

1 SUBMITTED 8 DEC 21
2 REVISIONS REQ. 21 FEB 22; REVISIONS RECD. 5 APR 22
3 ACCEPTED 19 APR 22
4 **ONLINE-FIRST: APRIL 2022**
5 DOI: <https://doi.org/10.18295/squmj.4.2022.034>

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7 **Recurrent scrotal Arteriovenous Malformation as a Slowly**
8 **Increasing Left Testicular Swelling**

9 *A case report*

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24 **Abstract**

25 Arteriovenous malformations (AVMs) are benign vascular lesions. Although, the
26 majority of AVMs occur in the central nervous system, there are published reports of
27 AVMs involving all systems including the scrotum, kidney, and uterus. Herein we
28 report a case of 37 years old male presented with recurrent gradual scrotal swelling
29 for 4 years attributed to scrotal AVM. Embolization was done but one year later his
30 symptoms reoccurred. As a result, left partial scrotal wall excision was carried out
31 without complications.

32 **Keywords:** arteriovenous malformation, AVM, scrotal swelling, scrotal malformation.

33

34 **Introduction**

35 Arteriovenous malformations (AVMs) are benign vascular lesions. They are described
36 as abnormal vessels fed by arteries and drained by veins without intervening
37 capillaries.^{1,2} Although the majority of AVMs occur in the central nervous system,
38 there are published reports of AVMs involving the scrotum, kidney, and uterus.^{2,3,4}
39 Few cases of scrotal AVMs have been described in the literature. Based on the
40 published reports, the clinical presentation of scrotal AVMs is highly variable,
41 ranging from an incidental finding on imaging for infertility to a bleeding mass.^{1,5}
42 Since scrotal AVMs have variable presentations and is rarely described in the
43 literature, we are reporting a case of a 37-year-old male presented with a slowly
44 increasing left testicular swelling attributed to scrotal AVM.

45

46 **Case Report**

47 37 years old male smoker presented to the urology clinic with a gradual scrotal
48 swelling that started four years ago. He complained of on and off scrotal pain,
49 occasional feeling of scrotal warmth, and scrotal discomfort. The patient was
50 diagnosed in another hospital with a testicular artery aneurysm and left testicular
51 varicocele. The patient denied any history of trauma, urinary tract infection, voiding
52 symptoms, previous surgeries, and his past medical history was unremarkable. Upon
53 physical examination, the testes were intra-scrotal. There were no signs of
54 inflammation, and the cremasteric reflex was intact bilaterally. Both epididymides
55 were palpable and non-tender. However, pampiniform plexus at the neck of the
56 scrotum was very pulsatile (figure A). Moreover, multiple skin varices over the left
57 scrotum were seen. Urinalysis was normal and urine culture was negative. Routine
58 laboratory tests were unremarkable. Abdominal and pelvis computed tomography
59 (CT) showed left scrotal arteriovenous malformation with enlarged small and
60 medium-sized serpiginous structures with a feeder artery arising from the proximal
61 superficial artery. Two months later, the patient was referred to interventional
62 radiology for embolization, which was successfully done utilizing Onyx 18% (figure
63 B). After one year, on follow-up, the pampiniform plexus were pulsatile again which
64 necessitated a CT angiogram. CT angiogram confirmed the recurrence of
65 arteriovenous malformation. The patient was counselled about the available treatment
66 options and given time to decide. Due to the risk of recurrence as well as the

67 possibility of technical failure with embolization, he decided to go with the surgical
68 treatment. The patient was booked for surgery, and partial scrotal wall excision was
69 done through an elliptical incision (figure C). Three arteries that feed into the
70 arteriovenous malformation were identified and controlled with vicryl ties. The
71 malformation and the skin that covering it were removed and sent to the pathology
72 lab. Dartos muscle was closed in a multi-fashion layer. The skin was closed by vicryl
73 rapide 4-0 in a vertical mattress. The histopathological study confirmed the diagnosis
74 by detecting vascular structures extending from fibrofatty tissues measuring 8x0.5 cm
75 grossly, and prominent subcutaneous large congested vascular spaces
76 microscopically. The patient was discharged one day after the surgery with no
77 complications. Two months postoperatively, the patient was doing fine with no active
78 complaint, and the wound healed properly.

79

80 The consent was obtained orally as the images were taken from the patient in the
81 clinic. We explained to him the importance of reporting and publishing his case for
82 educational purposes, and he agreed.

83

84 **Discussion**

85 AVMs are malformations in the circulatory system characterized by arteries and veins
86 that are not connected by capillaries leading to various degrees of ischemia and
87 pain.^{1,2} Even though Central nervous system cases represent the majority of AVMs,
88 there are published reports of AVMs involving the kidney, uterus, and scrotum.^{2,3,4}
89 AVMs are rarely present in the urinary tract.⁶ We reviewed four previously published
90 scrotal AVM cases (Table 1). All revealed ages ranging from 19 to 31 years while our
91 patient was 37 years old. Scrotal AVM embraces wide-ranging presentations
92 including infertility, acute recurrent pain in the hemiscrotum, pain and swelling on the
93 testicle, and progressive diffused swelling in the scrotum with flashing skin and local
94 warmth. Our case presented with gradually increasing left testicular swelling with on
95 and off scrotal pain, and occasional feeling of scrotal warmth and discomfort. Of the
96 four cases we have reviewed, three denied any history of trauma, and one had a
97 positive trauma history which was a severe pelvic fracture, and the patient indicated
98 that there is difficulty in maintaining erection since. Our patient denied any trauma
99 history. Varicocele was found in two of the cases and was seen by sonography
100 whereas our patient had multiple skin varices above the left scrotum that was seen

101 during physical examination. Each one of the four cases we reviewed diagnosed
102 scrotal AVM with a different modality. Some were challenging and required
103 orchiectomy for a diagnosis while others were simple and detected by pelvic
104 angiography. Our patient was diagnosed by abdominal and pelvic CT. Two studies
105 were able to find and embolize the feeding arteries. Our patient underwent
106 embolization but had recurrence one-year later. Similar to our case, surgical
107 intervention was eventually done in all four cases, and it varied from left scrotal AVM
108 excision, orchiectomy, ileo-femoral bypass, and resection of the whole left side of the
109 scrotum. In our case, partial scrotal wall excision was done. After the surgical
110 intervention, all patients were symptom-free.^{5,6,7,8} We believe that the difference in
111 the presentation could be attributed to the location of the AVM, onset, duration, and if
112 there is a history of trauma. A possible explanation for the differences in imaging
113 modalities used to diagnose scrotal AVMs is the availability of imaging techniques in
114 the hospitals that encountered those cases. The decision of surgical intervention is
115 mainly based on the symptoms and how symptoms negatively affect the patient's
116 quality of life.

117

118 **Conclusion**

119 Our case calls attention to a rare and challenging diagnosis that is scrotal AVM.
120 Recurrent scrotal pain, swelling, and warmth together with varicocele should raise
121 suspicion for scrotal AVM. Treatment varies depending on the symptoms present. We
122 believe that embolization of the feeding arteries is a possible option to start with, and
123 surgery should be preserved for recurrent cases.

124

125 **Authors' Contribution**

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

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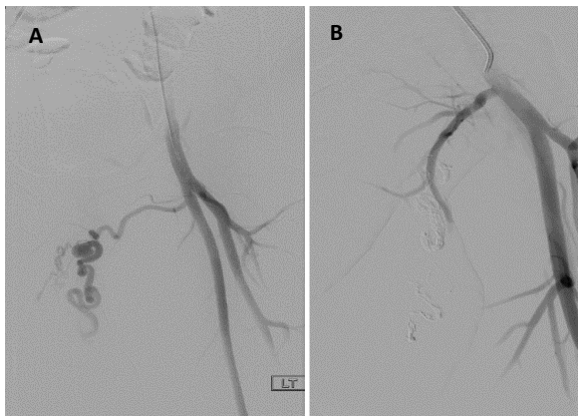
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159 **Figure 1:** a picture of the left scrotal swelling with clear multiple skin varices.

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161

162 **Figure 2:** the left image (A) showing a feeder artery supplying scrotal AVM. the right
163 image (B) angiogram following Onyx embolization through the AVM is almost
164 occluded.

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166

167 **Figure 3:** partial left scrotal wall with AVM excision.

Author	Age	Trauma History	Presentation	Semen analysis	Varicocele	Diagnosis method	Embolization	Treatment	Follow-up
Monoski et al. ⁵	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	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	22	No history of trauma	Acute recurrent pain in the right hemiscrotum	Not Performed	No evidence of varicocele	Post-orchietomy specimen	Not performed	Orchiectomy	2 years post-surgery, there was no complain
Mohammed et al. ⁸	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