JOURNAL OF NEUROSURGERY:
Case Lessons

J Neurosurg Case Lessons 3(24): CASE22105, 2022
DOI: 10.3171/CASE22105

Spinal glomus AVM presenting solely with groin pain: illustrative case

Natsumi Baba, MD, Ryo Horiuchi, MD, Takashi Yagi, MD, PhD, Kazuya Kanemaru, MD, PhD, Hideyuki Yoshioka, MD, PhD, and Hiroyuki Kinouchi, MD, PhD

Department of Neurosurgery, Interdisciplinary Graduate School of Medicine and Engineering, University of Yamanashi, Yamanashi, Japan

BACKGROUND Spinal glomus arteriovenous malformations (AVMs) are rare and can cause neurological morbidity due to spinal hemorrhage, venous hypertension, or mass effect.

OBSERVATIONS The authors presented a rare case of spinal glomus AVM presenting with groin pain due to nerve root compression by a feeder aneurysm. A 41-year-old woman was referred to the hospital with initial right groin pain that had worsened over 2 months. Magnetic resonance imaging showed intra- and extramedullary abnormal flow voids at the T11–12 level, and spinal angiography revealed an intramedullary AVM, with extramedullary protrusion of an aneurysm on the feeder vessel, which arose from the sulcal artery of the anterior spinal artery. Because compression of the right L1 nerve root by the aneurysm was the likely cause of the patient's pain, endovascular embolization was performed. The feeder aneurysm disappeared after partial n-butyl 2-cyanoacrylate embolization, and the groin pain disappeared immediately after treatment. Her clinical status has been stable with no recurrence during 1 year of follow-up.

LESSONS This is the first report of glomus-type AVM presenting with radiculopathy alone. One should not overlook the possibility of spinal AVM among patients with groin pain.

https://thejns.org/doi/abs/10.3171/CASE22105

KEYWORDS glomus AVM; spinal cord; groin pain

Groin pain is most commonly caused by hip disorders or sacroiliac joint dysfunction.[1] Although less frequent, lumbar radiculopathy due to upper-level lumbar disc hernia, synovial cyst, and epidural venous plexus is also known to cause groin pain.[2–6] However, the involvement of spinal cord arteriovenous malformation (AVM) has not been reported so far.

Spinal glomus AVMs are rare clinical disorders, and there have been limited reports on their natural history and optimal treatment. They typically cause myelopathy due to bleeding or its mass effect[7,8] but seldom cause radiculopathy. Although spinal aneurysm can cause radiculopathy, and 29% of spinal glomus AVMs are reported to have aneurysms,[7–10] no such case has yet been reported for a glomus-type AVM, whereas only one case of juvenile type that included radiculopathy was reported to have developed due to a prenidal aneurysm.[11]

Here, we report the first case of spinal glomus AVM in which groin pain was caused by a feeder aneurysm and selective embolization was effective in improving the symptoms.

Illustrative Case

A 41-year-old woman's first symptom was right groin pain, which worsened in the following 2 months. There were no other neurological deficits such as myelopathy or bladder or bowel dysfunction. Magnetic resonance imaging (MRI) revealed abnormal intra- and extramedullary flow voids at the T11–12 level, a round extramedullary flow void suggesting an aneurysm, and a diffuse intramedullary flow void with the appearance of a nidus (Fig. 1).

Selective left L1 segmental artery angiography revealed an AVM with an aneurysm at the thoracolumbar junction of the spinal cord. It was supplied by an abnormally dilated anterior spinal artery (ASA) from the artery of Adamkiewicz and drained caudally through the anterior spinal vein (Fig. 2A).

On cone beam computed tomography, the nidus occupied the intramedullary space and the aneurysm projected toward the right side, adjacent to the right L1 nerve root (Fig. 2B–D). The size of

ABBREVIATIONS ASA = anterior spinal artery; AVF = arteriovenous fistula; AVM = arteriovenous malformation; MRI = magnetic resonance imaging; NBCA = n-butyl 2-cyanoacrylate.

INCLUDE WHEN CITING Published June 13, 2022; DOI: 10.3171/CASE22105.
SUBMITTED March 8, 2022. **ACCEPTED** March 11, 2022.

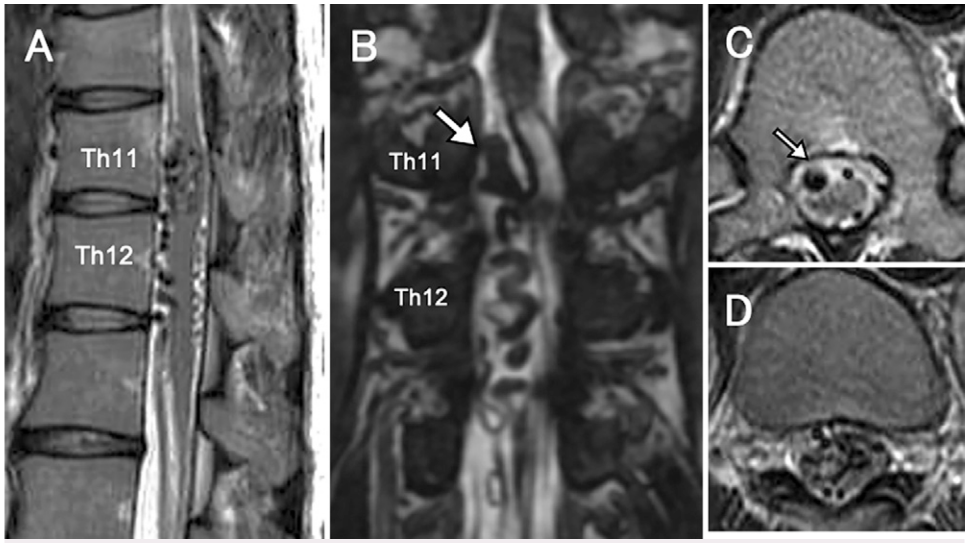

FIG. 1. Preoperative MRI: sagittal T2-weighted image (**A**), coronal 3D fast imaging employing steady-state acquisition (FIESTA) image (**B**), axial T2-weighted image at the level of the aneurysm (**C**), and axial T2-weighted image at the level of the nidus (**D**). Abnormal intra- and extramedullary flow voids can be seen at the T11–12 level. The *white arrow* indicates the extramedullary projection of the aneurysm (13 × 7 mm).

the aneurysm was 13 × 7 mm, and its lateral diameter was approximately half that of the spinal canal. Based on these findings, the patient was diagnosed with glomus AVM at the thoracolumbar junction, and compression of the right L1 nerve root by the feeding artery aneurysm was regarded as the cause of her groin pain.

Endovascular Treatment

Because the AVM occupied most of the area of the spinal cord and was fed by the ASA, we considered that complete embolization would carry a considerable risk of severe spinal cord infarction. Therefore, we planned partial embolization of the AVM, including

FIG. 2. Preoperative spinal angiography and cone beam computed tomography (CT). **A:** Angiography from the left L1 segmental artery. **B:** Coronal cone beam CT image. **C:** Axial cone beam CT image at the level of the aneurysm. **D:** Axial cone beam CT image at the level of the nidus. Angiography revealed a spinal AVM. An aneurysm on the feeder (*arrowhead*), nidus (*asterisk*), and draining veins to the caudal side (*black arrow*) are seen. Cone beam CT revealed the nidus localized in the lower thoracic spinal cord and extramedullary protrusion of the aneurysm (13 × 7 mm) on the right side of the spinal canal (*arrow*).

FIG. 3. Postoperative spinal angiography confirmed the disappearance of the aneurysm and a decrease in blood flow to the nidus.

the feeding artery aneurysm, to relieve the mass effect on the nerve root and decrease the risk of hemorrhage. A 4-Fr catheter (Michaelson, Medikit Co.) was inserted into the left L1 lumbar artery, and a Marathon catheter (Medtronic) was advanced into the nidus with the aid of a microguidewire (Chikai 008, ASAHI INTECC). We placed the catheter at the most distal part of the nidus, where the ASA running caudally was not visualized, and then injected 25% n-butyl 2-cyanoacrylate (NBCA). Another microcatheter was then placed just proximal to the first embolization point. A second embolization was performed by injecting 40% NBCA at this position where only the aneurysm was visualized. A final angiogram revealed no further filling of the aneurysm and reduction of the nidus flow, although there was residual AVM fed by the posterior spinal artery (Fig. 3).

Postoperative Course

There were no postoperative complications, and the patient's groin pain disappeared immediately after the procedure. Spinal angiography performed 4 months after the operation revealed the residual nidus but no recurrence of the aneurysm. MRI acquired at the same time confirmed thrombosis and shrinkage of the aneurysm (Fig. 4). The patient's clinical status has been stable during 1 year of follow-up.

Discussion

Observations

Spinal AVMs are generally classified into four types: dural arteriovenous fistulas (AVFs), glomus AVMs, juvenile AVMs, and pial AVFs.[12,13] The form of onset depends on the type of AVM. Dural AVFs typically develop myelopathy due to venous hypertension, whereas glomus AVMs usually present with bleeding or myelopathy.[7,14] Radiculopathy associated with spinal AVMs is extremely rare, and only one case of juvenile type AVM has been reported.[11] In the present case, a prenidal aneurysm of a radiculopial artery at the T12 level caused severe radicular pain, and symptoms were improved by endovascular embolization. Only two cases of radiculopathy due to an isolated spinal aneurysm that was not related to an AVM have been reported: a lateral sacral artery aneurysm that caused S1 nerve root compression and low back pain[15] and an artery of Adamkiewicz aneurysm of the 10th intercostal artery that caused mild low back pain and occasional right sciatica.[16] In both of these cases, the symptoms improved after clipping of the feeding artery. This is the first report of glomus-type AVM that presented with radiculopathy alone, caused by a coexisting feeder aneurysm.

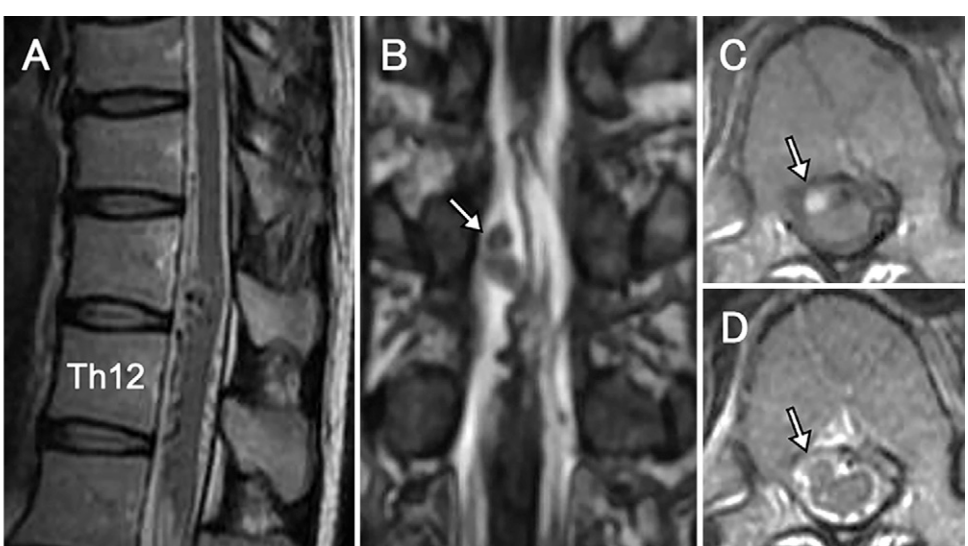

FIG. 4. Follow-up MRI 4 months after embolization. **A:** Sagittal T2-weighted image. **B:** Coronal 3D FIESTA image. **C:** Axial T1-weighted image. **D:** Axial T2-weighted image. The images show thrombosed aneurysm (*arrows*) and decreased size of the nidus.

TABLE 1. Summary of cases of spinal AVM that presented with symptoms due to mass effect of an aneurysm and were treated by endovascular embolization

Author & Year	Lesion Level	Lesion Type	Mechanism	Aneurysm Type	Aminoff Scale (before/after)	FU (mos)
Jung et al., 2018[11]	C4–5	Juvenile	Cord compression	Nidal	5/0	26
Jung et al., 2018[11]	C6	Perimedullary	Cord compression	Nidal	2/1	15
Jung et al., 2018[11]	T12	Juvenile	Root compression	Prenidal	2/0	54
Johnson and Petrie, 2009[18]	T9–11	Juvenile	Cord compression	Nidal	Lost to FU	–
Present case	T11	Glomus	Root compression	Prenidal	2/0	9

FU = follow-up.

Aneurysms associated with spinal AVMs have been reported to be localized approximately equally on the feeder and within the nidus.[17] Anatomically, the mass effect of an intranidal aneurysm causes myelopathy but rarely radiculopathy. In contrast, aneurysms on a perimedullary feeder can cause radiculopathy. However, approximately 80% of AVM-related spinal aneurysms are reported to present with hemorrhage,[17] and one of the reasons why radiculopathy due to a spinal aneurysm is rare is that the feeder aneurysm seldom grows large enough to exhibit mass effect.

The treatment options for intramedullary spinal AVMs include surgical, endovascular, or radiation therapy, but it is difficult to achieve preservation of spinal cord function as well as curative treatment by any method.[7] The ideal treatment strategy would be eradication of the AV shunt; however, reducing the bleeding risk while preserving spinal cord function is also a therapeutic option. Because symptomatic unruptured aneurysms associated with spinal AVMs are rare, appropriate treatment strategies have not been established. To date, only four cases treated by endovascular embolization have been reported (Table 1).[11,18] In these cases, the symptoms improved after treatment, which suggests that endovascular embolization is an effective treatment option for spinal AVM with aneurysms. In the present case, an ASA was involved as a feeding artery and the risk of spinal cord infarction was extremely high in the treatment strategy of complete occlusion of the shunt for radical cure. Therefore, we planned to occlude the aneurysm by feeder embolization alone, and disappearance of the aneurysm was confirmed after treatment. Although complete occlusion of the nidus could not be achieved, this treatment option was effective because the radicular pain disappeared immediately after the treatment without any complications, and a decrease in shunt flow was confirmed by follow-up angiography performed 4 months later. Further follow-up is necessary to assess the long-term risk of bleeding from this treatment.

Lessons

We report the first case of glomus AVM that presented with radiculopathy of groin pain due to nerve root compression by the feeder aneurysm. We should not overlook the possibility of spinal AVM among patients with radiculopathy. Endovascular treatment can be an optimal treatment strategy for glomus AVM.

References

1. Kurosawa D, Murakami E, Aizawa T. Groin pain associated with sacroiliac joint dysfunction and lumbar disorders. *Clin Neurol Neurosurg.* 2017;161:104–109.
2. Tokuhashi Y, Matsuzaki H, Uematsu Y, Oda H. Symptoms of thoracolumbar junction disc herniation. *Spine (Phila Pa 1976).* 2001; 26(22):E512–E518.
3. Yang SN, Kim DH. L1 radiculopathy mimicking meralgia paresthetica: a case report. *Muscle Nerve.* 2010;41(4):566–568.
4. Sangala JR, Uribe JS, Park P, Martinez C, Vale FL. Nerve root prolapse into a spinal arachnoid cyst: an unusual cause of radiculopathy. *Clin Neurol Neurosurg.* 2009;111(5):460–464.
5. Lee YS, Choi ES, Kim JO, Ji JH. A rare calcified thrombosis of the dilated epidural venous plexus presenting with lumbar radiculopathy: a case report. *Spine J.* 2011;11(2):e28–e31.
6. Jeong HJ, Sim WS, Park HJ, et al. Severe lumbar radiculopathy with epidural venous plexus engorgement in a morbidly obese pediatric patient: a case report. *Medicine (Baltimore).* 2019;98(33): e16842.
7. Gross BA, Du R. Spinal glomus (type II) arteriovenous malformations: a pooled analysis of hemorrhage risk and results of intervention. *Neurosurgery.* 2013;72(1):25–32.
8. Daou B, Atallah E, Al-Saiegh F, et al. Spinal glomus arteriovenous malformation manifesting with a subarachnoid hemorrhage. *World Neurosurg.* 2017;98:874.e1–874.e6.
9. Caragine LP, Halbach VV, Dowd CF, Ng PP, Higashida RT. Parallel venous channel as the recipient pouch in transverse/sigmoid sinus dural fistulae. *Neurosurgery.* 2003;53(6):1261–1267.
10. Lv X, Li Y, Yang X, Jiang C, Wu Z. Endovascular embolization for symptomatic perimedullary AVF and intramedullary AVM: a series and a literature review. *Neuroradiology.* 2012;54(4):349–359.
11. Jung SC, Song Y, Cho SH, et al. Endovascular management of aneurysms associated with spinal arteriovenous malformations. *J Neurointerv Surg.* 2018;10(2):198–203.
12. Anson JA, Spetzler RF. Interventional neuroradiology for spinal pathology. *Clin Neurosurg.* 1992;39:388–417.
13. Takai K. Spinal arteriovenous shunts: angioarchitecture and historical changes in classification. *Neurol Med Chir (Tokyo).* 2017;57(7): 356–365.
14. Gross BA, Du R. Spinal pial (type IV) arteriovenous fistulae: a systematic pooled analysis of demographics, hemorrhage risk, and treatment results. *Neurosurgery.* 2013;73(1):141–151.
15. Alberstone CD, Rupp FW, Anson JA. Spinal aneurysm of the lateral sacral artery. Case report. *J Neurosurg.* 2000;92(suppl 1): 101–104.
16. el Mahdi MA, Rudwan MA, Khaffaji SM, Jadallah FA. A giant spinal aneurysm with cord and root compression. *J Neurol Neurosurg Psychiatry.* 1989;52(4):532–535.
17. Madhugiri VS, Ambekar S, Roopesh Kumar VR, Sasidharan GM, Nanda A. Spinal aneurysms: clinicoradiological features and management paradigms. *J Neurosurg Spine.* 2013;19(1):34–48.
18. Johnson WD, Petrie MM. Variety of spinal vascular pathology seen in adult Cobb syndrome. *J Neurosurg Spine.* 2009;10(5): 430–435.

Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Yagi, Baba, Kinouchi. Acquisition of data: Yagi, Baba, Horiuchi, Kanemaru. Analysis and interpretation of data: Yagi, Baba, Kanemaru, Kinouchi. Drafting the article: Baba, Horiuchi, Kanemaru, Kinouchi. Critically revising the article: Yagi, Baba, Kanemaru, Yoshioka, Kinouchi. Reviewed submitted version of manuscript: Yagi, Baba, Horiuchi, Kanemaru, Kinouchi. Approved the final version of the manuscript on behalf of all authors: Yagi. Statistical analysis: Baba. Administrative/technical/material support: Yagi, Baba, Yoshioka. Study supervision: Yagi, Baba.

Correspondence

Takashi Yagi: Interdisciplinary Graduate School of Medicine and Engineering, University of Yamanashi, Japan. yagit@yamanashi.ac.jp.