CASE REPORT

Episodes of extreme lower leg pain caused by intraosseous varicose veins

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SUMMARY

We present a case of a 67-year-old man with episodes of extreme pain of the right lower extremity that prevented him from walking and sleeping. The patient had a history of varices in both legs. Physical examination showed a pretibial varix of the right leg inferiorly from a painful spot. X-ray of the right lower leg showed a cortex disruption at this spot. MRIs confirmed the disruption of the cortex of the right tibia and demonstrated an intraosseous vessel. The diagnosis intraosseous varices was made and the vein was surgically resected. Follow-up took place after 3 years and the patient was free from any symptoms related to the intraosseous varicose vein. The pathophysiology causing the pain symptoms is hard to understand, partly due to the limited cases presented with such anomalies. We demonstrate our case in the hope to generate more knowledge about this disorder.

BACKGROUND

Varicose veins are veins that are enlarged and distorted. Although any vein can become a varicose, varicose veins mostly appear on the legs. They are present in 10%–40% of the people between 30 and 70 years old. Symptoms associated with varicose veins are pain, pruritus and undesirable cosmetic appearance.^{1,2} More severe complications can be thrombosis, haemorrhage and ulcerations.³ Although the exact pathogenesis causing varicose veins is poorly understood, there are some assumptions made behind the causes of this disorder. Several studies have shown that venous incompetence resulting in varicose veins is caused by defects within the venous walls in endothelial cells, smooth muscle cells and extracellular matrix. Another assumption made is that direct valvular dysfunction resulting in varicose veins can be caused by inflammation and subclinical thrombosis. Some other causes can be pregnancy, obesity, hereditary, menopause and ageing.¹ The diagnosis is made clinically according to the clinical, aetiological, anatomical and pathological classification and with the help of duplex ultrasound.^{2–4} To avoid most of these symptoms, varicose veins can be treated by a surgical or non-surgical method. Some of the most common methods are compression stockings, sclerotherapy, phlebectomy, laser therapy, radiofrequency ablation and crossectomy.² In this paper, we describe a rare case of a 67-year-old patient that was diagnosed with pretibial intraosseous varicosity diagnosis by MRI and underwent successful surgical treatment.

CASE PRESENTATION

A 67-year-old man presented with episodes of severe pain in the right tibia, since several months. The pain always began after standing up and lasted up to 5 hours, after which the symptoms were completely gone. The severity of the pain was of such intensity that the patient was not able to walk during these episodes. There was no relation with activity, work out or trauma. The patient had a history of varices of both legs for which he wore compression stockings since 17 years. There was no other relevant medical history. Physical examination showed a pretibial varix, the varix itself was not painful on palpation; however, there was a painful spot just superior of the varix. Additional examinations were performed to investigate the symptoms the patient experienced. An X-ray was taken that showed a disruption of cortex at the location of the pain (figures 1 and 2). The disruption of cortex generated a suspicion for osteophytes/osteomas, and therefore an MRI of the right lower leg was performed. The MRI scan showed an intraosseous vessel pretibial at the place where there was disruption of cortex (figures 3 and 4). Because of the existing varices and the varix just inferior of the painful location, it was assumed that the vessel was a varix, and

Figure 1 Anteroposterior detailed view of the right tibia shows a circumscribed radiolucent spot (arrow).

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Figure 2 Lateral detailed view of the right tibia shows a circumscribed radiolucent spot (arrow) with some anterior increased bone formation.

therefore it would probably be of venous origin and the diagnosis of pretibial intraosseous varices was made.

TREATMENT

After discussing treatment options with the patient, we decided to perform surgery in the hope that this would relieve the patient from his episodes of pain. An incision was made just medially from the varix, the perforating vein was found. The periosteum was opened and the vein was cut out, this caused a bleeding that was stopped with wax. There was no sign of local thrombus formation. The periosteum was further excised, approximately 8 mm around the perforating vein and the skin was closed in layers. A compression bandage was applied and the stitches were left in place for 14 days. Follow-up 3 years after surgery demonstrated that the patient had not yet experienced a single episode of pretibial pain.

OUTCOME AND FOLLOW-UP

Follow-up 3 years after surgery demonstrated that the patient had not yet experienced a single episode of pretibial pain.

DISCUSSION

A total of 47 patients with intraosseous venous drainage of the varicose veins have been described in the literature,^{5–12} with one paper mentioning the perforating vein in the fibula instead of the tibia Diaz-Candamio *et al.*⁵ The first case that we found was described in 1962 by Schobinger. He described a 66-year-old man that suffered from swelling of the leg and phlebitis due to pretibial intraosseous varices.⁶ The pathophysiology of the disorder is still unclear and the cases reported in the literature are rare. There are several hypothesis about the pathophysiology, which could result in intraosseous venous drainage in varicosis. For example Peh *et al.*⁷ describes the intraosseous venous drainage anomaly as pre-existent because of the lack of symptoms in their young patient and the thought of the subcutaneous component of the venous anomaly as secondary to increased pressure. With

Figure 3 MRI in sagittal plane shows linear low-signal configuration intramedullary.

our patient, varicose veins of the lower legs pre-existed multiple years before the symptoms of pain began. The intraosseous vein could have been pre-existent to the symptoms as an anatomically present perforator that enlarged over time. It is hypothesised that this could be the cause of heightened venous flow due to deep venous thrombosis, venous insufficiency and venous hypertension.⁸ Peh *et al.* was the first to use sclerotherapy as treatment for the intraosseous varicose vein, instead of surgical removal.⁷ Although they mention that the treatment was successful, they do not mention follow-up after 5 months, and therefore it is unclear if the symptoms were gone over a longer period of time. Sclerotherapy was used many times by Ramelet *et al.* They described 35 cases in 32 patients that were diagnosed between 2006 and 2013.⁹ This was a collaboration between 6 countries and 13 phlebologists. The term ‘bone perforators’ is used to describe the intraosseous varicose veins defined as an abnormally large communicating vein between the superficial veins and the intraosseous venous network, through a round shaped bone defect, almost always located on the anterior aspect of the tibia. They treated 13 of their patients with sclerotherapy without any additional surgery. The rest of the cases were either surgery or a

Figure 4 MRI water sensitive sequence shows the vein in full extension. There is no bone marrow oedema visible, which indicates that the pain symptoms are not stress fracture or bone marrow oedema dependent (sagittal short-T1 inversion recovery sequence).

combination of surgery and sclerotherapy. Thirty of the 35 cases were symptomatic as were the cases described by other authors, including ourselves. It is not to say that most of the cases are symptomatic as the asymptomatic cases may never be found, because there is no need for additional diagnostic tests. The lack of symptoms in some of these patients does raise questions regarding the pathophysiology behind the episodes of pain that patients with intraosseous varicose veins experience. It is unclear whether the pain is being caused by pain in the vein wall (distension, ischaemia) or pain in the bone itself (periosteum, medullary canal). Ramelet *et al* describes the possibility of venous hyperpressure caused by reflux in the venous drainage of the tibia. We hypothesise those episodes of reflux cause high pressure in the vein resulting in venous distention triggering pain in the vein wall. Other vein-related pains could be ischaemia, caused by stasis, which is often seen in varicose veins. Pain in bone is thought to be caused by the periosteum that covers the bone.¹³ In the case of the venous anomaly, it is in constant contact with the periosteum even though the pain is not constantly present. It is possible that stasis raises the pressure in the vein causing it to push harder against the periosteum and causing more irritation, which results in pain. Another hypothesis could be that involvement of the medullary canal, which is also in contact with this varicose vein could be causing these symptoms. An example of pain caused by the medullary canal, without involvement of the periosteum is intraosseous engorgement syndrome.¹⁴ Although we hypothesise that the pain is most likely caused by stasis and distension of the varicose vein, it would be interesting to

examine these patients with duplex and perhaps MRI while they are experiencing these pain symptoms. This would help us to develop a better understanding of the pathophysiology behind the pain symptoms.

Learning points

- Intraosseous varices should be included in the differential diagnosis of patients that present themselves with severe episodes of pain of the lower extremity.
- MRI is a good tool to diagnose this disorder.
- Surgery could be a good treatment option for this disorder.

Contributors ESR had a part in patient treatment, follow-up and drafting the paper. MM was the radiologist that diagnosed the patient, he was responsible for the images in the paper and revised the paper critically. CMAMvdH was the plastic surgeon that performed surgery on the patient and performed follow-up visits. She revised the paper critically and is the scientific corresponding author of this manuscript.

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