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Factors Influencing the Prognosis in Intracranial Dural Arteriovenous Fistulas with Perimedullary Drainage

Abad Cherif El Asri, Brahim El Mostarchid, Ali Akhaddar, Okacha Naama, Miloudi Gazzaz, Mohamed Boucetta

Key words
- Angiography
- Brainstem
- Dural arteriovenous fistula with myelopathy
- Embolization
- MRI
- Surgery

Abbreviations and Acronyms
DAVF: Dural arteriovenous fistula
IDAVFPD: Intracranial dural arteriovenous fistulas with perimedullary venous drainage
MRI: Magnetic resonance imaging

Department of Neurosurgery, Military Hospital, Rabat, Morocco

To whom correspondence should be addressed: Abad Cherif El Asri, M.D. [E-mail: abad20031@hotmail.com]

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OBJECTIVE: **Intracranial dural arteriovenous fistulas with perimedullary venous drainage (IDAVFPD) are classified as type V dural arteriovenous fistulas. Publications are limited to single case reports and small case series. We conducted a systematic review of the literature for patients with IDAVFPD. The aim of this study is to identify the predictive factors of poor prognosis in patients with IDAVFPD.**

METHODS: **We present the case of a 48-year-old man who underwent surgical interruption of IDAVFPD. A complete MEDLINE search was then undertaken for all articles reporting outcomes data for IDAVFPD. According to the results we have divided the patient population into two groups: I, those patients who showed improvement after treatment, and II: those patients who did not show improvement. We conducted a comparative statistical analysis of the epidemiologic, clinical, radiologic, and therapeutic parameters between the two groups.**

RESULTS: **A total of 37 articles comprising with 58 cases were included for analysis with an average follow-up of 12 months. There were 36 patients in group I and 22 in group II. The average age was 57.8 years in group I and 54.3 years in group II ($P = 0.32$). Onset of symptoms was acute or subacute in 57% of patients in group I, and in 50% of patients in group II ($P = 0.62$). Bulbar signs were present in 28% of cases in group I and in 36% of cases in group II ($P = 0.49$). Hyperintensity of the brainstem on T_2-weighted sequence magnetic resonance imaging was more common in patients in group II (78%) compared with patients in group I (45%) ($P = 0.012$). Patients who underwent surgical procedure have shown good outcomes compared to patients treated with endovascular approach ($P = 0.039$).**

CONCLUSIONS: **The poor outcomes were correlated to the presence of brainstem signal abnormalities on magnetic resonance imaging, whereas the prognosis does not depend on age, sex, clinical presentation, or anatomic characteristics of the fistula.**

INTRODUCTION

A dural arteriovenous fistula (DAVF) is characterized by abnormal shunting of blood between the arterial system (meningeal branches and rarely cortical branches of the external and internal carotid artery, and vertebral arteries) and the venous system (dural sinus or close structures), without the presence of a normal intervening capillary bed (18). The venous drainage of intracranial DAVFs seems to be a dominant point for explaining most signs and symptoms. That is why some investigators have classified DAVFs according to their venous drainage (5). Among the different patterns of venous drainage, the Vth type corresponds to drainage into spinal perimedullary veins (7, 9). A significant delay in making the diagnosis of this type of DAVF has been observed, because of the clinical presentation presents as a slowly progressive ascending myelopathy. This is probably attributed to its rarity and unfamiliarity to most clinicians (7, 16). We conducted a systematic review of the literature with statistical analysis of outcomes parameters in patients with IDAVFPD to define prognosis factors and the adequate treatment options.

METHODS

A literature search of the electronic PubMed databases up to 1 July 2011 was conducted, using subject headings (MeSH) and keywords, and limited to human studies. The terms intracranial dural arteriovenous fistula and myelopathy, and combinations of the variables myelopathy, tetraparesis, intracranial, craniocervical, arteriovenous dural fistula, perimedullary venous drainage, and spinal medullary veins were used.

To identify additional eligible studies, the reference lists were also screened for articles. An analysis of the published studies concerning clinical and radiologic characteristics, treatment, and outcomes was performed. Articles written in a language other than English were excluded, as were abstracts, editorials, letters, or comments. All articles were analyzed, and we exclude those in which radiologic finding, treatment, or outcomes were not reported by investigators. Radiologic data of each reported case were reviewed and analyzed.

The data extraction included the following parameters: 1) year of publication and author; 2) number of reported patients; 3) age and gender; 4) onset of symptoms and signs on admission; 5) radiologic finding (myelography and/or magnetic resonance imaging [MRI]); 6) angiographic finding (feeding arteries and venous drainage); 7) modalities of treatment; and 8) outcomes.

The aim of our study was to identify the predictive factors of poor prognosis in patients with intracranial arteriovenous fistula with perimedullary venous drainage. According to the results reported by the investigators we divided the patient population into two groups:

- Group I: patients who showed improvement after treatment of the fistula (described by investigators as full recovery, subtotal recovery with slight disability, or moderate disability).
- Group II: patients who did not show improvement and who showed slight improvement after treatment (described by investigators as severe disability, no improvement, no change, or death).

We conducted a comparative statistical analysis of the epidemiologic, clinical, radiologic, and therapeutic parameters between the two groups. Qualitative parameters were compared using the χ^2 test, and quantitative parameters were compared using independent-samples t test. All statistical results were considered as significant if $P < 0.05$. The analysis was performed with the SPSS 11.5 package software (SPSS Inc., Chicago, Illinois, USA).

RESULTS

The MEDLINE search resulted in 58 cases of intracranial arteriovenous fistula with perimedullary venous drainage. The data of all 58 patients, including those of the current case, are summarized in **Table 1**. There were 36 patients in group I and 22 in group II.

Illustrative Case

A 48-year-old man presented with a rapidly progressive quadriparesis developing over 10 days. There was no history of recent trauma. Neurologic examination revealed severe quadriparesis, reduced sensation to pin prick below the neck, and hyperreflexia of the upper and lower limbs. He developed sudden onset of breathing difficulty, swallowing soft palate, dysphonia, and paralysis of the trapezius muscle. Cervical MRI performed for suspected cervical myelopathy showed swelling of the cervical spinal cord and medulla oblongata with high signal intensity on T_2-weighted imaging with serpiginous flow voids, suggestive of a vascular malformation (**Figure 1A** and **B**). The patient underwent spinal angiography, which failed to demonstrate an arteriovenous malformation. Cranial angiography demonstrated a left tentorial DAVF fed by tentorial artery or Bernasconi and Cassinari, with drainage into perimedullary veins throughout cerebellar veins (**Figure 2A** and **B**). The patient underwent an attempt at embolization with cyanoacryl glue; however, because of the anatomy of the carotid siphon, the microcatheter could not be safely positioned into the feeding artery. Surgery was performed by a left temporopterional approach. The temporal lobe was elevated to allow for inspection of the lateral wall of the cavernous sinus and the tentorium presented small dilated vessels on its subtentorial surface. The affected dura and the vein (draining proximally as it exited the arteriovenous shunt) were meticulously coagulated using bipolar electrocautery. The postoperative course was uneventful. Cervical MRI, obtained 2 months after surgery, showed resolution of the enlargement of the spinal cord and a decrease of high signal intensities in the cervicomedullary region. The left carotid angiograms showed obliteration of the fistula site and disappearance of spinal perimedullary venous drainage (**Figure 2C**). Despite closure of the fistula and normalization of MRI findings, the clinical condition of the patient did not significantly improve; he remained wheelchair-bound and dependent for daily activities.

Literature Review

Age and Sex. Age distribution of 58 patients with IDAVFPD is shown in **Table 2**. The mean age was 57.8 years for the patients in group I and was 54.3 years for those in group II. No significant difference of age was found between both groups ($P = 0.32$). The male-to-female ratio was 2.6 (26:10) in group I and 2.66 (16:6) in group II ($P = 0.96$). No significant difference of sex was found between both groups.

Clinical Presentation. The characteristics and clinical presentations of patients in each group are shown in **Table 2**. The average delay until onset of symptoms was 220 days in group I and 343 days in group II, without significant difference between both groups ($P = 0.18$). Mode of installation was acute or subacute in 57% of patients in group I, and in 50% of patients in group II ($P = 0.62$). Fifty-six patients presented signs of myelopathy. Tetraparesis was observed in 55% of patients in group I and in 73% of patients in group II ($P = 0.28$). Bulbar signs were present in 28% of cases in group I and in 36% of cases in group II ($P = 0.49$). Other clinical signs (nausea and vomiting, headache, unsteady gait, and speech disorder) were observed in two patients in group I.

Radiologic Finding. Craniospinal MRI was performed in 51 of the reported cases and in 7 cases the myelography and/or myeloscan was the only performed imaging (**Table 2**). MRI demonstrated signs of myelopathy as a hyperintensity of the cervical cord on T_2-weighted sequences, in 96% of patients except for the two patients who did not have clinical symptoms of myelopathy. Hyperintensity on T_2-weighted sequence of the brainstem (medulla oblonga) was significantly more common in group II patients (78%) compared with patients in group I (45%) ($P = 0.012$).

The feeding arteries arise from the internal and/or external carotid artery in 38 patients (22 in group I and 17 in group II) and from the vertebrobasilar system in 11 cases (**Table 3**). The dural fistula was irrigated by both branches of the carotid and vertebrobasilar arteries in seven patients. There was no significant difference between both groups ($P = 0.31$).

The venous drainage was direct in the perimedullary veins in 53% of cases in group I and in 45% of cases in group II. The drainage was in the perimedullary veins throughout a venous sinus in 47% of patients in group I and in 55% of patients in group II. There was no significant difference between both groups ($P = 0.40$).

Treatment Modalities. Surgical treatment was performed in 26 cases, embolization in 24 patients and in 7 cases, a combined open neurosurgical and endovascular approach was required to cure the dural fistula (**Table 3**). In those patients where the endovascular approach failed to exclude the fistula, then a surgical approach was undertaken. As a