Arteriovenous malformation of the scrotum: Is preoperative angioembolization a necessity?

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ABSTRACT

Arteriovenous malformations (AVMs) of the scrotum are uncommon lesions, usually picked up incidentally during the evaluation of scrotal masses or infertility. They have also been reported to present with acute bleeding. We present a case who presented with acute pain following an abandoned surgical attempt at excision, elsewhere. Diagnosis was confirmed by duplex ultrasound and magnetic resonance imaging. Angioembolization was deferred quoting concerns with radiation exposure. The patient underwent a near total excision of the scrotal mass. This is the first reported case, in the English literature, of a surgical resection of a scrotal AVM without a preceding angioembolization. Patients should be counselled about radiation exposure risks before angioembolization, and allowed to make an informed decision.

Key words: Arteriovenous malformation of scrotum, preoperative angioembolization, radiation exposure, sterility

INTRODUCTION

The most common arteriovenous malformations (AVMs) are intracranial, followed by extracranial head and neck, extremity, truncal and visceral sites. Rarely do AVMs involve the testes or other scrotal components, presenting mainly as paratesticular or intratesticular masses, and are detected during the evaluation of a swelling, infertility, and rarely, for bleeding. Scrotal AVM has been reported approximately only 12 times in the literature. To our knowledge, this would be the first reported case where a scrotal AVM was surgically excised without the safety net of preoperative angioembolization.

CASE REPORT

A 30-year-old man presented with a progressively increasing scrotal swelling of 10 years duration. He had an episode of an acute scrotal pain 4 months before, which subsided with analgesics. There was no history of recent or remote pelvic or scrotal trauma. He gave history of a failed surgical attempt at the local hospital 7 years ago due to uncontrolled bleeding, necessitating eight pints of blood transfusion. Examination revealed an ill-defined firm nonpulsatile swelling involving the entire scrotum and extending to the right buttock, with hyperpigmented and grossly thickened overlying skin. The anterior scrotal skin was more involved than the posterior. The spermatic cord was unremarkable and the right testes were at a lower level compared to the left.

A duplex ultrasound and MRI were done with a differential diagnosis of low flow vascular malformation and plexiform neurofibroma.

Duplex ultrasound showed a grossly thickened scrotal wall with prominent vessels exhibiting both venous and arterial components, with unusually high flow velocities. MRI was suggestive of AVM, showing an asymmetric heterogenous scrotal wall thickening with multiple flow voids.

Radiation exposure at levels of two Gy would have been sustained with angioembolization of the scrotal vascular malformation. 1–2 Gy has been documented to produce oligospermia with a prolonged recovery period of up to 3 years. A value above 2 Gy produces permanent azoospermia. The patient was not willing to risk radiation exposure. He was
therefore taken up for surgery under adequate blood cover.

An incision was made at the junction between abnormal and normal skin. There were multiple thickened and dilated vessels seen in the subcutaneous tissue. Testes appeared normal. The vascularity increased in density toward the midline and closer to the root of scrotum. The feeder vessels were probably from the pudendals and seen at the root of scrotum in the midline. The spermatic cord was first identified to trace the testes which were preserved. The scrotal vascular plexus were each ligated and divided, working our way up to the scrotal root. Near total excision of the lesion was done and the incision then closed in a T-shaped manner excising the redundant skin, in both the horizontal and longitudinal axes. In this case, our priority was good access for vascular control, and maximal resection of the AVM. This incision afforded us these advantages over a midline incision, with the benefit of better cosmesis.

The patient required no blood transfusion and had an uneventful postoperative recovery.

Histopathology confirmed the diagnosis of an angiomatous lesion, consistent with AVM; this was subsequently confirmed with elastin Van Gieson’s stain. Deep dermis and subcutis showed proliferation of varying calibered vessels, a few with ectatic features, of varying wall thickness lined by flattened to plump endothelial cells; occasional vessels showed smooth muscle in their wall [Figure 5].

**DISCUSSION**

Vascular lesions of the scrotum are rare. The most frequent condition by far is a varicocele, which appears as a tangle of dilated tortuous vascular structures on ultrasound. The other vascular lesions of the scrotum are hemangioma, lymphangioma, and AVMs, which are exceedingly rare. Twelve cases of scrotal AVMs have been reported thus far [Table 1]. Yilmaz et al. have reported an intrascrotal AVM simulating varicocele closely on ultrasound, but duplex evaluation showed the correct diagnoses of AVM. Although ultrasound and MRI play an important role in evaluation, angiography is essential to fully delineate feeder vessels, vascular take-offs, and draining veins.

The high flow within an AVM diverts blood from tissues, producing varying degrees of ischemia and consequent pain. Soultoulides et al. have reported a case of spermatic cord AVM presenting with acute scrotal pain, requiring...
Table 1: Clinical details of reported cases of high flow scrotal vascular malformations

| Author (year) | Age (years) | Age and presentation | Thrill/Bruit | Abnormal sperm analysis | Investigations | Management | Follow-up |
|---------------|-------------|---------------------|--------------|------------------------|---------------|------------|-----------|
| Hamid et al. (1992)<sup>[2]</sup> | 55 | Right scrotal swelling × 20 years. 3 months earlier, had scrotal pain with ulceration and bleeding | + | Azoospermia | Low s.testosterone Doppler stethoscope Arteriography-feeders from profunda femoris and internal iliac a | Angioembolization performed, but improved pain only for a few days, hence surgery had to be done | Not commented |
| Sule et al. (1993)<sup>[3]</sup> | 17 | Intermittently bleeding pulsatile left scrotal mass | + | - | Arteriography- Int&external pudendal a, circumflex branch of profunda femoris | Angioembolization with gelatine sponge and coils failed to eradicate the mass or control hemorrhage. Hence a complete surgical resection was done | No recurrence at 2 year follow-up |
| Konus et al. (1999)<sup>[4]</sup> | 8 | Progressively enlarging, intermittently bleeding, painful pulsatile scrotal mass | + | Not done | Doppler ultrasound. Arteriography-bilateral internal and external pudendal arteries. | Preoperative angioembolization with polyvinyl alcohol sponge. 2 days later, surgical excision of the embolized nidus | 1 year later, no residual disease on follow-up |
| Kang et al. (2004)<sup>[5]</sup> | 20 | Acute scrotal swelling detected 4 days after a trauma | - | Not done | Ultrasound and Doppler showed features of a hematoma, a multiseptated cystic mass and normal testes | Scrotal exploration and excision. Biopsy showed AVM confirmed by factor VIII & CD34 stains | Not mentioned |
| Bandi et al. (2004)<sup>[6]</sup> | 67 | Recurrent scrotal AVM-bleeding nonhealing ulcer 12 years after preoperative embolization and hemiscrotectomy | Not mentioned | Not done | None done at second presentation | Surgical excision. At pathological examination, AVM was confirmed with marked foreign-body giant cell reaction secondary to embolization Material | Not mentioned |
| Agarwal et al. (2006)<sup>[7]</sup> | 31 | Primary infertility and left scrotal fullness | - | Severe oligospermia | High normal FSH (33 mIU/mL), normal LH and s.testosterone. Arteriography (done after varicocelectomy)– hypertrophied internal pudendal and branch of superficial femoral a. | Underwent bilateral varicocelectomy, but there was no change in the scrotal mass or semen analysis. Angioembolization was done, but even 2 weeks later, scrotal mass size was unchanged. Hence scrotal exploration and excision of AVM were done | 3 months later, sperm count improved. FSH normalized (16 mIU/mL). 3 years later-successful spontaneous pregnancy. |
| Skiadas et al. (2006)<sup>[8]</sup> | 39 | Primary infertility. Intratesticular AVM | Oligospermia with low motility | Ultrasound and Colour Doppler showed left intratesticular 5 mm AVM. MRI confirmed diagnosis. The patient refused angiographic evaluation | Schistosoma periaclavicular palpation | The patient refused any intervention and was followed up at 6 monthly intervals | 7 year follow-up–lesion had increased to 8 mm |
| Sountoulides et al. (2007)<sup>[9]</sup> | 22 | Acute hemiscrotal pain. AVM of spermatic cord | Not done | - | Scrotal ultrasound Color Doppler. | A diagnostic scrotal exploration was negative. The patient presented again 4 months later with acute hemiscrotal pain; an orchiectomy was done with consent. Biopsy revealed spermatic cord AVM | 2 year follow-up–no recurrence of scrotal pain |

Table 1 (contd...)
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| Author (year) | Age (years) | Age and presentation | Thrill/Bruit | Abnormal sperm analysis | Investigations | Management | Follow-up |
|---------------|-------------|----------------------|--------------|-------------------------|---------------|------------|-----------|
| Yilmaz et al. (2009) | 51 | Pain and throbbing sensation in hemiscrotum. Intrascrotal AVM simulating varicocele | Pulsatile vessels + | Not done | Scrotal ultrasound. Confirmed at duplex Doppler | Not mentioned | Not mentioned |
| Bezirdjian et al. (1989) | 24 | Painless progressively enlarging right scrotal mass | + | Not done | Ultrasound initially diagnosed as varicocele. Arteriogram showed hypertrophied bilateral internal pudendal and a branch of right medial femoral circumflex arteries. | Angioembolization was done with polyvinyl alcohol sponge (Ivalon). On third postembolic day, entire scrotal skin necrosed with fever. At debridement, a stagnant AVM with extruding Ivalon particles was found. Biopsy confirmed scrotal AVM | Not mentioned |
| Jagannathan et al. (2011) | 2 | 2 cases, both presenting with scrotal swelling and bleeding | Not done | Not done | Ultrasound and Color Doppler Arteriography-left internal pudendal a feeders. | Selective angioembolization was done with polyvinyl alcohol. Parents refused surgery | 13 months follow-up—asymptomatic |
| Hatten et al. (2009) | 32 | A case of spontaneously bleeding scrotal AVM that developed after remote trauma | + | Not done | Article ahead of print. Further details not known. | | |
| Auman (1985) | 39 | Iatrogenic, postvasectomy Varicocele complicating arteriovenous fistula (AVF) | + | Not done | Surgical exploration. AVF between deferential artery (a branch of the hypogastric artery) and pampiniform plexus | Ligation (both artery and veins) | |
| Obadia et al. (1990) | 40 | Iatrogenic, following radical orchietomy for testicular cancer. Varicocele complicating arteriovenous fistula | + | Not done | Angiography—AVF between epigastric artery and pampiniform plexus | Surgical repair | |
| Romagnoli et al. (2004) | 44 | Spontaneous onset. Varicocele complicating arteriovenous fistula | + | Not done | Angiography—AVF between external pudendal artery and pampiniform plexus | Ligation (both artery and veins) | 1 year follow-up—no recurrence |
| Minei et al. (2008) | 40 | Spontaneous onset. Varicocele complicating arteriovenous fistula | - | Not done | CT angiography—AVF between testicular artery and pampiniform plexus | Laparoscopic ligation (both artery and veins) | 18 month follow-up—no recurrence |

Our patient also had a history of acute scrotal pain 4 months prior to presentation. There have been cases published of scrotal AVMs presenting either as painless paratesticular masses or as incidental findings during evaluation for infertility or as a combination of both infertility and scrotal swelling. The deleterious effect of scrotal AVM on spermatogenesis might be mediated by elevation in scrotal temperature causing oligo or azoospermia, which have been shown to improve following surgery. Scrotal AVMs appearing as masses can be detected by pelvic arteriography and managed...
by superselective embolization; however, this need not always be successful, \cite{2,3,7} making open surgical excision a necessity. Embolization’s primary benefit is as a preoperative procedure to reduce bleeding during surgery. Transcatheter embolization may be a useful procedure to control bleeding in an emergency setting \cite{13} and also as a permanent procedure in selected cases. However, embolization is not bereft of complications—necrosis of skin and/or gluteal muscles, bladder infarction, and, rarely, impotence—which can be avoided by superselective catheterization and embolizing as distal as possible, with the catheter close to the nidus. \cite{13}

Angioembolization was not curative in several reports of scrotal AVMs \cite{2,3,7} and warranted a subsequent surgical excision for resolution of symptoms. Preoperative angioembolization was done in three cases \cite{4,6,12} Bezirdjian et al. reported a complication of skin necrosis following transcatheter embolization with polyvinyl alcohol, which required debridement. All these reports claimed technical success seen by opacification of the feeder vessels after delivery of the embolizing material; however, clinical success was not attained by angioembolization alone, except in one report of two cases. \cite{13} Angioembolization carries the additional disadvantage of radiation exposure, the awareness of which is the purpose of this article. An elegant study on the radiation exposure sustained in varicocele embolization by Chalmers et al. \cite{18} estimated lifetime fatal cancer risk being of the order of 0.1%. They have commented that while most patients presenting for vascular interventions are elderly with severe vascular or other comorbid conditions, varicocele embolization is performed in healthy young men with normal life expectancy. Therefore, every effort must be made to minimize radiation exposure. It is not possible to discard direct testes exposure to the beam during the procedure, \cite{19} even though the option of using a gonad protector is available in the case of a varicocele. This option cannot be exercised in the face of a scrotal AVM, which directly overlies the testes. The stochastic radiological risk is relevant. The deterministic dose for temporary testicle sterility quoted by Santiago is 150 mGy. It is probable that many operators are unaware of the radiation doses incurred by varicocele embolizations in their own practice. Moreover, if enlightened about the level of radiation risk, it is likely that some patients would decline radiological intervention in favor of the safer alternative of surgical excision. \cite{18} These comments can be extrapolated for scrotal AVMs as well since they both occur in around the same anatomical location.

Surgical excision is the definitive management. Typically, embolization offers only transient improvement because of the recruitment of new collaterals by the nidus. A preoperative angioembolization must be followed by surgery within 24–72 h; \cite{20} it provides temporary occlusion of the nidus and facilitates surgery. The complication of surgery is the intraoperative blood loss, which can be minimized by a careful technique, particularly in superficial, easily accessible locations.

Recurrence is possible, therefore all patients should be made cognizant of this possibility and followed up. Our patient is now 4 months postsurgery and has had no recurrence till date.

**Limitations**

Preoperative semen analysis was not done as the patient refused to give a sample. The sample may have revealed abnormal counts. \cite{2,7,6} Preoperative embolization was not done as the patient refused to give consent once informed about the risks of radiation and periprocedural complications.

**CONCLUSION**

AVMs of the scrotum should be considered in the differential diagnoses of scrotal swellings, more so if coexisting with a history of spontaneous bleed or inexplicable recurrent scrotal pain. \cite{3} Radiological confirmation of the diagnosis should be done. Diagnostic angiography is the gold standard in imaging and recommended to identify the feeder vessels. Complete surgical excision is recommended as the definitive treatment. Preoperative angioembolization may be helpful for a subsequent surgical endeavor, but must not be relied upon for clinical resolution. \cite{3,6}

We wish to emphasize that the need of minimally invasive therapy is limited when safer surgical alternatives are available for easily accessible areas, especially superficial structures. Angioembolization, which has its reported complications \cite{12} and limitations in effecting a complete cure, \cite{2,3,7} maybe an unnecessary exercise in these sites. It may in addition cause foreign body granulomas, which have been documented in histopathological examinations. \cite{6,12} An added drawback is the real but underestimated risk
of radiation exposure that has to be explained to patients. Both the prescribing and interventional specialists should know the radiation doses associated with the procedure and their effects. Additionally, they are also legally required to provide the patient with this information, i.e., the informed consent document must reflect patient agreement to accept the derived radiological risks. Nevertheless, management must be individualized; if the surgeon feels a preoperative angioembolization would be helpful in reducing the intraoperative blood loss, particularly with AVMs in deep locations near critical structures, this is perfectly justifiable, and may be performed after an informed consent.

To our knowledge, this would be the first reported instance in the English literature, where surgery was attempted without cover of embolization for a scrotal AVM. The wide access incision used with excision of redundant skin definitely made surgical resection easier, and gave better cosmesis as an added benefit.

Key Messages
Arteriovenous malformations of the scrotum are a rare occurrence, and mandate complete surgical excision whenever possible. Preoperative angioembolization, although helpful, need not always be successful and carries a significant risk of under-reported radiation exposure and sterility. Patients must be appropriately counselled and allowed to make an informed choice of this therapeutic modality.

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