



Management of spinal dural arterio venous fistula with peri-medullar drainage. Experience of a north-African centre

Assoumane Ibrahim^{1,2}, Loucif Houari¹, Sanoussi Samuila³,
Sidi Said Abderahmane¹, Abdelhalim Morsli¹

¹ Department of Neurosurgery, CHU Bab El Oued, Algiers, ALGERIA

² Department of Neurosurgery, Maradi Reference Hospital, NIGER
REPUBLIC

³ Department of Neurosurgery, Niamey National Hospital, NIGER
REPUBLIC

ABSTRACT

Introduction: Spinal Dural Arterio Venous Fistula (SDAVF) is an arteriovenous communication on the spinal dura with peri-medullar venous drainage. It is a curable cause of myelopathy and the most common form of spinal arterio venous malformation (AVM). The average age of revelation is the fifth decade; it is a diagnostic and therapeutic emergency.

Materials and methods: This is a retrospective study conducted in the neuro surgical department of CHU Bab El Oued in Algiers during a five years time from November 2013 to September 2018. We assessed the clinical status of patients according to the Aminoff-Logue disability score before and after surgery. All patients did a total spine MRI followed by a Medullar angiography which facilitated the pin-pointing of the exact location of the dural fistula. The mean follow-up is 30 months.

Results: There were five males and two females, all of them older than 45 years of age. At the admission, patients presented with signs of neurological deficits. After the diagnosis of SDAVF the surgical intervention consisted of a disconnection of the arteriovenous communication by coagulation and section of the fistula at the foot of the vein after a laminectomy. Functional rehabilitation was prescribed for all patients and they were regularly followed-up.

Conclusion: Treatment of AVF is surgical or endovascular. Results depend largely on preoperative neurological status.

INTRODUCTION

The spinal dural Arterio Venous Fistula (SDAVF) with peri-medullar venous drainage is an abnormal communication within an afferent artery and an adjacent vein without interposition of capillary bed. This communication occurs at the spinal dura and is followed by a peri medullar venous drainage. Individualize by Kendall and Merland in the 1980 th , it is a rare pathology even if it represents the most frequent spinal arteriovenous malformation accounting about 60–80% of all spinal vascular malformations [1,2,3,4].

Keywords

arteriovenous fistula,
arteriovenous malformation,
myelopathy



Corresponding author:
Assoumane Ibrahim

Department of Neurosurgery, CHU
Bab El Oued, Algiers, Algeria

as_ibrah2006@yahoo.fr

Copyright and usage. This is an Open Access article, distributed under the terms of the Creative Commons Attribution Non-Commercial No Derivatives License (<https://creativecommons.org/licenses/by-nc-nd/4.0/>) which permits non-commercial re-use, distribution, and reproduction in any medium, provided the original work is unaltered and is properly cited. The written permission of the Romanian Society of Neurosurgery must be obtained for commercial re-use or in order to create a derivative work.

ISSN online 2344-4959
© Romanian Society of
Neurosurgery



First published
December 2019 by
London Academic Publishing
www.lapub.co.uk

Arterial blood flow is shunted directly into the venous plexus, under arterial pressure. The venous plexus subsequently becomes "arterialized," and obstruction of venous outflow leads to venous congestion, venous hypertension, and progressive ascending myelopathy [5]

The average age of revelation is the fifth decade. Clinical signs are not specific, that is the reason of the diagnosis delay causing sometimes severe disability; for that reason, SDAVF constitute a diagnostic and therapeutique emergency. Treatment of AVF is usually surgical or endovascular. The objective of our study is to assesses the surgical treatment of SDAVF and the clinical outcome of patients operated for SDAVF in the neurosurgical department in CHU BEO Algiers.

MATERIALS AND METHODS

This is a retrospective study conducted in the neuro surgical department of CHU Bab El Oued in Algiers during a five years time from November 2013 to September 2018. We analyze clinical data of patients operated for SDAVF.

We assessed the clinical status of patients according to the Aminoff-Logue disability score before and after surgery (Table 1).

Function & Score	Definition
gait	
0	normal
1	leg weakness, abnormal gait or stance but no restriction of activity
2	restricted activity but not requiring support
3	requiring 1 stick for walking
4	requiring 2 sticks, crutches, or walker
5	confined to wheelchair
urination	
0	normal
1	hesitancy, urgency, frequency, altered sensation, but continent
2	occasional urinary incontinence or retention
3	total incontinence or persistent retention
defecation	
0	normal
1	moderate constipation
2	severe constipation or occasional incontinence
3	total incontinence

Table 1: Aminoff-Logue disability score

This score has 4 grades: grade 1: 0-2; grade 2: 3-5; grade 3: 6-8; grade 4: 9-11

We performed a total spinal MRI which suspect the SDAVF and spinal digital subtraction angiography (DSA) that facilitated the pin-pointing of the exact location of the dural fistula.

All patients underwent microsurgical approach;

the day before surgery we perform a plan x ray on a metallic landmark to guide the skin incision. The microsurgical approach consisted of a disconnection of the arteriovenous communication by coagulation and section of the fistula at the foot of the vein after a laminectomy.

Patients presenting disabilities were send for functional rehabilitation.

RESULTS

During the time of our study we registered 7 patients suffering of SDAVF, there is predominance of male with a sex ratio of 5/2; The mean age of our patients is 56 years with extreme from 49 to 68 years. The time before consultation is fifty one month's with extreme from 6 to 60 months.

All patients suffered for motor disturbance (100%), a leg weakness is found in 5 patients representing 71, 42%, one patient require a stick for walking and another one require walker to move. Urination disturbance is present in all patients (100%) as occasional incontinence is found in 3patients (42, 85%), frequency urination is found in 2 patients (28, 57%) and occasional retention in 2 patients (28, 57%). Six patients presented defecation disorders representing 85, 75 % of patients, like moderate constipation for 4 patients (57, 14%), severe constipation for one patient, another one presented occasional incontinence. Three (3) patients present Sensibility disorder which represent 42, 85% of patients. The pre-operative disability scale of Aminoff-logue is III for 2 patients (28, 57%) and II for 4 patients (57, 14%), and I for one patient.

All patients did a total spine MRI, this investigation objectified serpiginous images and flow void sign of peri medullar vessels dilation; also, an intra medullar hyper intensity (figure 1).

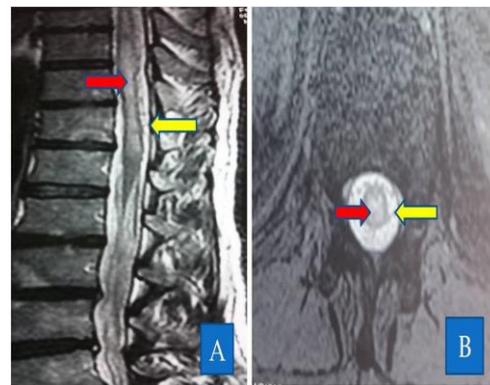


Figure 1: A: MRI sagittale T2 weithed showing flow void sign (yellow arrow) and intramedullar hyper intensity (red arrow).

To locate the communication, we performed medullar angiography for all patients; angiography has facilitated the pin-pointing of the exact location of the dural fistula. The investigation permits us to see the afferent pedicles, the peri medullar drainage vein which is dilated and serpiginous, also the dural fistula (figure 2). All our patients presented thoracic single fistula.

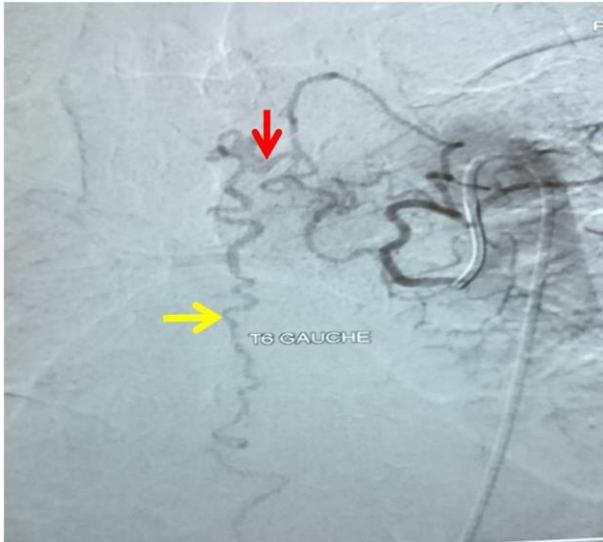


Figure 2: DSA: after catheterisation of Left T6 showing a fistula (red arrow) and an arterialized vein (yellow arrow).



Figure 3: Per operative image, after durotomy showing the fistula (yellow arrow) and the tortuous varterialized veins (red arrow).

For all our patients we performed microsurgical approach; under general anesthesia, on prone position, a median skin incision is made at the level of the fistula; we perform a laminectomy one level above and one below the fistula to expose the work

field before a median durotomy and suspension of the dural edges. Under microscope the arterialized veins are found tortuous and colored in red and the fistula is found communicating aradicular artery (figure 3). We performed the coagulation of the fistula and section of it at the foot of the vein. At the end of the procedure we spent about 30 minutes after the section of the fistula to make sure that the arterialized vein become progressively blue, which is the vein normal color. The dural closure is assured with continuous running suture.

Patients presenting disabilities were send for functional rehabilitation. The post-operative outcome 7 days and 6 months after surgery is summarized in the following table.

	Initial Aminoff-Logue Grade	Day 7 after surgery	6 Months later
Patient 1	III	IV	III
Patient 2	III	IV	II
Patient 3	II	III	I
Patient 4	II	II	II
Patient 5	II	II	I
Patient 6	I	I	I
Patient 7	II	II	I

Table 2: Post-operative outcome of patients at Day 7 and 6 months later.

The mean follow-up is 30 months; patients are seen in the clinic weekly for the first month and every month for three months and every trimester until one year.

DISCUSSION

In our study there is predominance of male with the Average age of revelation the fifth decade as reported by many authors [6, 7]. The time before consultation is long about 51 months in our series; it is comparable to the time reported in the literature [8, 9,]. This is due to the difficulty to diagnose the SDAVF because of lack specific signs. The diagnosis delay reduces the chance of recuperation even after the treatment.

The most frequent signs in our series is motor and sphincter disturbance representing 100% of patients followed by sensibility disturbance which represent 42, 85% of patients. Many authors

reported the predominance of motor disturbance follow by sensory disturbance and sphincter disturbance [2, 10, 26].

We performed MRI for all our patients, in case of SDAVF suspicion a DSA is performed to confirm and locate the level and number of fistula, the feeders as well. Yamaguchi [11] reported that Spinal CT angiography could be used to demonstrate the fistula localization. In practice it is not safe because of the radiation quantity, before getting the exact location of the fistula. MRI images signs On T1-weighted scans, the swollen cord is slightly hypointense and enlarged. Following contrast administration, diffuse enhancement may be seen within the cord as a sign of chronic venous congestion with a breakdown of the blood spinal cord barrier [13, 14]. On T2-weighted sequences, the cord edema is depicted as a centromedullary not well-delineated hyperintensity over multiple segments that is often accompanied by a hypointense rim, most likely representing deoxygenated blood within the dilated capillary vessels surrounding the congestive edema spin echo [3D-TSE]) compared with standard T2 TSE sequences [12].

The help of MR angiography in the positive diagnostic was emphasized by some authors. For Efrat Saraf-Lavi [15] the principal advantage of MR angiography is the improved detection of the fistula level, with the correct level \pm one level identified in 73% of true positive cases. They supported that Overall, the combination of MR imaging and MR angiography provides improved screening for dural AVF and benefits the subsequent radiographic DSA study by helping target the level of the fistula.

We performed for all our patients a total spine MRI, this investigation objectified suspicion signs of SDAVF but to confirm the diagnostic the DSA was used, this investigation allow us to locate the fistula, the number of afferent arteries. Many authors recognize the Digital angiography as a helpful tool that can give a map of the vascularisation and help to plan the endovascular treatment. It is the gold standard for the diagnostic of SDAVF [16, 17, 18, 19]. All our patients presented thoracic single fistula, many authors reported that the thoracic level is the most frequently affected [2,26,34] and most of patients present a single fistula, the multiple are rarely reported[2,9].

All patients underwent microsurgical approach;

the surgical technique consisted of a disconnection of the arteriovenous communication by coagulation and section of the fistula at the foot of the vein after a laminectomy. Permanent vascular clips could be used [22]. Authors reported the use of microsurgery or endovascular to be effective [7, 20, 21]. Many authors reported a rate of recanalization after embolization with polyvinyl alcohol, in up to 83% of cases [27,28].but with the evolution of material use Endovascular embolization with liquid adhesive material is reported to have a very low rate of recanalization [29,30]. The success rates of endovascular therapy have been reported to vary between 25% and 75% [31, 32] but For J. Marc et al the Advantages of the endovascular technique are its noninvasiveness and the possibility of an immediate angiographic control of the treatment [10]. Steinmetz MP et al in a recent meta-analysis suggested complete occlusion of the fistula following surgery in 98% [33].

The outcome was marqued by a worsening of the Aminoff-Logue disability scale in three patients in the first week after the surgery. But after six months of occupational therapy five patients (71.42%) improved their Aminoff-Logue disability scale while two patients remain stable. They did not improve their disability after two years. Approximately 75% of patients get a degree of satisfaction as report in the literature [22].

In their series of 17 patients treated by microsurgery Jakub w et al reported that functional improvement or good stable condition was achieved in 65% of the patients at discharge and in 76% in long-term follow-up [34]. Chibbaro S et al advocated multidisciplinary approach based on direct cooperation between endovascular specialists and neurosurgeons before the choice of the treatment option. They obtained the excellent result of 100% cure rate with no procedural complications after patient's selection for microsurgical treatment [21] The most complications of the microsurgical treatment are CSF leak, infection, haematoma, and neurological sequelae [23, 24, 25]. In our study we did not experiment such complications.

CONCLUSION

SDAVF is a rare pathology, with unspecific clinical signs leading to diagnostic difficulties. To avoid misdiagnosis a multidisciplinary collaboration is required. The neuroradiologist help is very

important using MRI and DSA to get the exact location of the fistula and guide the treatment, either surgical or endovascular. After our series we conclude that microsurgery is a good option for the treatment but the post-operative outcomes depend largely on preoperative neurological status.

Conflict of interest: none

REFERENCES

- Houdart E, Chapot R, Boissonnet H et Merland JJ. Fistules artérioveineuses dures rachidiennes. *Encycl Méd Chir (Editions Scientifiques et Médicales Elsevier SAS, Paris, tous droits réservés), Neurologie*, 17-490-B-20, 2000, p. 4.
- Lee J et al. Clinical presentation, imaging findings, and prognosis of spinal dural arteriovenous fistula. *J Clin Neurosci* 2015 Elsevier.
- Robert F. Spetzler, M. Yashar S. Kalani, Peter Nakaji, *Neurovascular Surgery*. Thieme Publishers New York 2015
- Nozar Aghakhani; Fabrice Parker; Philippe David; Pierre Lasjaunias; Marc Tadie, Curable Cause of Paraplegia Spinal Dural Arteriovenous Fistulae. American Heart Association, Inc. 2008.
- Jennifer E. Fugate, D.O. Giuseppe Lanzino and Alejandro A. Rabinstein. Clinical presentation and prognostic factors of spinal dural arteriovenous fistulas: an overview. *Neurosurg Focus* 32 (5):E17, 2012.
- Muralidharan R, Saladino A, Lanzino G, et al. The clinical and radiological presentation of spinal dural arteriovenous fistula. *Spine (Phila Pa 1976)* 2011;36:E1641-7.
- Saladino A, Atkinson JL, Rabinstein AA. Surgical treatment of spinal dural arteriovenous fistulae: a consecutive series of 154 patients. *Neurosurgery* 2010;67:1350-7 [discussion 1357-8].
- Narvid J, Hetts SW, Larsen D, Neuhaus J, Singh TP, McSwain H, et al. Spinal dural arteriovenous fistulae: clinical features and long-term results. *Neurosurgery*. 2008;62:159-67.
- Donghai W, Ning Y, Peng Z, Shuo X, Xueen L, Peng Z, et al. The diagnosis of spinal dural arteriovenous fistulas. *Spine*. 2013;38: E546-53.
- J. Marc C. van Dijk; Karel G. TerBrugge; Robert A. Willinsky; Richard I. Farb; M. Christopher Wallace. Multidisciplinary Management of Spinal Dural Arteriovenous Fistulas Clinical Presentation and Long-Term Follow-Up in 49 Patients. 2002 American Heart Association, Inc.
- Yamaguchi S, Eguchi K, Kiura Y, et al. Multi-detector-row CT angiography as a preoperative evaluation for spinal arteriovenous fistulae. *Neurosurg Rev* 2007;30:321-26, discussion 327.
- Krings T, Lasjaunias PL, Hans FJ, et al. Imaging in spinal vascular disease. *Neuroimaging Clin N Am* 2007;17:57-72.
- Chen CJ, Chen CM, Lin TK. Enhanced cervical MRI in identifying intracranial dural arteriovenous fistulae with spinal perimedullary venous drainage. *Neuroradiology* 1998;40:393-97.
- Terwey B, Becker H, Thron AK, et al. Gadolinium-DTPA enhanced MR imaging of spinal dural arteriovenous fistulas. *J Comput Assist Tomogr* 1989;13:30-37.
- Efrat Saraf-Lavi, Brian C. Bowen, Robert M. Quencer, Evelyn M.L. Sklar, Alan Holz, Steve Falcone, Richard E. Latchaw, Robert Duncan, and Ajay Wakhloo. Detection of Spinal Dural Arteriovenous Fistulae with MR Imaging and Contrast-Enhanced MR Angiography: Sensitivity, Specificity, and Prediction of Vertebral Level. *AJNR Am J Neuroradiol* 23:858-867, May 2002.
- Morris JM. Imaging of dural arteriovenous fistula. *Radiol Clin North Am* 2012;50:823-39.
- Muralidharan R, Saladino A, Lanzino G, et al. The clinical and radiological presentation of spinal dural arteriovenous fistula. *Spine (Phila Pa 1976)* 2011;36:E1641-7.
- Marcus J, Schwarz J, Singh IP, et al. Spinal dural arteriovenous fistulas: a review. *Curr Atheroscler Rep* 2013;15:335.
- Hacein-Bey L, Konostas AA, Pile-Spellman J. Natural history, current concepts, classification, factors impacting endovascular therapy, and pathophysiology of cerebral and spinal dural arteriovenous fistulas. *Clin Neurol Neurosurg* 2014;121:64-75.
- Patsalides A, Santillan A, Knopman J, et al. Endovascular management of spinal dural arteriovenous fistulas. *J Neurointerv Surg* 2011;3:80-4.
- Chibbaro S, Gory B, Marsella M, et al. Surgical management of spinal dural arteriovenous fistulas. *J Clin Neurosci* 2015;22:180-3.
- Antoine Nachanakian, Antonios El Helou, Ghassan Abou Chedid and Moussa Alaywan. Spinal Dural Arteriovenous Fistulas — Treatable Cause of Myelopathy. 2013, <http://dx.doi.org/10.5772/56725>.
- Schick U and Hassler W. Treatment and outcome of spinal dural arteriovenous fistulas. *Eur Spine J*. 2003; 12(4): 350-355.
- Song JK, Vinuela F, Gobin YP, Duckwiler GR, Murayama Y, Kureshi I, Frazee JG, Martin NA. Surgical and endovascular treatment of spinal dural arteriovenous fistulas: long-term disability assessment and prognostic factors. *J Neurosurg*. 2001; 94(2 Suppl): 199-204.
- Ropper AE, Gross BA, Du R. Surgical Treatment of Type I Spinal Dural Arteriovenous Fistulas. *Neurosurg Focus* 2012; 32(5): e3
- Ortega-Suero G, Etesam JP, Gamazo MM, Rodríguez-Boto G. Fístulas arteriovenosas espinales del adulto. Manejo de una serie de casos desde una planta de Neurología. *Neurología*. 2018. <https://doi.org/10.1016/j.nrl.2016.12.001>
- Hall WA, Oldfield EH, Doppman JL. Recanalization of spinal arteriovenous malformations following embolization. *J Neurosurg*. 1989;70: 714-720.
- Morgan MK, Marsh WR. Management of spinal dural arteriovenous malformations. *J Neurosurg*. 1989;70:832-836.

29. Song JK, Gobin YP, Duckwiler GR, Murayama Y, Frazee JG, Martin NA, Vinuela F. N-butyl 2-cyanoacrylate embolization of spinal dural arteriovenous fistulae. *AJNR Am J Neuroradiol.* 2001;22:40–47.
30. Anson JA, Spetzler RF. Spinal dural arteriovenous malformations. In: Awad IA, Barrow DL, eds. *Dural Arteriovenous Malformations.* ParkRidge, Ill: American Association of Neurological Surgeons; 1993:175–191.
31. Van Dijk JM, TerBrugge KG, Willinsky RA, et al. Multidisciplinary management of spinal dural arteriovenous fistulas: clinical presentation and longterm follow-up in 49 patients. *Stroke* 2002;33:1578–83
32. Niimi Y, Berenstein A, Setton A, et al. Embolization of spinal dural arteriovenous fistulae: results and follow-up. *Neurosurgery* 1997;40:675– 82, discussion 682–83
33. Steinmetz MP, Chow MM, Krishnaney AA, et al. Outcome after the treatment of spinal dural arteriovenous fistulae: a contemporary single-institution series and meta-analysis. *Neurosurgery* 2004;55:77– 87, discussion 87–88
34. Jakub Wojciechowski, Przemysław Kunert *, Arkadiusz Nowak, Tomasz Dziedzic, Tomasz Czernicki, Katarzyna Wójtowicz, Kamil Leśniewski, Andrzej Marchel. Surgical treatment for spinal dural arteriovenous fistulas: Outcome, complications and prognostic factors. *Neurologia neurochirurgia polska* (2017) 446-453. <http://dx.doi.org/10.1016/j.pjnns.2017.07.001> 0028-3843/.