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Case Report

Spinal arteriovenous fistula in the lipoma of the filum terminale: A report of two cases and literature review

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Received : 27 January 2021
Accepted : 17 February 2021
Published : 17 March 2021

DOI
10.25259/SNI_80_2021

Quick Response Code:

ABSTRACT

Background: Filum terminale arteriovenous fistulas (FTAVFs) are rare and their pathogenesis remains unknown. The authors report two cases of FTAVF that arose in the lipoma of the filum terminale.

Case Description: The two patients were 72 and 76 years of age, and both presented with a progressive paraparesis. The first patient had an arteriovenous fistula (AVF) located at L5 that was supplied by the anterior spinal artery originating from the left T10 intercostal artery. The second patient's AVF at L3-4 was fed by the anterior spinal artery originating from the left T8 intercostal artery. Both patients underwent partial resection of the filum terminale at the location of the shunts. The pathological examinations revealed that both the AVFs were embedded in the adipose tissue of the filum terminale, revealing the fatty fila that were not visible in preoperative magnetic resonance images.

Conclusion: Two cases of FTAVF were successfully treated by obliterating the fistulas through partial resection of the affected fatty filum terminale. The literature review revealed 13 cases of FTAVF concomitant with the lipomas of the filum terminale. Resection and histological evaluation of the filum terminale should be performed to treat and elucidate the pathogenesis of FTAVF.

Keywords: Fatty filum terminale, Filum terminale arteriovenous fistula, Pathology

INTRODUCTION

Filum terminale arteriovenous fistulas (FTAVFs) represent 3.1% of all intradural AVFs and <5% of spinal intradural arteriovenous malformations.[3,6] FTAVFs are categorized as Type IV, that is, intradural perimedullary arteriovenous fistulas (AVFs). They develop in the pia mater of the filum terminale.[3] They are further classified into three groups according to their angioarchitecture: type I consists – a single feeder of the artery of the filum terminal (AFT), Type II – multiple feeders including the AFT, and Type III – associated with conus arteriovenous malformations.[3] FTAVFs are generally considered to be acquired lesions because of their prevalence in adults.[3,6] However, congenital factors might play some role in the development of FTAVFs, considering that more than 10 cases of coexistent FTAVFs and spinal lipomas have been reported previously.[6] In this paper, we report two cases of FTAVFs that arose in the fatty filum terminale. We first reviewed such cases in the existing literature and then discussed the possible pathogenesis of FTAVFs.

CASE REPORT

Case 1

A 72-year-old male presented with a 5-year history of a progressive lower extremity paraparesis and hypesthesia below the L2 dermatome level. The preoperative T2-weighted magnetic resonance imaging (MRI) revealed edema plus flow voids in the lower spinal cord including the conus medullaris accompanying lumbar canal stenosis from L3 to L5 [Figure 1]. In addition, the preoperative T1-weighted MRIs did not show any high signal intensity suggestive of the fat tissue along the filum terminale [Figure 1]. Spinal digital subtraction angiography (DSA) of the left T10 intercostal artery revealed AVF at L5 [Figure 1]. Surgery included decompressive lumbar laminectomy with obliteration of the AVF. At surgery, we found dilated arteries and veins running over the edematous filum terminale at L5 level [Figure 2]. Intravenous indocyanine green (ICG) showed that the AFT had a caudal inflow, and directly connected to the dilated veins of the filum terminale draining rostrally [Figure 2]. The transitional point from the caudally running artery to the cranially running vein was considered a shunting point; and a temporary aneurysm clip was applied on the arterial side immediately proximal to the shunting point [Figure 2]. Intravenous ICG revealed that another minor feeding artery with a rostral inflow was connected to the AVF, and another temporary aneurysm clip was applied to the minor feeder near the shunting point [Figure 2]. Since intravenous ICG and DSA confirmed the disappearance of the AVF, a part of the filum terminale was resected with the AVF. Histopathology revealed an arterialized vein surrounded by adipose tissue confirming the AVF arose in the lipoma of the filum terminale despite the absence of the lipoma on preoperative MRIs. The patient's symptoms improved and 6 month postoperative MRIs and DSA scans confirmed complete resolution of the AVF.

Case 2

A 76-year-old female also presented with a progressive paraparesis and gradual onset of urinary retention. Sagittal T2-weighted MRI of the thoracic and lumbar spine revealed edema of the spinal cord with flow voids around it [Figure 3]. The T1-weighted MRIs did not indicate any evidence of fat deposition in the filum terminale [Figure 3]. Spinal DSA of the left T8 intercostal artery showed an AVF at the L3-4 level [Figure 3]. L3-4 laminectomy was performed and revealed dilated abnormal vessels running over a swollen filum terminale [Figure 4]. Micro-Doppler ultrasonography confirmed the area where the blood flows turned its direction from caudal to rostral (i.e., identifying the transitional point for the AVF). Part of the filum terminale and the AVF was resected along with the feeding/draining vessels [Figure 4]. The postoperative histological study confirmed that the arterialized vein was embedded in the adipose tissue [Figure 4]. These findings proved

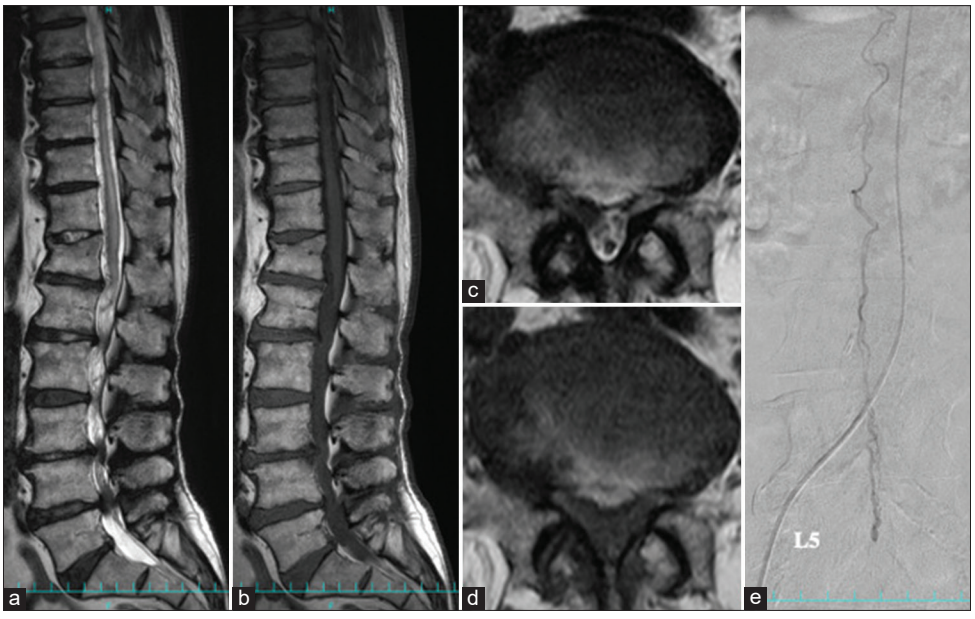

Figure 1: Filum terminale arteriovenous fistula at L5 (Case 1). (a) Sagittal T2-weighted magnetic resonance image (MRI) shows the swollen spinal cord surrounded by flow voids. It also shows redundant cauda equina and spinal canal stenosis from L3 to L5. (b) Sagittal T1-weighted MRI does not show high signal intensity in the filum terminale. (c and d) Axial T1- and T2-weighted MRIs at L5 indicate thickened filum terminale due to edema, although the fatty component is not evident. (e) Selective catheter angiography from left T10 intercostal artery shows an arteriovenous fistula located at L5 that is fed by a caudally running artery in the filum terminale. The drainer is a vein in the filum terminale with an ascending flow.

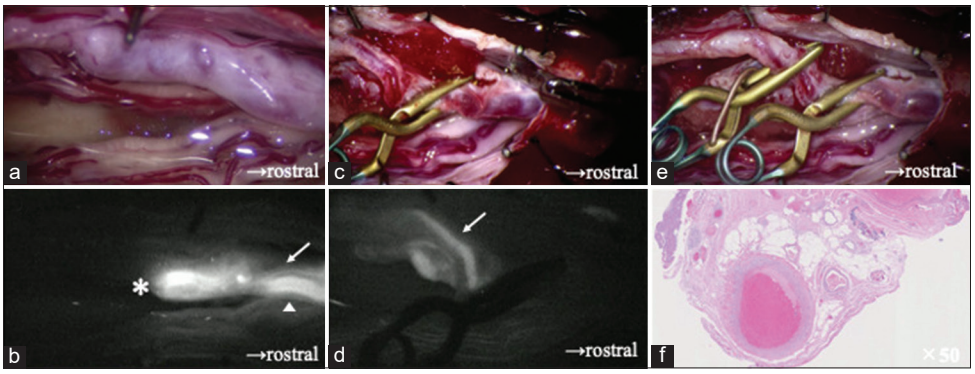

Figure 2: Case 1 continued. (a) Intraoperative findings show edematous filum terminale with dilated arteries and veins, (b) Intravenous indocyanine green (ICG) video angiogram shows the artery of the filum terminale (AFT) with a caudal inflow (arrow), connecting directly with the vein in the filum terminale drained rostrally (arrowhead), The transitional point from the artery to the vein was considered as the arteriovenous fistula (AVF) (asterisk), (c) A temporary aneurysm clip is applied on the AFT immediately proximal to the shunting point, (d) intravenous ICG angiogram shows another minor feeding artery (arrow) joining the AVF with a caudal inflow, (e) one more temporary aneurysm clip is applied to the minor feeding artery, (f) the pathological finding of resected filum terminale shows an arterialized vein embedded in the adipose tissue (hematoxylin and eosin stain, ×50).

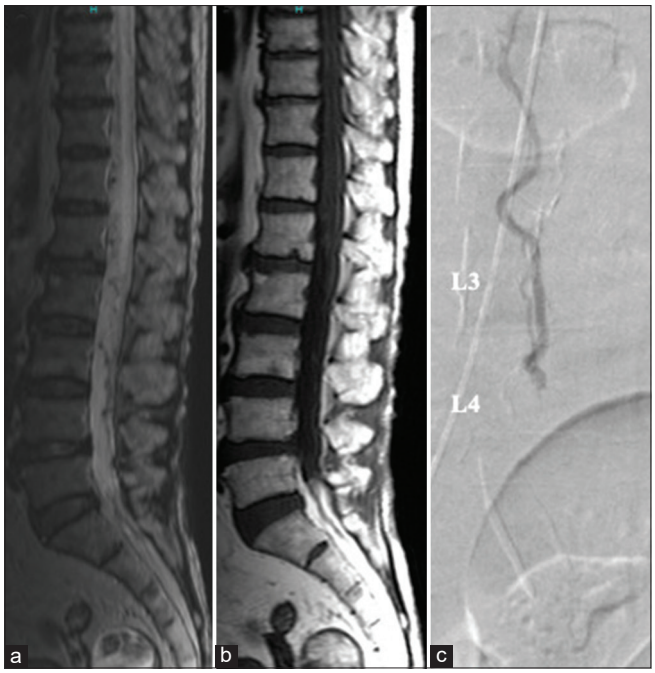

Figure 3: Filum terminale arteriovenous fistula at L3-4 (case 2). (a) Sagittal T2-weighted magnetic resonance image (MRI) shows edematous spinal cord and perimedullary flow voids, (b) spinal lipoma is not recognized on sagittal T1-weighted MRI, (c) selective catheter angiography from left T8 intercostal artery shows an arteriovenous fistula located at the L5 level supplied by an artery that originated from the anterior spinal artery.

that the AVF arose in the lipoma of the filum terminale despite the radiologically negative findings of fatty filum terminale. Although the immediate postoperative MRIs confirmed the disappearance of the abnormal flow voids, the postoperative course was complicated by patient's sustaining an intracerebral hemorrhagic. The patient was

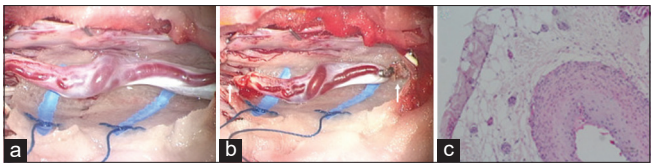

Figure 4: Case 2 continued. (a) Intraoperative finding shows edematous filum terminale with dilated arteries and veins. (b) The filum terminale is resected with the arteriovenous fistula (arrow). (c) The histopathological study of the resected filum terminale shows an arterialized vein surrounded by the adipose tissue.

then transferred to a rehabilitation facility bedridden and with aphasia.

DISCUSSION

Review of the literature regarding FTAVF concomitant with the lipomas of the filum terminale

We reviewed a total of thirteen cases with concomitant FTAVF and fatty fila; this included our two cases reported above [Table 1].[5-8] Patients averaged 55.5 years of age with a preponderance of males (i.e., 74.6%). Eight out of the 13 AVFs (61.5%) were fed by a single feeder. The fatty component in the filum terminale was radiographically evident on preoperative MR studies in nine cases. In two cases,[7,8] fatty fila were not referred to in the article; however, the histopathological figures clearly show the adipose tissue around the shunt vessels. Lumbar canal stenosis was observed in four cases. Two of the 13 cases were treated endovascularly, while the remaining 11 required surgery; one was surgically treated after a failed embolization, and resection of the filum terminale was performed in eight of the surgically treated cases.

Table 1: Reports of filum terminale arteriovenous fistula concomitant with fatty fila.

S. No.	Authors (years)	Gender	Age (years)	Location of the Fistula	Feeder Level	Number of Feeders	LCS	Treatment
1	Djindjian, 1989	Male	53	Sacral	Bil IIA	Multiple	NA	Embolization with NBCA+*En bloc* resection of the filum
2	Cheung, 2005	Male	42	S2	Rt T12	Single	No	Surgical excision of the fistula
3	Trinh, 2011*	Male	57	NA	NA	Single	Yes†	*En bloc* removal of the fistula and the filum
4	Macht, 2012	Male	57	S3-4	Lt IIA	Single	No	Embolization with NBCA
5	Chanthanaphak, 2013	Female	57	S2	Lt T4	Single	No	Surgical disconnection of the fistula and untethering
6	Sharma, 2014	Male	48	L5	T10	Single	L4-5	Surgical excision of the fistula
7	Takeuchi, 2014*	Male	71	L4	Lt T9	Single	L3-5	*En bloc* removal of the fistula and the filum
8	Giordan, 2017	Male	46	S3-4	Bil IIA	Multiple	NA	Embolization
9	Takai, 2019	Male	83	S4	Rt IIA	Single	NA	*En bloc* removal of the fistula and the filum
10		Male	54	S4	Bil IIA	Multiple	NA	*En bloc* removal of the fistula and the filum
11		Male	40	S1	Lt L3, MSA	Multiple	NA	*En bloc* removal of the fistula and the filum
12	Present cases, 2021	Male	72	L5	Lt T10	Multiple	L3-5	*En bloc* removal of the fistula and the filum
13		Female	76	L4	Lt T8	single	No	*En bloc* removal of the fistula and the filum

Bil: Bilateral, IIA: Internal iliac artery, LCS, Lumbar canal stenosis, Lt: Left, MSA: Median sacral artery, NA: Not available, NBCA: n-butyl-cyanoacrylate, Rt: Right. *Presented specimen showed adipose tissue around the shunt vessels, †Two cases were reported to be complicated with LCS at L4-S1 or L4-5 level; however, there was no detailed description of which case corresponds to which

Etiology of FTAVF

Given that FTAVF itself occurs rarely, the number of 13 cases with two coexistent pathologies led us to believe that there might be an association between the two pathologies.[6] Our two cases pathologically revealed the fatty filum terminale that was hidden in radiographically normal-appearing filum terminale; therefore, the presence of fatty filum terminale around the FTAVF might have been unrecognized in the past. The development of *de novo* FTAVF with spinal lipoma has been also reported.[6] The hypervascularization induced by spinal lipoma has been speculated to contribute to the etiology of FTAVF.[2] In addition, lumbar canal stenosis might be a predisposing factor for the development of AVFs, which may cause repetitive microtrauma and subsequent inflammation in the pinched filum terminale.[3] To summarize, the lipoma of the filum terminale may be a predisposing factor for the occurrence of AVFs, and lumbar canal stenosis may secondarily trigger the actual development of AVFs.

The role of resection and histological evaluation of the filum terminale as a background of FTAVFs

Brown *et al*. found lipomas in the filum terminal with an incidence of 4% from randomly selected lumbosacral spine MRIs.[1] Any adipose tissue observed in the intradural filum terminale in adults implies a congenital abnormality.[4] MRI appears to be less sensitive than a histopathologic examination in detecting fat tissue in the filum terminale.[4] Fat deposition in the filum terminale, therefore, can only be found with a histopathological evaluation. In this regard, histopathological examination of the filum terminale as a background of AVF is encouraged to disclose more cases similar to ours, and to elucidate the pathogenesis of this relatively rare vascular disorder.

CONCLUSION

This study provides indicate a possible association between FTAVF and fatty filum terminale. Resection is appropriate curative treatment, and histological evaluation of the filum

terminale should be performed to elucidate the pathogenesis of AVF.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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How to cite this article: Shimizu, Mitsuhara T, Takeda M, Yamaguchi S. Spinal arteriovenous fistula in the lipoma of the filum terminale: A report of two cases and literature review. Surg Neurol Int 2021;12:103.