CLINICAL IMAGES

Popliteal adventitial disease in a triathlete — a case report

S E Verkuijl, R V P de Villiers, D Revelas, D Pantanowitz

A 37-year-old recreational long-distance triathlete complained of right leg pain during uphill running since early 2002. The pain was cramp-like with tightness, loss of power and numbness in the foot. After rest running was possible for a short period. He had previously had three deep venous thromboses (DVTs) in the right leg, which leg showed signs of chronic venous incompetence with good peripheral pulsations. Factor V-Leiden had been demonstrated and treated with coumarine anticoagulants. The patient did not smoke.

In August 2002, during an Ironman triathlon, the patient was unable to run because of pain in the anterior compartment and numbness in the foot. Chronic anterior compartment syndrome was diagnosed. Duplex sonography revealed no arterial abnormalities and a grossly abnormal venous system. In September 2002 an anterior fasciotomy was performed; thereafter running was possible for 2 months, after which time the symptoms returned. In February 2003 lateral compartment syndrome was diagnosed and a lateral fasciotomy was performed, whereafter the patient was symptom-free for about 1 month. Investigations demonstrated incompetence of the right greater saphenous vein and several perforators. The greater saphenous vein was stripped in June 2003 to improve the venous outflow, but the symptoms remained.

Duplex sonography revealed a cystic structure measuring 21 mm x 14 mm in the right popliteal fossa, compressing the popliteal artery. The popliteal fossa was explored in November 2003 and a popliteal entrapment, caused by an aberrant insertion of the medial gastrocnemius head was demonstrated and resected. A cystic structure was not seen and a popliteal aneurysm was ruled out. Weeks afterwards the symptoms persisted, with disappearance of the pedal pulses after running, which symptoms worsened until walking uphill became impossible in December 2003. Duplex examination showed that the cystic structure was still present, compressing the popliteal artery during dorsiflexion. Magnetic resonance imaging (MRI) showed that the cyst was multiloculated, 2 cm x 3 cm in size, and in direct relation to the popliteal artery, which was narrowed by half (Figs 1 and 2). MR angiogram demonstrated a 1 cm stenotic segment of the popliteal artery and extensive collateral formation in the thigh and calf (Fig. 3).

In January 2004 the popliteal fossa was re-explored and a large adventitial cyst was found and excised. A femoro-popliteal bypass was necessary to bypass the damaged popliteal artery. Histological examination showed that the cyst was composed of a fibrous connective tissue wall, with evidence of myxoid degeneration.

Eight weeks after the bypass the patient finished an Ironman triathlon in 12 hours and 13 minutes, without experiencing any problems. The chronic eczema on his right ankle had healed completely for the first time since 1996. In the next 2 years he achieved personal best times over the Ironman distance as well as the standard marathon (sub 3 hours).

Discussion

Cystic adventitial disease of the popliteal artery is a rare cause of intermittent claudication, with less than 350 cases reported worldwide.[1] The aetiology includes repeated trauma, tracking of capsular synovial structures along vascular branches and incorporation of a joint-related ganglion-like structure into the developing vessel during embryological development.[1,2] It typically affects men between 20 and 50 years of age without risk factors for atherosclerotic disease.[2,3] Popliteal entrapment and cystic adventitial disease of the popliteal artery can occur together,[4] suggesting that popliteal entrapment can cause

Dr Sabine Verkuijl (MMed (Fam Med)) is a family physician, qualified in the Netherlands in 1995. She came to South Africa in June 1998 and worked in a rural district hospital in the former Transkei until October 2001. She is currently employed by the Health Systems Trust as Eastern Cape co-ordinator for the TASC Tuberculosis project. She enjoys middle and long distance triathlon.

Dr Richard de Villiers qualified as a radiologist in 1997 at the University of Stellenbosch, and is currently with Drs Van Wageningen and Partners, in Vergelegen and Stellenbosch Mediclinics. He is a consultant at the South African Sports Science Institute, and honorary consultant at Tygerberg Academic Hospital, both in Cape Town. His special interest is in sports imaging and musculosketetal radiology.

Dr Dimitri Revelas is a vascular surgeon at the Linksfield Park Clinic in Johannesburg and an honorary lecturer in surgery at the Helen Joseph Hospital.

Prof. Desmond Pantanowitz is a professor of surgery at the University of the Witwatersrand and Head of Surgery at the Helen Joseph Hospital in Johannesburg. He is a vascular surgeon with a particular interest in paragangliomas, vasculitis and adventitial cystic disease of the popliteal artery.

Corresponding author: *S E Verkuijl (sabine@hst.org.za)*

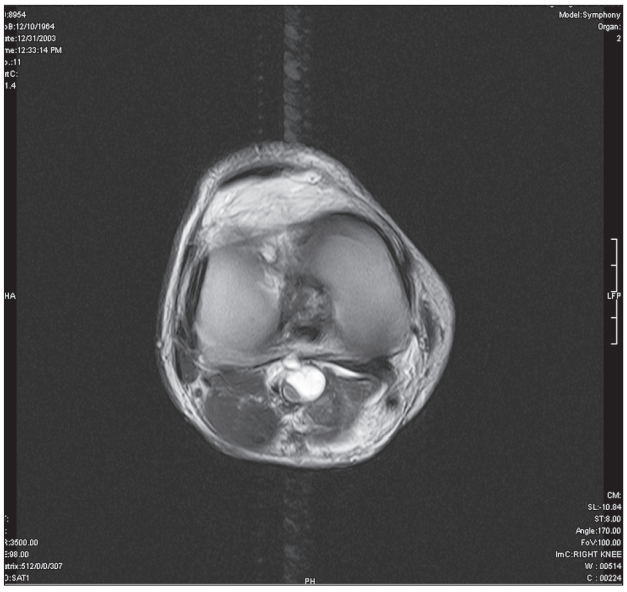

Fig. 1. MRI of the right knee showing the cyst compressing the popliteal artery.

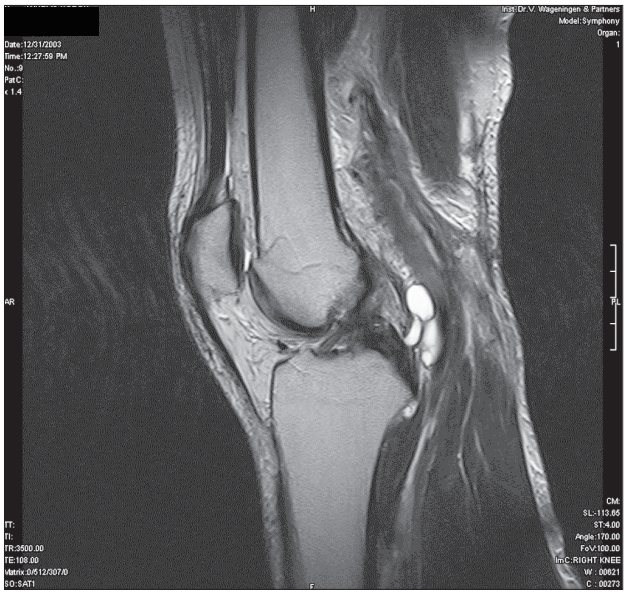

Fig. 2. MRI of the right knee showing the lobulated cyst in relation to the popliteal vessels.

adventitial cyst formation through micro-trauma. Our case illustrates that diagnosis is often difficult; symptoms developed gradually and the diagnostic focus was on the pre-existing venous outflow problems, while initially arterial disease was not demonstrated.

Chronic compartment syndrome (CCS) is also an uncommon cause of claudication in young adults, and venous insufficiency has been described as a cause of CCS. Symptoms associated with CCS include muscle cramping or swelling in isolated muscle groups with plantar paraesthesiae. Symptoms are often longstanding and disappear after extended rest, only to reappear when exercise is resumed. Functional popliteal entrapment has been described as one of the least common variants of CCS. Treatment of choice for CCS is fasciotomy.[5] In this patient the clinical history suggests that there was indeed a CCS before the arterial problem became clear. It is possible that the adventitial cyst compressed the popliteal vein which, together with the damaged venous system, resulted in the antero-lateral CCS. The aberrant insertion of the gastrocnemius muscle may have contributed to the development of the adventitial cyst.

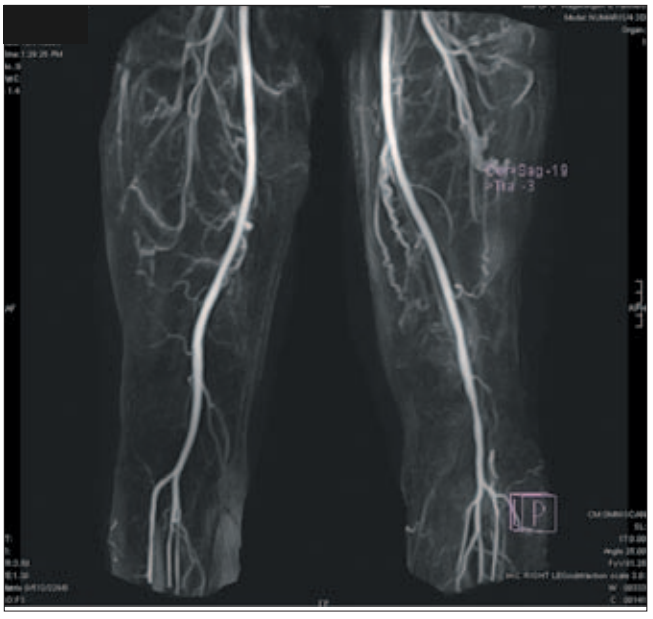

Fig. 3. MR angiogram showing narrowing of the right popliteal artery (arrow) and collaterals proximal to the knee.

Non-invasive diagnostic studies including colour duplex sonography, computed tomography (CT) and MRI are important to reach a final diagnosis and decide on appropriate management in most cases.[3] Sonography differentiates between cystic adventitial disease and a partially thrombosed aneurysm.[6] MR imaging and angiography enable evaluation of popliteal fossa anatomy and vascular compromise without the use of ionising radiation or iodinated contrast material.[2]

1. Levien LJ, Benn CA. Adventitial cystic disease: a unifying hypothesis. *J Vasc Surg* 1998; **28:** 193-205.
2. Elias DA, White LM, Rubenstein JD, Christakis M, Merchant N. Clinical evaluation and MR imaging features of popliteal artery entrapment and cystic adventitial disease. *Am J Roentgenol* 2003; **180:** 627-632.
3. Tsolakis IA, Walvatne CS, Caldwell MD. Cystic adventitial disease of the popliteal artery: diagnosis and treatment. *Eur J Vasc Endovasc Surg* 1998; **15:** 188-194.
4. Altehoeffer C, Blum U, Ebert D. Cystic adventitial degeneration and entrapment syndrome of the popliteal artery as a differential diagnosis of popliteal stenosis or occlusion in the younger age group. *Vasa* 1998; **27:** 179-182.
5. Turnipseed WD. Diagnosis and management of chronic compartment syndrome. *Surgery* 200; **132:** 613-619.
6. Brodmann M, Stark G, Pabst E, *et al*. Cystic adventitial degeneration of the popliteal artery – the diagnostic value of duplex sonography. *Eur J Radiol* 2001; **38:** 209-212.