CASE REPORT

A case of scrotal venous malformation mimicking a failed varicocelelectomy

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SUMMARY

A 21-year-old man presented to interventional radiology department with several years of left scrotal pain, which exacerbated by prolonged standing and walking. The patient had undergone a left varicocelelectomy at the age of 10, after which he had a persistent scrotal mass. As he grew older, the left scrotal mass continued to increase in size, and symptoms progressively worsened. Physical examination revealed a non-tender, firm left scrotal mass which remained unchanged in size after Valsalva manoeuvres. Initial ultrasound examination revealed an extratesticular, intrascrotal mass with minimal internal flow. MRI revealed a heterogeneously enhancing, low-flow venous malformation centred in the midscrotum involving multiple tissue planes. Two sessions of percutaneous sclerotherapy failed to relieve his pain. Three months after the second sclerotherapy, the patient underwent surgical resection. At the time of his postoperative visit, his left scrotal pain had significantly improved and pain with prolonged standing and walking had resolved completely.

BACKGROUND

Scrotal venous malformation (SVM) is an exceedingly rare congenital entity which has not been well published in the literature. Venous malformation (VM) is a subtype of vascular malformation, and is not a true neoplasm. VMs have an estimated incidence of 1–2 in 10 000, most commonly located in the head and neck (40%), extremities (40%) and trunk (20%).¹ They are usually diagnosed at birth, but may be discovered at any age. VMs typically will not involute, unlike infantile haemangiomas. Patients often present with pain and swelling and can have clinical features that overlap with other vascular neoplasms and malformations. In patients with atypical history and deeper lesions, appropriate diagnostic imaging may be warranted for diagnosis.^{1 2}

Vascular malformations are categorised using flow dynamics into high-flow and low-flow lesions based on their presence or absence of arterial components under the Mulliken and Glowacki classification system.³ Low-flow lesions are subcategorised based on the components into venous, capillary, lymphatic or mixed malformations. VMs are known to be the most common peripheral vascular malformations.^{1 4} They are typically compressible and non-pulsatile on examination with a characteristic bluish, purple discolouration when involving the skin.^{5 6}

Given that VMs comprise almost half of all referrals for treatment in large vascular anomaly centres,^{5 7} and that they are largely misdiagnosed

initially owing to diagnostic unfamiliarity,² it is likely that scrotal VMs are overlooked or misdiagnosed as haemangiomas or other vascular malformations. Correct diagnosis and categorisation is essential because treatment options for various vascular malformations and neoplasms are significantly different.

CASE PRESENTATION

A twenty-one-year-old man presented to interventional radiology department with several years of left scrotal pain, which exacerbated by prolonged standing and walking. The patient was diagnosed with left varicocele during his childhood and was treated with left retroperitoneal gonadal vein clipping at the age of 10. However, his scrotal mass continued to gradually increase in size, and symptoms progressively worsened, particularly after puberty. He underwent a laparoscopic ligation of enlarged left retroperitoneal veins, which on venography appeared to feed the VM. This procedure also failed to relieve his symptoms. The patient denied any history of trauma or recent infections, and was otherwise healthy without additional medical problems. Laboratory tests were unremarkable. Physical examination revealed a non-tender, firm, left scrotal mass with irregular borders. The mass remained unchanged in size with Valsalva manoeuvres and postural changes. Initial testicular ultrasound examination prior to clinic visit revealed an infiltrative, intrascrotal, extratesticular, heterogeneously hypoechoic mass with anechoic channels measuring 7.6×3.8×5.1 cm (figure 1A). Colour Doppler image revealed minimal internal flow (figure 1B).

INVESTIGATIONS

MRI of the scrotum was subsequently performed. This revealed a 7.8×7.0×5.2 cm lobulated mass centred in the scrotum involving multiple tissue planes and extending to the corpus cavernosum and perineum. T1 sequences demonstrated diffuse foci of fat interspersed within the lesion, and fat-suppressed T2 sequences revealed fibrous striations without flow voids (figure 2A,B). Postcontrast images revealed diffuse, heterogeneous internal enhancement with ectatic draining veins (figure 2C,D).

Further characterisation of flow dynamics and venous mapping prior to intervention was obtained by means of digital subtraction venograms, performed after selection of the left common iliac vein (figure 3A) and after direct percutaneous access of scrotal venous sinusoids (figure 3B). There were multiple tortuous draining varices emanating from

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Figure 1 Scrotal mass. Longitudinal grey-scale image (A) revealing a heterogeneous, hypoechoic, extratesticular mass with anechoic channels (calipers). Colour Doppler image (B) showing minimal internal flow.

the scrotal VM and venous collaterals draining into the left renal vein.

DIFFERENTIAL DIAGNOSIS

Differential considerations for low-flow vascular malformations include VM, lymphatic malformation, and capillary malformation. The patient's history, physical examination, MRI and digital subtraction venogram findings were consistent with a low-flow VM. Lymphatic malformations commonly demonstrate characteristic fluid/fluid levels or cystic spaces without internal enhancement, and are usually non-compressible on examination.^{1 2 8 9} Capillary malformation typically manifests as a port-wine stain (cutaneous red discolouration), and the diagnosis is usually made based on history and physical examination alone.

TREATMENT

We performed two sessions of percutaneous sclerotherapy approximately 1 month apart. On our first treatment, we injected 3.7 mL of 98% dehydrated ethanol into the VM until there was decreased filling (figure 3C). After the initial therapy,

the patient reported that there was no symptom relief or considerable interval change in size. Approximately 1 month after the initial treatment, we performed additional sclerotherapy with 5 mL of 98% dehydrated ethanol (figure 3D).

OUTCOME AND FOLLOW-UP

The patient's symptoms failed to improve after two sessions of percutaneous sclerotherapy, and MRI of the scrotal VM performed 2 months after the second sclerotherapy appeared essentially unchanged in size and enhancement characteristics from the pretreatment MRI (figure 4A,B). At this point, the patient elected to undergo surgical resection by urology.

Surgery revealed that spermatic cord vessels were unremarkable in venous drainage of the scrotal VM. There were areas of scarred tissue adjacent to the areas of prior sclerotherapy. Significant amounts of the scrotal VM were resected (figure 5A,B). Histopathology revealed multiple venous channels with various luminal diameters, confirming VM.

At the time of his postoperative visit, left scrotal pain had significantly improved and pain with prolonged standing and walking had resolved.

Figure 2 Scrotal venous malformation. Axial fat-saturated T2-weighted (A) and axial postcontrast T1-weighted (B) images revealing an infiltrative mass with fibrous septations. Sagittal T1-weighted (C) image revealing punctate fat interspersed between vascular spaces (D) and sagittal postcontrast T1-weighted image showing dilated draining veins.

Figure 3 Digital subtraction venograms. Left common iliac venogram (A) and direct percutaneous scrotal venogram (B) revealing multiple draining varices and collateral veins. Varicocele clips are visualised along the left gonadal vein (arrowheads). Postembolisation digital subtraction venograms after the first sclerotherapy (C) and second sclerotherapy (D) revealing diminished flow in the venous malformation and draining veins.

DISCUSSION

SVM is an exceedingly rare entity, and this is the first case report of its kind in the literature, to the best of our knowledge. An additional complicating factor which makes our case unique was the patient's history of prior varicocele from an outside facility. Incidence of varicocele recurrence is not uncommon^{10 11} and can manifest with similar symptoms and clinical examination findings as with our patient, which can make an accurate diagnosis difficult, if not impossible, without imaging.

MRI is the most effective, non-invasive tool for diagnosis and characterisation of vascular malformations.^{8 12 13} Haemodynamic flow characteristics, anatomical extent and enhancement patterns are essential findings for an accurate diagnosis.^{8 12 13} Flow characteristics and drainage patterns of the vascular malformation can be further delineated by catheter-based digital subtraction angiography. Colour and duplex Doppler ultrasound can be useful for initial assessment of flow.

Treatment options for VM include percutaneous sclerotherapy, surgical excision, and laser photocoagulation.^{7 14 15} Surgical excision may be used in combination with embolotherapy. Use of the injected scleroagent, absolute ethanol, for treating scrotal VM has not been reported in the literature, to our knowledge. Absolute ethanol may provide potentially definitive treatment in VM with relatively low rate of recurrence.^{7 16} However, risks involved with non-target embolisation include skin necrosis, nerve damage, muscle fibrosis, venous thrombosis, and pulmonary embolism.^{7 16}

Although the extent of our patient's VM and location made our case challenging, we decided that our patient was a suitable candidate for percutaneous sclerotherapy after imaging and careful venous mapping via direct percutaneous injection of venous sinusoids. Therapeutic response was likely diminished because the VM components did not intercommunicate after direct percutaneous injection of ethanol. Our case suggests that

Figure 4 Scrotal venous malformation after percutaneous sclerotherapy. Axial fat-saturated T2-weighted (A) and sagittal postcontrast T1-weighted (B) images revealing no significant change after two sessions of ethanol sclerotherapy.

Figure 5 (A and B) Intraoperative images of scrotal venous malformation.

surgical resection is more efficacious and a potentially definitive treatment for scrotal VM, compared with treatment with sclerotherapy alone.

In summary, scrotal VM is an exceedingly rare entity that may mimic symptoms and physical examination findings of failed varicocelectomy. Appropriate imaging is recommended to obtain the diagnosis and to guide further management decisions. Ultrasound, contrast enhanced MRI, and catheter-based digital subtraction venography can serve as essential tools prior to percutaneous sclerotherapy and surgical resection.

Learning points

- ▶ Vascular malformations are categorised using flow dynamics into high-flow and low-flow lesions based on their presence or absence of arterial components. Venous malformations are low-flow vascular malformations.
- ▶ Scrotal venous malformation (SVM) is an exceedingly rare entity that may mimic symptoms and clinical examination findings of other vascular malformations, to include failed varicocelectomy, as in our patient. Appropriate imaging is vital for accurate diagnosis and guidance of further management decisions. This holds particularly true for patients with atypical clinical history and deep lesions.
- ▶ Correct differentiation and characterisation based on flow characteristics are extremely important because treatment options are significantly different.
- ▶ SVM may be treated with percutaneous sclerotherapy, surgical resection or a combination of both. However, surgical resection may be the definitive therapy.

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