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CASE REPORT

Recurrent Scrotal Arteriovenous Malformation as a Slowly Increasing Left Testicular Swelling

A case report

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ABSTRACT: Arteriovenous malformations (AVMs) are benign vascular lesions. Although, the majority of AVMs occur in the central nervous system, there are published reports of AVMs involving all systems including the scrotum, kidney, and uterus. We report a 37-year-old male patient who presented to the urology clinic of a tertiary care hospital in Riyadh, Saudi Arabia, in 2021 with recurrent gradual scrotal swelling for four years attributed to scrotal AVM. Embolisation was performed; however, one year later his symptoms reoccurred. As a result, left partial scrotal wall excision was carried out without complications.

Keywords: Arteriovenous Malformation; Urology Varicocele; Case Report; Saudi Arabia.

Arteriovenous malformations (AVMs) are benign vascular lesions. They are described as abnormal vessels fed by arteries and drained by veins without intervening capillaries.[1,2] Although the majority of AVMs occur in the central nervous system, there are published reports of AVMs involving the scrotum, kidney and uterus.[2–4] Based on the published reports, the clinical presentation of scrotal AVMs is highly variable, ranging from an incidental finding on imaging for infertility to a bleeding mass.[1,5] Since scrotal AVMs have variable presentations and are rarely described in the literature, we are reporting a case of a patient who presented with a slowly increasing left testicular swelling attributed to scrotal AVM.

Case Report

A 37-year-old male smoker presented to the urology clinic of a tertiary care hospital in Riyadh, Saudi Arabia, in 2021 with a gradual scrotal swelling that started four years ago. He complained of on and off scrotal pain, occasional feeling of scrotal warmth, and scrotal discomfort. The patient was diagnosed in another hospital with a testicular artery aneurysm and left testicular varicocele. The patient denied any history of trauma, urinary tract infection, voiding symptoms, previous surgeries and his past medical history was unremarkable. Upon physical examination, the testes were intra-scrotal. There were no signs of inflammation, and the cremasteric reflex was intact bilaterally. Both epididymides were palpable and non-tender. However, pampiniform plexus at the neck of the scrotum was very pulsatile [Figure 1]. Moreover, multiple skin varices over the left scrotum were seen. Urinalysis was normal and urine culture was negative. Routine laboratory tests were unremarkable. Abdominal and pelvis computed tomography (CT) showed left scrotal arteriovenous malformation with enlarged small and medium-sized serpiginous structures with a feeder artery arising from the proximal superficial artery. Two months later, the patient was referred to interventional radiology for embolisation, which was successfully performed utilising Onyx 18% [Figure 2]. On the one-year follow-up, the pampiniform plexus were pulsatile again which necessitated a CT angiogram. CT angiogram confirmed the recurrence of arteriovenous malformation. The patient was counselled about the available treatment options and given time to decide. Due to the risk of recurrence as well as the possibility of technical failure with embolisation, he decided to go with the surgical treatment. The patient was booked for surgery and partial scrotal wall excision was done through an elliptical incision [Figure 3]. Three arteries that feed into the arteriovenous malformation were identified and controlled with vicryl ties. The malformation and the skin that covering it were removed and sent to the pathology lab. The *dartos* muscle was closed in a multi-fashion layer. The skin was closed by vicryl rapide 4-0 in a vertical mattress. The histopathological study confirmed the diagnosis by detecting vascular structures extending from fibrofatty tissues measuring 8 × 0.5 cm grossly and prominent subcutaneous

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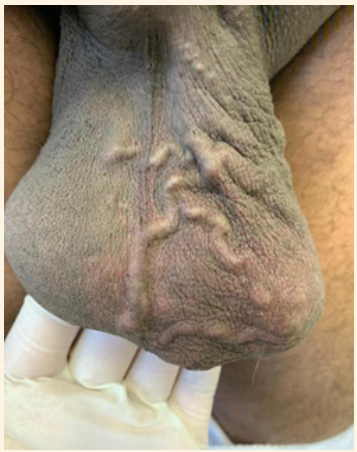

Figure 1: A photograph of the left scrotal swelling of a 37-year-old male patient with clear multiple skin varices.

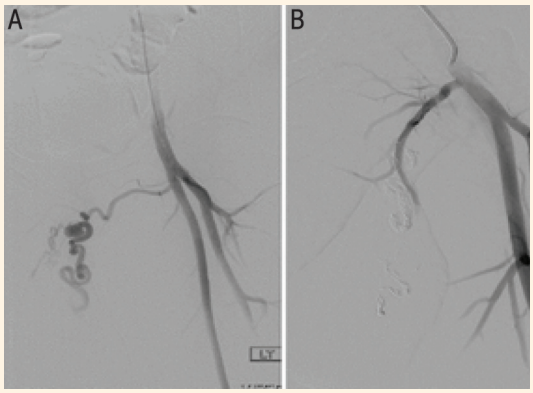

Figure 2: Computed tomography angiogram of a 37-year-old male patient showing (**A**) a feeder artery supplying scrotal arteriovenous malformation (AVM) and (**B**) an angiogram following Onyx embolisation through the AVM is almost occluded.

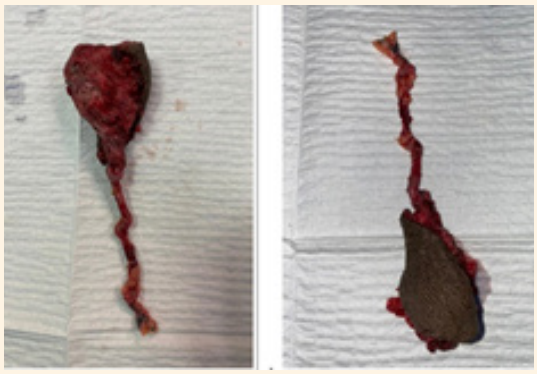

Figure 3: Photograph of the excised partial left scrotal wall with arteriovenous malformation from a 37-year-old male patient.

large congested vascular spaces microscopically. The patient was discharged one day after the surgery with no complications. Two months postoperatively, the patient has recovered well with no active complaint and the wound healed properly.

Table 1: A brief summary of the reported cases of scrotal arteriovenous malformation

Author	Age	Trauma History	Presentation	Semen analysis	Varicocele	Diagnosis method	Embolisation	Treatment	Follow-up
Monoski *et al.*[5] (2006)	31	No history of trauma	Infertility	Severe oligospermia	A left varicocele	Pelvic angiography	Performed	Surgical left scrotal AVM excision	3 years post-surgery, successful spontaneous pregnancy
Agrawal *et al.*[6] (2006)	25	Positive-severe pelvic fracture 4 years ago	Pain associated with a soft swelling on his right testicle	Not Performed	No evidence of varicocele	Histopathological examination	Not performed	Ileo-femoral bypass surgery	Not mentioned
Sountoulides *et al.*[7] (2007)	22	No history of trauma	Acute recurrent pain in the right hemiscrotum	Not Performed	No evidence of varicocele	Post-orchiectomy specimen	Not performed	Orchiectomy	2 years post-surgery, there was no complain
Mohammed *et al.*[8] (2020)	19	No history of trauma	Progressive diffused swelling in the scrotum with flashing skin and local warmth	Not Performed	Varicocele with 1 cm dilated veins	CT arteriography	Performed	The whole left side of the scrotum was removed, and the left testicle was fixed to the right side	12 months post-surgery, there was no complain
Current case	37	No history of trauma	Gradually increasing scrotal mass with on and off scrotal pain and discomfort	Not performed	Multiple skin varices over the left scrotum were seen	Abdominal and pelvis CT	Performed	Partial scrotal wall excision	Two months post-surgery, there was no complain

AVM = Arteriovenous malformation; CT = computed tomography.

Consent was obtained from the patient as the images were taken in the clinic for publication purposes.

Discussion

AVMs are malformations in the circulatory system characterised by arteries and veins that are not connected by capillaries leading to various degrees of ischaemia and pain.[1,2] Even though central nervous system cases represent the majority of AVMs, there are published reports of AVMs involving the kidney, uterus and scrotum.[2–4] AVMs are rarely present in the urinary tract.[6] We reviewed four previously published scrotal AVM cases [Table 1]. All reported ages ranging from 19 to 31 years while the current patient was 37 years old. Scrotal AVM covers a wide-range of presentations including infertility, acute recurrent pain in the hemiscrotum, pain and swelling on the testicle and progressive diffused swelling in the scrotum with flashing skin and local warmth. The current case presented with gradually increasing left testicular swelling with on and off scrotal pain as well as occasional feeling of scrotal warmth and discomfort. Of the four cases we have reviewed, three denied any history of trauma and one had a positive trauma history, which was a severe pelvic fracture, where the patient indicated that there was difficulty in maintaining erection since. The current patient denied any history of trauma. Varicocele was found in two of the cases and was seen by sonography whereas the current patient had multiple skin varices above the left scrotum that was seen during physical examination. Each one of the reviewed four cases diagnosed scrotal AVM with a different modality. Some were challenging and required orchiectomy for a diagnosis while others were simple and detected by pelvic angiography. The current patient was diagnosed by abdominal and pelvic CT. Two studies were able to find and embolise the feeding arteries. The current patient underwent embolisation but had recurrence one-year later. Similar to the current case, surgical intervention was eventually done in all four cases that varied from left scrotal AVM excision, orchiectomy, ileo-femoral bypass and resection of the whole left side of the scrotum. In the current case, partial scrotal wall excision was done. After the surgical intervention, all patients were symptom-free.[5–8] The difference in the presentation could be attributed to the location of the AVM, onset, duration and if there is a history of trauma. A possible explanation for the differences in imaging modalities used to diagnose scrotal AVMs is the availability of imaging techniques in the hospitals that encountered those cases. The decision of surgical intervention is mainly based on the symptoms and how symptoms negatively affect the patient's quality of life.

Conclusion

The current case calls attention to a rare and challenging diagnosis that is scrotal AVM. Recurrent scrotal pain, swelling and warmth together with varicocele should raise suspicion for scrotal AVM. Treatment varies depending on the symptoms present. Embolisation of the feeding arteries is a possible option initially and surgery should be preserved for recurrent cases.

AUTHORS' CONTRIBUTION

FMA, MSA and SIA were responsible for conceiving the idea, literature search, data acquisition and manuscript drafting and revision. AA, SA, YA and AB were primarily involved in the management of the case and critically reviewed the final version of the manuscript. All the authors have read and approved the final version of this manuscript.

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