**Popliteal adventitial disease in a triathlete**

A 37 year old recreational long distance triathlete complained of right leg pain during running since early 2002. The problems mainly occurred when running uphill, with a cramp-like pain and tightness in the anterior and lateral aspect of the right lower leg, associated with a loss of power and numbness in the foot. After periods of rest and treatment with physiotherapy, acupuncture and nutritional supplements running was possible for a short period until the problems started again. Previous medical history revealed three deep venous thromboses (DVT) in the right leg since 1985, the last one in 1995. A factor V mutation had been demonstrated (Factor V-Leiden), for which the patient was on life-long coumarine anti-coagulants. He did not smoke. The right leg showed signs of chronic venous incompetence (oedema, extensive scarring from previous venous ulcers, chronic eczema) with good peripheral pulsations. There were no other abnormal findings.

In August 2002, during an Ironman triathlon (3.8km swimming, 180km cycling and 42.2km running), running became impossible due to pain in the anterior compartment and numbness in the foot. The compartment appeared swollen and the symptoms decreased temporarily with massaging it at intervals. A diagnosis of chronic anterior compartment syndrome was made. A duplex-sonography revealed no arterial abnormalities and a grossly abnormal venous system. In September 2002 an anterior fasciotomy was performed.

After this procedure running was possible for two months, when the symptoms returned, especially during uphill running and mainly on the lateral side of the right lower leg. In February 2003, when the problems worsened, a diagnosis of lateral compartment syndrome was made and a lateral fasciotomy was performed.

Again, the patient was symptom-free for about one month, but thereafter the same symptoms returned progressively. Further investigations demonstrated incompetence of the right greater saphenus vein and several perforators. In order to reduce the venous pressure and improve the outflow, the greater saphenus vein was stripped and the perforators ligated in June 2003.

After this procedure the symptoms remained. The maximum symptom-free running distance was 2 km even at a slow pace. Walking was not affected. The patient consulted several doctors and was treated with physiotherapy without any improvement. The diagnosis of a posterior compartment syndrome was considered. A duplex-sonography now revealed a cystic structure of 21 by 14 mm in the right popliteal fossa, compressing the popliteal artery, suggestive of a popliteal artery aneurysm. The popliteal fossa was explored in November 2003 and a popliteal entrapment, caused by an aberrant insertion of the medial head of the gastrocnemius muscle was demonstrated. A cystic structure was not seen and a popliteal aneurysm was ruled out. The medial head insertion of the gastrocnemius was resected during this procedure.

Weeks after this operation the same symptoms were still persisting, with a disappearance of the pedal pulses and a prolonged capillary refill after running, returning after about 3 minutes. Symptoms became worse and in December 2003 walking uphill became also impossible. On duplex examination the cystic structure was still present, which was clearly compressing the popliteal artery during dorsiflexion. Magnetic Resonance Imaging (MRI) showed that the cyst was multi-loculated, 2 by 3 cm in size and located in direct relation to the popliteal artery. The popliteal artery was significantly compressed by the cyst and was narrowed to approximately half its normal size.
A standard contrasted peripheral MR angiogram demonstrated a stenotic segment of the popliteal artery of approximately 1 cm. There was extensive collateral formation in the thigh as well as the calf proximal and distal to the stenotic segment. There was no occlusion of the popliteal artery.
In January 2004 the popliteal fossa was re-explored and a large adventitial cyst, containing a mucoid substance, was found and excised. A femoro-popliteal bypass using a vein graft was necessary to bypass the damaged popliteal artery and to ensure adequate flow. Histology showed that the cyst was composed of a fibrous connective tissue wall, with evidence of myxoid degeneration.
Fig.5. Adventitial cyst at surgery.

Eight weeks after the bypass the patient finished the Ironman triathlon in New Zealand 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.

Discussion
Cystic adventitial disease of the popliteal artery is a rare cause of intermittent claudication. Since its first description in 1947 less than 350 cases have been reported worldwide. The etiology is controversial and the literature reports various hypotheses for its origin, including repeated trauma (such as caused by entrapment), tracking of capsular synovial structures along vascular branches, systemic disorder and incorporation of a joint-related ganglion-like structure into the developing vessel during embryological development. The incidence is estimated at one in 1200 cases of claudication. The disease typically affects males between 20 and 50 years of age without risk factors for atherosclerotic disease. Popliteal entrapment and cystic adventitial disease of the popliteal artery have been reported to occur together, which suggests that popliteal entrapment can cause adventitial cyst formation through micro-trauma. Our case study illustrates that the diagnosis is often difficult and leads to significant delays. In this athlete, symptoms developed gradually and the clinical 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 for claudication in young adults, and venous insufficiency has been described as a cause for CCS. Symptoms associated with CCS include muscle cramping or swelling in isolated muscle groups with plantar paresthesias, seriously affecting athletic performance. Symptoms are often longstanding and disappear after extended rest, only to reappear when exercise is resumed. Turnipseed describes functional popliteal entrapment as one of the least common variants of CCS. Most patients have been treated for other forms of CCS before this functional problem is discovered. In this condition, the medial head of the gastrocnemius muscle compresses the neurovascular bundle against a fibrous band of fascia at the entrance of the soleal canal during plantar flexion. Treatment of choice for CCS is fasciotomy.

In this patient the MRI angiogram suggests that the arterial problem existed for quite some time. 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, in combination with the poor venous outflow due to damage to the venous system after the repeated DVT’s, resulted in the antero-lateral CCS. It is not clear what role the aberrant insertion of the gastrocnemius muscle has played in the development of the adventitial cyst, but this might also have been a contributory factor.
Imaging was important in reaching the final diagnosis. Non-invasive diagnostic studies include colour duplex sonography, computed tomography (CT) and Magnetic Resonance Imaging (MRI), which can assist in establishing correct recognition of the disease in most cases.\textsuperscript{3,5} It is crucial to know the morphological background of the disorder, namely that it is a cyst of the adventitia of the artery which leads to an exercise-dependent flow inhibition.\textsuperscript{12} Sonography allows differentiation between cystic adventitial disease and a partially thrombosed aneurysm. The cystic lesion is often of high signal on T\textsubscript{1}-weighted MR images due to its mucoid content.\textsuperscript{4}

Percutaneous US-guided aspiration has been described as an easy, safe, efficacious method for treating adventitial cystic disease. In symptomatic patients who do not have thrombotic occlusion, it may be considered the treatment of choice.\textsuperscript{13}

Colour duplex sonography may show posture-dependent popliteal artery narrowing, changes in colour flow or increased peak systolic velocity in popliteal entrapment syndrome. Popliteal fossa anatomy and vascular complications may be evaluated as well. MR imaging and angiography enable evaluation of popliteal fossa anatomy and vascular compromise without the use of ionizing radiation or iodinated contract material. Two-dimensional time-of-flight MR sequences at rest and during active plantar flexion may show posture-dependent vascular compression.\textsuperscript{4}

Conclusion
Cystic adventitial disease of the popliteal artery, whether or not in combination with popliteal entrapment syndrome and/or chronic antero-lateral compartment syndrome, is a difficult diagnosis that needs to be considered in young athletes presenting with leg pain during exercise. Duplex-sonography and MR imaging are important to reach a final diagnosis and decide on appropriate management.
References: