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Original Article

Treatment of 23 spinal perimedullary arteriovenous fistulas in a single center: A simple and practical treatment strategy

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ABSTRACT

Background: The aim of the study is to present our strategy for stratifying patients with spinal perimedullary arteriovenous fistulas (PMAVFs) and apply the appropriate treatment.

Methods: This retrospective study included 23 patients with PMAVF. We divided the patients into three groups according to the location of the fistula and size of the predominant feeder: Group 1 (dorsal PMAVF, $n = 4$), Group 2 (nondorsal PMAVF having a predominant feeder through which the smallest coil-deploying microcatheter could pass, $n = 6$), and Group 3 (nondorsal PMAVF having no feeder through which the smallest available microcatheter could pass, $n = 13$). Group 1 underwent surgical treatment. All patients in Groups 2 and 3 underwent endovascular treatment with a liquid embolic agent, except one in Group 3, who opted for surgical treatment. Coil was used as a supplementary tool for treating lesions in Group 2. Patients' basic and clinical characteristics, treatment, and outcome data were recorded.

Results: Six patients were aged <15 years. Overall, patient fistulas were located in the thoracic region ($n = 11$), conus region ($n = 7$), and cervical spine ($n = 5$). Of the 18 PMAVFs who underwent endovascular treatment, 100% occlusion was observed in 14, 90% in 3, and 75% in 1. Nineteen patients had complete or partial recovery of neurological deficits. Six patients experienced temporary worsening immediately after treatment but recovered within 3 months. No bleeding or rebleeding was noted after either treatment.

Conclusion: Our simple strategy for stratifying PMAVF for treatment is easy to apply in clinical practice and results in favorable outcomes.

Keywords: Endovascular treatment, Outcome, Patient selection, Perimedullary fistula, Spinal vascular malformation

INTRODUCTION

A spinal perimedullary arteriovenous fistula (PMAVF), or Type IV spinal arteriovenous malformation (AVM), is an uncommon vascular malformation. PMAVF, a direct intradural and extramedullary arteriovenous fistula (AVF), is typically supplied by the anterior spinal artery, posterolateral artery, radiculomedullary artery, or the artery to the terminal filum. Its drainage is accomplished through the superficial perimedullary veins or pial venous network. Approximately 20–31% of spinal vascular malformations are PMAVFs. Spinal medullary AVMs and PMAVFs have similar clinical presentations.[2,21] However, spinal medullary AVMs are associated with a higher incidence of subarachnoid hemorrhage (37.9%) compared with PMAVFs.

Previous studies have arbitrarily classified PMAVFs according to the size and number of feeders, fistula size, and morphology of draining veins.[2,23] Some studies have used N-butylcyanoacrylate (NBCA) as the sole embolic material, whereas others have used both NBCA and platinum coils; without a standard method of treatment, outcomes have differed.[2,23] According to an analysis of a Japanese national registry,[26] endovascular treatment of PMAVF was successful in 83.6% of cases, with total occlusion achieved in 29.0% and a complication rate of 16.3%. Neurological improvement was achieved in 34.9% of cases, and modified Rankin Scale scores worsened in 27.9% of cases at 30-day follow-up. The discrepancies in success rates, complications, and outcomes observed among these studies can be attributed to variations in patient selection, treatment methods, and endovascular treatment protocols. The classifications described in these studies are subjective rather than quantitative. Further, no correlation was found between the classification of PMAVF and the management strategy applied to it, including the surgical approach.

Locating and eradicating the PMAVF shunt can be highly challenging in microsurgical treatment because PMAVFs are often located between congested veins, thus obscuring the fistula.[15,22,25] The goal of endovascular management of the PMAVF is to occlude the distal portion of the feeders, the arteriovenous shunt point, and the proximal portion of the draining vein.[21] Given the advancements in embolic materials and catheter technology, PMAVF treatment should be tailored according to a more practical and quantitative mode of measurement. This article thus proposes a strategy for stratifying patients with PMAVF and applying appropriate treatment.

MATERIALS AND METHODS

This retrospective study examined prospectively collected data and used the neuroradiology database to identify patients with a diagnosis of PMAVF. The ethics committee of National Taiwan University Hospital approved this study. Patients' basic and clinical characteristics, treatment, and outcome data were recorded.

All patients with PMAVF underwent magnetic resonance imaging examination and catheter angiography. The patients with PMAVF were stratified according to the location of PMAVF. We divided the patients into three groups [Figure 1] according to the location of the fistula and size of the predominant feeder: Group 1 (dorsal PMAVF), Group 2 (nondorsal PMAVF having a predominant feeder through which the smallest coil-deploying microcatheter could pass, n = 6), and Group 3 (nondorsal PMAVF having no feeder through which the smallest available microcatheter could pass). In regard to catheter technology, the available size of coil-deploying microcatheter was between 0.7 mm and 1.0 mm. The classification is illustrated in Figure 2.

All patients in Group 1 underwent surgical treatment. All patients in Groups 2 and 3 underwent endovascular treatment, except one patient who opted for microsurgical treatment.

In Group 2, we administered an intravenous bolus of hydrocortisone (5.3 mg/kg) before embolization. This was

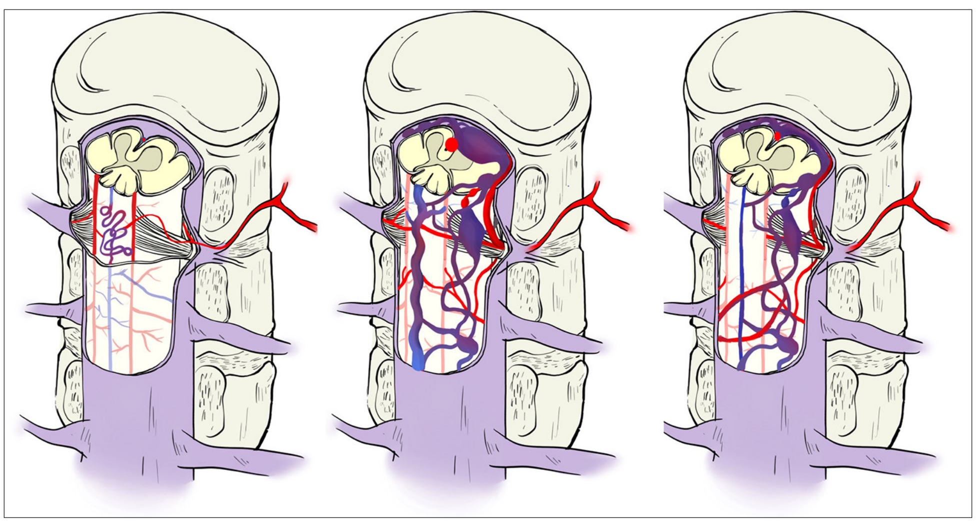

Figure 1: An illustration contrasting the different groups of perimedullary arteriovenous fistulas (PAMVF). Group 1 (left) is a PAMVF that occurs solely dorsal to the spinal cord. Group 2 (central) is a nondorsal PMAVF having a predominant feeder through which the smallest coil-deploying microcatheter could pass. Group 3 (right) is a nondorsal PMAVF having no feeder through which the smallest available microcatheter could pass.

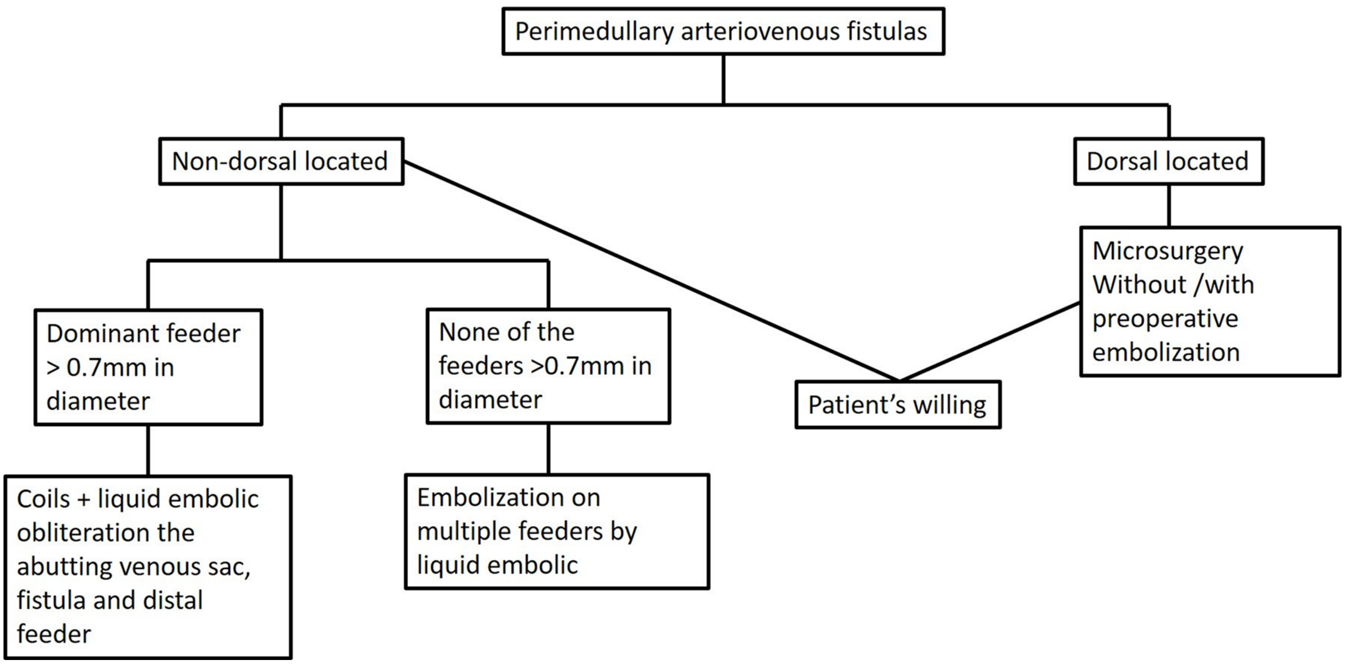

Figure 2: Algorithm used to stratify the patients with perimedullary arteriovenous fistulas.

repeated hourly for 3–7 days.[10] The microcatheter was inserted into the largest feeder and guided into the venous aneurysm just beyond the fistula. Subsequently, coils (manual push or electrically detachable coils) were deployed inside the venous aneurysm, fistula, and distal feeding artery until the flow slowed or was completely occluded. However, in some cases, the microcatheter could not pass to the venous aneurysm due to challenging anatomy, in which case, detachable coils were deployed at the distal feeding artery, and to prevent incomplete obliteration and augment the embolic material, the injection of liquid embolic followed this until it reached the proximal draining vein. Liquid embolic, such as NBCA, isobutyl 2-cyanoacrylate (Ethicon), or Onyx (ev3, Irvine, CA, USA), were used at different times. We avoided embolizing only the proximal feeding arteries. If obvious stasis of the contrast medium was noted in large draining veins after embolization, heparin was administered for 3 days, followed by an oral anticoagulant.[10,21] Embolization was deemed successful when the liquid embolic agent obliterated the distal feeding artery and proximal draining veins. Group 3 received multiple sessions of embolization of the feeding arteries. Before and after each session, the patient received intravenous hydrocortisone. The largest artery close to the fistula and proximal draining veins were embolized first.

Postembolization angiography results were classified as follows: complete occlusion, nearly complete occlusion (only slow residual arteriovenous shunting on angiography), and partial occlusion (with obvious residual arteriovenous shunting on angiography).

Patient outcomes were classified as excellent (complete recovery), improved (partial recovery of neurological signs), unchanged (same neurological status as before treatment), or worse (development of any new neurological signs) at clinical follow-up conducted at least 2 years after treatment.

RESULTS

We identified 156 patients diagnosed as having a spinal vascular malformation, of whom 23 (15 male and 8 female patients) had catheter angiography-confirmed PMAVF. Among them, 3 patients were 1–3 years old, 3 were 3–14 years old, 14 were 15–40 years old, and 3 were 40–56 years old. All patients had muscle weakness (sudden in 9; progressive in 14) as their primary symptom. Of these patients, nine had additional sensations or pain symptoms, and four had urine incontinence. The fistulas were located in the thoracic region (n = 11), conus region (n = 7), and cervical spine (n = 5). Of the included patients, 4 patients had Group 1 lesions, 6 patients had Group 2 lesions, and 13 patients had Group 3 lesions. In five lesions in Group 2, the main feeder of the PMAVFs was either the anterior spinal artery or the artery of Adamkiewicz. All lesions in Group 2 were associated with a dilated venous aneurysm near the fistula site.

Three of the four patients in Group 1 had a single feeding artery. All patients in Group 1 underwent surgery; the outcomes were excellent in two patients, improved in one, and unchanged in one. One patient had spontaneous closure of the fistula during the operation, which was confirmed through postoperative angiography [Figure 3]. No recurrence of clinical symptoms was noted in this patient during the 6-year follow-up. One patient in Group 3 underwent surgical treatment, which resulted in improved outcomes.

A liquid embolic agent was used in 18 patients in Groups 2 and 3 (NBCA in 12, Onyx in 3, and both in 2). Two patients received both NBCA and Onyx because of failed occlusion of the proximal drainage veins when using NBCA, and Onyx was used as salvage. Coiling was used in Group 2 [Figures 4 and 5].

Group 3 patients received 1–3 sessions of treatment [Figure 6]. Of the 18 PMAVFs managed endovascularly, complete occlusion was observed in 14 (77.8%), nearly complete occlusion in 3 (16.7%), and partial occlusion in 1 (5.5%). The outcome in the endovascular group (n = 18) was excellent in 10 patients (55.6%), improved in 5 (27.8%), and unchanged in 3 (16.6%); temporary neurological deficits were observed immediately after treatment in 6 patients

Figure 3: A 45-year-old man had mild bilateral lower leg weakness and upper extremity numbness for months. Sagittal T2-weighted magnetic resonance imaging revealed (a) an abnormal flow void (arrow) on the dorsal aspect of the C5/6 cord associated with some fine void spots in the ventral aspect of C3/4. (b and c) Lateral view of left vertebral angiography showing a Group 1 perimedullary arteriovenous fistula predominantly supplied by the left C5 radiculomedullary artery (yellow arrow) and posterior spinal artery (white arrow). (d and e) During the surgical treatment of the lesion, the abnormal vasculature and fistula spontaneously disappeared. The follow-up angiography confirmed it. The patient had no more neurological signs after the operation and has remained symptom-free for 6 years.

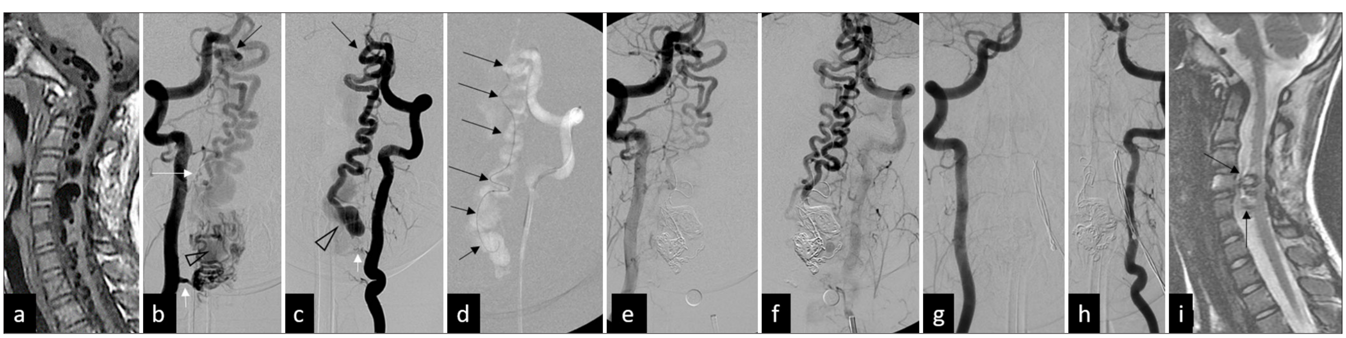

Figure 4: (a) A 4-year-old boy with left hemiplegia was diagnosed as having a Group 2 perimedullary arteriovenous fistula at the C4-5 level by magnetic resonance (MR). (b and c) On bilateral vertebral angiograms (frontal view b; right, c; left), the main feeding arteries are the bilateral anterior spinal artery (black arrows), with multiple small feeders from segmental radiculomedullary arteries (white arrows). A large venous aneurysm is noted near the fistula site (arrowheads). (d) Through the left anterior spinal artery approach, the microcatheter (black arrows) is navigated into the venous aneurysm, multiple detachable coils are deployed, followed by an Onyx injection, and (e and f) the shunting is completely obliterated. (g and h) Six-month follow-up angiography indicates no evidence of local residual or recurrent disease and (i) is confirmed by 2-year follow-up MR. Coil mass (arrows) is noted without abnormal void signal. The patient became symptom-free at the 3-year follow-up.

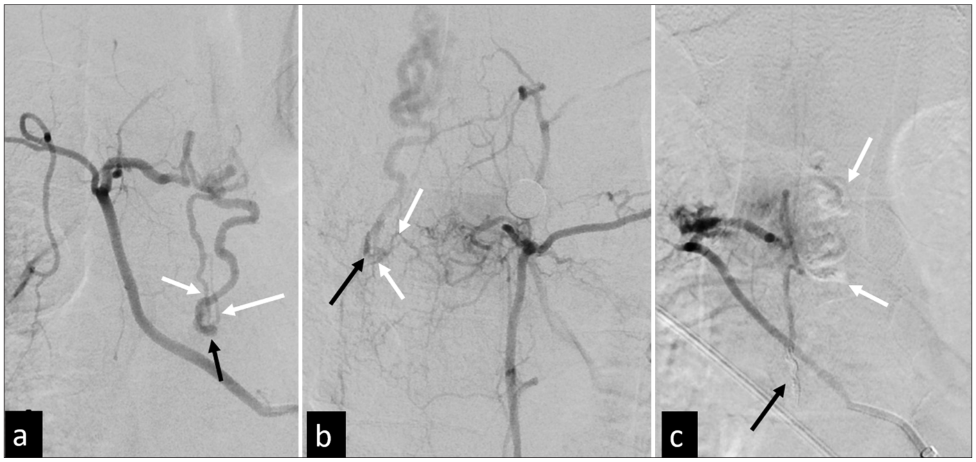

Figure 5: (a and b) A 34-year-old man presented with sudden-onset hemiparesis and loss of sensation in the lower extremities. The clinical diagnosis was Brown–Sequard syndrome below T6. The left T8 intercostal artery angiography reveals a group 2 perimedullary arteriovenous fistula with multiple radiculomedullary feeders (a and b; white arrows) sharing a common drainage (black arrows). (c) The lesion is treated with Onyx (white arrows) with complementary coiling (black arrow) at the distal main feeder. The patient experienced complete recovery and remained symptom-free at the 7-year follow-up.

(33.3%), which resolved or improved within 3 months. The mean follow-up time was approximately 7.3 years (2.5–15 years). This finding indicates that in long-term follow-up after endovascular treatment, none of the patient's condition worsened. No cases of bleeding or rebleeding after either endovascular or surgical management were noted.

DISCUSSION

In this report, we have presented our strategy for stratifying patients with PMAVF. We allocated patients to different groups, which received either surgical or endovascular treatment depending on the location of the lesion and the availability of endovascular materials.

We focused on the management strategy and classified PMAVF into three groups according to the location and size of the main feeder and the accessible treatment. Group 1 lesions, which involve dorsal PMAVFs, are more easily and directly approached using surgical treatment, whereas the fistula sites in Group 2 and Group 3 lesions are often difficult to locate, as they develop between congested veins, making localization and eradication of the shunt challenging.[15,25] In such scenarios, the fistula can be located using intraoperative near-infrared indocyanine green videoangiography[4,13] or intraoperative ultrasound.[24,27]

The key difference between the lesions in Groups 2 and 3 is the size of the dominant feeding artery. The diameter of the main arteries characterizes group 2 lesions according to the currently available smallest coil-deploying microcatheter (0.51–0.7 mm), such as the Excelsior SL-10 microcatheter (Stryker Neurovascular, USA) or Marathon microcatheter (Medtronic, Minneapolis, Minnesota, USA). This was done for two reasons. First, to date, the smallest microcatheter used in clinical practice for coil deployment has a diameter of approximately 1.5–1.7 F (0.51–0.68 mm). This is a limitation of the current technique, but the tool is available and suitable for our approach and planning in treating type 2 PMAVF lesions. Second, PMAVF lesions with a feeder larger than 0.7 mm are always associated with a large venous aneurysm close to the fistula; the endovascular technique is suitable in this case. With advances in microcatheter technology, even smaller microcatheters may be available for use in the future without changing this classification system.

Before Onyx was available, high concentrations (approximately 50%) of NBCA were used to treat brain AVM and AVFs using the "sandwich pushing technique" to manage fistulous components. However, the outcome was unpredictable due to the lack of control over the injection, which is a major concern when treating high-flow lesions, such as a PMAVF. One study by Hsu *et al.*[6] reported that the venous pouch abutting the brain AVM nidus can be approached from the arterial side and that prioritizing venous coiling can facilitate fistula occlusion. In this study, we coiled the venous aneurysm first, followed by the fistula, and ended at the distal feeding artery. Subsequently, a liquid embolic was injected at the distal feeding artery and inside the coil frame, which catches NBCA or Onyx. Coils offer greater stability compared to liquid embolic agents and are less likely to be washed away during the procedure.[29] In addition, coils can act as a framework to reduce the speed of

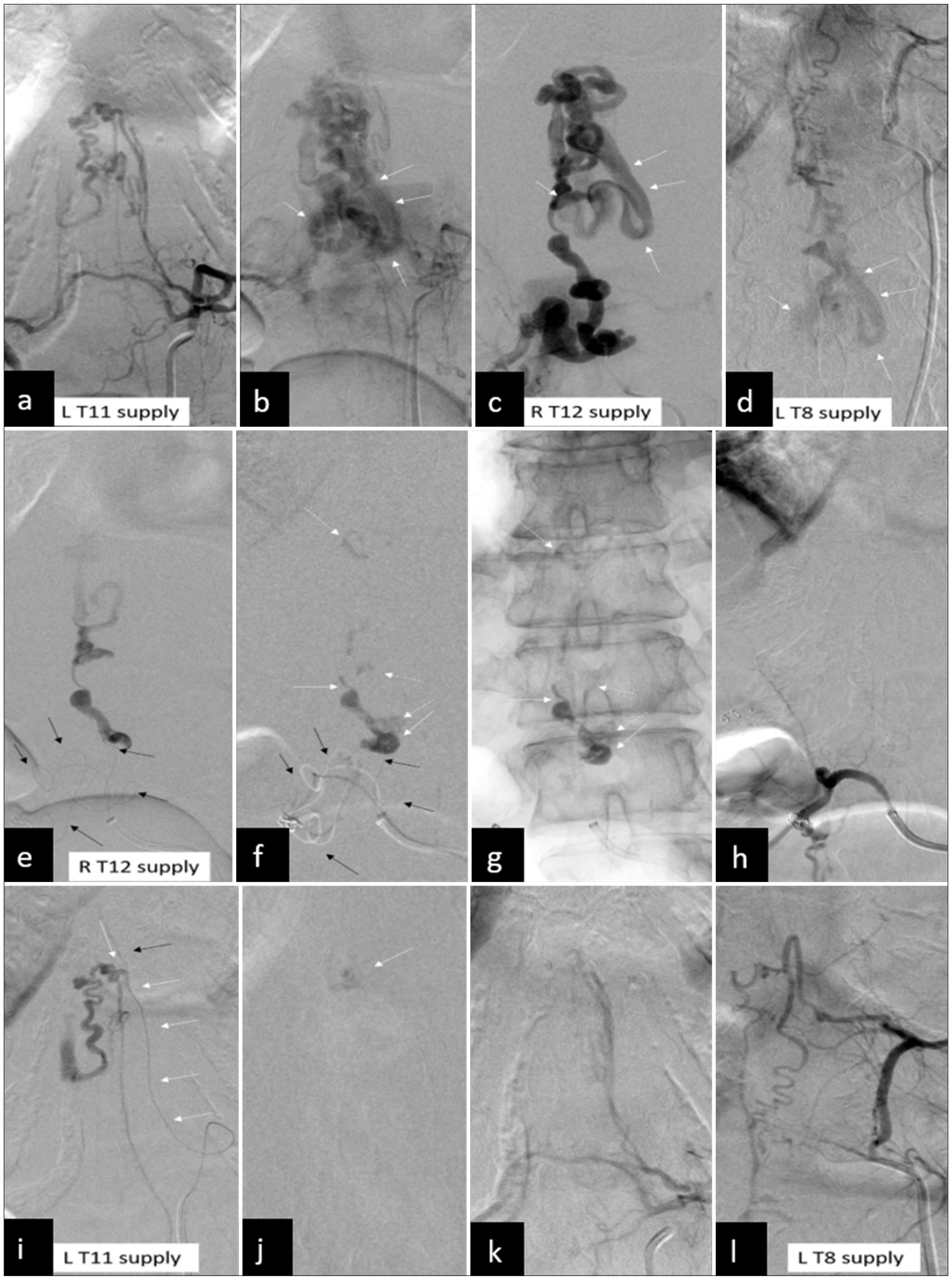

Figure 6: (a and b) A 37-year-old woman suffered from right lower leg numbness for 1 year. The symptoms then gradually progressed to bilateral lower limb associated with weakness and dysuria. A group 3 perimedullary arteriovenous fistula with multiple feeders from left T11, (c) right T12, and (d) left T8 intercostal arteries. A large common venous drainage (b-d; white arrows) is noted. (e and f) The 1st session of embolization is conducted at the right T12 intercostal artery with a microcatheter (black arrows) advancing to the fistula site as close as possible. (f and g) The feeder (f and g, white arrows) and part of the venous drainage (dashed white arrows) are obliterated by N-butylcyanoacrylate (NBCA). (h) The postembolization angiography (h) shows a complete obliteration of this compartment. (i) In the second session, the microcatheter (i; white arrows) is advanced distally to the posterolateral spinal artery (black arrow). (j) The fistula is obliterated by NBCA (j, white arrow). (k) The postembolization angiography (k) shows a complete obliteration of this compartment. (l) Finally, no further treatment is performed at the left T8 intercostal artery because no or minimal arteriovenous shunting is found. The patient experienced stability of symptoms at the 15-year follow-up.

the flow of or catch the liquid embolic agent, thus preventing its flow beyond the coil mass. In this study, when the coils were packed with appropriate density, liquid embolic flow beyond the coil mass was not observed during the procedure. The advantage of this method in treating PMAVF is that it can also occlude the smaller feeding arteries from other radiculomedullary branches; this is because they typically share a common drainage venous aneurysm. When the feeding artery and fistula were too tortuous to allow safe guidance to the venous sac, we deployed a small coil at the largest distal feeding artery, which facilitated a reduction of the flow rate during NBCA or Onyx injection. We applied this technique in Group 2 and obtained favorable outcomes.

Conversely, we also have the option of using Onyx, which can be injected slowly and in a repeated slow "push–pause–push" technique under fluoroscopic guidance. We do not have a preference between NBCA and Onyx, although some centers do not use Onyx for the treatment of PMAVF[9] because of concerns about potential hemorrhage, permanent cord compression, and the toxicity of dimethyl sulfoxide. Complications of Onyx embolization include inadvertent embolization or embolic reflux into the artery supplying the spinal cord, the development of perimedullary phlebitis, and vasospasm due to perivascular inflammation.[5,7] In cranial dural AVF, balloon catheters can be used to occlude the parent artery before liquid embolic injection proximally. However, guiding the occlusive balloon catheter into a safe position in the small spinal artery is difficult. The use of coils does not raise any mass effect concerns because they are loosely packed, and progressive thrombosis and shrinkage of the venous aneurysm should alleviate any pressure.

For Group 3 lesions, the embolization of multiple small feeders can only be accomplished using small microcatheters, and liquid embolic is the only suitable embolic material. Typically, 2 or 3 sessions are required for an acceptable result. However, the tortuosity and size of the feeding arteries in Group 3 PMAVFs pose challenges that make techniques such as the "pressure cooker" and the "double catheter" impractical for the embolization of PMAVFs.

Although postangiographic spontaneous closure of a PMAVF is rare, some studies have reported such closure.[1,3,8,18] Several mechanisms of spontaneous thrombosis of vascular malformation have been proposed, including anatomical, such as single venous drainage, and vessel dissection or hemodynamic factors, such as venous thrombosis due to dehydration, trauma, or the use of angiographic contrast medium. How contrast medium is involved remains unclear.

Previous studies on the endovascular treatment of PMAVFs have reported clinical improvements in 65–100% of patients, with various complete occlusion rates.[11,12,14,16,17,19,20,21,26,28] However, the sample size in most studies has typically been relatively small. In this study, of the 18 PMAVFs managed endovascularly, complete and near occlusion was achieved in 17 (94.5%). The outcomes in the endovascular group (n = 18) were excellent and improved in 15 patients (83.3%). In a study of 51 pediatric PMAVFs, after transarterial embolization, 46 (90.2%) were completely occluded, and patients' clinical status was improved in 42 (82.4%).[9] In this study, two-thirds of the patients were older than 15 years and had longer periods of disease, but the result was comparable to previous studies' results. Clinical deterioration after treatment was reported in 0–10%.[19,20,28,29] None of our patients were in worse condition after endovascular treatment in long-term follow-up. The findings were comparable to or better than previous findings, with our classification approach for patient selection for both surgical and endovascular treatment being relatively simple.

This was a retrospective analysis of prospective data. Although the sample size was relatively small, PMAVF is a rare disease. A clinical trial comparing the outcomes of all reported techniques is impractical. Therefore, we proposed a simple strategy for managing PMAVF according to the fistula location and size of the dominant feeder. The key difference between Group 2 and Group 3 lesions is the size of the dominant feeder and the currently available smallest-sized microcatheter, which can deploy detachable coils and inject liquid embolic. The typical diameter of a microcatheter is approximately 0.7 mm. If microcatheters continue to trend smaller in the future, the definitions of these groups may be revised accordingly.

CONCLUSION

We proposed a simple but practical strategy for managing PMAVF according to the location of the fistula and the exact size of the dominant feeder. The strategy can be easily applied in clinical practice.

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Use of artificial intelligence (AI)-assisted technology for manuscript preparation: The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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