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CASE REPORT

Rectal Venous Malformation Treated by Superior Rectal Artery Embolization

J. El-Sheikha[1] · M. W. Little[1] · M. Bratby[1]

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Abstract A 25-year-old female was referred to the Interventional Radiology Department for investigation and treatment of a rectal venous malformation (RVM) causing large recurrent episodes of rectal bleeding and chronic anaemia. Magnetic resonance imaging (MRI) demonstrated a large venous malformation affecting the rectum, lower pelvis and left thigh. After three failed attempts at injection foam sclerotherapy using fluoroscopic colonoscopy, a multidisciplinary team proposed an embolization procedure of the arterial inflow to the venous malformation. Following discussion with the patient, embolization of the superior rectal arteries was undertaken with immediate on-table fluoroscopic improvement in the RVM. Post-treatment, a significant reduction in bleeding, was reported by day 10 with subsequent return to activities of daily living and full employment. Follow-up MRI at 1 year demonstrated significant reduction in bowel-wall thickening.

Keywords Klippel–Trenaunay syndrome · KTS · Embolization · Venous malformation · Varicose

Introduction

Venous malformations of the rectum (RVM) are rare but often present in early childhood with recurrent rectal bleeding and severe chronic anaemia [1]. It is also common to find RVMs affecting the abdominal and lower limb venous system, with pelvic, vulval and lower limb varicose veins with enlarged symptomatic haemorrhoids frequently reported [2]. The anatomical location of RVMs make them a significant challenge to treat. Treatment options for RVMs are limited and especially challenging. Bowel resections are often successful but may require stoma formation, problematic in younger patients, and can result in long-term morbidity [3]. Minimally invasive treatments, such as injection sclerotherapy, have increased in popularity as these avoid the risks of surgery but may require several treatments and can struggle to treat large malformations [4]. This paper describes a novel approach using superior rectal artery embolization to treat an RVM, a technique based on previous studies using arterial embolization or transanal dearterialization (THD) in the treatment of haemorrhoids [5].

Case

A 25-year-old female with iron deficiency anaemia and a history of daily rectal bleeding was referred to the interventional radiology department for treatment planning of a known venous malformation affecting of the rectum, vulva and thigh. Magnetic resonance imaging (MRI) examination demonstrated an extensive abnormality of the rectum with gross thickening and high signal on T2 and STIR sequences. The thickening and increased signal extended from the

✉ J. El-Sheikha
Jelsheikha@doctors.org.uk

[1] Interventional Radiology Unit, Radiology Department, John Radcliffe Hospital, 2nd Floor, Oxford OX3 9DU, UK

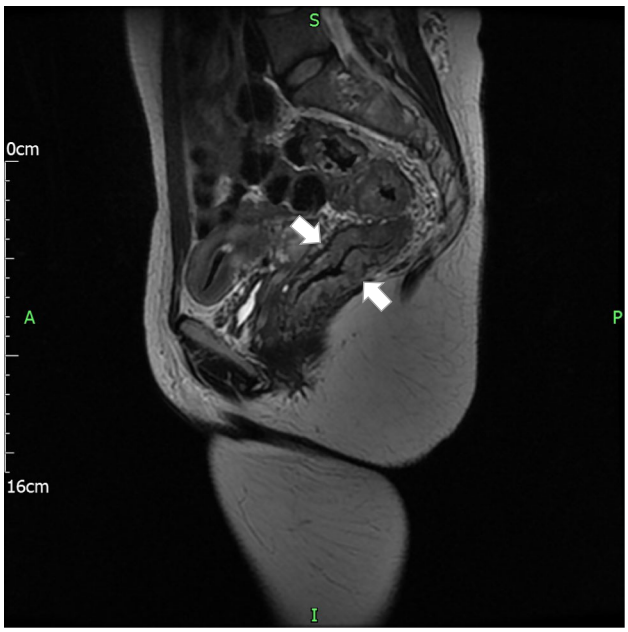

Fig. 1 T2 Sagittal. Arrows demonstrate the rectal venous malformation with bowel-wall thickening

anal verge for approximately 15 cm proximally to involve a segment of distal sigmoid colon, with the colon above this level appearing normal. There were also multiple draining veins in the mesorectal fat but no overt involvement of the bladder, cervix or uterus was demonstrated (Fig. 1). In addition, it was noted that there was further high-signal involving the vulva, subcutaneous tissues of left lower back, left gluteal muscles and into the posterior compartment of the left leg. Hypertrophy of left sciatic vein into a grossly dilated left internal iliac vein was noted. The status of left SFV was undetermined from this study but is often hypoplastic rather than absent [6]. The diagnosis of Klippel–Trenaunay syndrome (KTS) with predominant rectal involvement was made, and treatment using endoscopic sclerotherapy was planned.

Three sessions of combined fluoroscopic-colonoscopy-guided injection sclerotherapy were attempted. The first session was abandoned due to significant mural thickening preventing needle access into the RVM. The second and third sessions both used a longer 22G Chiba needle (Cook Medical ®) to successfully puncture through the thickened rectal wall. Injection of foam sclerotherapy of 3% sodium tetradecyl sulphate (Fibrovein®) diluted to 3:1 in room air was performed but was unfortunately not successful. Following discussion at a regional Multidisciplinary Team Meeting (MDT) and review of related medical literature, it was proposed that embolization treatment of the superior rectal arteries was recognized as an emerging and successful treatment for symptomatic haemorrhoids. Following a discussion with the patient and informed written

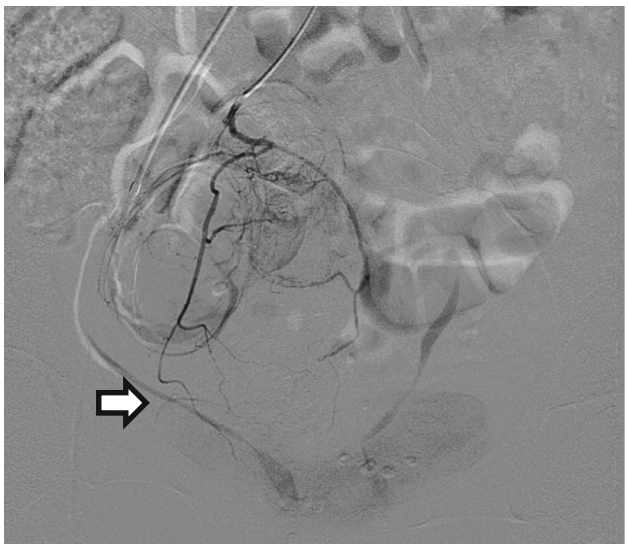

Fig. 2 Superior rectal artery angiogram demonstrating the superior rectal artery anatomy and early contrast filling of the rectal venous lakes

consent, angiography and embolization of the superior rectal arteries was performed. From an ultrasound-guided retrograde right common femoral artery approach, a SOS® catheter (Angiodynamics®) was navigated into the inferior mesenteric artery. A Progreat (Terumo®) 2.7 microcatheter was then used to super-selectively catheterize the superior rectal arteries. Delayed angiography from the distal aspect of the superior rectal arteries revealed marked para-rectal venous engorgement (Fig. 2). Bilateral superior rectal artery embolization (SRAE) was performed using a combinations of 2, 3, and 4 mm Concerto coils, and an excellent angiographic result was noted (Figs. 3, 4).

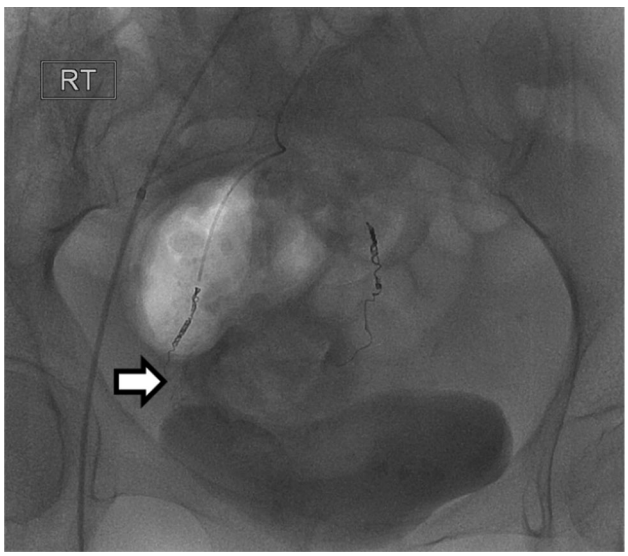

Fig. 3 Angiogram post-coiling demonstrating contrast stasis within the venous lakes of the rectal venous malformation

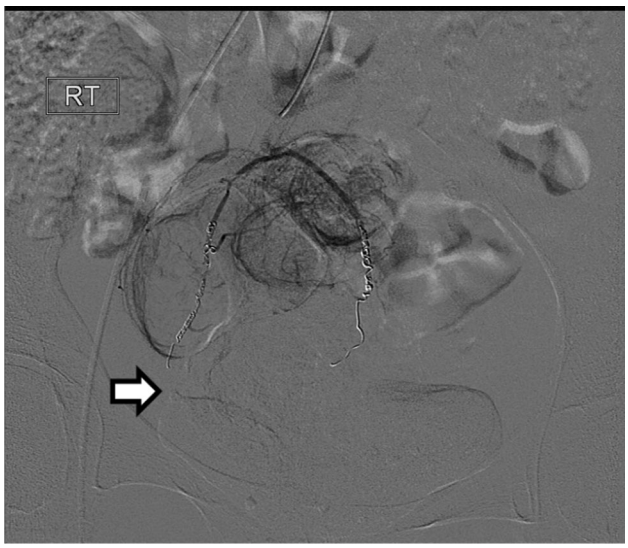

Fig. 4 Angiogram post-coiling demonstrating SRA anatomy and marked reduction in the early contrast opacification of the rectal venous malformation

Following treatment, she had pain and a little more bleeding for 7 days. However, by day 10, her symptoms had settled and there was significant reduction in bleeding, with subsequent return to activities of daily living and full employment. Follow-up MRI at 1 year (Fig. 5) demonstrated a marked reduction in the thickness of the rectal wall. A remainder of some thickening was noted in the lower rectum which presumably was supplied by the middle/lower rectal arteries, and the remainder of malformation in pelvis and left buttock and thigh was unchanged.

Discussion

This is the first paper to describe superior rectal artery embolization (SRAE) for a large RVM. By reducing the arterial inflow and consequently the venous pressure of the RVM, a significant reduction in the venous engorgement and the patient's symptoms was achieved in a single embolization procedure. This approach is conceptually similar to the "Emborrhoid" technique [7] that uses superior rectal artery embolization to treat symptomatic haemorrhoids and was itself a technique based on transanal dearterialization (THD) and ligation of the haemorrhoidal arteries. Embolization has several advantages over minimally invasive treatments such as foam sclerotherapy. Primarily, the large size and difficulty in percutaneously accessing RVMs through thickened rectal mucosal walls, as well as the large volumes of sclerosant required to adequately treat the venous walls, drastically reduces the effectiveness of foam sclerotherapy. It is therefore likely that surgical resection would have been the only option available to the patient if neither minimally invasive technique was successful [8]. Large volumes of foam sclerotherapy also increase the risk of developing deep vein thrombosis, pulmonary embolism and even death [9]. While no complications arose from this case specifically, Vidal et al. reported that the risks following the Emborrhoid technique in 14 patients were limited to haemorrhoid recurrence ($n = 4$) and transient perianal reaction ($n = 1$). No pain or ischaemic reactions were reported [7].

This case also shows the importance of a multidisciplinary approach to the management of complex vascular malformations, especially in patients with Klippel–

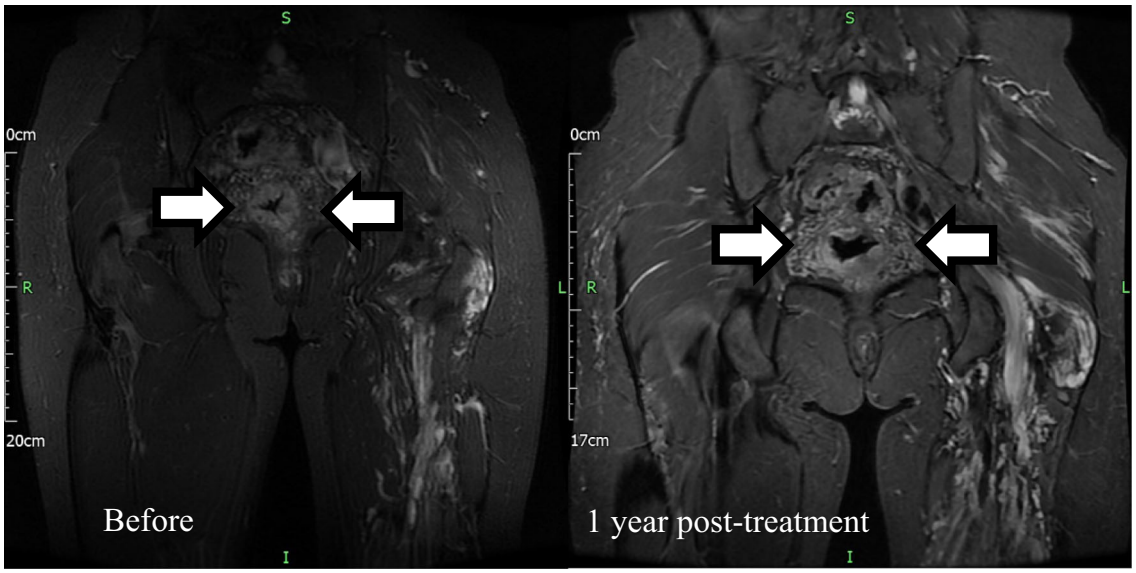

Fig. 5 STIR Coronal. Arrows demonstrate significant reduction in the RVM volume and bowel-wall thickening

Trenaunay syndrome (KTS), a disease which is characterized by a triad of port wine nevi, bony or soft tissue hypertrophy of an extremity (localized gigantism) and varicose veins or venous malformations of unusual distribution. Such patients often present in childhood and are often challenging to treat. It is not uncommon to find unusual venous anatomy, such as a persistent sciatic vein instead of a superficial femoral vein, or multiple varicose veins thought the pelvis or lower limb with resulting risks of vaginal or rectal bleeding and even pulmonary thromboembolism [10]. Selective embolization should be considered in patients with large rectal venous malformations following MDT discussion as in our patient. We have shown that careful embolization can produce dramatic clinical benefit and should be included in one's armamentarium.

Compliance with Ethical Standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical Statement All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards.

Informed Consent Informed consent was obtained from all individual participants included in the study.

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