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Case Report

A case of spinal arteriovenous fistula complicated with congenital spinal lipoma and successfully treated with endovascular therapy *,**

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ABSTRACT

Spinal cord lipomas concurrent with spinal arteriovenous fistulas (SAVFs) are rare, and their natural history and optimal treatment remain unclear. We report the case of a 44-year-old woman with a history of surgical intervention at 2 months of age for myelomeningocele, with the possibility of lipmyelomingocele based on the surgical history, who presented with progressive paralysis and paresthesia in both lower extremities over 3 years. She was diagnosed with an SAVF concurrent with a congenital spinal lipoma, and transarterial embolization was performed after confirming the absence of neurological symptoms during a provocation test. Postoperative imaging showed reduced T2-weighted high-intensity signals in the spinal cord, and 3 months after the procedure, her neurological symptoms improved significantly, allowing her to transition from nonambulatory wheelchair dependence to ambulation with a double cane. This case highlights the need for detailed imaging and vascular evaluation in patients with spinal lipomas and SAVFs, especially in cases involving the filum terminale type, which is closely associated with the anterior spinal artery. Proximal occlusion may occur if feeders have multiple branches or a long distance to the drainer, increasing the risk of incomplete treatment. A provocation test before embolization can enhance both safety and efficacy.

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Introduction

Spinal lipomas concurrent with spinal arteriovenous fistula (SAVF) are rare, and only a few cases have been reported

to date. In this report, the patient experienced neurological symptoms exacerbated by a spinal arteriovenous shunt, which was likely attributable to a congenital spinal lipoma. Herein, we report a favorable outcome achieved through transarterial embolization (TAE) with endovascular treatment

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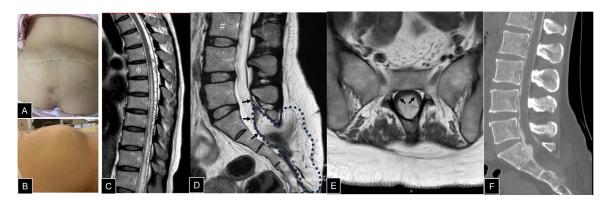


Fig. 1 – (A, B) Superficial image of her buttocks show surgical scar and a sacral dimple. (C, D) T2WI of spinal MR imaging reveal intramedullary high-intensity signal changes from the level of the sixth thoracic vertebra to conus medullaris as well as a flow void around the spinal cord. Arrows indicates the area of continuity of spinal cord to the spinal lipoma. Dots indicate the periphery of spinal lipoma. *,*Represent sixth thoracic vertebra and third lumber vertebra, respectively. (E) Axial image of T1WI of MR imaging shows a flattened spinal cord with smooth borderline (black arrows). (F) A contiguous spinal cord and spinal lipoma is seen on CT myelography.

and present a review of literature on similar cases. The research within our submission has been approved by the ethics institutional review board of our hospital.

Case presentation

A 44-year-old woman with a history of myelomeningocele surgery at 2 months of age, with the possibility of lipomyelomenigocele based on the surgical history, presented with bladder and bowel dysfunction managed with selfcatheterization and laxatives. Three years ago, she developed paresthesia in both lower limbs, which progressed to her upper limbs 2 months prior to presentation. Her gait disturbance worsened, rendering her wheelchair-dependent. Following detailed imaging and neurological assessment, she was referred to our hospital by her primary care physician for evaluation of her progressing symptoms. Neurological findings upon presentation to our hospital revealed muscle weakness of her lower limbs with pain and paresthesia with accompanying dysesthesia. The manual muscle testing (MMT) of the lower limbs showed the following results: iliopsoas 2/2, quadriceps femoris 3/3, tibialis anterior 0/0, extensor hallucis longus 2/2, flexor hallucis longus 2/2, and gastrocnemius 0/0. The Aminoff-Logue disability scales for gait and micturition were G5 and M3, respectively. Physical findings included a surgical scar on the lumbar region present since infancy, a bulging lipoma in the same region, and a sacral dimple (Fig. 1A, B). T2WI of spinal MR imaging revealed intramedullary highintensity signal changes from the level of the sixth thoracic vertebra to conus medullaris as well as a flow void around the spinal cord (Fig. 1C). Presence of a lipoma extending from her buttocks to the first sacral vertebra level of the spinal canal and continuity with the spinal cord is observed at the same elevation (Fig. 1D). T1WI of MR imaging at the level of the first sacral vertebra showed the lipoma was located of the dorsal side of the spinal cord (Fig. 1E). There was contiguous alignment with the spinal lipoma and conus medullaris, but the cauda equina was not observed (Fig. 1F). Contrast-enhanced CT and DSA revealed first and third sacral artery branching from the left lateral sacral artery, and median sacral artery, formed a shunt onto the spinal lipoma, and a draining vein ascending ventral to the spinal cord (Fig. 2A-F). The third sacral artery exhibited a vascular architecture overlying the spinal lipoma. It divided into 3 major branches, each forming distinct shunt points. These branches ultimately drained into the common vein that was previously identified and ascended on the ventral side of the spinal cord (Fig. 2G-H). Based on these findings, we diagnosed SAVF with a shunt overlying the spinal lipoma and contributed mainly by a branch from the left third sacral artery. After discussing the treatment plan with the spinal surgery group, the patient opted for endovascular treatment due to the inherent high risk of damaging spinal nerve fibers traversing within the lipoma during an open surgery.

Endovascular treatment was performed through a puncture of the right femoral artery under local anesthesia, and the Guiding catheter (Axcelguide Guiding System 4Fr 75 cm Mori-1, Medikit, Tokyo, Japan) was inserted. We used a microguidewire (Chikai10 Micro Guide Wire 0.010" 200cm, ASAHI INTECC, Aichi, Japan) to guide and an intermediate catheter (Guidepost 3.2/3.4Fr 120cm, Tokai Medical Products, Aichi, Japan) to support the microcatheter. The intermediate catheter was guided to the left third sacral artery, and a provocation test with 10 mg of lidocaine was performed to confirm that no neurological symptoms had developed. A flow-guide microcatheter (Marathon Flow Directed catheter 2.7/1.5Fr, 165cm, Medtronic, Minnesota, USA) was guided into one of the feeders from the left third sacral artery, and N-butyl cyanoacrylate (NBCA) liquid adhesive (Histoacryl, B.Braun, Melsungen, Germany) diluted to 20% with ethyl ester of iodinated poppy-seed oil fatty acid (Lipiodol, Guerbet Japan, Tokyo, Japan) was used for TAE. Injection of 0.09 ml NBCA resulted in partial feeder embolization due to the backflow. A more distal feeder was selected using a flow-direct microcatheter (DeFrictor Nano Flow Directed catheter 2.2/1.3Fr 165 cm, Medico's Hirata, Osaka, Japan), and TAE was performed again with 20% NBCA. TAE was performed with 0.33 ml of

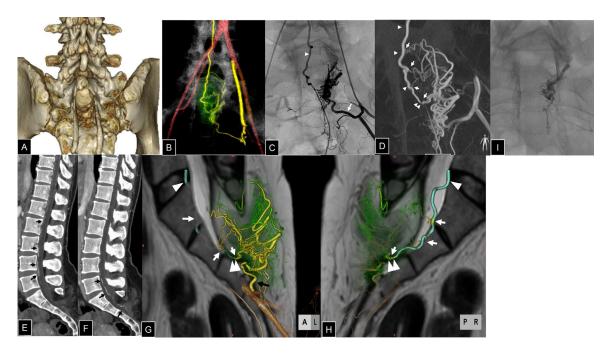


Fig. 2 – (A) 3D reconstructed image of bone CT reveals spinal bifida at sacrum. (B) Contrast-enhanced CT shows the third sacral artery branching from left lateral sacral artery was the feeder involved in the arteriovenous fistula. (C) Anterior-posterior view of the angiography of the third sacral artery (arrow) and the draining vein (arrowhead). (D) 3D rotational angiography of the third sacral artery reveals origin of the draining veins (double arrowheads) and developed and ascended ventral to the spinal cord (arrowheads). Multiple fistulas are shown in arrows. (E, F) Lateral view of contrast-enhanced CT shows draining vein ascending ventral to the spinal cord. (black arrows). (G) Fusion image of sagittal MRI T2WI and 3D rotational angiography seen from left anterior oblique. Black arrow indicates the main feeder from third sacral artery. The draining vein is shown with arrowheads and its origin is shown with double arrowheads. Shunt points are labeled with arrows. (H) Fusion image of sagittal MRI T2WI and 3D rotational angiography seen from right posterior oblique. The draining vein is shown with arrowheads and its origin is shown with double arrowheads. Shunt points are labeled with arrows. I: Embolization from the third sacral artery showing the feeder is partially embolized by 0.33 ml of 20% N-butyl-cyanoacrylate.

NBCA to achieve partial embolization, and the embolization was terminated as NBCA reflux was observed reaching up to the third sacral artery (Fig. 2I). Immediately after surgery, the patient experienced improvement in ankle paralysis and pain caused by dysesthesia but, since the treatment was limited to arterial embolization, there was a risk of recurrence of the AVF and the plan was to follow up with imaging studies.

T2WI of MR imaging on the second postoperative day revealed a flow void around the spinal cord and weakening of the intramedullary T2 high-intensity signal (Fig. 3A, B). Contrast-enhanced CT on the third postoperative day showed decreased venous drainage through the fistula; however, residual blood flow persisted and a feeding artery from the left first sacral artery was evident (Fig. 3C, D). The patient was retreated 9 days postoperatively. Similar to the first procedure, angiography was performed under local anesthesia through a puncture of the right femoral artery. This confirmed the presence of an AVF arising from the left first sacral artery, which was not observed and became apparent in the previous procedure. A shunt was formed from the left lateral aspect of the artery to the base of the lipoma and draining into the ventral vein ascending from the right lateral side (Fig. 4 A, B). The flow-direct catheter was guided to the S1 radicular artery at first sacral foramen level (Fig. 4C). Embolization was performed with 16.7% NBCA because of the large distance between the catheter and recipient vein. NBCA (0.13 ml) was injected into the fistulous tract, but extravasation occurred near the first sacral foramen at the end of the procedure due to continuous pressure infusion (Fig. 4D). The extravasation site was immediately embolized with additional NBCA infusion, and no symptoms occurred during this procedure.

Two days after retreatment, T2WI of MR imaging confirmed the disappearance of the flow void and intramedullary high-intensity signal (Fig. 4E). CT angiography revealed casts of NBCA in the first sacral artery and the partially in the recipient vein, confirming that the vein had lost its angiographical delineation (Fig. 4F, G). The postoperative evaluation of the lower limb MMT revealed the following improvements: iliopsoas 4/2, quadriceps femoris 4/4, tibialis anterior 1/1, and gastrocnemius 0/0. Additionally, the pain level associated with dysesthesia improved to one-tenth of the preoperative level. The patient showed clinical improvement and was transferred to a rehabilitation hospital on the 17th postoperative day. Three months after discharge from our hospital, T2WI of MR imaging showed decreased volume of spinal lipoma and findings of attenuated T2 high-intensity signal changes in the spinal

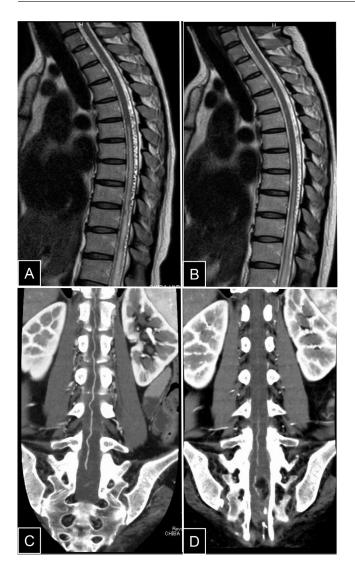


Fig. 3 – The upper row shows T2WI of MR imaging preoperatively (A), 2 days postoperatively (B), respectively. The lower rows are contrast-enhanced CT images preoperatively (C), 3 days postoperatively (D), respectively.

cord (Fig. 4H). The patient showed improvement in ambulation from preoperative wheelchair level to walking with a double cane; moreover, the patient advanced from G5 to G4 on the Aminoff–Logue scale, although abnormal sensation persisted. Since only partial casts were identified on the drainer side, the patient will continue to be followed on an outpatient basis for possible AVF recurrence.

Discussion

Spinal lipoma is a developmental abnormality due to infiltrated fatty tissue during neural tube formation, occurring in approximately one in 4000 births, with a male-to-female ratio of approximately 1:1.5 [1]. In 1982, Chapman et al. classified spinal lipomas into 3 types: dorsal type, caudal type, and

transitional type. Further, in 2001, Arai et al. added the filar type and lipomyelomenigocele type, expanding the classification to 5 types [2]. Pang et al. placed complex and challenging cases that did not follow the criteria for either dorsal or transitional type in a separate category known as chaotic type [3,4]. Recently, Morota et al. proposed a new classification system for spinal lipomas based on embryological stages. Based on Morota's classification, type 1 refers to a primary neural tube formation failure, and the key feature is that the conus medullaris is formed caudal or ventral to the lipoma. Therefore, a case in which a lipoma extends from the dorsal to the caudal side of the conus medullaris, if conus medullaris is formed, it is classified as a transitional type based on conventional classification and is classified as type 1 based on Morota's classification. The current case meets the criteria for being type 1 according to Morota's classification [5].

Spinal arteriovenous fistulas (AVFs) have historically been classified based on angiographic findings and surgical observations, such as the location and morphology of the shunt [6]. However, no standardized classification system currently exists. Additionally, no classification has been proposed for spinal AVFs coexisting with tumors or spinal lipomas. Myelopathy is generally a chronic and progressive condition due to impaired venous perfusion. It can lead to acute spinal cord paralysis due to intramedullary or subarachnoid hemorrhage. Concomitant cases of SAVM and spinal lipoma are rare, with only 23 cases reported since 1989, when Djindjian et al. first reported the combined case [7]. However, their natural history, pathogenesis, classification and treatment strategies are yet to be established. SAVFs may also occur below the conus medullaris without complications of spinal lipoma. Hong et al. reported 3 types of AVFs: filum terminale AVF, spinal dural AVF, and radicular AVF but, did not classify the cases associated with lipoma [8]. In the present case, we reviewed 24 previously reported cases of spinal lipomas complicated with SAVF, including the present case (Table 1) [9–16].

The published reports predominantly involved patients who were middle-aged males (median age, 50.5 years), and the shunt location in most cases was the lumbosacral region (86%). Spinal lipomas of the dorsal and filum terminale types were common (37.5% and 41.6%, respectively). Endovascular embolization combined with surgical treatment or surgical treatment alone was performed in 45.8% and 41.6% of cases, respectively, whereas endovascular treatment alone was performed in only 12.5% of cases. Endovascular treatment involved embolization of the shunt, whereas surgical treatment involved removal and/or coagulation of the AVF. The management of lipomas varies depending on procedures, ranging from partial or total excision to meticulous untethering of the lipoma from the cord. Notably, in the filum terminal type, the anterior spinal artery was involved in AVF formation in 6 of the 8 cases (75%) in which the vascular structure was evaluated (Table 2). Takai et al. indicated that the Adamkiewicz artery, which normally branches from the ninth thoracic to the second lumbar level, branched from a lower level to supply the anterior spinal artery because of spinal cord tethering [14]. Therefore, endovascular treatment of the filum terminal type is associated with a higher risk of embolic material migrating into adjacent critical vessels, and surgical treatment is considered to be safe. Horiuchi et al. administered li-

atient	Year	Authors	Age	Gender	Fistula level	Lipoma type	Feeder	Treatment	Endovascular	Surgical	Outcome	ASA involved
	1989	Djindjian	53	М	S2-3	Terminal	LSA	Embolization, surgical		Complete resection of lipoma	Improved	No
!	1999	Konig	50	M	L5	Lipomyelo menigocele	L3 RMenA	Surgical		•	Unknown	
1	2000	Lee	44	M	Th11-12	Dorsal	Th10, 12, L1 RMenA	Embolization, surgical	NBCA	Subtotal rection of lipoma, untethering of the cord	Complete recovery	
•	2005	Weon	30	M	L4-5	Dorsal	L3, L4 RA	Embolization, surgical	NBCA, coil	Complete resection of extradural lipoma, partial removal of intradural lipoma	Improved	No
i	2005	Cheung	42	M	S1-2	Terminal	ASA	Surgical		Partial resection of lipoma, AVF resection	Improved	Yes
	2005	Rajeav	44	F	L1-2	Dorsal	L1 RA	Surgical		Decompression of lipoma, cord detethered, AVF coagulation	Improved	No
	2007	Erdogan	40	M	L3	Dorsal	L2, L3 RA	Embolization, surgical	Onyx	Lipoma resection and detethering of the cord	Complete recovery	No
	2013	Sato	72	M	L3	Dorsal	L2 RA	Embolization	NBCA	_	Improved	No
	2014	Mavani	29	M	L5-S1	Transitional	L4 RA	surgical		Detethered from sacral lipoma, AVF resection	Improved	No
0	2015	Krisht	58	F	sacral	Dorsal	LSA	Embolization, surgical	Onyx	Lipoma resection, detether cord, AVF ligation	Improved	No
1	2016	Horiuchi	51	M	L5	Dorsal	LSA	Embolization, surgical	coil	AVF ligation	Improved	
.2	2016	Horiuchi	53	M	L2-3	Dorsal	L2 RA	Embolization, surgical	NBCA	Lipoma resection, AVF resection	Improved	

(continued on next page)

Table 1 (continued)

Patient	Year	Authors	Age	Gender	Fistula level	Lipoma type	Feeder	Treatment	Endovascular	Surgical	Outcome	ASA involved
13	2017	Talenti	19	M	S1-3	Lipomyelo menigocele	LSA, MSA, HGA	Embolization, surgical	Onyx, PVA	Lipoma resection and detethering of the cord	Worsen but improved gradually	No
14	2017	Talenti	53	F	S1	Lipomyelocele	MSA	Embolization, surgical	Onyx	Partial resection of lipoma, AVF coagulation	Unchanged	
15	2018	Whitaker- Lea	57	F	L5-S1	Lipomyelo menigocele	LSA	Embolization	Onyx	, and the second	Improved	
16	2018	Przepiórka	30	F	sacral	terminal	N/A	surgical		AVF coagulation and resection	Improved	
17	2018	Przepiórka	33	M	S2-3	Terminal	LSA	Embolization, surgical	Onyx, Phil	Partial resection of lipoma, AVF coagulation	Considerable improvement	
18	2018	Takai	83	M	S4	Terminal	IIA	Surgical		Detethered from sacral lipoma, AVF resection	Improved	Yes
19	2018	Takai	54	M	S4	Terminal	IIA	Surgical		Detethered from sacral lipoma, AVF resection	Improved	Yes
20	2018	Takai	40	M	S1	Terminal	MSA	Surgical		Detethered from sacral lipoma, AVF resection	Unchanged	Yes
21	2021	Shimizu	72	M	L5	Terminal	ASA	Surgical		Partial resection of lipoma, AVF resection	Improved	Yes
22	2021	Shimizu	76	F	L3-4	Terminal	ASA	Surgical		Partial resection of lipoma, AVF resection	Worsen due to ICH	Yes
23	2023	Yamazaki	70	M	S3	Terminal	LSA	Surgical		Lipoma resection, AVF	Improved	No
24	2024	Kubota	44	F	S1-3	Dorsal	LSA	Embolization	NBCA		Improved	No

Abbreviations: ASA, anterior spinal artery; AVF, arteriovenous fistula; F, female; HGA, hypogastric artery; ICH, intracerebral hemorrhage; IIA, internal iliac artery; LSA, lateral sacral artery; M, male; MSA, median sacral artery; N/A, not applicable; NBCA, n-butyl-2-cyanoacrylate; PVA, polyvinyl alcohol; RA, radicular artery; RMenA, radiculomeningeal artery.

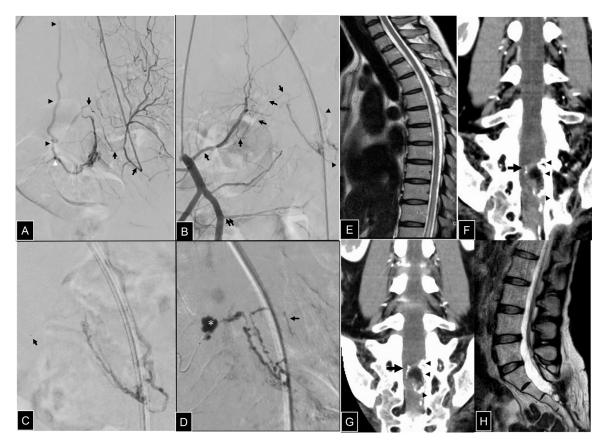


Fig. 4 – (A) Anterior-posterior view of angiography from left lateral sacral artery shows newly developed arteriovenous fistula arising from the left sacral artery (arrows). Draining vein is shown as black arrowheads. The white arrowhead indicates the site of the probable shunt point. (B) Lateral view of angiography from left lateral sacral artery. The arrows indicate the first sacral artery and draining vein is shown as black arrowhead. The white arrowhead indicates the site of the probable shunt point. The double arrows represent the third sacral artery which was embolized in the initial treatment. (C) Lateral view of angiography from the microcatheter prior to the embolization procedure. The arrow indicates the tip of the microcatheter, where embolization was initiated. (D) Embolization was performed with 0.13 ml of 16.7% N-butyl cyanoacrylate (NBCA) and it was injected into the fistulous tract, but extravasation occurred near the first sacral foramen at the end of the procedure (*). Black arrow indicates the NBCA casts have partially reached the draining vein. (E) Sagittal T2WI of MR imaging 11 days postoperatively (2 days after the second treatment). (F, G) Contrast-enhanced coronal CT image 17 days postoperatively (7 days after the second treatment). Black arrow indicates the NBCA casts have partially reached the draining vein. Black arrowheads show the vessels where the feeding artery was embolized with NBCA. (H) Sagittal T2WI of spinal MR imaging 3 months postoperatively.

docaine via a feeder under general anesthesia to 2 patients with spinal lipomas and determined whether embolization was feasible based on the amplitude of motor evoked potentials (MEP) [10]. In cases where there is a decrease in amplitude, embolization with liquid embolic material is considered high-risk and proximal embolization with coils is selected. In case of transient MEP amplitude reduction, NBCA embolization is performed followed by resection. Although we did not use MEPs, we confirmed intraoperatively that no motor or sensory deficits occurred during the awake provocation test, and there were no postoperative neurological symptoms due to embolization. Pre-embolization provocation tests may help in risk assessment by awake conditions or using MEPs.

Angiography of the segmental arteries from Th7 to L4 and contrast-enhanced CT angiography in this case could

not identify a radiculomedullary or radiculopial artery, and the involvement of the anterior and posterior spinal artery in the lipoma could not be assessed. The left lateral sacral artery fed within the lipoma and then formed a shunt to a single draining vein that drained ascending the ventral side of the spinal cord. The feeding artery branched into multiple branches within the lipoma, and 3 separate shunts to the draining vein were identified. The endovascular approach requires that the embolization material reach this draining vein to achieve radical cure. However, the distance from the point where the microcatheter could be guided to the draining vein is long because the vein passes through the lipoma, and even low concentrations of NBCA carry the risk of proximal occlusion and recurrence. Of the 12 cases reviewed in the literature, only 4 were cured by endovascular treatment alone, of which

Table 2 – Characteristics, management and outcomes of spinal arteriovenous fistulas associated with spinal lipoma.

Age (median)	50.5						
Male (%)	17 (70.8)						
Level (%)							
Thoracic	1 (3.4)						
Lumber	11 (37.9)						
Sacral	14 (48.3)						
Lipoma type (%)							
Dorsal	9 (37.5)						
Caudal	0 (0)						
Terminal	10 (41.6)						
Transitional	1 (4.1)						
Lipomyelocele/Lipomyelomenigocele (%)	4 (16.7)						
Treatment (%)							
Embolization	3 (12.5)						
Surgical	11 (45.8)						
Both	10 (41.6)						
Outcome (%)							
Improved	16/19 (84.2)						
Complete recovery	2/19 (10.5)						
Worsened	1/19 (5.2)						
ASA involved in terminal type (%)	6/8 (75.0)						
Abbreviation: ASA, anterior spinal artery.							

Erdogan et al. achieved complete occlusion by embolization but added surgical treatment to untether the spinal cord. In other cases, surgical treatment was added in cases where the embolic material did not reach the draining vein, resulting in proximal occlusion, or in cases where neurological symptoms appeared during the provocation test. The decision to proceed to surgical treatment should be made when endovascular access becomes difficult. In the surgical approach, a radical cure can be achieved by isolation of the draining vein. On the other hand, since the safety and invasiveness of surgery depend on the size and extent of the lipoma and the surrounding nerves and vascular structures, the treatment options should be evaluated in each case.

The spinal cord was not tethered in 9 of the 20 cases who were treated surgically. Both tethered cord syndrome and SAVF present with various symptoms, such as paraplegia in both lower extremities, bladder and rectal functional disturbances, and low back pain, making it critical to distinguish the 2 conditions and determine the presence of concomitant lesions. Indeed, Cheung et al. reported a case of delayed worsening of paraplegia after spinal cord detachment, which was first identified to be an AVF and later improved when the AVF was treated surgically [9]. This study supports establishing a standard for performing preoperative MRI or contrast-enhanced CT in all patients with suspected tethered cord syndrome to determine the presence of a concurrent AVF.

The etiology of AVF associated with spinal lipoma has not yet been elucidated. One hypothesis is that angiogenic factors such as fibroblast growth factor 2, vascular endothelial growth factor, and platelet-derived growth factor B are released from the lipoma, resulting in abnormal angiogenesis and localized vascular growth within the Dural defects [17,18]. It has also been proposed that venous congestion and thrombosis caused by veins stretched by tethering of the spine may contribute

secondarily to the development of AVF. Given the median age of onset of 50 years based on case reports, it is possible that angiogenic factors, venous thrombosis, and postoperative inflammation are involved in the contribution. In this case, we hypothesize that the development of the AVF was attributable to a combination of factors, including the infantile surgery, chronic inflammation caused by the lipoma, and tethering of the spinal cord. Considering the patient's preference, endovascular treatment was chosen. However, if surgical intervention were performed, the risk of neurological injury would be high due to adhesion from the infantile surgery and the difficulty in identifying the course of nerves within the lipoma, even with the use of intraoperative neuromonitoring.

In the present case, favorable results were achieved with endovascular treatment alone. Detailed vascular evaluation and provocation studies are critical for identifying and treating cases of combined spinal lipoma and AVF because the type, morphology, and feeding vessels of AVF may vary. It is important to determine whether to perform endovascular treatment, surgical treatment, or a combination of both based on thorough preoperative assessment of each patient.

Conclusion

Patients with spinal lipoma concurrent with spinal arteriovenous fistula exhibit diverse localizations of the lipoma and feeding arteries. In particular, the filum terminale type is strongly associated with the anterior spinal artery, necessitating detailed imaging and vascular evaluation for effective embolization and excision. During embolization, caution is required as feeders with multiple branches and long distances between the catheter and the draining vein may lead to incomplete treatment due to proximal occlusion. Performing a provocation test prior to embolization may contribute to safer and more effective treatment outcomes.

Patient consent

Written informed consent was obtained from the patient for the publication of this case report, including all associated images and information. The authors confirm that the consent documentation is retained in accordance with institutional and journal requirements.

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