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A sacral filum terminale arteriovenous fistula fed by a left T9 artery of Adamkiewicz

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ABSTRACT

This is a case of 62-year-old patient presenting with tingling and numbness extending from the buttocks area towards the lower extremities and gait instability. Contrast Magnetic Resonance Imaging (MRI) and time-resolved imaging of contrast kinetics Magnetic Resonance Imaging (TRICKS MRI) raised suspicion for a possible dural arteriovenous fistula. Diagnostic Digital Subtraction Angiography (DSA) showed a filum terminale arterio venous fistula (AVF) fed by the left T9 from the anterior spinal artery of Adamkiewicz all the way down to the fistulous point at L5-S1. The patient underwent successful laminectomy of L5-S1 and clipping of the filum terminale arteriovenous fistula.

1. Introduction

Vascular lesions of the spine are divided into arteriovenous fistulae and arteriovenous malformations and account for 1–2% of all vascular malformations of the central nervous system [1,2]. The majority of these lesions are spinal dural arteriovenous fistulae (sDAVF) and are mostly located between T6 and L2 [2]. Filum terminale arteriovenous fistulae (FTAVF) are a rare type of sDAVF characterized by an abnormal single connection between the anterior spinal artery and its distal portion(the artery of the filum terminale), with a draining vein [3]. The arterialization of the vein causes venous congestion and this increased venous pressure may cause decreased tissue perfusion, vascular steal, ischemia or hemorrhage [4]. Symptoms are usually related to progressive myelopathy but because of the vague presentation, the average patient will present with symptoms 1–3 years prior to diagnosis [5]. The main presenting symptom is motor weakness which is seen in 80% of cases followed by sensory loss, back pain, bladder and bowel disturbances and sexual dysfunction [3]. Digital Subtraction Angiography is the gold standard for accurate diagnosis [6]. In this case report, we present a rare case of a FTAVF fed by the artery of Adamkiewicz originating at T9 and going all the way caudally to the fistulous point at L5-S1.

2. Case report

2.1. Clinical presentation

This is a case of a 62-year-old patient, presenting with tingling and numbness extending from the buttocks area towards the lower extremities and gait instability. A non-contrast spinal Magnetic Resonance Imaging (MRI) was performed and showed a hemorrhage in the spinal cord (T8-T9) with flow voids raising concern for a spinal dural arteriovenous fistula (sDAVF). (Fig. 1).

2.2. Physical exam

Upon admission, the patient's neurological exam showed muscle strength at 4/5 in the bilateral psoas and quadriceps muscles and otherwise 5/5 in all four extremities. Sensation was intact to fine touch and pinprick in all extremities and equal bilaterally. Reflexes were 2 + overall and the rectal tone was normal.

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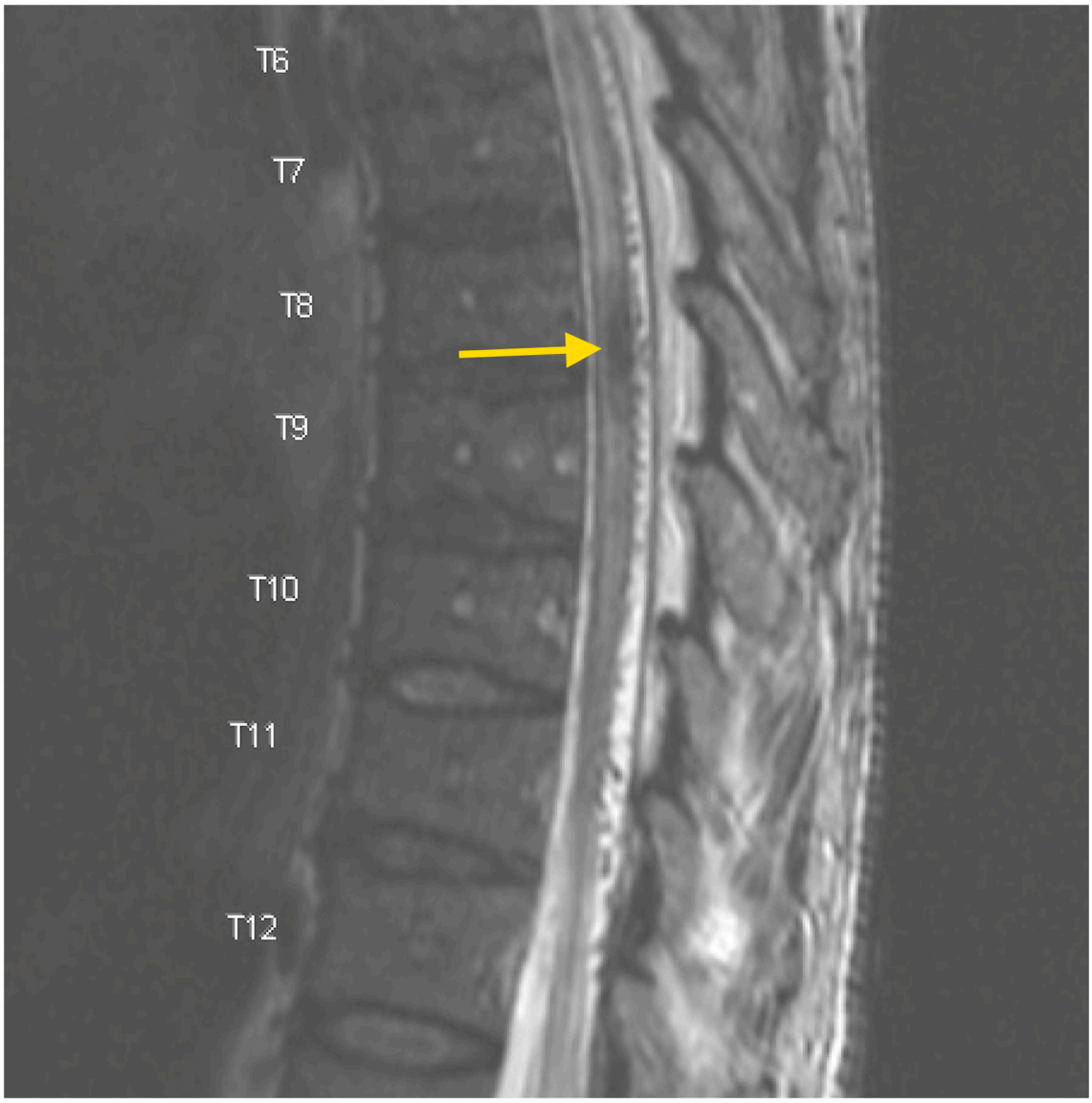

Fig. 1. Sagittal T2 Weighted MRI showing a spinal cord hemorrhage (T8-T9) and flow voids. (Yellow arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

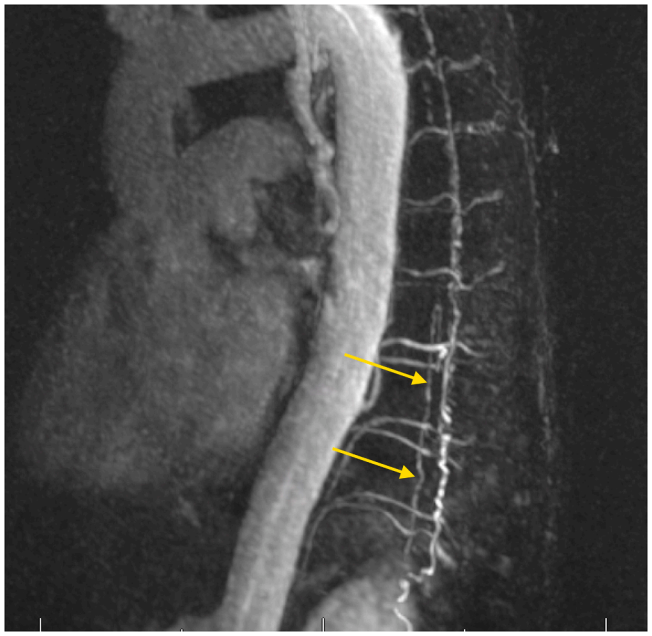

Fig. 2. TRICKS MRI showing a filum terminale AVF fed by the left T9 from the anterior spinal artery of Adamkiewicz all the way down to the fistulous point at L5-S1. (Yellow Arrows). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

2.3. Diagnosis and imaging

The patient underwent a Time-Resolved Imaging of Contrast Kinetics Magnetic Resonance Imaging (TRICKS MRI) which showed a prominent left radiculomedullary branch at T10 emanating from a branch of the aorta at T11 extending through the T10-T11 neural foramen and supplying both the sDAVF and the artery of Adamkiewicz. (Fig. 2) The findings of the TRICKS MRI necessitated a diagnostic digital subtraction angiography (DSA) which showed a filum terminale AVF fed by the left T9 from the anterior spinal artery of Adamkiewicz all the way down to the fistulous point at L5-S1, with a single draining vein filling caudal/rostral going side to side with the artery of the filum. (Fig. 3, Fig. 4).

2.4. Procedure and hospital course

Weighing the risk of super selectively catheterizing the artery of Adamkiewicz, with the risk of dissection/occlusion or retrograde thrombosis, the decision was made to perform a laminectomy and clipping of the fistula. The patients underwent an L5-S1 laminectomy with intradural ligation of the sDAVF using a straight 7 mm Lazik clip. Neurophysiological monitoring did not show any changes after clipping and imaging with Indocyanine green showed disconnection of the fistula and absence of an early draining vein. (Video 1) During the post-operative period, the patient did not experience any complications, was able to ambulate and his symptoms improved. A follow-up DSA showed 100% occlusion of the sDAVF and patency of the artery of Adamkiewicz. (Fig. 5, Fig. 6) The patient was discharged home.

3. Discussion

The filum terminale is a 15 cm long connective tissue filament composed of collagen fibers and covered by pia matter that connects the conus medullaris at the level of L1-L2 to the end of the dural tube at the level of S2 [3]. The filum terminale vascular supply is provided mainly by the filum terminale artery, a caudal extension of the anterior spinal artery running anteriorly, in addition to the lateral and middle sacral arteries and drains in the vein of the filum terminale running posteriorly [7,8]. Initially described by Djindjian et al. in 1977, FTAVF's are very rare lesions and comprise 3% of all spinal AVF's [8,9]. Increased blood flow through the fistulous point causes increased venous pressure resulting in congestive myelopathy or conus medullaris syndrome [3]. Because of the unspecific symptoms and the similar pathophysiology of FTAVF's to other vascular lesions, these entities are usually misdiagnosed [10]. On MRI, FTAVF's are associated with high T2 signals extending from the conus up to the spinal cord [3,10]. DSA is the gold standard diagnostic modality as it provides the most accurate description of the lesion, identifies all the feeding arteries, and reveals any associated spinal vascular malformations [6]. On DSA, FTAVF's present as single hole fistulas below the conus and the fistulous point can be identified at the point of transition from a small artery to the large congested vein [3].

80% of FTAVF's are located between L3 and L5 and are supplied by a single feeder, the filum terminale artery [7]. Going back in the literature, Giordan et el were able to identify 39 cases of FTAVF's in a 40-year literature review [3]. This does not only highlight the rarity of such lesions, but interestingly, also shows that FTAVF's with a wide distance between the feeder and fistula are even rarer [3,8,11-13]. In our case report, the distance between the feeder (T9) and the fistula (L5/S1) was 9 vertebral levels [14].

The primary goal of treating FTAVF's is the complete obliteration of the fistula, while maintaining the patency of the spinal circulation. Treatment can be achieved via open surgery or endovascular approaches safely and effectively [15]. Several factors, some related to the fistula and others related to the patients, play a major role in deciding the

Fig. 3. Anterior-Posterior angiography showing the feeder originating from the artery of Adamkiewicz at the level of left T9.

modality of treatment. Generally, surgical techniques have been the mainstay of treatment throughout the literature and is preferred when the feeder is far away from the fistula [16]. During the procedure, a laminectomy and removal of the ligamentum flavum at the level of the fistula is performed to expose the dura [3].Once the fistula is exposed, the feeding artery can be easily identified due to its relatively small size compared to the draining vein. When the aberrant connection is identified, it is clipped to occlude the fistula. All this is done while somatosensory and motor evoked potentials are monitored to guarantee good clinical outcomes and avoid spinal cord infarction [7,8,17,18]. Intraoperative Indocyanine green angiography is performed before and after clipping to make sure the fistula was disconnected [3]. The success rate of the surgical approach are close to 100% and complications rates tend to be less than 5% [19,20]. Endovascular embolization has been performed in 33% of the cases and the success rate is close to 76% [10,15]. this modality of treatment is preserved for cases where the feeder is close to the fistula due to the high risk of injuring the anterior spinal artery in cases of long distance between the feeding artery and fistula [17].

4. Conclusion

FTAVF's are rare vascular entities that may have detrimental effects on the spinal cord. In most cases, diagnosis is late due to the similarity of

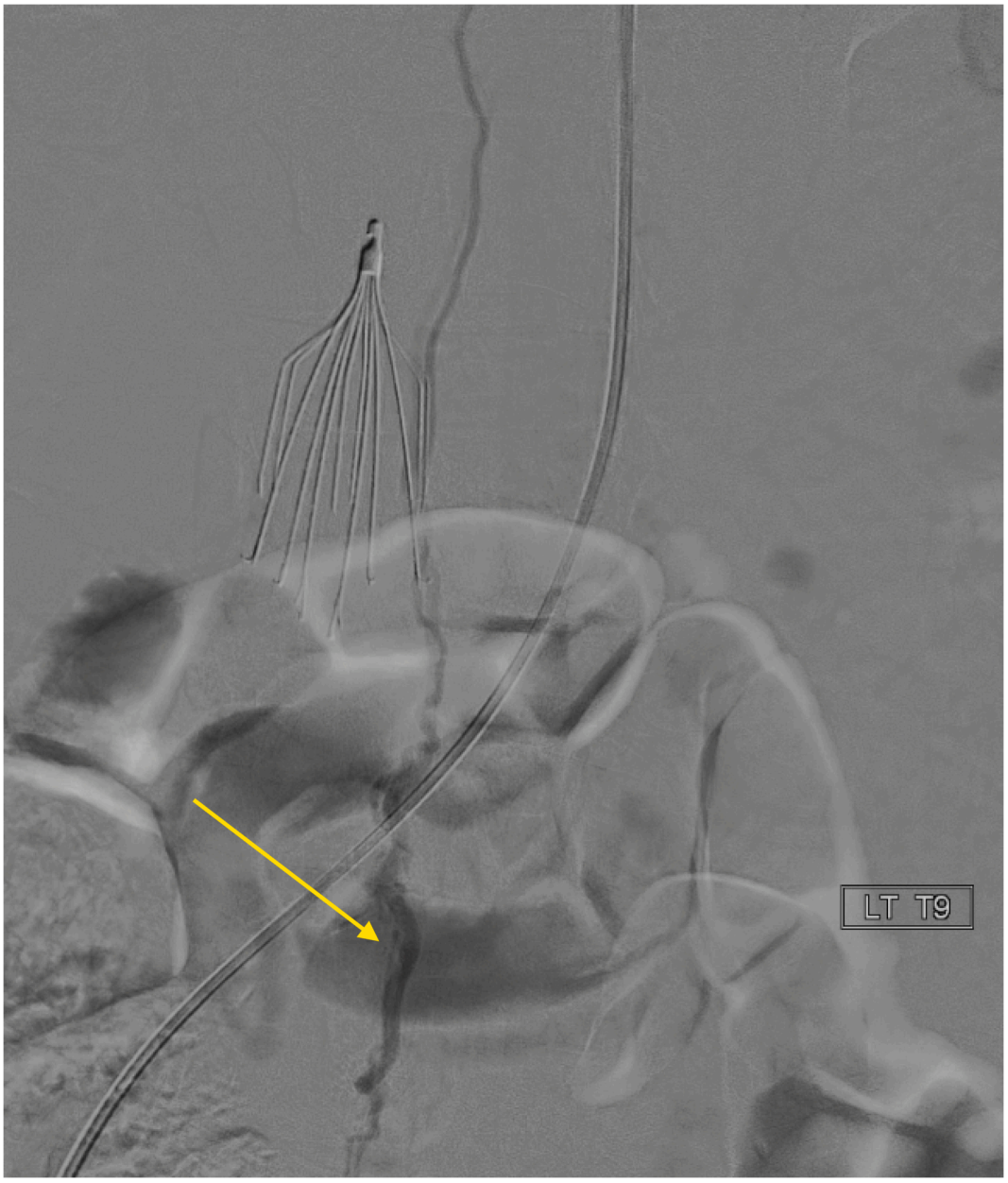

Fig. 4. Anterior-Posterior angiography showing the filum terminale fistula at L5-S1. (Yellow Arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

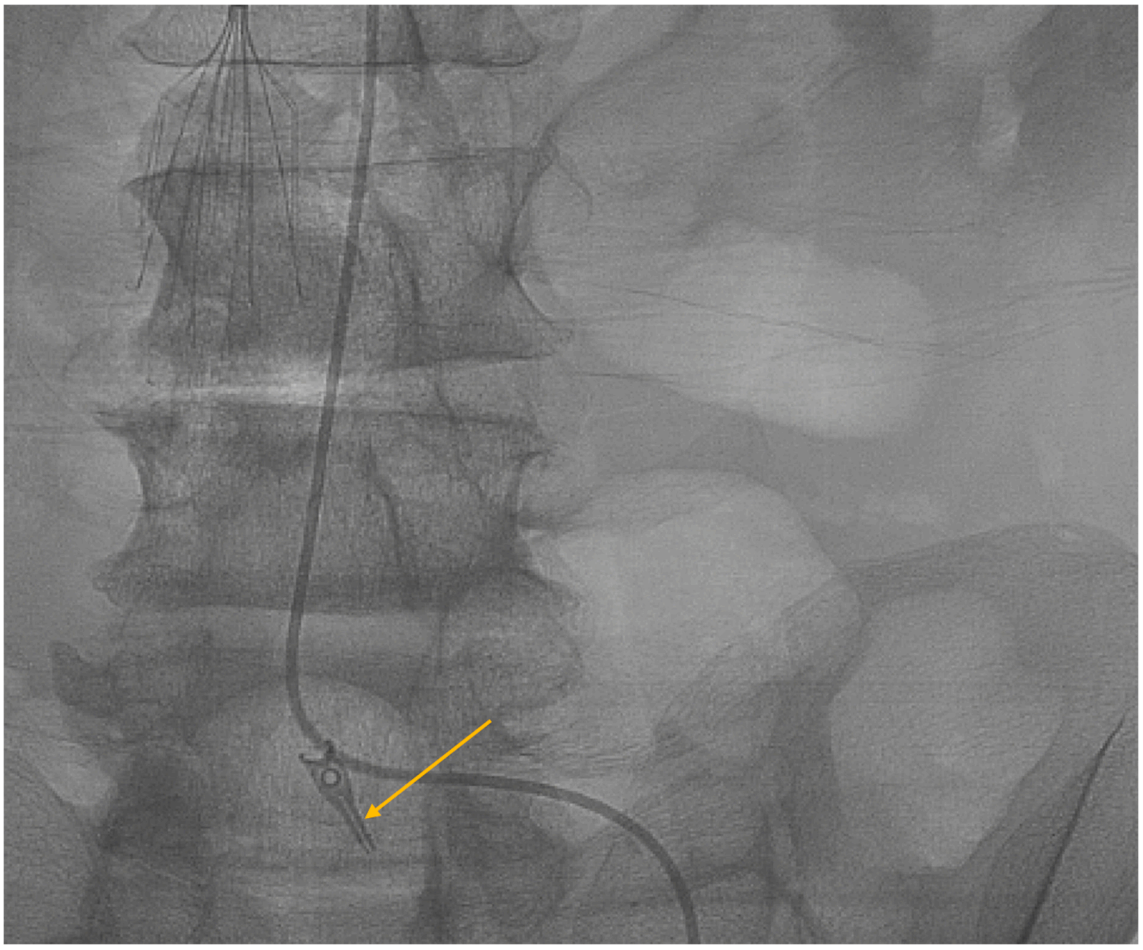

Fig. 5. Post-operative anterior-posterior fluoroscopy showing the clip used to obliterate the FTAVF. (Yellow arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

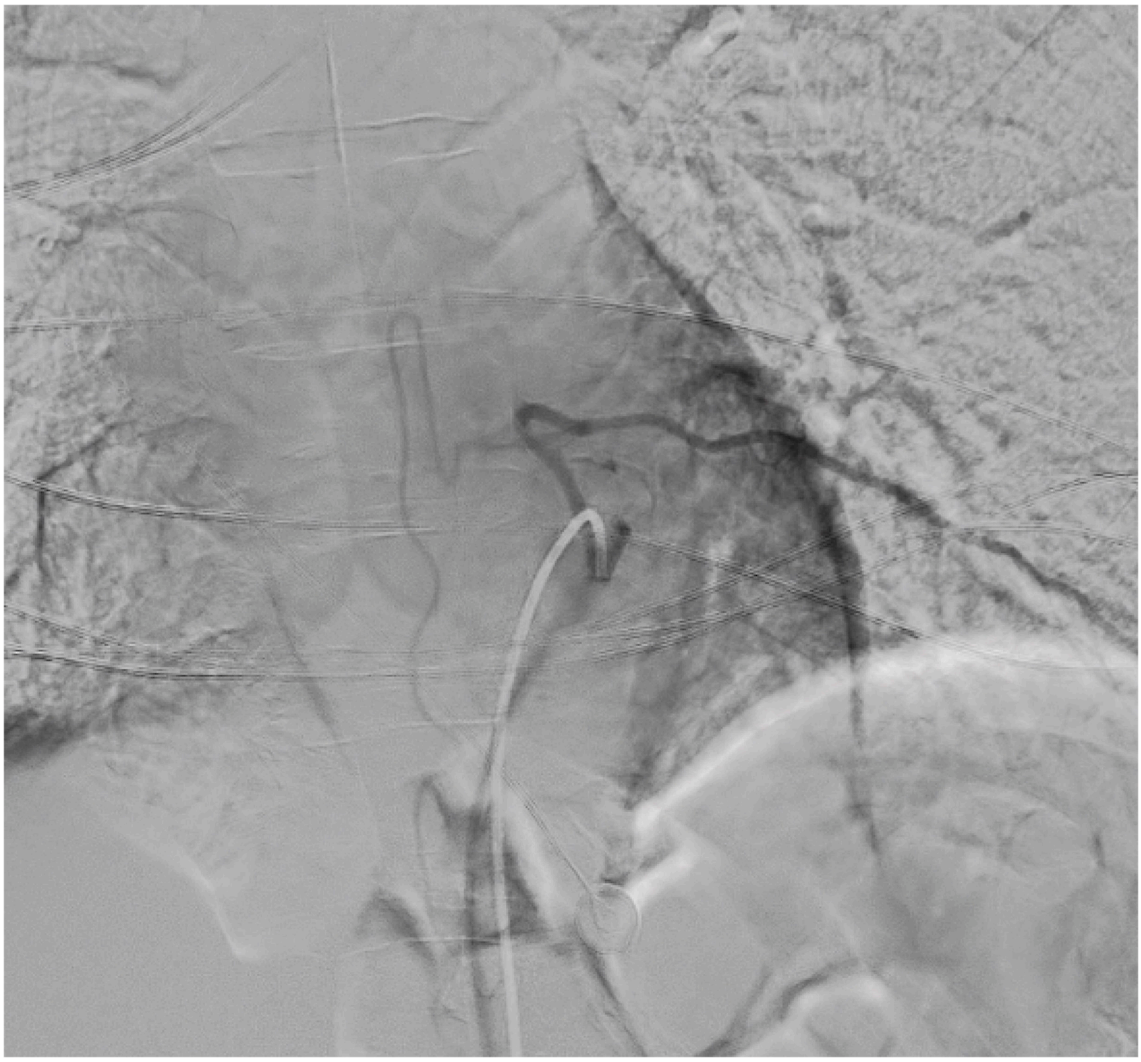

Fig. 6. Post-op anterior-posterior angiography showing obliteration of the FATVF.

clinical symptoms and radiological characteristics of FTAVF's with other vascular lesions. Once, diagnosed, treatment can be achieved via surgery or endovascularly depending on several factors. In our case, the large distance between the feeding artery and the fistulous point and the fact that the feeder was the artery of Adamkiewicz made the surgical option the preferred one.

5. Disclosures

Data sharing statement: The relevant anonymized patient-level data are available on reasonable request from the authors.

Ethical approval: All procedures performed in the studies involving human participants were per the Institutional Review Board (IRB) ethical standards and national research committee and the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Informed consent: The study protocol was reviewed and approved by the University Institutional Review Board. Following our institutional guidelines, all protected health information was removed, and individual patient consent was not required in the analysis of the case report.

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Declaration of Competing Interest

The authors declare the following financial interests/personal relationships which may be considered as potential competing interests: Pascal Jabbour reports a relationship with MicroVention Inc that includes: consulting or advisory. Pascal Jabbour reports a relationship with Medtronic Inc that includes: consulting or advisory. Pascal Jabbour reports a relationship with Cerus endovascular that includes: consulting or advisory. Pascal Jabbour reports a relationship with Balt that includes: consulting or advisory. Stavropoula I Tjoumakaris reports a relationship with MicroVention Inc that includes: consulting or advisory. Stavropoula I Tjoumakaris reports a relationship with Medtronic Inc that includes: consulting or advisory. Michael R Gooch reports a relationship with Stryker that includes: consulting or advisory

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.inat.2022.101531.

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