Cystic adventitial disease of the popliteal artery with spontaneous regression

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ABSTRACT

Cystic adventitial disease (CAD) of the popliteal artery with spontaneous regression is rare. We describe a 44-year-old man with rapid onset of severe intermittent claudication who is currently undergoing conservative follow-up. CAD was diagnosed, and resection of the lesion with autologous vein replacement was scheduled. However, the claudication suddenly improved at 5 weeks after onset. Computed tomography and ultrasound revealed that the cystic lesion in the adventitia had nearly disappeared. This case report describes the imaging findings and the possibility of conservative treatment. CAD can occur in the popliteal artery and is usually treated surgically. However, reports of spontaneous regression are rare. We report our experience with a case of CAD that eventually spontaneously regressed. (J Vasc Surg Cases and Innovative Techniques 2018;4:136-9.)

CASE REPORT

A 44-year-old man had rapid onset of severe claudication after walking approximately 50 m. He was referred to our hospital because his ankle-brachial index (ABI) was 0.9 on the right and 1.2 on the left. The patient had no history of smoking or any other medical history or injury in the lower limbs. No abnormalities were found on a serum chemistry examination. We considered popliteal artery entrapment, Buerger disease, popliteal artery aneurysm, acute arterial embolism, and lower extremity vasculitis[1] in the differential diagnosis. Contrast-enhanced computed tomography (CT) revealed a cystic lesion in the right popliteal artery wall occupying the intravascular lumen (Fig 1) as well as multilocular cysts between the popliteal artery and femur (Fig 2, *1A-1C*). Therefore, he was diagnosed with cystic adventitial disease (CAD) because of these typical CT findings and was scheduled to undergo resection with vein replacement surgery.

The patient was hospitalized to prepare for surgery as scheduled 7 weeks after symptom onset. However, the claudication suddenly improved 5 weeks after initial presentation. Therefore, contrast-enhanced CT was repeated and revealed that the CAD had nearly disappeared. Whereas ring-thickened adventitia was observed at a more distal site, the popliteal artery was not stenotic and had no cystic compression (Fig 2, *2A-2C*). The multilocular cysts between the popliteal artery and femur were also less prominent (Fig 2, *2A* and *2B*), and the ABI was 1.2 on the right and 1.2 on the left. We therefore canceled the operation and decided on outpatient follow-up. CT performed 10 months later showed that the CAD had drastically diminished, and the multilocular cysts were also less prominent (Fig 2, *3A-3C*). During 24 months of follow-up, no marked changes were observed in the ABI, and the patient remained asymptomatic; conservative follow-up is therefore being continued.

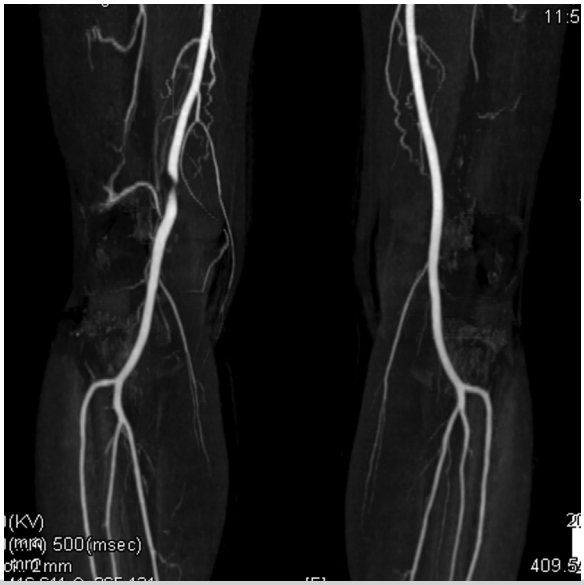

Fig 1. Contrast-enhanced computed tomography (CT) on the initial visit revealed a so-called hourglass appearance, which is a typical finding of cystic adventitial disease (CAD).

Informed consent was obtained from the patient for publication of this case report.

DISCUSSION

In 1947, Atkins and Key[2] first reported CAD in the external iliac artery. CAD is a relatively rare vascular

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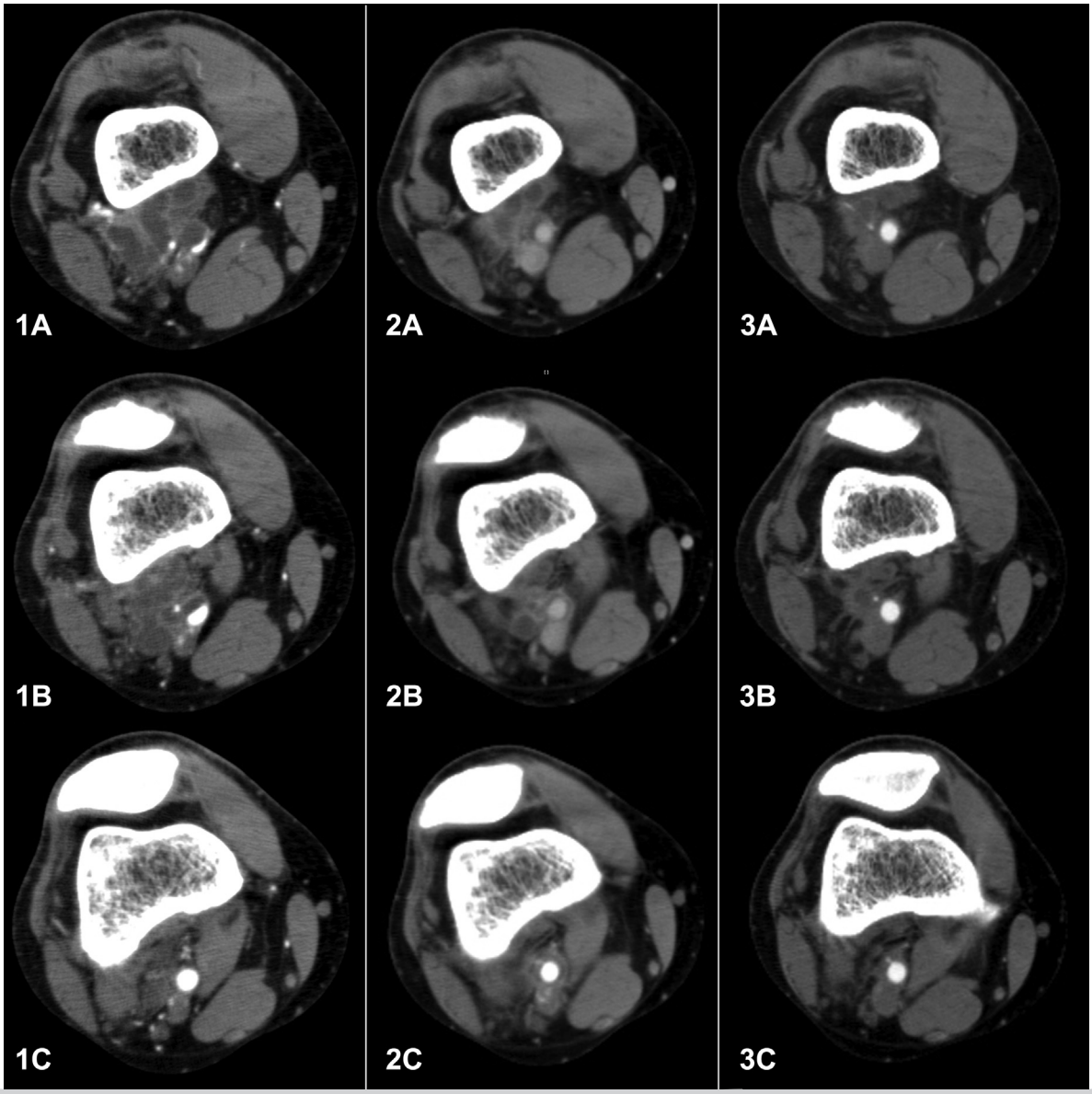

Fig 2. Contrast-enhanced computed tomography (CT) on the initial visit **(1A-1C)**. CT revealed a cystic lesion in the right popliteal arterial wall occupying the intravascular lumen as well as multilocular cysts between the popliteal artery and femur. Seven weeks later, contrast-enhanced CT was performed again, revealing that cystic adventitial disease (CAD) of the popliteal artery had nearly disappeared. Although ring-thickened adventitia was observed, the popliteal artery was not stenotic and now showed cystic compression. Furthermore, the multilocular cysts between the popliteal artery and femur had become less prominent **(2A-2C)**. Follow-up CT 10 months later showed that CAD had drastically diminished, and the multilocular cysts were also less prominent **(3A-3C)**.

disease in which a colloidlike substance accumulates between the adventitia and the media, resulting in stenosis or occlusion of the arterial lumen.[2] Whereas a number of cases have been reported, the etiology remains unknown. However, several theories have been proposed: the trauma theory,[3,4] the systemic disorder theory,[5,6] the developmental theory,[3-5] and the articular (synovial) theory.[7] The trauma theory implicates repetitive truma.[3,4] The developmental theory implicates mesenchymal cells that appear in the adventitia of the vessels during embryogenesis.[4,5] The prevailing opinion and current body of evidence largely support the developmental theory as the most rational explanation.[8,9] However, Desy and Spinner[7] supported the articular (synovial) theory, noting that the developmental theory could not explain CAD with an axial vessel. Moreover, they suggested that adventitial cyst formation begins with a capsular tear or defect that leads to the tracking

of synovial fluid along a vascular articular branch. Therefore, these theories will require further investigation and analysis.

Resection with vein replacement, surgical cyst excision, and imaging-guided cyst aspiration have been used in treatment. Percutaneous imaging-guided aspiration is associated with initial success,[10] but recurrence due to rapid enlargement of CAD after aspiration has been reported.[11] Therefore, considering the likelihood of recurrence, resection with vein replacement is preferred.[3,5,9,12]

Owen et al[13] first reported a case of conservative follow-up in 1990. Eight cases of spontaneous regression, including ours, have been reported since then.[13-18] Reasons for regression were proposed in each case. Two studies suggested that the cystic contents were absorbed by surrounding tissue when the cyst collapsed because of increased internal pressure.[17,18] One report suggested that the mechanism of regression was likely related to a connection between the cyst and synovium.[19] Moreover, one author suggested that the cyst's content might have passed through the periarterial space or through a communication with the articulation of the knee.[16] The other four articles did not suggest explanations. Communication with the knee joint capsule through a ductlike structure was reported in one case.[19] No typical imaging findings or clinical course was observed in the eight cases as far as we simply reviewed the selected and published images. Therefore, the precise reason for spontaneous regression remains unclear. However, in five cases, including ours, rapid regression was seen within 5 weeks after onset.[14-16,18] In addition, spontaneous regression occurred in one case each at 10 months and 15 months after onset. However, claudication recurred 2 months after onset in one case, leading to resection with vein replacement.[19]

In our case, although CAD and multilocular cysts were present, CAD disappeared along with reduction in the peripheral cysts between the popliteal artery and bone. Communication with either the peripheral multilocular cysts or articular capsule was not noted on CT or magnetic resonance imaging in this patient (images not shown). However, we believe that the CAD's content may have passed through the periarterial space and through a communication with the articulation of the knee, and the CAD and multilocular cysts collapsed because of a rise in internal pressure. Ring-thickened adventitia was observed at a more distal site, and the popliteal artery showed no cystic compression and was not stenotic (Fig 2, *2A-2C*). When CAD communicates with the articular capsule, the mechanism of spontaneous resolution may be cystic rupture or drainage into the joint capsule.[20] We could not clarify the reason for spontaneous regression because communication between CAD and the knee was not found. Thus, even in cases in which communication between CAD and the articular capsule cannot be confirmed, the CAD in the popliteal artery may regress spontaneously in some patients, although this is rare. Even when CAD spontaneously regresses, however, long-term follow-up is mandatory because of reported recurrence after spontaneous regression.

CONCLUSIONS

Whereas reports of spontaneous regression are rare, this case demonstrated spontaneous regression within several weeks of onset, and CT revealed interesting changes in CAD along with changes in the peripheral cysts. It is preferable to plan for the treatment of CAD of the popliteal artery with spontaneous regression in mind.

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