Septic True Aneurysm of the Posterior Tibial Artery Diagnosed after Anterior Arthroscopic Debridement of a Septic Ankle following Infective Endocarditis: A Case Report

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Learning Point of the Article:
Magnetic resonance imaging should be performed before arthroscopic surgery to rule out aneurysm, especially in a patient with aseptic ankle and/or a history of infective endocarditis.

Abstract
Introduction: There have been reports of true aneurysm of the posterior tibial artery, but only three reports have described true aneurysm of the posterior tibial artery at the ankle, and there has been only one report of tarsal tunnel syndrome caused by true aneurysm of this artery. In this case report, we describe a rare case of true septic aneurysm of the posterior tibial artery presenting as tarsal tunnel syndrome which was found after anterior arthroscopic debridement of a septic ankle in a 55-year-old man.

Case Report: 13 years earlier, this patient had undergone aortic valve replacement for severe aortic regurgitation caused by infective endocarditis with aortic valve vegetations. Since then, the patient had been treated with the oral anticoagulant warfarin. The aneurysm was successfully treated by a saphenous vein graft and administration of antibiotics. The patient likely developed septic ankle and aneurysm as a consequence of infective endocarditis.

Conclusions: Magnetic resonance imaging should be performed before arthroscopic surgery to rule out aneurysm, especially in a patient with a septic ankle and/or a history of infective endocarditis.

Keywords: Anterior arthroscopy, infective endocarditis, posterior tibial artery, septic ankle, tarsal tunnel syndrome, true septic aneurysm.

Introduction
The incidence of joint sepsis has been estimated as 2–10 per 10,000 individuals annually [1]. The knee is the most common site of septic arthritis, followed by the shoulder, hip, and ankle [2]. Sepsis involving the ankle joint is rarer, accounting for only 3.4%–7% of all cases [3, 4, 5]. The most common causative organism in a septic ankle joint is Staphylococcus [6, 7]. Infrapopliteal aneurysms are uncommon but often appear as false aneurysms associated with trauma, infection, iatrogenic injury, changes in the collagen matrix, and inflammation [8, 9, 10, 11, 12]. True aneurysms of the posterior tibial artery are rare [13, 14, 15, 16, 17]. True aneurysms around the ankle are even rarer, with only three cases described in the English language literature [18, 19, 20]. Here, we describe the rare case of a 55-year-old man with tarsal tunnel syndrome and a history of surgery for infective endocarditis who was found to have a true septic aneurysm of the posterior tibial artery after anterior arthroscopic debridement of a septic ankle joint using standard anteromedial and anterolateral portals, in whom a reversed saphenous vein graft was successfully performed to reconstruct the damaged posterior tibial artery.

Case Report
A 55-year-old man presented to our university hospital complaining of general fatigue for the past several months. 13 years earlier, he had undergone aorta valve replacement at our...
hospital for severe aortic regurgitation caused by infective endocarditis with aortic valve vegetations and had been receiving oral warfarin as an anticoagulant since that time. He was 167 cm tall, weighed 60 kg, and had a body mass index of 21.7. Laboratory investigations revealed severe anemia with hemoglobin of 4.4 g/dL, so he was admitted to the internal medicine department of our hospital for blood transfusion. He also reported a 5-year history of untreated right ankle pain that had worsened in the previous month. During this hospitalization, he was referred to our department for the assessment of the right ankle pain. On physical examination, there were tenderness, local warmth, and redness over the right ankle, especially on the anterior aspect, and range of motion at the ankle joint was limited to 0° of dorsiflexion and 5° of plantarflexion. Laboratory investigations revealed evidence of inflammation (white blood cell count 8600 cells/µL and C-reactive protein 8.44 mg/dL). Clinical examination showed no features suggestive of congenital connective tissue disorder. Radiography and computed tomography images revealed osseous destruction and diffuse narrowing of the right ankle joint space (Fig. 1a and b and Fig. 2a, b, c). Synovial fluid culture from the right ankle was negative. After 18 days of hospitalization, when the anemia had improved (hemoglobin 10.7 g/dL), we performed anterior ankle arthroscopic surgery with heparin cover. The procedure included irrigation, extensive debridement, synovectomy with the removal of loose fragments, and curettage using standard anteromedial and anterolateral portals. Intraoperatively, dense fibrous tissue, aggressive hypertrophic synovitis, extensive severe chondral fibrillation, and subchondral erosion were seen in the arrowed ankle joint space (Fig. 3a and b). A sample of synovial tissue was obtained for culture and pathologic examination (Fig. 3c). Postoperatively, he was allowed to bear weight on his right foot as tolerated. Histological examination indicated chronic synovitis with infiltration of neutrophils and lymphocytes, angiogenesis, and accumulation of hemosiderin, suggesting chronic inflammation with infectious etiology (Fig. 4). Culture yielded Staphylococcus capitis (subspecies capitis), and appropriate antibiotic therapy was started based on the results of sensitivity. 6 days after ankle arthroscopy, the patient accidentally twisted the right leg, and his full body weight became suddenly supported entirely by the right foot. Soon afterward, he developed pain and swelling behind the medial aspect of the ankle (Fig. 5) and reported decreased sensation over the plantar aspect of the right foot. 7 days postoperatively, we noticed a painful pulsatile mass behind the medial malleolus. A Tinel’s-like sign was positive at the level of this pulsating mass, although motor function of the toes and ankle was normal. The patient was then referred to the cardiovascular surgery team at our hospital. Color and duplex Doppler ultrasonography showed dilation of the posterior tibial artery behind the medial malleolus (Fig. 6). Enhanced three-dimensional computed tomography showed a dilated posterior tibial artery that measured 25mm × 13mm (Fig. 7). The final diagnosis was true aneurysm of the posterior tibial artery with paresthesia due to nerve compression similar to tarsal tunnel syndrome. The decision was taken to perform arterial reconstruction to prevent rupture of the aneurysm and progression of the ankle pain.
swelling, and paresthesia. Thus, 19 days after arthroscopic surgery, cardiovascular surgery team performed reconstruction of the posterior tibial artery again under heparin cover. Curved incision was made along the posterior tibial artery behind the medial malleolus. The artery was found to be dilated (Fig. 8a) with severe adhesion between the aneurysm and the surrounding connective tissue, likely due to inflammatory reaction to infection. The expanded aneurysm was seen to be compressing the tibial nerve, but the flexor hallucis longus was intact, as was the tibial nerve, which excluded the possibility of the aneurysm in the posterior tibial artery being caused iatrogenically during the previous anterior arthroscopic debridement procedure. The posterior tibial artery was isolated and clipped proximally and distally, followed by resection of the aneurysm and interposition of a reversed saphenous vein segment while preserving blood flow in the posterior tibial artery (Fig. 8b, Fig. 9). 3 weeks after the vein graft surgery, the patient was allowed to bear weight on his right foot as tolerated. Histological examination of the aneurysm wall showed fibrosis, loss of elastic tissue, and formation of capillaries, along with infiltration of neutrophils and lymphocytes into the dilated aneurysmal wall with thrombus (Fig. 10), suggesting a true septic aneurysm that already existed before the anterior ankle arthroscopic surgery rather than a pseudoaneurysm caused during anterior arthroscopic debridement. Subsequently, the post-operative course was uneventful, and the pain and paresthesia on the plantar aspect of the foot disappeared. Antibiotic treatment was continued for 6 weeks after the vein graft surgery and white blood cell count decreased to 3300 cells/μL and C-reactive protein decreased to 0.05 mg/dL. The patient was discharged on antibiotics 4 weeks after the vein graft surgery, but still occasionally felt minor pain during prolonged activity and had a limited range of motion at the ankle. However, he unfortunately suddenly died because of acute ruptured infectious intracranial aneurysm after approximately 6 months after vein graft surgery.

Discussion

To the best of our knowledge, only three cases of true aneurysm of the posterior tibial artery at the ankle have been reported in the literature (Table 1) [18, 19, 20]. Our patient also had peripheral neuropathy, similar to tarsal tunnel syndrome, because of compression of the tibial nerve within the tarsal tunnel by a true septic aneurysm. The main etiologic causes of tarsal tunnel syndromes are the presence of a ganglion, osseous prominence with tarsal bone coalition, trauma, varicose veins, neuroma, hypertrophy of the flexor retinaculum, foot deformities, and idiopathic [21, 22, 23, 24, 25, 26, 27]. Recently, there have been reports of tarsal tunnel syndrome caused by pseudoaneurysm [28], suture material [29], posterior tibial vein aneurysm [30], aneurysm of a branch of the posterior tibial artery [31], and a perforating branch from the posterior tibial
artery [32]. However, to the best of our knowledge, only one case of tarsal tunnel syndrome caused by true aneurysm of the posterior tibial artery, which showed a positive Tinel’s-like sign similar to our case, has been described in the literature (Table 1) [19]. Open arthrotomy is often needed to effectively eradicate infection in a septic joint [4,5]. However, arthroscopy allows a direct magnified view of the intra-articular anatomy, joint lavage, synovectomy of septic pannus formation without the morbidity of extensive surgical incisions, more rapid recovery, and a lower incidence of iatrogenic injury, with outcomes similar to those of the more traditional open approach and fewer complications [33, 34, 35, 36, 37, 38]. Therefore, we opted to perform arthroscopic surgery to treat the septic ankle in our case. Arthroscopic surgery with concomitant antibiotics might be an acceptable treatment method for this condition. Infection weakens the arterial wall and an infected aneurysm enlarges [39, 40]. Infected aneurysms are one of the most serious complications of infective endocarditis [41]. The incidence of infected aneurysm is approximately 0.9%–1.3% among all cases of arterial aneurysm [42, 43]. Staphylococcus species are the most common pathogens involved in an infected aneurysm [43]. In this study, Staphylococcus species was detected in the ankle. Therefore, in a patient with endocardial vegetations, an infected embolus might be carried to the posterior tibial artery and the ankle joint, and the posterior tibial artery might enlarge as a result of bacterial invasion. Anderson et al. [44] and Johnson et al. [45] reported that arterial repair or vein grafting should be avoided when treating septic aneurysm because of a high incidence of persistent infection and late rupture. However, Patel et al. [46] and Sakai et al. [31] reported that autologous vein graft could be used to bypass an excised aneurysm, although infection-related graft failure remains a significant complication. Our patient had a very serious medical condition and there was a high probability of rupture of the aneurysm. Therefore, we performed a vein graft and kept the patient on long-term antibiotic therapy, as recommended by Patel et al. [46]. Fichelle et al. [47] and Heyd et al. [48] reported that medical treatment for an infected aneurysm requires at least 6 weeks of appropriate parenteral and/or oral antimicrobial therapy, and we treated our patient accordingly. In our patient, inflammatory markers, white cell count and C-reactive protein, remain within normal limits, indicating no recurrence until his death due to acute ruptured infectious intracranial aneurysm.

One of the limitations of this case report was the short follow-up period. At present, the patient is stable, the septic ankle is settled, but extensive open ankle debridement may be needed in the future. Another limitation is that magnetic resonance imaging was not performed before arthroscopic surgery. The aneurysm seemed to be preexisting and enlarged only after he supported his full body weight on the right foot. Due to thrombus or septic embolism at the distal level of the septic true aneurysm of the posterior tibial artery. In retrospect, we should have obtained magnetic resonance images before ankle arthroscopic surgery to check for aneurysm, given that the patient had a septic ankle joint and a history of infective endocarditis. Moreover, we should have taken brain MR images because cerebral complications are still frequent, occurring in up to 30% of infectious endocarditis patients, with stroke complicating 12% of endocarditis cases [49, 50, 51], and mortality approaches 80% in patients with ruptured infectious intracranial aneurysm [52].

**Conclusion**

We have encountered a rare case of true septic aneurysm of the posterior tibial artery found after anterior arthroscopic debridement of a septic ankle in a 55-year-old man who presented with tarsal tunnel syndrome and had undergone aortic valve replacement for severe aortic regurgitation caused by infective endocarditis with aortic valve vegetations 13 years earlier and had been receiving anticoagulant therapy since then. The aneurysm was successfully treated by a saphenous vein graft.

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**Table 1: Cases of true aneurysm of the posterior tibial artery around the ankle reported in the literature**

| Author, year | Number of cases | Age (years) | Sex | Comorbidity | Symptoms | Period of symptom | Etiology | Size of aneurysm | Treatment for PTA aneurysm |
|-------------|-----------------|-------------|-----|-------------|----------|------------------|----------|-----------------|-----------------------------|
| Danes et al., 2006 [18] | 1 | 60 | Male | Hypertension/diabetes-controlled DM | Painless swelling | 1 month | Mycotic | 1.12 cm | Reversed saphenous vein graft |
| Tshomba et al., 2006 [19] | 1 | 54 | Male | None | Painless swelling | 6 months | Atherosclerotic | 2.5×1.4 cm | End-to-end reconstruction (resection and end-to-end anastomosis) |
| Sagar and Button, 2014 [20] | 1 | 64 | Male | Diet-controlled diabetes | Painless swelling | Over 1 year | Unknown | 1.4 cm | Reversed vein graft |
| This case | 1 | 55 | Male | Aortic valve replacement for IE | Painful swelling | 1 day (until diagnosis) | Mycotic | 2.5×1.3 cm | Reversed saphenous vein graft |

DM: Diabetes mellitus, IE: Infective endocarditis, PTA: Posterior tibial artery
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Clinical Message
Magnetic resonance imaging should be performed before arthroscopic surgery to rule out aneurysm, especially in a patient with a septic ankle and/or a history of infective endocarditis.
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