	6.0 ×10 ³ /μL	Total protein	6.4 g/dL
Neutrophil	83.5 %	Albumin	2.8 g/dL
Lymphocyte	4.5 %	BUN	99.1 mg/dL
Red blood cell	245 ×10 ⁴ /μL	Creatinine	10.5 mg/dL
Hemoglobin	8.6 g/dL	Total bilirubin	0.4 mg/dL
Hematocrit	26.4 %	Glucose	209 g/dL
MCV	107.8 fL	AST	16 U/L
MCH	35.1 %	ALT	18 U/L
MCHC	32.6 %	LDH	225 U/L
Platelet	17.4 ×10 ⁴ /μL	ALP	103 U/L
Congestive System		CK	67 U/L
PT-INR	1.1	Sodium	139 mEq/L
APTT	36.6 s	Potassium	3.7 mEq/L
Fibrinogen	825.0 mg/dL	Chlorine	96 mEq/L
D-dimer	9.2 μg/mL	CRP	9.4 mg/dL

MCV: mean cell volume, MCH: mean corpuscular hemoglobin, MCHC: mean corpuscular hemoglobin concentration, PT-INR: prothrombin time-international normalized ratio, APTT: activated partial thromboplastin time, BUN: blood urea nitrogen, AST: aspartate aminotransferase, ALT: alanine aminotransferase, LDH: lactate dehydrogenase, ALP: alkaline phosphatase, CK: creatine kinase, CRP: C-reactive protein

cause of repeated compression of the PSA from the patient regularly sitting on it in his daily life. However, reports on PSAA complicated with bacterial infection (infected PSAA) are extremely scarce (8, 10-13). Furthermore, only two cases of massive infected PSAA that were as large as that reported in the present case have previously been reported (10, 13). Because mycotic aneurysms are generally prone to rupture due to their rapid increase in size (14), the infected PSAA in the present case was naturally considered to be at an extremely high risk of rupture. Therefore, although infected PSAA is extremely rare, it is desirable to treat PSAA, especially when its maximum diameter is ≥5 cm in patients with pre-existing conditions, which increase susceptibility to infection (4).

An infected aneurysm is generally caused by the hematogenous dissemination of infectious microemboli, intimal injury of blood vessels due to infectious organisms circulating in the blood stream, invasion from an adjacent infection, or direct bacterial entry into the blood stream at the time of vascular injury, including those that occur during some medical procedures (14). Our patient was obviously a compromised host because he had been undergoing chronic hemodialysis due to terminal-stage CKD caused by type 2

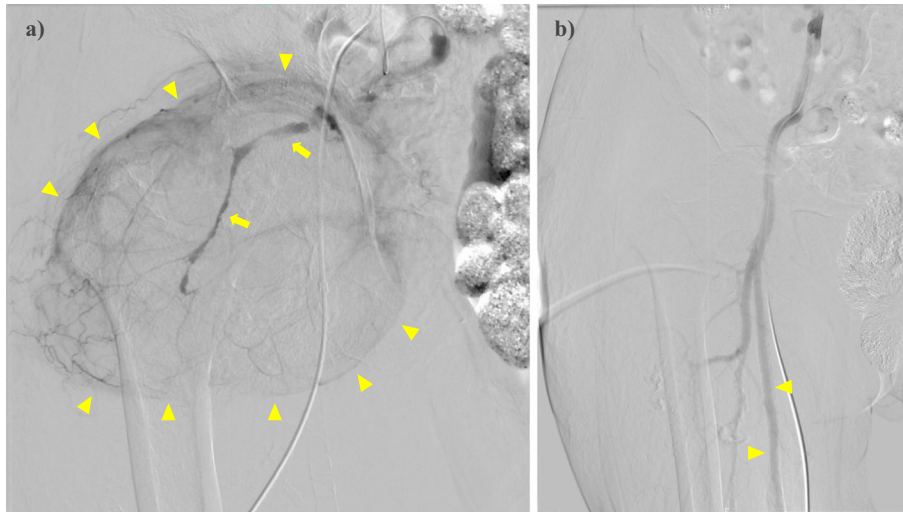


Figure 2. An angiographic examination of the right persistent sciatic artery on admission. (a) Angiography of the right persistent sciatic artery shows the presence of an extremely massive persistent sciatic artery aneurysm with a maximum diameter of 10 cm (arrowheads) with a sustained blood flow through the aneurysm (arrows). (b) An incomplete-type persistent sciatic artery is present, which lacks a connection to the right popliteal artery, with the right superficial femoral artery (arrowheads) apparently being connected to the popliteal artery.

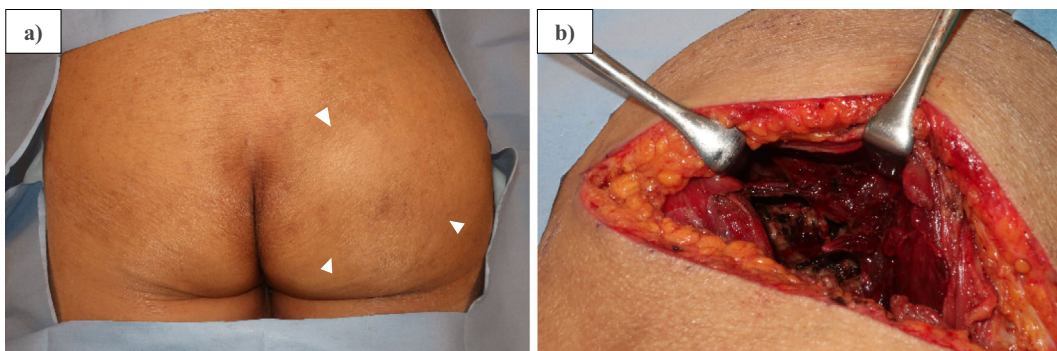


Figure 3. Preoperative and intraoperative findings of infected hematoma drainage by the incision of the right persistent sciatic artery aneurysm. (a) Before incision: Physical findings of the right buttock before the incision for drainage. The right buttock was swollen with mild tenderness (arrowheads). (b) After incision: A mixture of large amounts of old and fresh hematoma in the aneurysm is shown.

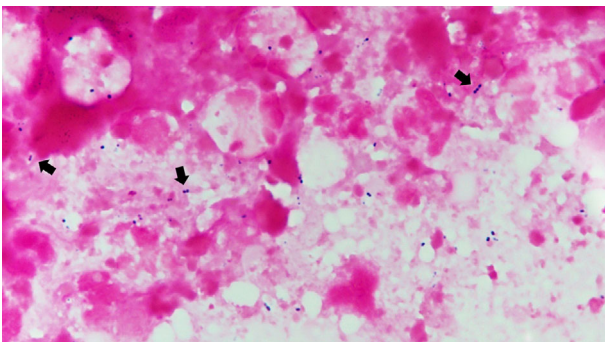


Figure 4. The findings of gram staining of drainage specimen of infected hematoma ($\times 1,000$ microscopic field). Gram staining of the drainage specimen from the hematoma, obtained by the incision of the right persistent sciatic artery aneurysm, showed gram-positive cocci compatible with *Streptococcus* spp. (arrows).

DM; thus, he was therefore extremely susceptible to bacterial infection. The causative bacterium was assumed to be *Streptococcus* spp. from the findings of gram staining, without yielding any clear findings by blood culture due to previously administered antibiotics (15, 16). We were unable to perform polymerase chain reaction to detect bacterial 16S rDNA. The route of bacterial invasion was suspected to be direct invasion at the time of vascular injury accompanying hemodialysis, based on the lack of any abnormalities at other possible sites of invasion, such as the skin and the soft tissue surrounding the right buttock, or the oral cavity. However, as the causative bacteria was *Streptococcus* spp., which is part of the oral bacterial flora, it is also possible that bacteria from the oral cavity had entered the blood stream and damaged the PSAA wall, even without a history of any invasive dental procedures or dental diseases (such as peri-

odontitis or dental caries). In addition, infective endocarditis (IE) may have contributed to the development of infected-PSAA in the present case. Because our patient fulfilled the four minor modified Duke criteria (m-DC), including fever, underlying heart disease, rheumatoid factor, and mycotic aneurysm (17), our patient would have been diagnosed with “definite” IE according to the m-DC if the blood cultures had been positive. However, the blood cultures failed to yield any bacteria due to previously administered antibiotics (15, 16), and TTE and TEE failed to reveal any cardiac valve vegetations.

Generally, mycotic aneurysms are treated surgically by extensive aneurysmal resection with the resection of surrounding infected and necrotic tissues, followed by vascular reconstruction (18). However, adhesion of surrounding tissues, including the sciatic nerve or relatively large blood vessels with infected PSAA, is likely to be present, which could lead to serious complications, such as severe bleeding or sciatic nerve injury, including troublesome neuralgia (11). One case of infected-PSAA was successfully treated by drainage of infected hematoma through a small incision and this treatment method was reported to be one of the useful and much less invasive treatment strategies without requiring revascularization surgery (13). Because the infected PSAA of our patient was massive (maximum diameter: 10 cm), gravely invasive surgical resection of the aneurysm and surrounding infected tissues with vascular reconstruction was highly likely to cause serious bleeding or sciatic nerve injury. Thus, we performed drainage of the infected PSAA through aneurysmal incision as the main treatment strategy for our patient. Although an angiographic examination showed the presence of a sustained blood flow in the infected PSAA, we were able to address this problem with TAE to prevent serious bleeding. Fortunately, incomplete-type PSA rather than complete-type PSA was present in this case; thus, it was not connected to the popliteal artery (2). After drainage, the residual infected PSAA gradually decreased in size over time after the consecutive administration of antimicrobial agents, without requiring any additional surgical procedures. Because the control of the infection by drainage of the infected PSAA through an aneurysmal incision may be insufficient without complete aneurysmal resection or resection of the surrounding infected or necrotic tissues, the long-term administration of antimicrobial agents is essential (13). A case of massive infected PSAA controlled by the long-term administration of antibiotic agents after sufficient drainage of an infected aneurysm through a small incision has been previously reported (13). The duration of antibiotic administration has not yet been clearly defined, but a duration of >6 months is recommended after the surgical treatment of a mycotic aneurysm (19). Despite this, there is one case report of an extremely massive infected PSAA requiring antibiotics for >12 months after drainage (13). A lack of definitive evidence due to an extremely small number of reported cases means that the duration of antimicrobial agent administration must be determined on a case-by-

case basis, especially in patients not undergoing the complete removal of infected tissues by radical surgery. In these patients, long-term antibiotic use may therefore be required.

Conclusion

The drainage of an infected PSAA through an aneurysmal incision after TAE with the subsequent long-term administration of antimicrobial agents to prevent serious bleeding may therefore be a useful treatment strategy for massive infected PSAA, which requires rapid intervention due to a high risk of rupture and a tendency to demonstrate rapid growth. This approach can help to avoid the complications associated with highly invasive resection of PSAA, which requires revascularization surgery.

Because the patient had dementia, written informed consent was obtained from one of the relatives responsible for this patient, namely his daughter.

The authors state that they have no Conflict of Interest (COI).

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