Case Report

A giant pelvic arteriovenous malformation

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Abbreviations & Acronyms
AVM = arteriovenous malformation
CT = computed tomography
DOV = Dominant outflow vein
NBCA = n-butyl-2-cyanoacrylate
pAVM = pelvic arteriovenous malformation
TAE = transcatheter arterial embolization

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Introduction: Pelvic arteriovenous malformations are rare in male patients. We present a case of pelvic arteriovenous malformation involving the seminal vesicle.

Case presentation: A 58-year-old man was diagnosed with pelvic arteriovenous malformation that involved the left seminal vesicle by angiography. The patient underwent three embolization procedures and made favorable progress after the embolizations.

Conclusion: Herein, we report a rare case of pelvic arteriovenous malformation involving the seminal vesicle treated by embolizations with good outcome.

Key words: angiography, dehydrated ethanol, embolization, pelvic arteriovenous malformation, seminal vesicle.

Keynote message

Pelvic arteriovenous malformations (pAVMs) are rare in male patients. Here, we present a case of the pAVM involving one of the seminal vesicles. The patient underwent three embolizations and made favorable progress.

Introduction

AVMs are uncommon vascular lesions characterized by abnormal connections (an abnormal capillary network) between the arterial and venous systems. Most of these lesions are congenital, while some result from trauma or surgery.[1] AVMs predominantly affect the extremities, head and neck, and lungs, with a higher incidence in women.[2] pAVMs are particularly rare, accounting for less than 2% of all AVMs, especially in men.[2–4] Only two case reports describe pAVM localized to the seminal vesicle.[1,5] In the pelvic region, most AVMs arise following trauma or surgical procedures.[1] pAVMs typically exhibit slow growth over an extended period, and their symptoms are often nonspecific.

Herein, we report the third case in world literature of pAVM involving the seminal vesicle and its treatment with three sequential embolization procedures.

Case

A 58-year-old man with no significant past medical history except for varices of the left lower extremity was referred to our hospital for the diagnosis of a growing pAVM involving the left seminal vesicle, which was incidentally found on CT (Fig. 1a). The physical examination was normal. Angiography revealed a pAVM located adjacent to the left seminal vesicle, with multiple feeding vessels arising from branches of the left internal iliac artery and draining into the dilated left internal iliac vein with a large venous sac (Fig. 1b,c). The patient was diagnosed with pAVM (type II). Although the patient had no subjective symptoms, he requested treatment due to the progressive increase in pAVM volume. Eventually, three sessions of TAE with dehydrated ethanol were necessary to treat the pAVM.

In the first session, two main feeding arteries arising from the left internal iliac artery were selectively embolized with 15.1 mL of dehydrated ethanol and 25% NBCA (Fig. 2a–c). In the

Fig. 1 (a) CT image showing the parapelvic AVM involving the left seminal vesicle. (b) Arterial phase angiographic image of the AVM with multiple feeding vessels arising from branches of the left internal iliac artery. (c) Venous phase angiographic image of the AVM, demonstrating a drainage vessel connected to the left iliac vein.

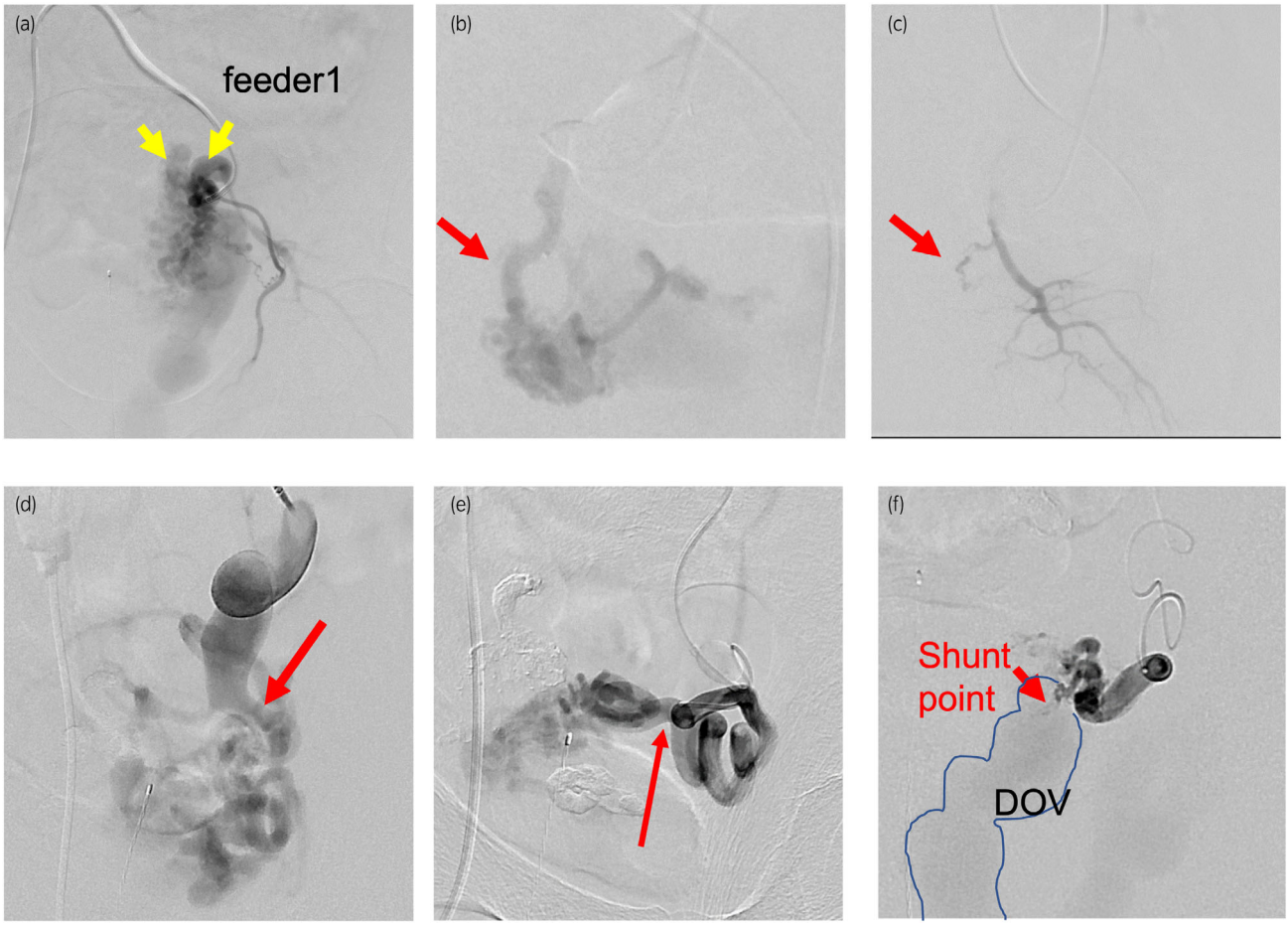

Fig. 2 (a) Arterial phase angiographic image highlighting the two main feeding arteries originating from the left iliac artery (indicated by yellow arrows). (b) Angiographic image of the two main feeders before treatment. The arrow indicates the area to be embolized. (c) Angiographic image of the feeders after the first treatment, shows the absence of contrast agent downstream of the arrow. (d) The arrow indicates a feeder that was not embolized during the initial treatment. (e) Area injected with ethanol during treatment. (f) Angiographic image revealing the main shunt point, which became apparent after the second treatment.

second session, another feeding artery arising from the left iliac artery, which was not treated in the previous treatment, was selectively embolized with 13 mL of dehydrated ethanol and 33% NBCA (Fig. 2d,e). After the second treatment, the main shunt point was recognized at the top of the venous sac (Fig. 2f). Temporary hematospermia was noted after the

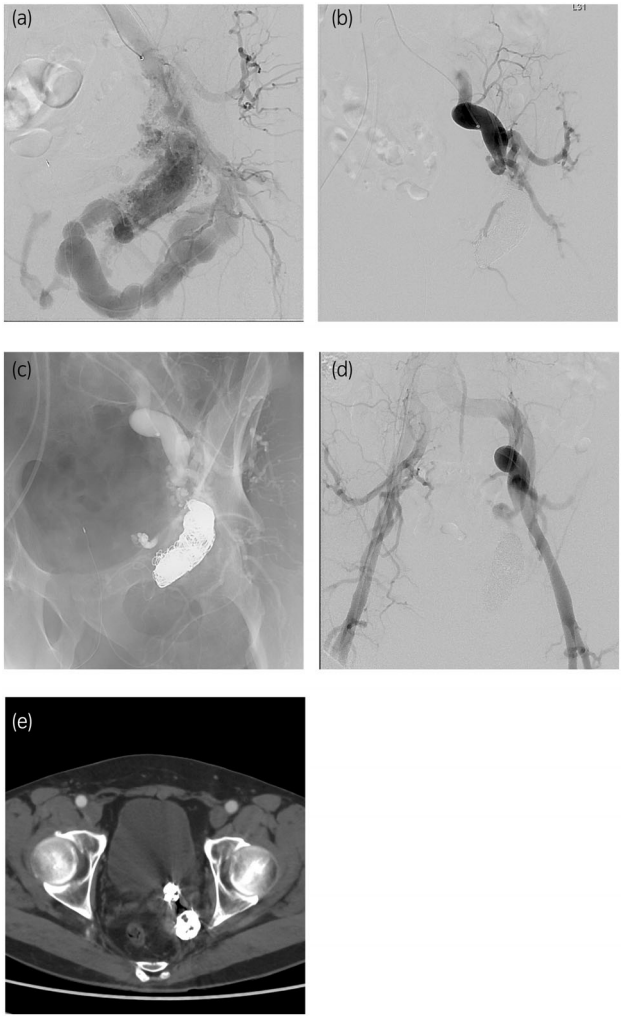

Fig. 3 (a) Venous phase angiographic image before the third session. (b) The main shunt point at the top of the venous sac, embolized with dehydrated ethanol and coils. (c) Angiographic image showing the coils used during the third session. (d) Angiographic image after the third session, where the pAVM is no longer detectable. (e) Contrast-enhanced CT image of the pAVM after the third session. The pAVM involving the left seminal vesicle is not detectable.

second session but disappeared spontaneously. In the third session, the main sac was embolized with a coil and dehydrated ethanol, and then another shunt point on the caudal side became apparent, which was embolized with coils and dehydrated ethanol (total 12.5 mL of dehydrated ethanol) (Fig. 3a–d). After the three treatments, the disappearance of the shunt flow was confirmed in the final angiography (Fig. 3e).

The post-embolization course was uneventful, and the patient remains free from recurrence at 1 year postoperatively.

Discussion

In general, pAVMs are more often due to pelvic trauma, tumors, surgical procedures, or pregnancy. Congenital pAVM, especially in male patients, are rare.[1,6] pAVMs account for approximately 3% of all AVMs, with 68% of congenital cases reported in women.[7]

Within the genitourinary tract, the major AVM lesions involve the kidney and the bladder, with six cases reported in the ureter.[8,9] Interestingly, there have been only two reported cases of pAVMs localized to the seminal vesicles, making this the third documented case worldwide (Table 1).[1,5]

AVMs must be differentiated from arteriovenous fistulas and hemangiomas.[1] An arteriovenous fistula is a solitary, irregular connection between an artery and a vein, typically acquired due to erosion or trauma.[6] On the other hand, a hemangioma is a congenital neoplasm of vascular tissue involving small vessels (vessels with small lumen size rather than wall thickness). In cases of hemangiomas, the feeding and draining vessels usually appear normal in histological and arteriographic examinations.[6]

Congenital pAVMs tend to grow slowly and become symptomatic later in life.[10,11] Common subjective symptoms include abdominal or pelvic discomfort, pain, rectal pain, tenesmus, and genitourinary complaints such as hematuria, hydronephrosis, hematospermia, impotence, orchitis, and sciatica. Remarkably, 20% of patients with pAVM remain asymptomatic.[1,11–13] In cases of large AVMs, cardiac failure may occur due to hemodynamic changes.[8]

Table 1 Three cases of pAVM localized to the seminal vesicle

Case	Reported year	Age	Chief complaint	Urological symptoms	Laterality	Feeder	Drainage	Treatment	Outcome
1	1999	39	Lower abdominal and pelvic pain	None	Right	Internal iliac artery	Pelvic vein	Selective arterial embolization by polyvinyl alcohol and surgical resection	Additional embolization was needed
2	2002	48	None	None	Right	Internal iliac artery and superior hemorrhoidal artery	Common iliac vein and inferior vena cava	Selective arterial embolization by histoacryl	Success with five sessions of treatment
Our case		58	None	None	Left	Internal iliac artery	Internal iliac vein	Selective arterial embolization by ethanol and NBCA	Success with three sessions of treatment

Diagnosis typically involves angiography. AVMs in the trunk and extremities are categorized based on the angiographic morphology of the nidus.

- Type I: No more than three separate arteries shunting to the initial part of a single venous component.
- Type II: Multiple arterioles shunting to the initial part of a single venous component (dominant outflow vein).
- Type IIIa: Fine multiple shunts between arterioles and venules presenting as a blush or fine striation on angiography.
- Type IIIb: Fine multiple shunts between arterioles and venules presenting as a complex vascular network on angiography.[14]

Indications for catheter intervention include symptomatic disease and growing lesions. Asymptomatic or mildly symptomatic lesions generally do not require treatment but should be monitored with ultrasound or CT scan studies every 6–12 months.[6]

Organ ischemia is a complication that must be anticipated during embolization. Even if the intended area can be embolized, the possibility of organ ischemia in the dominant area should be considered. The main causes of organ ischemia are overflow of embolic material to the central part of the vessels above the embolic zone, outflow to the periphery beyond the embolic zone, and inflow into critical vessels via collateral blood vessels.[15] The seminal vesicles are primarily nourished by the seminal artery and the branches of the internal iliac artery. Because of the dual vascular dominance of the seminal vesicle, it is considered to be at lower risk of organ ischemia associated with embolization compared to the single vascular dominant organs, and more aggressive therapeutic intervention could be considered.

Because of their complex vascular structures, pAVMs can be therapeutically challenging. However, in cases where the pAVM is localized in the perivesical space, catheter intervention may be effective.[4] Patients typically required a mean of 2.4 embolization procedures (range, 1–11) over an average period of 23.3 months (range, 1–144 months).[16] There was no difference in the mean number of embolizations between male and female patients.[16]

In this case, the patient was indeed asymptomatic and follow-up was an option, but he requested treatment. Considering the growing lesions which is in the indication for treatment and the possibility of symptoms appearing in the future, we decided to treat the patient. The possibility of organ ischemia in the seminal vesicle due to treatment was considered, but since the seminal vesicle is anatomically dual vascular dominant organ by the seminal artery and the branches of the internal iliac artery, complications were considered less likely compared to single vascular dominant organs. Three embolization procedures were needed, which is comparable to the 2.4 sessions reported in the literature.[16]

pAVM is a relatively rare disease with a wide range of subjective symptoms. Diagnosis is relatively easy if imaging studies are performed. Although this case was asymptomatic and discovered incidentally, in cases of unexpected pain or discomfort in the abdominopelvic region, hematuria or

hematospermia, it is necessary to treat the patient with the possibility of this disease in mind.

Conclusion

We present the third documented case of pAVM involving the seminal vesicle. The diagnosis was confirmed as a type II pAVM. The patient underwent three embolization procedures, resulting in favorable progress.

Author contributions

Taro Izumi: Writing – original draft. Marie Osawa: Writing – original draft. Ibuki Tsuru: Writing – review and editing. Akihiro Ono: Writing – review and editing. Haruki Kume: Supervision; writing – review and editing. Yoshiyuki Shiga: Writing – review and editing. Masaki Nakamura: Writing – original draft; writing – review and editing.

Conflict of interest

The authors declare no conflict of interest.

Approval of the research protocol by an Institutional Reviewer Board

Not applicable.

Informed consent

Written informed consent for publication was obtained from the patient.

Registry and the Registration No. of the study/trial

Not applicable.

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