Case Report

J Korean Soc Radiol 2020;81(6):1517-1522
https://doi.org/10.3348/jksr.2020.0029
pISSN 1738-2637 / eISSN 2288-2928

대한영상의학회지

Bilateral Persistent Sciatic Arteries Complicated with Aneurysmal Dilatation and Thrombotic Occlusion: A Case Report

동맥류와 혈전성 폐색이 동반된 양측 잔류좌골동맥: 증례 보고

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Persistent sciatic artery (PSA), a rare congenital vascular anomaly, increases susceptibility to aneurysms and accounts for 40–61% of the cases. Here, we describe a case of PSA in a 70-year-old man with a history of alcoholic liver cirrhosis. Bilateral complete PSAs were detected incidentally on computed tomography angiography during evaluation for spontaneous intramuscular bleeding in the thigh due to cirrhosis-related coagulopathy. Selective angiography of the left PSA revealed aneurysmal dilatation and thrombotic occlusion of the PSA, which was partially removed with aspiration thrombectomy. Intramuscular bleeding was succesfully managed with empirical embolization of the deep femoral artery.

Index terms Lower Extremity; Computed Tomography Angiography; Endovascular Procedures; Aneurysm; Thrombosis; Congenital Abnormalities

Received February 26, 2020
Revised March 29, 2020
Accepted April 17, 2020

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INTRODUCTION

Persistent sciatic artery (PSA) is a rare congenital vascular anomaly. The incidence of PSA is 0.03–0.06% in the general population, and the mean age of detection is 57 years (1). It is strongly associated with aneurysms, which can be complicated by thromboembolisms, radiculopathy, or rupture of the lower extremity arteries (2). We present a case of bilateral PSAs, which were found incidentally during the evaluation of intramuscular bleeding of the thigh in a patient with cirrhosis-related coagulopathy.

CASE REPORT

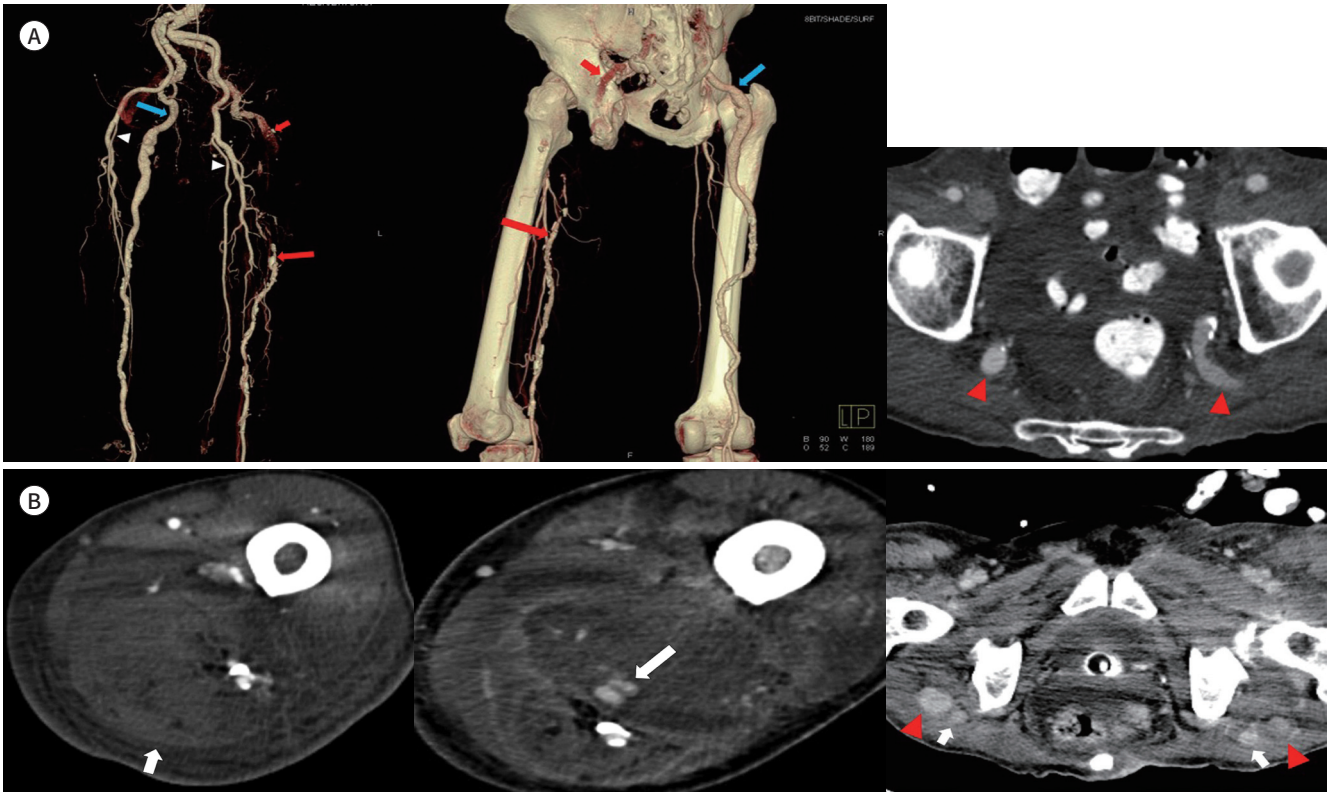
A 70-year-old man with alcoholic liver cirrhosis presented with swelling and bruising over the posterior left thigh. Physical examination revealed a normal left femoral arterial pulse but decreased left popliteal and dorsalis pedis arterial pulses. Laboratory investigations revealed anemia, with a hemoglobin level of 6.1 g/dL (normal, 13.0–16.5 g/dL); thrombocytopenia, with a platelet count of 43000/μL (140000–400000/μL); and increased prothrombin time, with an international normalized ratio of 1.48 (0.87–1.12). The coagulopathy was interpreted as a consequence of the underlying liver cirrhosis.

We did a computed tomography angiography (CTA) for lower extremity to identify the cause of the left thigh swelling. The equipment used was: Somatom Definition AS+ (128 slices; Siemens Medical Solutions, Erlangen, Germany); 120 kV/474 effective mAs;, slice thickness 2 mm; contrast material dose 120 cc; injection speed 4 mL/sec. The CTA incidentally revealed bilateral PSAs. These anomalous arteries originated from the bilateral internal iliac arteries, passed through the sciatic foramina and the posterior thighs, and finally communicated with the popliteal arteries. We observed aneurysmal dilations of the PSAs from the exits of the pelvis to the lesser trochanters. Furthermore, we also noted long segmental thrombotic occlusion of the left PSA (Fig. 1A). The superficial femoral arteries (SFAs) were hypoplastic bilaterally and did not reach the popliteal arteries, but we noted multiple collateral vessels around the left femoral veins. Therefore, we diagnosed the condition as type 2a PSA according to the Pillet-Gauffre classification. Hematomas were observed in the left biceps femoris and adductor muscles without active bleeding focus on CTA (Fig. 1B, 1st). We did an arteriography to find the focus of bleeding of the intramuscular hematomas; however, an active focus of bleeding was not detected.

Anemia and thrombocytopenia did not improve after the patient had received transfusion of 4 units of packed red blood cells, 4 units of fresh frozen plasma, and 6 units of platelet concentrate. Moreover, the patient presented with an increase of approximately 1 cm in the left thigh circumference. Two weeks after the initial CTA and arteriography, we did a follow-up CT venography (CTV) to find the venous source of bleeding. The equipment used was the same as CTA. The protocols for CTV was: 120 kV/541 effective mAs;, slice thickness 5 mm; contrast material dose 120 cc; injection speed 3 mL/sec. The CTV revealed increased size and multifocal enlargement of the hematomas and venous contrast extravasation in the hematomas on delayed phase (Fig. 1B, 2nd).

The patient was referred to the interventional radiology department for the management of intractable intramuscular bleeding of the thigh. Angiography of the left external iliac artery showed no definite focus of bleeding in the left upper (Fig. 1C, 1st) and lower thigh. Reconstitution of the left PSA via left femoral artery branches at mid thigh was noted. Angiography of the left PSA revealed aneurysmal dilatation of the proximal part at the level of the pelvis, with many thrombi in the dilated portion (Fig. 1C, 2nd). First, we planned to do aspiration thrombectomy to recanalize the obstructed segment of the PSA and empirical embolization of the SFA and deep femoral artery (DFA). A small amount of thrombi was removed in the proximal portion of the obstructed PSA. However, most of the thrombi remained; these were organized thrombi (Fig. 1C, 3rd).

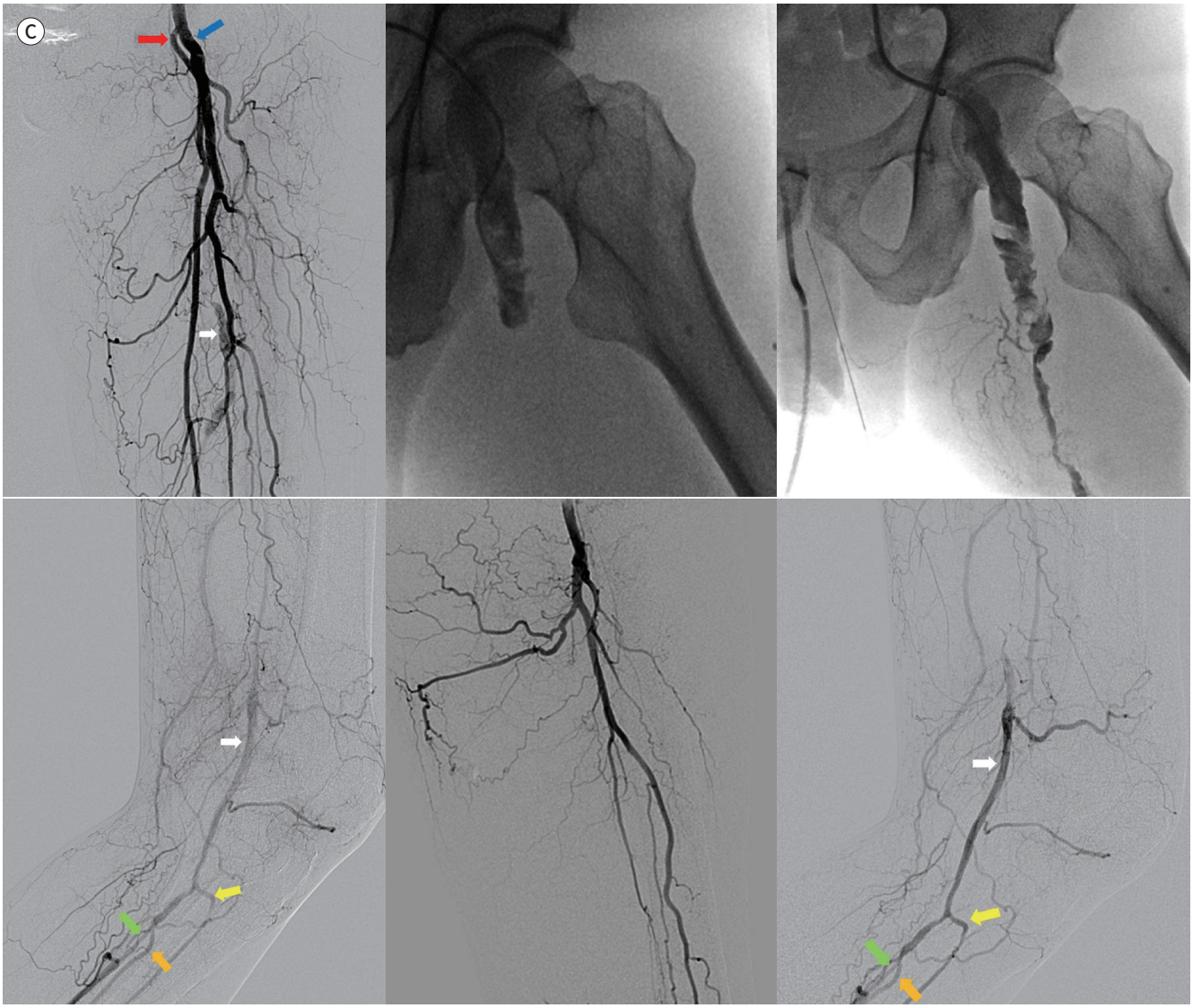
Fig. 1. A 70-year-old man with bilateral PSAs, incidentally detected on CTA and treated with angiographic embolization.
A. Initial CTA with the 3D volume-rendering technique with and without bone subtraction (1st and 2nd) shows bilateral PSAs (arrows) with incompletely developed SFAs (white arrowheads). Right PSA (blue arrows) shows aneurysmal dilatation. Left PSA (red arrows) shows proximal aneurysmal dilatation and long segmental thrombotic occlusion (short red arrows) with reconstitution of flow at the mid-thigh level (long red arrows) via multiple collateral branches from the left femoral arteries. The axial image (3rd) reveals aneurysmal dilations of both PSAs (red arrowheads) at the level of the greater sciatic foramen.
B. Initial CTA shows a hematoma (arrow, 1st) in the biceps femoris muscle without evidence of active bleeding. Subsequent arteriography shows no focus of bleeding (not shown). Despite the transfusion therapy, the patient's signs and symptoms worsened. Follow-up CT venography shows venous contrast extravasation (arrow, 2nd) in the hematoma on delayed phase. Also, the sciatic arteries (red arrowheads, 3rd) and veins (white arrows, 3rd) are still visible.
CTA = computed tomography angiography, PSAs = persistent sciatic arteries, SFAs = superficial femoral arteries



Second, we planned to do an empirical embolization of the left DFA with preservation of SFA supplying below-the-knee arteries. The delayed phase of angiography of the left SFA showed multiple collaterals along the course of the left SFA. The popliteal artery and the below-the-knee arteries were reconstituted at the distal thigh level (Fig. 1C, 4th). Successful embolization of the left DFA was carried out with Gelfoam particles (1500–2000 μm; Pharmacia & Upjohn, Kalamazoo, MI, USA) (Fig. 1C, 5th). After the embolization of the left DFA, the left SFA angiography showed patent SFA and below-the-knee arteries (Fig. 1C, 6th). A day after the embolization, the circumference of the left thigh decreased by approximately 1 cm, and the hemoglobin level was maintained without transfusion. The patient had an uneventful course because of hypovolemic shock secondary to rectal ulcer bleeding two months after the embolization.

Written informed consent could not be obtained from the patient, because he had died before this manuscript was written. The Ethics Committee of our institution has waived the approval.

Fig. 1. A 70-year-old man with bilateral PSAs, incidentally detected on CTA and treated with angiographic embolization.
C. Angiography of the left external iliac artery shows no definite bleeding foci in the left upper (1st) or lower (not shown) thigh. Reconstitution of left PSA (white arrow, 1st) via the left femoral artery branches at the mid-thigh level is noted (red arrow in 1st, SFA; blue arrow in 1st, DFA). Pre-thrombectomy angiography of the left PSA (2nd) shows aneurysmal dilatation of the proximal PSA with multiple filling defects in the dilated portion, suggesting thrombi, and long segmental occlusion at the upper-thigh level of the left PSA. Post-thrombectomy angiography (3rd) of the left PSA still shows a partially obstructed segment. Delayed-phase angiography (4th) of left SFA shows no definite bleeding foci. Note the collaterals along the course of the left SFA. The popliteal artery (white arrow, 4th) and below-the-knee arteries (yellow arrow in 4th, posterior tibial artery; orange arrow in 4th, anterior tibial artery; green arrow in 4th, peroneal artery) are reconstituted at the distal-thigh level. Angiography of the left DFA (5th) shows no definite bleeding foci. We performed empirical Gelfoam embolization of the left DFA (not shown). Post-embolization angiography of the left SFA (6th) shows patent SFA and below-the-knee arteries (yellow arrow in 6th, posterior tibial artery; orange arrow in 6th, anterior tibial artery; green arrow in 6th, peroneal artery) with reconstitution of the popliteal artery (white arrow, 6th) at the distal-thigh level.
CTA = computed tomography angiography, DFA = deep femoral artery, PSAs = persistent sciatic arteries, SFA = superficial femoral artery



DISCUSSION

The sciatic artery is an embryologic axial artery of the lower limb that originates from the umbilical artery. It usually runs along the dorsal aspect of, and provides the arterial supply

to, the lower limb bud during early development (2, 3). In the third trimester of gestation, the femoral artery matures into a major artery and communicates with the popliteal artery; then the sciatic artery begins to involute. The anomaly in this process determines the type of PSA (from type 1 to type 5), according to the Pillet-Gauffre classification (2, 4). Type 1 represents complete PSA with normal SFA. Type 2 refers to complete PSA with incomplete SFA, which is subdivided into 2a and 2b. In type 2a, the SFA is present but does not reach the popliteal artery. In type 2b, the SFA is absent. Type 3 refers to incomplete PSA in which its proximal part has persisted with normal SFA, while type 4 refers to incomplete PSA in which its distal part has persisted with normal SFA. In type 5, the PSA originates from the median sacral artery, which is subdivided into subtypes 5a and 5b according to the presence or absence of complete SFA (4).

Generally, multiple diagnostic tests are used. Recently, CTA has replaced conventional angiography, because CTA allows for a detailed evaluation of the relationship between PSA and surrounding vessels, nerves, and musculoskeletal structures, which can help surgeons to better plan the intervention; moreover, this procedure has low invasiveness (4, 5). In this case, CTA played a major role in the detection of PSA and in deciding on the treatment strategy.

There is no formal consensus about the management of PSA, since it depends on the individual symptoms, presence of aneurysm or vascular occlusion, and anatomic variations. Intervention is indicated if a PSA becomes symptomatic or aneurysmal (1, 6). It is reported that 40–61% of cases of PSA are complicated by aneurysms, which can develop into thromboembolism, radiculopathy due to sciatic nerve compression, and rupture (1, 4).

In the incomplete type of PSA, ligation or embolization of PSA is sufficient. In contrast, the complete type, in which the sciatic artery mainly supplies blood to the lower extremity, requires revascularization of the distal extremity and multiple surgical procedures, such as bypass, ligation, stent grafting, and amputation (7). However, the surgical procedure carries the risk of sciatic nerve injury because of the proximity. Endovascular interventions, such as coil or detachable balloon embolization and endovascular stent graft, can be used to avoid this risk (1, 7). In this patient, surgical treatment was not recommended, despite the complete type of PSA with aneurysmal changes and thrombosis, because of the underlying liver cirrhosis.

Van Hooft et al. (8) found that 20% of PSA patients were unexpectedly diagnosed during evaluations for other medical problems, such as varices, gun-shot injury, hemi-hypertrophy of the leg, and postoperative bleeding. To the best of our knowledge, this is the first report of unexpected diagnosis of PSA during the evaluation and management of spontaneous intramuscular bleeding in the thigh.

In conclusion, PSA is a rare congenital vascular anomaly strongly associated with aneurysm formation and occasionally detected as an incidental finding. PSA can remain asymptomatic for many years. Nonetheless, when detected as an incidental finding, the precise anatomic variant type and presence of complications, such as aneurysmal change of thrombosis, must be evaluated carefully to prevent life-threatening complications.

Author Contributions

Conceptualization, H.Y.; data curation, J.S.J.; investigation, J.S.J., H.Y.; methodology, J.S.J., H.Y.; project administration, H.Y., K.J.H.; resources, J.S.J., H.Y.; supervision, H.Y., K.J.H.; visualization, J.S.J., H.Y.; writing—original draft, J.S.J.; and writing—review & editing, H.Y., K.J.H.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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동맥류와 혈전성 폐색이 동반된 양측 잔류좌골동맥: 증례 보고

장서진 · 한윤희* · 권재현

양측 잔류좌골동맥은 드문 선천성 혈관 기형으로 동맥류를 잘 동반하며, 이는 사례의 40~61%에서 발생한다. 본 증례는 알코올성 간경화가 있는 70세 남자에서 발생한 좌골동맥 잔존의 1예이다. 간경화와 관련된 응고 병증으로 인한 대퇴부 출혈을 평가하기 위해 시행한 컴퓨터단층사진 혈관조영술 검사에서 양측 완전 좌골동맥 잔존이 우연히 발견되었다. 환자는 선택적 좌측 잔류좌골동맥 혈관조영술에서 좌측 잔류좌골동맥의 동맥류 및 혈전성 폐색이 있어 흡인 혈전제거술로 혈전 부분 제거를 시행하였으며 심부 대퇴 동맥의 경험적 혈관조영색전술을 통해 대퇴근육 내 출혈을 성공적으로 치료하였다.

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