Received: 21 June 2022 | Revised: 25 March 2023 | Accepted: 29 March 2023

DOI: 10.1002/ccr3.7202

CASE REPORT

Clinical Case Reports Open Access WILEY

Dorsal intradural spinal arteriovenous fistula associated with giant intradural spinal aneurysm, a case report

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Abstract

Arteriovenous fistula and spinal aneurysms like other vascular malformations can mimic radiculopathy and low back pain. Precise imaging work combined with a hybrid endovascular-microsurgical approach is the key element for the best clinical outcome.

KEYWORDS

arteriovenous fistula, radiculopathy, spinal aneurysm, spinal arteriovenous fistula, spine vascular malformation

1 | INTRODUCTION

A 50-year-old male with bilateral L4 radiculopathy was consulted. Lumbosacral magnetic resonance imaging and angiography showed a simultaneous dorsal intradural arteriovenous fistula and a large spinal aneurysm posterior to L4. The lesions were treated with a hybrid endovascular-microsurgical approach and excellent postoperative outcome.

Among the vertebral column vascular lesions, spinal arteriovenous fistulas (SAVF) and spinal arteriovenous malformations (SAVM) are two major vascular lesions. In general these lesions are thought to be developmental but there are reports on acquired cases, especially in SAVF.[1,2]

These lesions are usually presented with various nonspecific clinical presentations myelopathy, subarachnoid hemorrhage, hematomyelia, radiculopathy, and back pain are cardinal manifestations of spinal vascular malformations but any sensory-motor presentation can be the first clinical picture of these lesions. A large proportion of vascular lesions are detected incidentally on MRI. SAVM is considered a developmental disease and sometimes is associated with other simultaneous vascular abnormalities. Nidus formation, Aneurysmal lesions, and giant varices are among the most reported concurrent lesions associated with SAVM; however, these coincidences are still considered to be exceedingly rare.[2,3] Spinal aneurysms are rare vascular lesions arising from arterial branches supplying the spinal cord or roots. In terms of histologic features, these lesions are homologues to intracranial aneurysms with true three-wall angio-architecture. Due to its rarity, less is known about real clinical manifestations, natural course, and best treatment strategy.[4] In this rare case, we report dorsal intradural SAVF associated with a simultaneous intraspinal aneurysm that was treated in a hybrid vascular approach.

2 | CASE

A 50 years old male with a history of bilateral lower limb radicular pain was consulted with our department. He was conscious, and cooperative with stable vital signs. Physical examination showed bilateral lower limb radicular pain originating from the lower back and radiating to great toes in the L4 root distribution pathway. The pain was sharp, exacerbated by physical activity, and had more severity in the left lower limb. His past medical history was negligible. The rest of the physical examination had no specific findings. The lab results were in the normal range. A lumbosacral MRI was obtained and showed no considerable disc herniation, canal stenosis, or spinal cord or root abnormality attributable to his radicular pain. There were multiple signal voids in the lumbar spine T2-weighted images accompanied by large spherical lesions in the thecal sac at the level of the L4 vertebra. By the interpretation of MRI and myelogram images, the lesions were in favor of intradural, extramedullary tumoral, and vascular pathology (Figure 1). A complementary spinal angiography was performed for vascular workup. Spinal angiography revealed an incidental dorsal intradural SAVF (Kim-Spetzler type II SAVF) in the left L4 foramen and a large right spinal aneurysm. The SAVF was on the L4 nerve root, while the aneurysm was between the cauda equina rootlets. Based on spinal digital subtraction angiography (DSA) images, the aneurysm originated from lateral sacral artery branches and drained into the coronal venous plexus of the cord (Figure 2). The clinical picture was best matched with a SAVF lesion. Therefore, SAVF rather than the aneurysmtook the treatment priority. Informed medical consent was obtained and the patient was scheduled for a SAVF endovascular treatment.

2.1 | SAVF endovascular obliteration

After the patient transfer to an angiography suit, under general anesthesia and in a supine position, after skin preparation and draping in a sterile fashion, utilizing DSA guidance, with internal iliac artery-to-lateral sacral artery catheterization (Marathon™ microcatheter, Medtronic, Minneapolis, MN, USA) we gained access to the SAVF. Feeder vessel and draining veins were reconfirmed with intra-operative DSA, then under fluoroscopic visualization, 1 cc of 25% n-butyl cyanoacrylate (NBCA) glue (glue: Lipiodol ratio, 1:3)was injected into the feeder artery and fistula. Intraoperative DSA confirmed complete obliteration of the SAVF. The patient was transferred to the general neurosurgery ward. Subsequent MRI was obtained 24 h after the embolization, there were no detectable signal voids in the lumbar region in post embolization MRI. However, the aneurysm had no considerable change in size. Due to the considerable mass effect of the aneurysm and persistent residual symptoms, he was scheduled for spinal aneurysm microsurgery.

2.2 | Spinal aneurysm microsurgery

Under general anesthesia and in a prone position, after skin preparation and draping, in a sterile fashion, using C-arm navigation, L4 skin was incised in the midline plane and paravertebral muscles were stripped off. Using a high-speed drill, L4 modified bilateral laminectomy was performed, the dura was opened in the midline plane and we gained access to the cauda equina zone. There was a large and thrombosed aneurysm in between cauda equina roots that was adherent to the nerves from its anterior wall (Figure 3). In terms of angiographic findings, an aneurysm originated from a small branch of the lateral sacral artery and drained into the coronal venous plexus.

Using microsurgical techniques, the aneurysm feeder artery was temporarily clipped. SSEP (somatosensory evoked potential monitoring) and MEP (motor evoked potential monitoring) showed no detectable neurological decline, so the aneurysm feeder and draining vessels were coagulated. We tried to free the nerve roots from anterior wall of the aneurysm without significant success and SSEP-MEP findings did not support further manipulations. The aneurysm was cut through circumferentially, clots were removed and irrigated and the aneurysm was excised. The anterior wall of the aneurysm remained adherent to nerve roots without mass effect. The surgical field was irrigated with copious amount of warm, sterile normal saline, the dura was repaired with a 4/0 nylon continuous suture. A submuscular drain was inserted, and paravertebral muscles, fascia, and skin were repaired in anatomic layers. The patient was transferred to the neurosurgery ward.

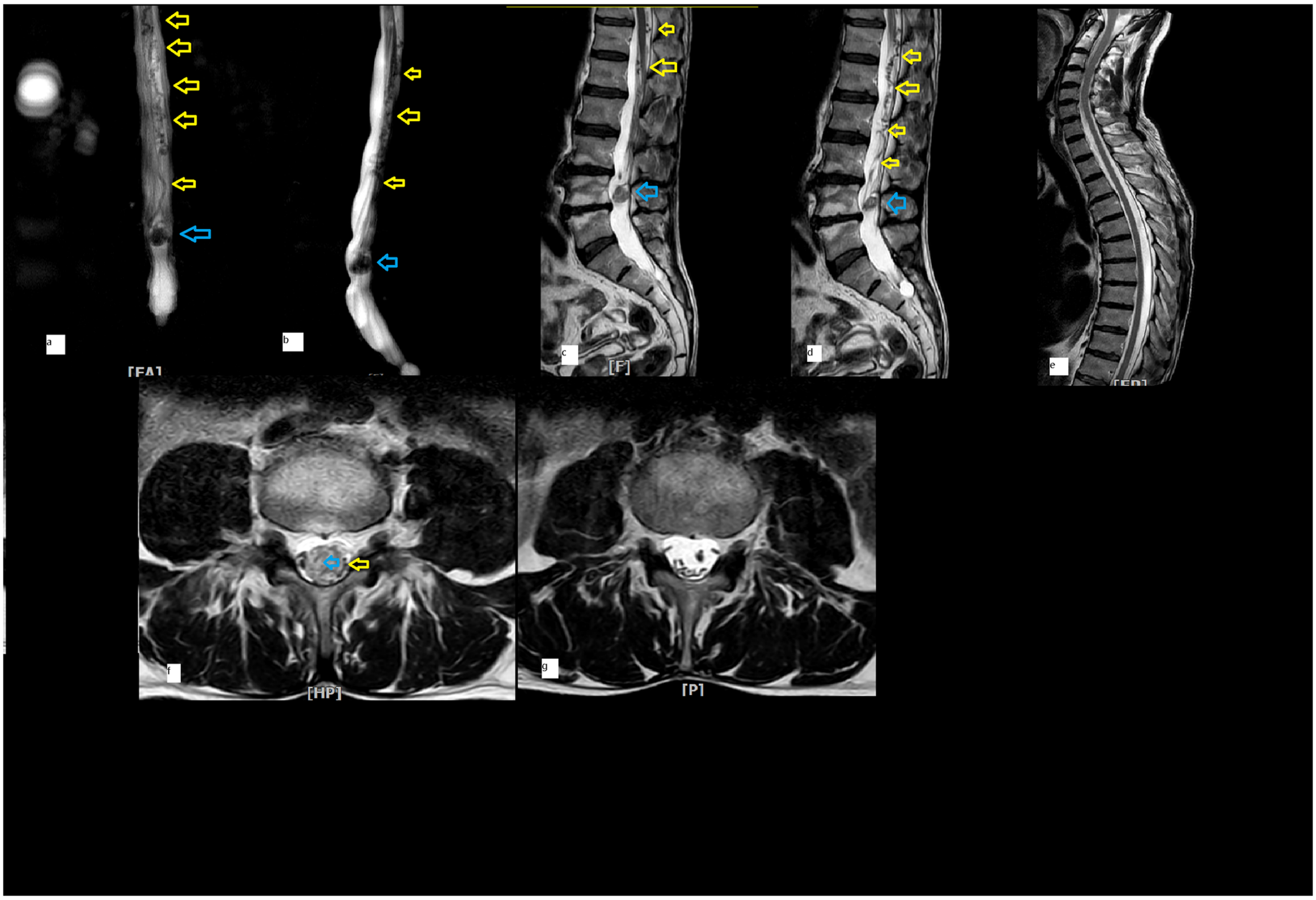

FIGURE 1 Preoperative MRI images showa round-shaped intradural lesions posterior to L4 with multiple signal voids in the lumbar intrathecal space. (A, B) Anterior and lateral myelograms showing vascular tufts and round intradural lesions in the lumbar region. (C, D) Sagittal and parasagittal T2-weighted sequences demonstrating vascular malformation associated with a large intrathecal mass adjacent to L4 posterior surface. Cervicothoracic MRI (E) showed no other nervous system lesions. (F, G) Axial T2-weighted sequences showing the round vascular lesion had compressed roots dorsally. yellow arrow: vascular malformation, blue arrow: spinal aneurysm.

2.3 | Postoperation and follow-up

Postoperative neurological examination revealed resolution of the radicular pain in both lower limbs with intact neurological status (ASIA E). The patient received routine post-laminectomy careand was discharged in a few days. He was visited in the university hospital clinic 2 weeks after surgery. Neurological status was excellent and stable. Follow-up DSA showed complete resolution of the lesions. Follow-up MRIs were taken 1 month and 1year postoperation and showed no significant finding (Figure 4).

3 | DISCUSSION

Spinal vascular lesions encompass a large diversity of lesions. Hemangioma, cavernous malformations, aneurysms, arterio-venous fistula (AVF), and arteriovenous malformations (AVM) are among the most encountered spinal vascular lesions. AVF consists major proportion of spine vascular lesions with an annual incidence of 5–10 per million.[5] Due to SDAVF rarity and technology dependency for a definite diagnosis, it is under-reported and less is known about it. There are multiple classification systems for spinal arteriovenous fistulas and malformations. Each system has tried to delineate anatomical differences in each type of spinal AVF and AVM lesionswhile guiding clinicians for the best treatment strategy. Kim and Spetzler tried to re-introduce a new classification system based on the anatomical location of spinal AVF and AVM lesions. Extradural AVF, intradural dorsal AVF, intradural ventral AVF, extradural-Intradural AVM, intramedullary AVM, and conus medullaris AVM are their main type categorization systems (type I-VI, respectively).[6] In intradural dorsal AVF (IDDAVF), the feeder artery is usually a segmental radicular artery that is fistulized with radicular or medullary vein. These veins are connected to the coronal venous plexus, causing segment veins and/or coronal venous plexus engorgement thus causing congestive

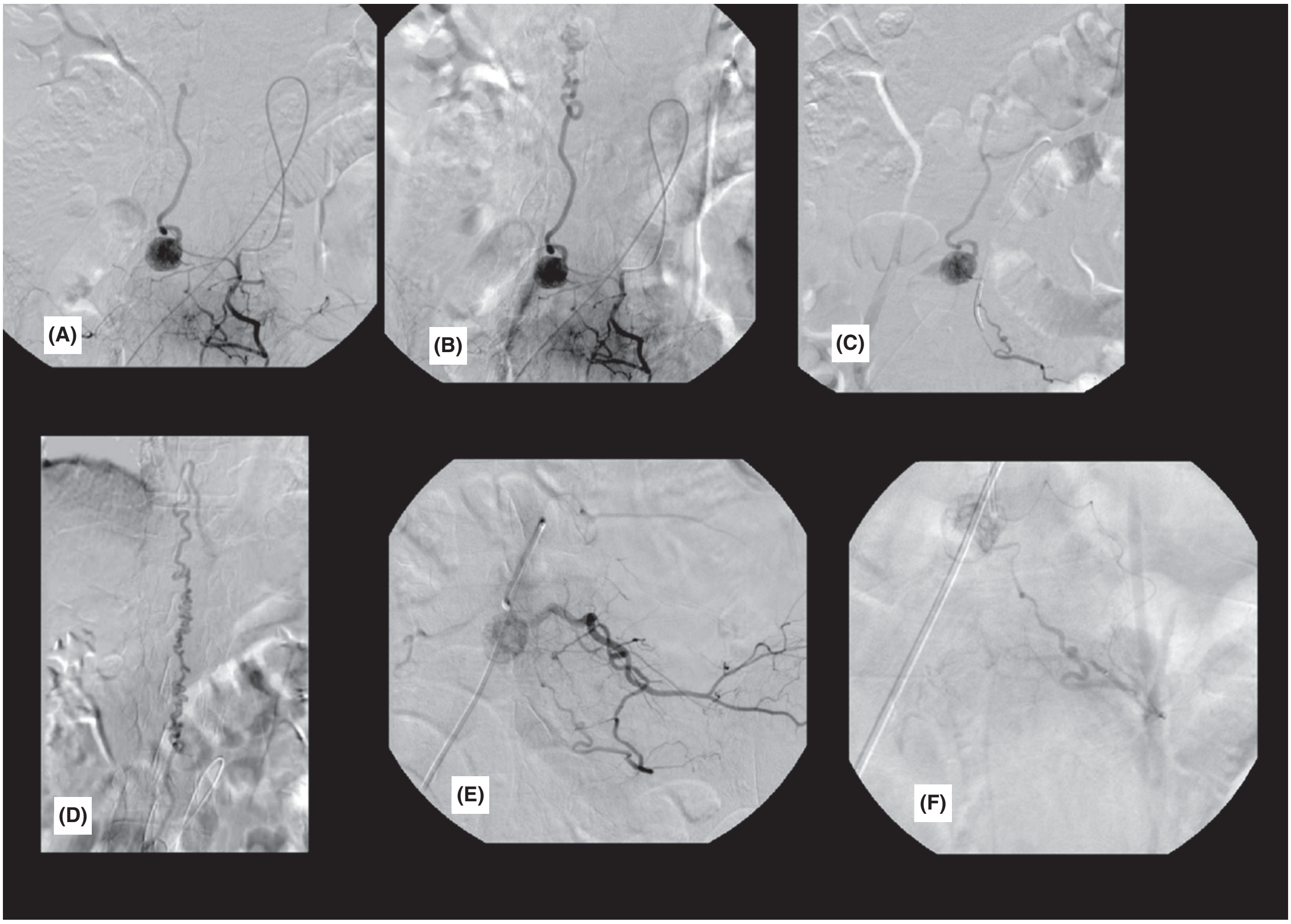

FIGURE 2 Preoperative DSA demonstrating a DIDAVF in L4 neural foramen and a large aneurysm posterior to L4 originating from lateral sacral artery branches. (A, B) Lateral sacral artery and branching arterioles selective angiography in a timely fashion to reveal fistula, aneurysm, and draining vein, (C) super selective aneurysm feeder injection, (D) venous phase, tortuous draining vein, (E, F) selective NBCA injection and AVF obliteration, reduction in aneurysm blush is significant.

myelopathy and hemorrhage. Endovascular interventions and microsurgery are the mainstays of treatment in spinal AVF and AVM. Microsurgery plays a more curative role in spinal AVF and AVM treatment. Low recurrence rate, direct visualization of vascular beds, neural decompression, and better availability of microsurgery are the major advantages of an open surgical approach for spinal AVF and AVM.[7] Endovascular interventions are less-invasive, fast, and efficient in the treatment of AVF and AVM. However, higher recanalization rates, post-embolization residues, high radiation exposure, technology dependency, and lack of decompressive properties in comparison to open surgical approaches, are major disadvantages of endovascular treatments for spinal AVF and AVM.[8,9]

Lee et al. reported a large series on 71 patients with spinal dural arteriovenous fistula (SDAVF). They reported only a 66% successful occlusion rate after endovascular treatments of their spinal AVF without further clinical improvement after re-endovascular therapy or expectant management. They also had reported a 21% recurrence rate after endovascular therapy. Fourteen patients of their failed endovascular treatments ($n=19$), were undergone microsurgical treatment. Thirteen of them (92.9%) had improved or stabilized clinical outcome.[10]

Each modality has its own specific patient selection strategy and in some cases, the clinicians take advantage of both techniques.[11] In large vascular lesions such as metameric or extradural-intradural AVMs, endovascular intervention helps to obliterate feeder vessels and minimize the intra-operative bleeding making microsurgery more feasible.[5,12,13]

Spine aneurysms are exceedingly rare intrathecal vascular lesions. Our main knowledge about spinal aneurysms is based on few reports and case series published in the literature. AVM, pseudoxanthoma elasticum, fibromuscular dysplasia, Behcet's disease, and rheumatoid arthritis are reported in association with spinal aneurysms. However, except for AVM, the exact and reliable association between these diseases and the spinal aneurysm is not well established.[14,15] Still there are multiple obscurities

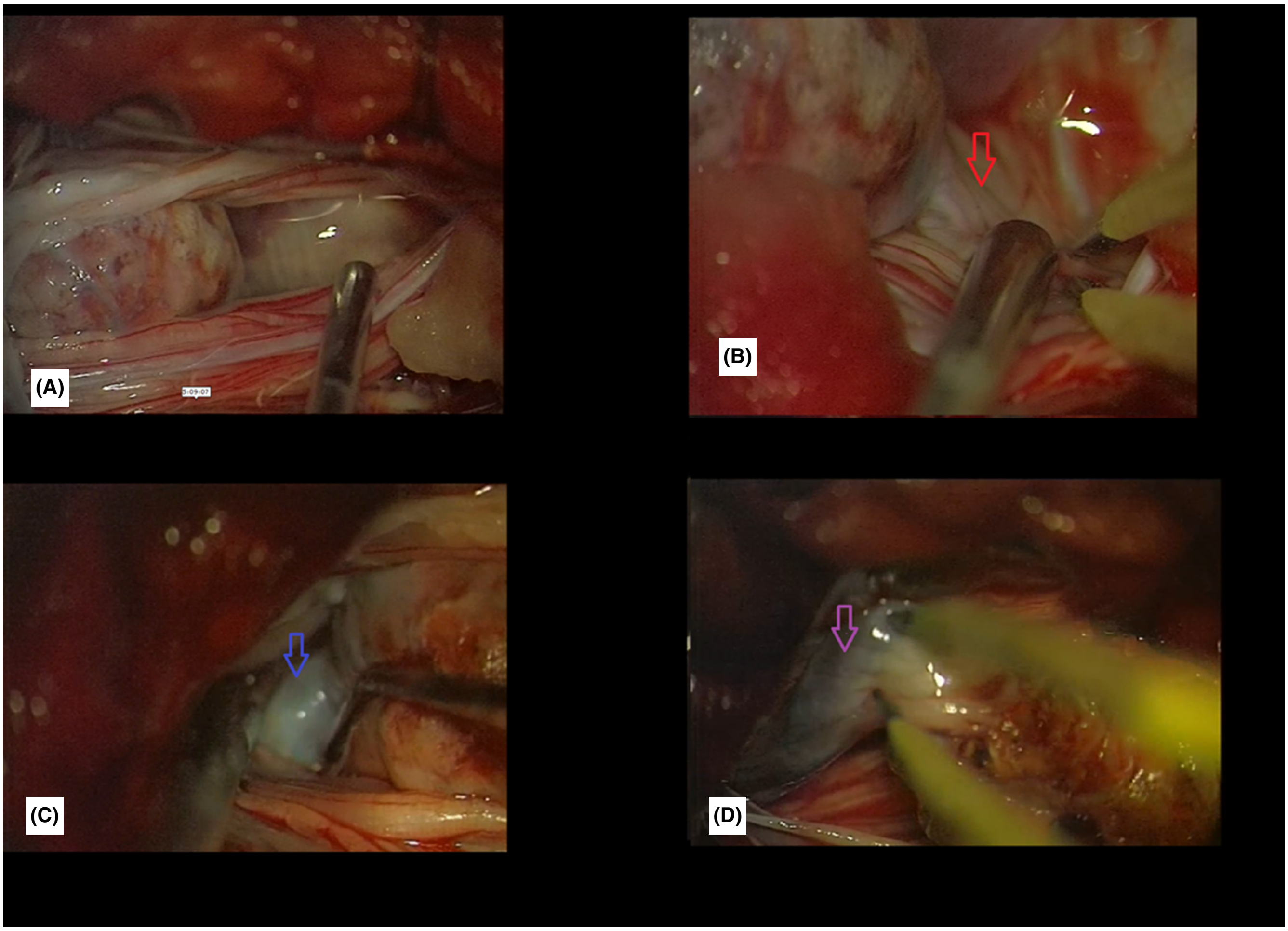

FIGURE 3 Intraoperative photographs demonstrate an intrathecal large aneurysm between cauda equina roots(A). Lateral sacral artery branch (B, red arrow), draining vein to coronal venous plexus (C, blue arrow) and thrombosed venous side of AVF (D, purple arrow) are noticeable.

in terms of pathophysiology, population-based incidence, clinical approach, and best medical treatments.

Endovascular treatments for spinal aneurysm are an available option but there are clinical and radiological obstacles. Parent vessel preservation specially in eloquent spinal vesselterritory such as anterior, posterior, and radicular arteries is of paramount importance. On the other hand, feeder vessel spasm, injury, or occlusion, make the parent vessel catheterization extremely difficult and risky.[16] Reineri reported a case series on spinal aneurysms and found radiculo-pial feeder arteries are relatively safe in terms of endovascular occlusion for the spinal artery.[17] It is worth mentioning that we had to choose NCBA over endovascular coiling for this case since the intradural aneurysm was so big causing the mass effects on the surrounding roots. On the other hand, we had to choose NCBA over other compounds like Onyx® because it gets hard faster than Onyx® and had better maneuverability for such a complex case.

Mass effect and acute neurological deficits are the main indications for microsurgical intervention for spinal aneurysms while smaller lesions with a silent clinical course can be treated via endovascular approaches. Microsurgery provides better parent vessel preservation and decompressive benefits rather than an endovascular approach.[4,15,16] Madhugiri et al. conducted a systematic review of spinal aneurysms and concluded that AVM and cord dysfunction-associated spinal aneurysms will benefit from the microsurgical approach.[18] Mansour et al. reported a craniovertebral junction AVF and aneurysm in the anterior spinal artery (ASA) territory that was microsurgically treated.[19] Nakagawa et al. reported endovascular obliteration of C1-C2 AVF led to simultaneous ASA aneurysm shrinkage during follow-up imaging without any rebleeding.[20] Inci reported a 61-year-old man presented with paraparesis having a spinal aneurysm around the conus medullaris with multiple signal voids around. According to their report, due to

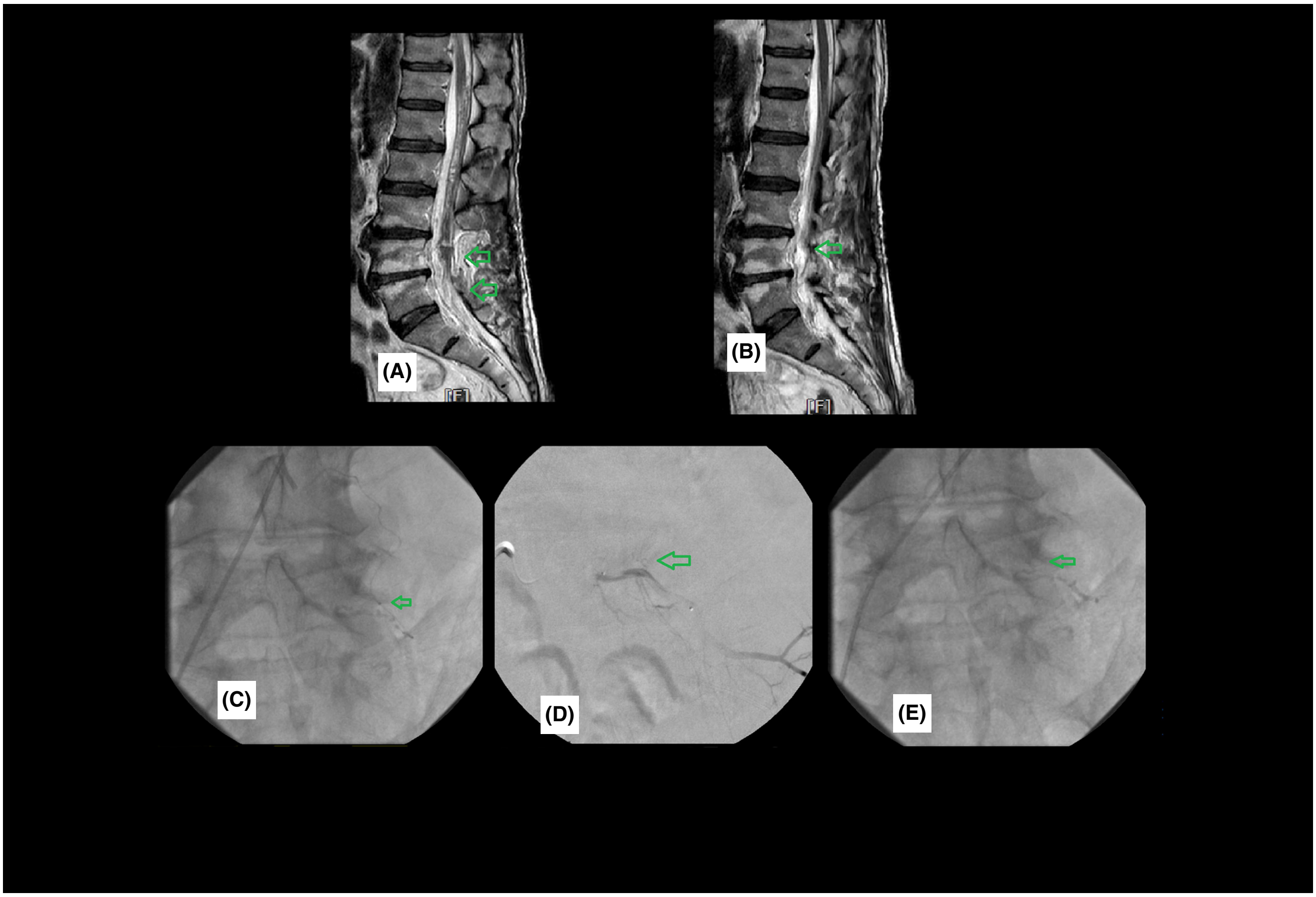

FIGURE 4 Postoperative MRI in mid (A) and parasagittal (B) T-weighted images and DSA (C–E) showed no obvious vascular lesions in follow-up studies. Green arrows: postoperative vascular treatment scar with minimal residues. Please note the complete feeder and draining occlusion.

atherosclerotic vessels, pre-operative diagnosis of AVF could not be confirmed but intraoperative exploration made the aneurysm-AVF concurrency clear. In Inci et al. report, due to mass effect, the patient underwent surgical exploration, and incidentally, a low-flow spinal AVF was detected near the L1 pedicle. AVF was coagulated and cut and then the aneurysm was resected with excellent surgical outcomes.[20]

Tenorio et al. reported a case of a thoracolumbar region spinal aneurysm presented with thalamic infarct, abdominal pain, and lower extremities weakness that was treated via microsurgical approach. They reviewed recent literature in this regard and recommended surgical interventions versus conservative management. Their recommendation was based on the hemorrhagic nature of aneurysms and multiple reports of death in expectant management patients.[21] Son et al. reported a case of a spinal aneurysm in Adamkiewicz artery territory who refused surgical and endovascular treatment and developed arachnoiditis and deteriorated neurological status over follow-up.[16] Romero et al. reported two cases of expectant management in patients with mild neurological symptoms. The reasons for expectant management were the mild neurological symptoms with lack of feeder artery detection in DSA and eloquent parent vessel perfusion territory. One of the patients experienced transient deterioration of neurological status over the hospitalization period but recovered slowly over the follow-up period.[14]

4 | CONCLUSION

Spinal vascular malformations are insidious lesions that can present in different ways. It would be rational to recommend spinal DSA for all the suspected vascular lesions for better anatomical understanding and optimum medical treatment. The Surgical approach should be tailored to clinical priority and radiological findings of each lesion.

AUTHOR CONTRIBUTIONS

Ehsan Mohammad Hosseini: Conceptualization; data curation; investigation; methodology; project administration; supervision; validation; visualization; writing – original draft; writing – review and editing. **Alireza Rasekh:**

Data curation; investigation; software; supervision; validation; writing – original draft. **Noushin Vahdat:** Data curation; investigation; methodology; project administration; software; supervision; validation; visualization; writing – original draft; writing – review and editing. **Keyvan Eghbal:** Supervision; validation; visualization; writing – original draft. **Mohammad Jamali:** Supervision; validation; writing – original draft. **Abdolkarim Rahmanian:** Data curation; methodology; project administration; validation; visualization; writing – original draft. **Arman Sourani:** Conceptualization; data curation; investigation; methodology; project administration; resources; software; supervision; validation; visualization; writing – original draft; writing – review and editing. **Mina Foroughi:** Data curation; investigation; methodology; validation; visualization; writing – original draft; writing – review and editing. **Sadegh Baradaran Mahdavi:**Methodology; project administration; supervision; validation; visualization; writing – original draft; writing – review and editing.

ACKNOWLEDGMENTS

The authors would like to thank the patient and his family for their cooperation.

FUNDING INFORMATION

None.

CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request. Some data may not be made available because of privacy or ethical restrictions.

ETHICS STATEMENT

All procedures performed were under the institutional and/or national research committee's ethical standards and the 1964 Helsinki Declaration and later amendments or comparable ethical standards. Shiraz University neurosurgery department board members supervised and approved this report on behalf of the Ethical Committee of Shiraz University of medical sciences.

CONSENT FOR PUBLICATION

All authors agreed that the manuscript will be submitted and published in Wiley's Clinical Case Report journal.

CONSENT STATEMENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

CONSENT TO PARTICIPATE

Informed consent was obtained from all individual participants included in the Study.

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REFERENCES

1. Brinjikji W, Ahn ES, Patterson MC, Lanzino G. Challenging dogma: report of a spinal cord arteriovenous malformation as an acquired lesion in a pediatric patient. *J Neurosurg Spine*. 2019;32(2):302-304.
2. Krings T, Lasjaunias PL, Geibprasert S, et al. Classification of spinal vascular malformations. *Neuroradiol J*. 2009;22(suppl. 1):97-106.
3. Baldassarre B, Balestrino A, D'Andrea A, et al. Management of spinal aneurysms associated with arteriovenous malformations: systematic literature review and illustrative case. *Eur Spine J*. 2021;30(10):2767-2774.
4. Nguyen NH, Le VC, Nguyen TQ, Nguyen TH. Subarachnoid hemorrhage due to ruptured spinal artery aneurysm: a diagnostic challenge. *Case Rep Neurol*. 2020;12(suppl. 1):169-175.
5. Koch C. Spinal dural arteriovenous fistula. *Curr Opin Neurol*. 2006;19(1):69-75.
6. Kim LJ, Spetzler RF. Classification and surgical management of spinal arteriovenous lesions: arteriovenous fistulae and arteriovenous malformations. *Neurosurgery*. 2006;59(5 suppl. 3):S195-S201. discussion S3-13.
7. Naga Shravan Kumar K, Visvanathan K, Swamiyappan SS, Dhanasekaran J, Joseph S, Krishnamurthy G. Surgical management of spinal dural arteriovenous fistula—a single centre experience. *Interdiscip. Neurosurg*. 2022;28:101500.
8. Lenck S, Nicholson P, Tymianski R, et al. Spinal and paraspinal arteriovenous lesions. *Stroke*. 2019;50(8):2259-2269.
9. Schick U, Hassler W. Treatment and outcome of spinal dural arteriovenous fistulas. *Eur Spine J*. 2003;12(4):350-355.
10. Lee HS, Kang H-S, Kim SM, et al. Treatment strategy to maximize the treatment outcome of spinal dural arteriovenous fistula after initial endovascular embolization attempt at diagnostic angiography. *Sci Rep*. 2021;11(1):10004.
11. Ropper AE, Gross BA, Du R. Surgical treatment of type I spinal dural arteriovenous fistulas. *Neurosurg Focus*. 2012;32(5):E3.

12. Phadke RV, Bhattacharyya A, Handique A, et al. Endovascular treatment in spinal perimedullary arteriovenous fistula. *Interv Neuroradiol*. 2014;20(3):357-367.
13. Endo T, Endo H, Sato K, Matsumoto Y, Tominaga T. Surgical and endovascular treatment for spinal arteriovenous malformations. *Neurol Med Chir*. 2016;56(8):457-464.
14. Gutierrez Romero D, Batista AL, Gentric JC, Raymond J, Roy D, Weill A. Ruptured isolated spinal artery aneurysms. Report of two cases and review of the literature. *Interv Neuroradiol*. 2014;20(6):774-780.
15. Jiarakongmun P, Chewit P, Pongpech S. Ruptured anterior spinal artery aneurysm associated with coarctation of aorta. Case report and literature review. *Interv Neuroradiol*. 2002;8(3):285-292.
16. Son S, Lee S-G, Park C-W. Solitary ruptured aneurysm of the spinal artery of adamkiewicz with subarachnoid hemorrhage. *J Korean Neurosurg Soc*. 2013;54(1):50-53.
17. Renieri L, Raz E, Lanzino G, et al. Spinal artery aneurysms: clinical presentation, radiological findings and outcome. *J Neurointerv Surg*. 2018;10(7):644-648.
18. Madhugiri VS, Ambekar S, Roopesh Kumar VR, Sasidharan GM, Nanda A. Spinal aneurysms: clinicoradiological features and management paradigms: a systematic review. *J Neurosurg Spine*. 2013;19(1):34-48.
19. Mansour A, Endo T, Inoue T, et al. Clipping of an anterior spinal artery aneurysm using an endoscopic fluorescence imaging system for craniocervical junction epidural arteriovenous fistula: technical note. *J Neurosurg Spine*. 2019;31(2):279-284.
20. Nakagawa I, Park H-S, Hironaka Y, Wada T, Kichikawa K, Nakase H. Cervical spinal epidural arteriovenous fistula with coexisting spinal anterior spinal artery aneurysm presenting as subarachnoid hemorrhage—case report. *J Stroke Cerebrovasc Dis*. 2014;23(10):e461-e465.
21. Tenorio A, Holmes BB, Abla AA, Amans M, Meisel K. An isolated ruptured spinal aneurysm presents with a thalamic infarct: case report. *BMC Neurol*. 2021;21(1):52.

How to cite this article: Mohammad Hosseini E, Rasekhi A, Vahdat N, et al. Dorsal intradural spinal arteriovenous fistula associated with giant intradural spinal aneurysm, a case report. *Clin Case Rep*. 2023;11:e7202. doi:10.1002/ccr3.7202