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CASE REPORT
Reconstructive

Resection and Reconstruction of Giant Abdominoscrotal Arteriovenous Malformation

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Summary: Genital arteriovenous malformations are rare and present unique surgical challenges in preserving urogenital function, abdominal wall integrity, and lower limb perfusion. A 32-year-old man with a giant abdominoscrotal arteriovenous malformation presented with recurrent heavy bleeding. Due to the high risk of rebleeding and fatal hemorrhage, surgery with curative intent was proposed and the patient was counseled on the risks of ischemia to the lower limb, testes, and penis. Preoperative embolization of the feeding vessels was performed. Three days later, surgical excision of the mass with the affected scrotum, left rectus muscle, sheath, and overlying abdominal skin followed. The testes were dissected from the malformation and preserved along with the right internal pudendal artery. The left thigh skin was advanced to the scrotal remnants and a neoscrotum created. The resulting large abdominal wall defect was reconstructed in layers with a pedicled anterolateral thigh flap, including innervated vastus lateralis muscle, to prevent herniation. Recovery was uneventful, and a 4-year follow-up revealed no significant clinical or radiological recurrence with recovery of flap sensation, retained erectile function, and no herniation. We report this case due to rarity of giant abdominoscrotal arteriovenous malformations and present preoperative embolization, surgical resection, and functional anterolateral thigh flap reconstruction as a valuable treatment option of this life-threatening illness. *(Plast Reconstr Surg Glob Open 2020;8:e2725; doi: 10.1097/GOX.0000000000002725; Published online 26 March 2020.)*

Giant arteriovenous malformations (AVMs) cause serious symptoms and may progress to cause heart failure or fatal bleeding.[1] Management is often multidisciplinary and individualized with options, including conservative therapy, endovascular treatments, and surgery. We report the case of a rare giant abdominoscrotal AVM managed by surgical resection and function-preserving reconstruction with an anterolateral thigh (ALT) flap.

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CASE REPORT

A 32-year-old man suffering from scrotal AVM since childhood was presented with a 3-year history of recurrent heavy scrotal bleeding seeking surgical treatment. Bleeding was often due to trivial trauma such as scratches and had progressively worsened over the last 6 months requiring multiple emergency hospital admissions. On examination, the AVM mass was seen over the left abdominal and inguinal regions extending to the scrotum, which was enlarged with dilated tortuous vessels, pushing his penis to the right. Contrast-enhanced computed tomography (CT) scan revealed an AVM from the scrotum to 10 cm above the umbilicus extending into the left rectus abdominis muscle with 4 main feeding vessels (Fig. 1 and **see Video 1 [online]**, which displays the preoperative CT angiography showing the arteriovenous malformation and the dilated feeding vessels. The left-side vessel diameters: deep inferior epigastric artery 10 mm, veins 5 mm; superficial inferior epigastric artery 5 mm, vein 5 mm; external pudendal artery 3 mm, and internal pudendal artery 3 mm): the left, deep inferior epigastric artery (DIEA),

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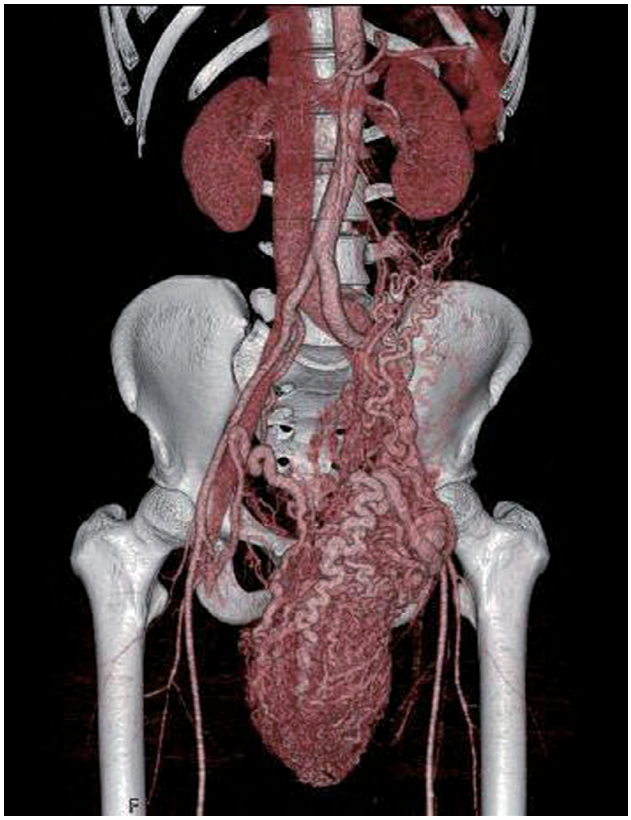

Fig. 1. Preoperative CT angiography showing the arteriovenous malformation and dilated feeding vessels, from the scrotum to 10 cm above the umbilicus.

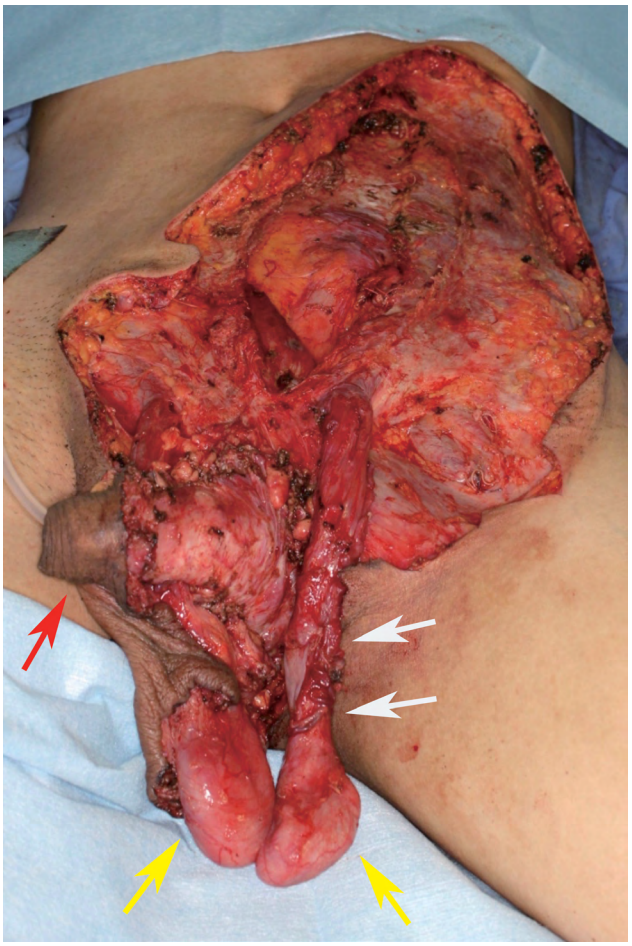

Fig. 2. The defect after resection of the arteriovenous malformation with preserved vascularity of the penis and testes. Red arrow: penis, yellow arrows: testes, and white arrows: spermatic cord.

superficial inferior epigastric artery (SIEA), external pudendal artery, and internal pudendal artery (IPA).

A multidisciplinary approach was taken with consultation of vascular surgery, urology, radiology, and orthopedic surgery. Due to the high risk of rebleeding and fatal hemorrhage, surgery with curative intent was proposed (**see figure, Supplemental Digital Content 1**, which displays preoperative photograph showing the extent of the AVM, the planned resection, and ALT flap design, http://links.lww.com/PRSGO/B349). The patient and his family were counseled on the proposed surgery and the risk of loss of function or amputation of the left lower limb, testes, or penis due to blood flow interruption. Preoperative semen analysis revealed azoospermia; however, his testosterone level was normal.

Preoperative embolization of the feeding blood vessels was performed using sixty 5 mm coils and *N*-butyl cyanoacrylate glue. Three days later, surgical excision of the AVM and overlying skin followed. Intraoperatively, the left rectus muscle (length 15 cm, width 5 cm, thickness 3 cm) and anterior rectus sheath from the level of the umbilicus and below were excised. The external iliac and femoral arteries were identified for control, following which the dilated DIEA, SIEA, and accompanying veins were ligated.

Preservation of the posterior scrotal skin was planned; however, most of the left scrotum skin had to be resected due to bleeding. The left IPA, which had a palpable thrill, was resected leaving the right IPA intact to preserve erectile function. The suspensory ligament of the penis and dorsal penile nerve were retained. The right testis was partially surrounded by the AVM but was separated with ease using blunt finger dissection (see **figure, Supplemental Digital Content 2**, which displays intraoperative photograph showing the AVM completely surrounding the left testis and cord and the right testis separated from the AVM. Dashed lines indicate right and left boundaries. http://links.lww.com/PRSGO/B350). The left testis and spermatic cord were completely covered by the AVM with multiple adherent small blood vessels. We freed the left testis and spermatic cord by careful release and electrocautery, gently separating the AVM using Kelly forceps while cutting using bipolar or monopolar as needed for hemostasis (Fig. 2). Both testes were atrophic, suggesting reduced function. Nevertheless, both were fixed to the remaining scrotum skin to prevent torsion, and the upper inner left thigh skin was then advanced to the right scrotal skin remnants to create a neoscrotum.

An 18 × 8 cm pedicled ALT flap including 20 × 12 cm fascia and 15 × 6 cm vastus lateralis (VL) with branch of femoral nerve preserved was used to reconstruct the abdominal wall (Fig. 3). A tunnel with a width of 8 cm was

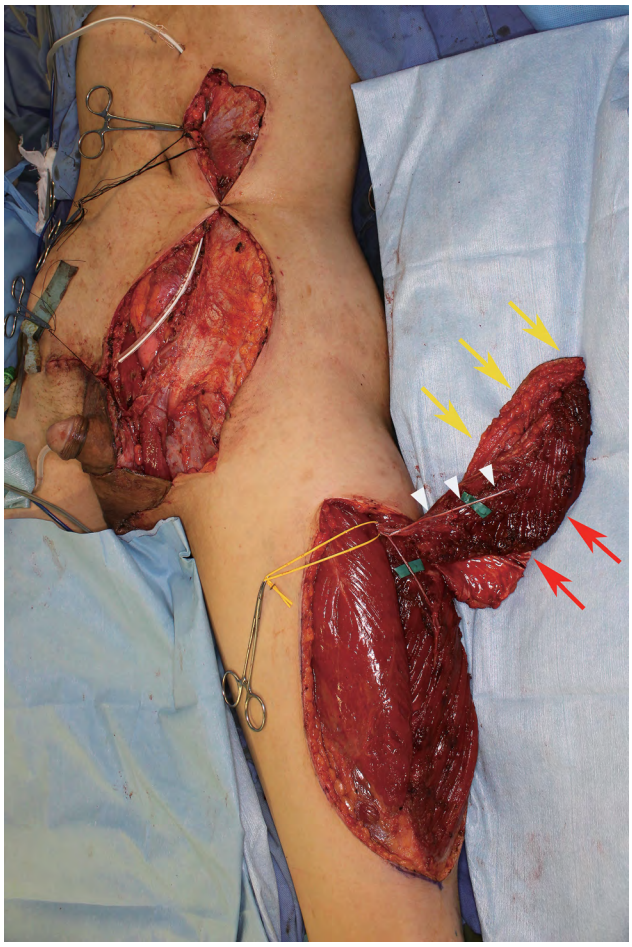

Fig. 3. The ALT flap including vastus lateralis and its fascia. Yellow arrows: ALT flap, red arrows: vastus lateralis, and white arrowheads: a branch of femoral nerve.

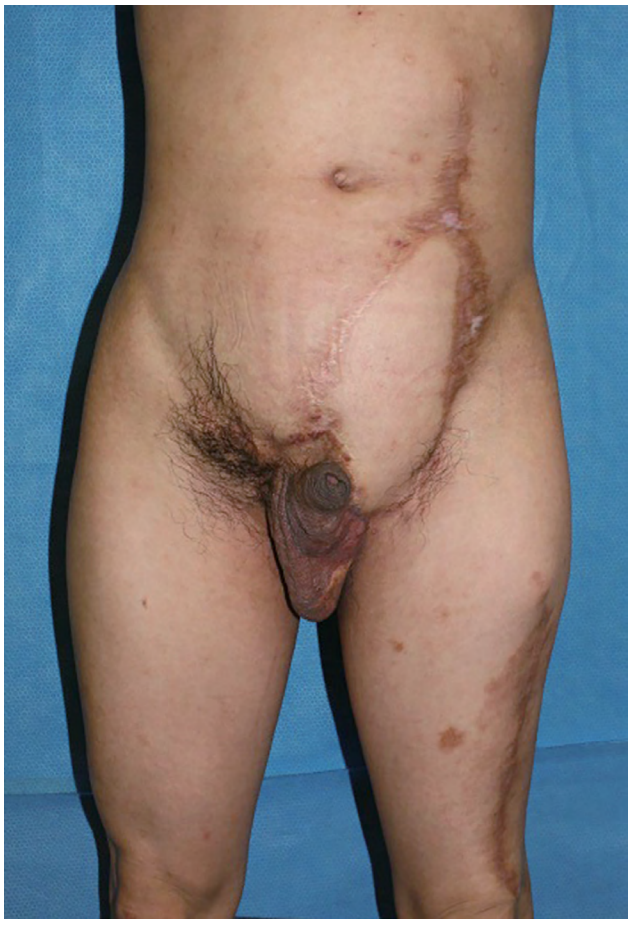

Fig. 4. Result 2 years after surgery showing the reconstructed abdominal wall and scrotum with no significant clinical or radiological recurrence of the AVM.

created below the rectus femoral muscle and the sartorius muscle, and the flap was delivered through the tunnel. The VL fascia lata was used to reconstruct the anterior rectus sheath defect and the muscle used to fill the rectus abdominis muscle defect, while the descending branch of lateral femoral cutaneous nerve was sutured to an intercostal nerve to enhance sensory recovery of the ALT flap (see **figure, Supplemental Digital Content 3**, which displays the ALT flap insertion for the reconstructed abdominal wall. (White arrows: fascia of ALT flap; http://links.lww.com/PRSGO/B351). Total blood loss was 705 mL, and no transfusion was required.

Recovery was uneventful, and 4-year follow-up revealed no significant recurrence on CT scan (**see Video 2 [online]**, which displays the postoperative CT angiography taken 3 years after operation showing the remaining preserved vessels with no significant arteriovenous malformation recurrence) with recovery of flap sensation (Semmes-Weinstein Monofilament 3.22), retained erectile function, and no herniation (Fig. 4). There was no lymphorrhea or lymphedema, and the patient was extremely satisfied with the overall outcome.

DISCUSSION

Genital and abdominal AVMs are rare and present unique surgical challenges in managing hemorrhage, preventing recurrence, and preserving urogenital function, aesthetics, abdominal wall integrity, and lower limb perfusion.[2–8] Zachariah et al[9] reported and reviewed several relatively smaller, localized scrotal AVMs managed by excision and direct closure with and without preoperative embolization. In contrast, only one previous report of giant scrotal AVM was found, which was managed nonoperatively, perhaps in view of the challenges mentioned above.[2] To the best of our knowledge, this case presents the first complete resection and flap reconstruction of a giant abdominoscrotal AVM.

Firstly, control of the main contributing vessels DIEA, SIEA, external pudendal artery, and IPA (**see Video 1 [online]**, which displays the preoperative CT angiography showing the arteriovenous malformation and the dilated feeding vessels. The left-side vessel diameters: deep inferior epigastric artery 10 mm, veins 5 mm; superficial inferior epigastric artery 5 mm, vein 5 mm; external pudendal artery 3 mm, and internal pudendal artery 3 mm) was essential for safe surgical excision. To attain this, extensive preoperative embolization was performed, minimizing

intraoperative blood loss by reducing the blood flow to the AVM.[3,5] This enabled block excision with the associated skin, fascia, rectus muscle, and anterior sheath. Due to the location of the AVM encircling the testes and the base of the penis, there was a risk of inadvertent injury to these structures with the testicular vessels most vulnerable during release of the spermatic cord. In such an event, temporal ectopic replantation of the testes in the axilla was planned. However, vascularity of both testes and the penis was maintained after AVM resection (Fig. 2). Additional conservation of the dorsal penile nerve and right IPA ensured preserved erectile function. Aboseif et al[10] reported marked reduction in erectile function when both IPAs were damaged, hence we retained the right-sided one. AVMs often occur on 1 ipsilateral side of the body. Interestingly, in our case, though the AVM seemed to infiltrate the right scrotum from its location on the left, separation of structures on the right was done with ease and we could preserve the right-sided vessels. Vienet-Legué et al[7] and Brotherton and Yazdany[8] reported surgical resection of large vulvar AVM in adolescent girls, both of whom developed recurrence 2 months later, resulting in further surgery and embolization therapy. These cases emphasize the importance of careful dissection and adequate AVM resection in preventing recurrence. Additionally, they also highlight the importance of perioperative patient counseling in genital AVM as was seen in our case, as genital function or cosmesis may need to be sacrificed to attain adequate AVM resection.[7,8]

Following AVM resection, the resulting abdominal wall defect was too large for direct closure (Figs. 2, 3). Major complications following such resection of rectus muscle and sheath include hernias, wound dehiscence, surgical site infection, imbalance of truncal muscular support, and unsightly abdominal contour.[11] The pedicled ALT with VL flap provided well-vascularized tissue for reconstruction of the structural and contour defects. This flap has been shown to provide good functional recovery and quality of life (mean Hernia-Related Quality-of-life Survey: 73.44%) with minimal donor site morbidity when used for reconstruction of complex abdominal defects.[11] However, the benefits of preserved VL innervation are disputed and require further study.[12] The flap design did not allow for inclusion of the intact lateral femoral cutaneous nerve, but microsurgical coaptation to the intercostal nerve on flap insertion contributed to successful sensory recovery and overall patient satisfaction.

CONCLUSIONS

This case reports the successful resection of a rare giant abdominoscrotal AVM and presents preoperative embolization, surgical resection, and functional ALT flap reconstruction as a valuable treatment option for this life-threatening illness.

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