

Research article

The recovery management of patients with operated extramedullary spinal arteriovenous fistula, evolution and socio-professional reintegration: case report and review of the literature

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ABSTRACT: Adequate therapeutic intervention performed in the case of extradural spinal AVM and an intensive recovery program allow the amelioration of neurological manifestations in a very high percentage. With the ultimate goal of practicing a trade, a good biological recovery is needed for social recovery. The effectiveness of strategies for professional integration and reintegration depends largely on the patient's experiences before the onset of the disease. The paper aims to review the treatment, the evolution of patients with extradural spinal AV and the possibilities of socio-professional reintegration. **Methods and analysis.** A case presentation of a patient diagnosed with extradural spinal AV fistula is proposed, along with a review of the current literature on the treatment of this pathology, the evolution and the possibilities of vocational recovery. The studies will be analysed and selected in two stages, in the first stage the titles and abstracts, in the second stage, the articles with full text will be analysed, selected and a narrative synthesis of the included studies will be made. **Summary case.** The 51-year-old urban patient, a professional driving instructor who underwent endovascular and surgical treatment for extradural spinal AV fistula, is hospitalized for a moderate motor deficit, such as paraparesis, back pain, mechanical pain in the knees and gait disorders. The objectives of recovery are represented by neuromotor recovery and socio-professional reintegration. **Conclusions.** Spinal EAVFs are rare lesions with a low risk of bleeding; the clinical manifestations are determined by the compression of the bone marrow; these being significantly improved after the endovascular and surgical treatment. Studies show a good long-term prognosis, which is determined by the absence of recurrences. An essential role in the integration of patients with disabilities in the socio-professional life is the identification of their deficiencies and their reorientation according to the outstanding abilities, the stimulation of the preserved skills.

Keywords: *rehabilitation, disc hernia, low back pain, paraplegia*

1. INTRODUCTION

Spinal cord irrigation consists of four arteries, with a path parallel to the axis of the spinal cord, 2 anterior and 2 posterior spinal arteries (1). Their starting point is the vertebral artery. The root arteries leave the spinal arteries, with a metameric distribution. Between the two vascular systems, anterior and posterior, there are numerous anastomoses. The

veins are also represented by two vascular systems, the intramedullary system and the perimedullary system. They drain into the anterior and posterior internal vertebral venous plexus from the epidural space. Venous plexuses also have numerous anastomoses.

Spinal cord malformations are a fairly rare cause of spinal cord compression. There are no valves in the venous plexuses (internal and external), so the blood flows back into the plexuses if the pressure in the jugular veins increases. Spinal arteriovenous malformations (AVMs) have a low incidence of about 4% of all tumours at this level (2, 3). They are the prerogative of adulthood. Over time, several MAV classifications have been made (4). These classifications are based on the location of the condition, the imaging techniques used, the treatment methods. From 1967 to 2015, seven classifications were reported. Spinal vascular malformations (MVS) are classified according to location (intradural, perimedullary, radicular, extradural) and type of flow (high flow, low flow, without arteriovenous shunt) (5). An essential aspect in the classification is the differentiation of shunt lesions from non-shunting lesions (spinal cord cavernous). In shunt lesions, the classification specifies the supply artery, the type of transition between the artery and the vein (plexiform or nidus type) as well as the large or small volume of the fistula (2). This classification is important for the therapeutic approach to AVM (6). High-flow MVS are generally congenital lesions diagnosed in children and young patients without gender predominance. They have hemodynamic disorders, mass effects or bleeding, but can also be discovered by chance. Low-flow SVMs tend to be acquired lesions that occur in older men with progressive myelopathy caused by spinal venous hypertension. They are rarely associated with vascular syndromes but may accompany prothrombotic conditions. Extradural arteriovenous fistulas (EAVFs) are the direct connection between an extradural artery and a vein, resulting in a high-flow fistula that communicates with the epidural venous system. Myelopathy is caused by direct compression or retrograde venous hypertension (7). Dural fistulas are the most common, accounting for 70% of total AVM. According to literature, only 15% are symptomatic. The most common location was the thoracic spine (61%), followed by the cervix (22.7%), lumbar (14.5%) and sacral (1.8%). Spinal extradural AVFs are much rarer than other types of spinal AVM.

The clinical picture may be, in the early stages non-specific (8), dominated by up to 85% of cases of low back pain (51.8%), in evolution with sensitivity disorders and paresis of the lower limbs (75.5%), with slow evolution (months - years), paraesthesia's (60%), intestinal / bladder dysfunction (41.8%) and myelopathy (36.4%) (9). Between 10 and 20% of cases may present with sudden onset myelopathy, especially in patients under 30 years of age, due to bleeding malformation, bleeding that may occur, depending on the location of the malformation, hematomyelia, subarachnoid haemorrhage or extradural spinal hematoma. It describes a particular syndrome, called subacute necrotic myelopathy (Foix-Alajouanine syndrome), characterized by paraplegia with anaesthesia below the level of the lesion and loss of sphincter control. Imaging, the medullary ischemia caused by spontaneous thrombosis of the malformation is detected (in cases with fulminant, irreversible evolution). Table 1 shows the classification of AVM, incidence, type of shunt and symptoms.

From an imaging point of view, the following are used: dynamic magnetic resonance angiography and three-dimensional angiography with computed tomography (CTA), necessary to establish the method of treatment is the method of choice; MRI can detect certain types of spinal vascular malformations with results close to angiography, in the case of high-flow sounds, but with unclear sensitivity in the case of low-flow sounds (6); myelography, can highlight the vascular trajectory (5).

Table 1. Classification of arterial-venous malformations

Type of malformation/ Incidence/ Age	Characteristics / Localization	Symptomatology
Type I: Dural arteriovenous malformations (Dural fistulas) /70-80%/elderly	Fed by a root arteriole which forms an arteriovenous shunt at the level of the spinal root located in the intervertebral foramen and which subsequently drains into a dilated vein in the posterior part of the spinal cord (10)/ Thoracolumbar	Lumbar pain and progressive myelodradiculopathy by affecting the nerve root at the level of the foramen and the spinal cord below the level of vascular malformation by medullary venous congestion secondary to high venous pressure existing in the drainage vein.
Subdural malformations		
Type II: intramedullary arteriovenous malformations/ 15%/any age	Consisting of a compact vascular nest fed by medullary arteries and may associate as in the case of cerebral arteriovenous malformations aneurysms located at the level of the medullary arteries/ Cervical-thoracic-lumbosacral	Low back pain, tenderness, and paresis of the lower limbs
Type III: juvenile spinal arteriovenous malformations/-/any age	Consisting of a voluminous vascular glomus that encloses the spinal cord and adjacent vertebral bodies thus producing / Cervical-lumbosacral, Predominantly thoracic (11)	low back pain, tenderness, and paresis of the lower limbs
Type IV: subdural, extramedullary/-/any age	These are direct arterio-venous fistulas between a perimedullary artery (most commonly Adamkiewicz' s artery) and a drainage vein; they appear at a younger age compared to type I / Thoracic spine (61%), cervix (22.7%), lumbar (14.5%) and sacral (1.8%)	They can be deduced by massive haemorrhages in the spinal subarachnoid space, causing paresis/plegia (75.5%), paraesthesia's (60%), pain (51.8%), bowel/bladder dysfunction (41.8%) and myelopathy (36.4%) (9).

Catheter angiography remains the gold standard for assessing spinal vascularity and disorders. The application of the 4D-CTA method in spinal cord injuries is limited, it is the third choice for non-invasive angiography, after dynamic ARM and three-dimensional CTA, highlighting the accuracy of the diagnosis in the Dural spinal fissures (5, 12).

Therapeutic management consists of a surgical or endovascular approach. Diagnostic angiography may be associated with embolization in the same session (13). Endovascular treatment is primarily intended for Dural fistulas. Type II may benefit from endovascular and surgical treatment. Type IV benefits from multimodal treatment (embolization and then surgical treatment), while type III is outside the current therapeutic resources (Table 2).

Table 2. Treatment indicated according to the type of spinal arteriovenous malformation

Type MAV	Surgical Treatment	Endovascular Treatment
Type I	Second intention	First intention
Type II	First/Second intention	First/Second intention
Type III	After embolization	First intention, association with surgical treatment
in addition to therapeutic resources, multiple recurrences		
Type IV	Multimodal treatment, after embolization	First intention

Initially affected by high recurrence rates due to inadequate embolization material, endovascular techniques are nowadays a viable alternative to surgery, mainly due to the introduction of liquid embolic agents (13).

This proposed review focuses on the identification of therapeutic interventions in patients diagnosed with spinal malformations with extradural arterial-venous fistula, the evolution, and the possibilities of vocational reintegration.

Studies referring to the clinical manifestations determined by the presence of extradural AV malformations, the treatment methods approached, the evolution of patients and the possibilities of socio-professional reintegration will be eligible and included in the evaluation. To identify the possibilities of socio-professional reintegration, we identified the studies that were addressed to patients with neurological impairment, such as tetraparesis and paraparesis. No age or sex restrictions will be imposed.

We have included all research published from 1983 to December 31, 2021, that refer to the proposed objectives, not restricting the studies by any type of settings (using as well books and documents, clinical trials, meta-analysis, randomized controlled trials, reviews/systematic reviews, etc. No language restrictions were applied, but most of the studies were in English.

For data collection, the PubMed platform was accessed, being searched different literature sources (i.e., extensive papers, dissertations, abstracts, conference papers, posters, and reports, etc.).

The search terms have been modified and refined to achieve the proposed objectives: spinal arteriovenous fistula, spinal arteriovenous fistula extradural, spinal arteriovenous fistula extradural treatment, vocational reinsertion, vocational neurological reintegration (Table 3).

Table 3. Search strategy for PubMed from 1983 – December 2021

Search items	Identified no. of papers
Spinal arteriovenous fistula extradural	72
Spinal arteriovenous fistula	1,417
Spinal arteriovenous fistula extradural treatment	60
Spinal arteriovenous fistula extradural treatment, free full text	16
Spinal arteriovenous fistula extradural treatment Reviews/Systematic Reviews	13
Meta-analysis	2
Clinical trials	1
Reinsertion vocational	49
Vocational reintegration neurological	58

As references management tool, EndNote was used to insert bibliographies and references. Duplicate articles will be identified and removed from the EndNote library.

RESULTS AND DISCUSSION

A centralized analysis of PubMed databases on the incidence and clinical manifestations of patients with AVM shows that 1% of lesions were incidental; 93% of patients had neurological deficits and 36% of cases started with bleeding. Some studies show an increased incidence among male patients (14). A study performed exclusively on patients with extradural AV fistulas shows that the patient's average age was 45.9 years and that there was no significant sexual predilection. Only 3% of the lesions were incidental, while 10% occurred in patients with bleeding (15). Studies performed on a total of 321 patients with extradural spinal AV fistula who received endovascular and surgical concomitant treatment show a neurological improvement of between 89 and 97.7%. Rangel-Castilla, in a study of 110 patients with AVM, of whom 44 had extradural spinal AV fistulas, reported gaining independence in 97.7% of patients included in the study with a recurrence of 13.6% in patients with extradural (Table 4).

Table 4. Summary of study treatment and evolution

Study	Patients No.	Treatment Surgical / endovascular	Remote clinical outcomes
Huang et al (15)	101	Associated Treatment, Complete cancellation 91% of cases	Neurological improvement 89%, Stationary 9%, 2% worsening
Akgun et al (8)	78	Associated Treatment, Complete cancellation 97.4% of cases	Improvement
Vázquez (16)	3	Associated Treatment, Complete cancellation	Improvement, no recurrences
Singh (11)	74	52.7% embolization, 21.6% surgery, Associated 5.4%	Neurological improvement in 26 patients, 25 stabilized, the rest transferred.
Clarke (17)	6	Associated Treatment	Neurological improvement
Zhang et al (18)	1	Associated Treatment	Neurological improvement
Takai (19)	14	Combined treatment, regardless of location, customized surgical technique depending on venous drainage	Improvement, no recurrences
Rangel-Castilla (9)	44/110	42% embolization Resection 86.4% 12.7% embolization only	97.7% independent for those with extradural, 86.4% other MAVs; No deaths Recurrence 13.6% in patients with extradural, 15.2% in MAV

Several strategies have been implemented in Europe, classified into "Policies", "Systems" and "Services", which aim to integrate and reintegrate people with chronic illnesses into work. The policies consist of national strategies spanning 5 or 10 years. The systems are represented by pension programs, incentives, allowances. The services address the specific needs of people with a certain deficit. The unemployment rate for people with disabilities is high and incomes are lower compared to people without chronic illnesses (20). Work has a positive impact on health and well-being (20).

Any recovery process aims at obtaining biological recovery, along with the social one. adaptation of the person to the workplace, ensuring professional security (21). According to the official data of the National Authority for the Rights of Persons with Disabilities, Children and Adoptions, as of June 30, 2020, the total number of persons with disabilities was 853,465, representing 3.85% of the Romanian population (21).

Handicap means the automatic limitation of the person to social life. In recent years, special emphasis has been placed on the rehabilitation of people with disabilities. They are often considered maladapted and stigmatized. The socio-professional reintegration of these people is a general problem, the unemployment among them is double compared to the general population, active (22). The European Strategy on Social Inclusion presents a series of measures and proposals for the preservation and identification of jobs for people with disabilities. The incomes of these people are lower than the general population. Until the * 80s, the medical model was approved, which ensured a minimum financial income, following the replacement of this model with the social one.

Chronic neurological pathologies cause a series of functional deficits with repercussions in the fulfillment of ADLs and the profession, especially since the onset is at a younger age. Therapy of any kind is of utmost importance (22, 23). The psycho-social consequences of the neurological patient (24), paraplegic or tetraplegic, are dependent on the level of injury, the patient's profession, family support, age and social environment at the onset of disability (23, 25). In 1997, Germany had 48 institutions dedicated to

the vocational rehabilitation of people with physical disabilities, sensory disabilities, and developmental disabilities.

Of the 651 paraplegic patients who went to socio-professional rehabilitation centres, 80% are men, 45% have returned to their previous activity or started another paid activity (23). Socio-professional reintegration is becoming easier with the development of technology. Requests to vocational reorientation centres are lower for people over 30, from married people. 86% of applicants are unmarried (table 5).

Table 5. Results of studies on vocational reorientation of paraplegic patients.

The study published by P. Calmels (27), performed on 58 patients with SCI (50 men), with a mean age of 41.38 ± 13.55 years, the average score of the Barthel index being on average 73.88 ± 21.87 , shows that the return to a professional activity is correlated with age, degree of independence, Barthel Index, ASIA score, level of training, family support.

Of particular importance are the occupational therapy and vocational therapy workshops, the presence of a mechanism to finance the (re) integration of people with disabilities (28). It is also appropriate to raise awareness of work-related issues among people with disabilities, to increase the motivation and educational level of these people.

Driving does not appear to be important in some studies (29), but some authors show the importance of driving to increase the quality of life (27).

SUMMARY CASE

Material and method. Patient, 51 years old, from the urban environment, professional driving instructor, diagnosed with paraparesis after extramedullary arteriovenous spinal fistula D5 operated, neurological lesion ASIA C, motor level D12, neurogenic bladder, neurogenic colon, hospitalized for the moderate motor deficit, paraparesis type, back pain, mechanical pain in the knees, gait disorders.

Informed consent statement: informed consent was obtained from the patient for the publication of this case report.

Disease history. The current disease started suddenly, two years ago, with paraesthesia's in the lower limbs and sphincter disorders. In the next three days, the functional deficit appears in the upper and lower limbs, the patient being completely immobilized. He was referred to the neurosurgery clinic, where, at the neurological examination, spastic paraparesis 2/5 gr, hypoesthesia under the bilateral D5 dermatome, thermal anaesthesia and fine proprioceptive below the level of the right D6 dermatome, ROT live lower limbs, without clonus, Babinski „+“ bilateral. On MRI examination of the cervical spine, dorsal congestive myelopathy was described, extensive vascular dilatations subdural extramedullary at the level of the vertebral body D5, which extends to the lumbar level, with the character of subdural spinal fistula. To elucidate the diagnosis, spinal angiography was performed, which revealed at the D5 level an arterio-venous fistula with extended extramedullary subdural venous drainage. Following the interdisciplinary clinical evaluation, the MRI and angiographic examination decided the surgery, which consisted of bilateral laminectomy D5-D6, durotomy, coagulation of the arterial feeder of the fistula and resection of the fistulous area. Control MRI examination revealed a reduction in the size of the venous dilatations, with no other signs of spinal cord compression. Control spinal angiography performed by injection of the D5 artery no longer showed the venous dilatations present in the anterior arteriography.

Clinical examination at discharge reveals paraparesis grade 2/5 predominantly left, improving sensitivity under dermatome D6. At discharge, neuromotor recovery, analgesic treatment, change of bladder catheter at 14 days are recommended.

The patient followed a sustained recovery program, 3 courses/year, in specialized centres and physical therapy at home, 2-3 sessions/week. He is currently addressing the outpatient recovery office for: motor deficit in the lower limbs, sphincter disorders. The objective examination reveals: wheelchair ambulance, possible walking with two brachial crutches over short distances; muscular hypotrophy in the gluteal muscles and lower limb muscle groups, osteotendinous hyperreflexia MI, lower limb vascular-trophic

disorders, pain in the mobilization of the knees with instability in orthostatic and gait, urination by catheterization, neurogenic colon.

For the evaluation of spasticity, the Ashworth scale and the modified Ashworth scale (MAS) have been used, with values between 0 and 4 (0 = without accentuating the tone, 1 = slight accentuation of the tone, manifested by grip and release, or by minimal resistance to flexion and extension, (1 +) = slight increase in grip, followed by minimal resistance in the range of motion, 2 = increase in muscle tone in the range of motion, but the affected limb is easily mobilized, 3 = considerable increase in muscle tone, passive movements are difficult, 4 = the affected joint is rigid in flexion and extension) (30).

The neuro-sensitive evaluation was performed, with the ASIA score which is considered as being the gold standard (31, 32). The motor score was achieved by testing the muscles with functional importance on each myotome, right-left, on a scale from 0-5; the figures add up to the overall engine score. This score is clinically essential, it is objective, allowing the clinician to assess the patient's progress between two consecutive evaluations, with an important role in the prognosis. The motor level is defined by "key muscle" (C5 – Biceps, C6 – Long and short radial extensor of the carpus, C7 – Brachial triceps, C8 – Deep flexor of the fingers, D1 – Abduction of the finger V, L2 – Iliopsoas L3 – Quadriceps L4 – Anterior tibial, L5 – Long extensor of the toe + extension of the fingers, S1 – Plantar flexion of the ankle), depending on the muscle testing, it must have a value of 3, and the muscles of the overlying segment must have values of 4-5. The sensitive level is achieved by testing the painful, superficial sensitivity on each dermis right and left. As in the case of determining the motor score, in determining the sensory score key points can be used for each lesion level: occipital protuberance, supraclavicular fossa, acromioclavicular joint, antecubital fossa, police, Medius, auricle, axillary tip, intercostal spaces, xiphoid appendix, sciatic tuberosity, perianal area, distal femoral shaft, popliteal area, medial malleolus, lateral heel.

The examined patient presents both active and passive mobility normal limits on the muscle groups of the upper limbs, ADL-normal limits, normal grip, evaluation of digital dexterity normal limits. Regarding the evaluation of the lower train, the following data can be mentioned: maintain the position at the edge of the bed, unassisted transfer, possible verticalization and short distance walking with 2 brachial crutches resulting in ASIA motor-50 score for upper limbs (28 for lower limbs and sensitivity level L4). According to the Aminoff-Logue Disability Scale (ALS), the patient had a G4M3 score at the time of evaluation. The recovery objectives are the following: Improving neuropathic pain; Decreasing spasticity of the lower limbs; Improving motor deficit; Re-educating sensitivity; Improving functional status; Increasing quality of life; Prevention of urinary tract infections; Socio-professional reintegration of the patient.

The ability to walk is the target of neuromotor rehabilitation (21). The patient followed a complex recovery program, consisting of physiotherapy and hydrokinetic therapy, electrotherapy (Huffs Schmidt currents), massage. The objectives of physical therapy are combating vicious attitudes, limiting motor deficit, and increasing mobility in all joints, maintaining muscle strength in the upper train, increasing stability in orthostatism, initiating gait, improving ADL, cardio-respiratory re-education. The walking training was performed with the Lokomat robotic (21) device that uses a physiological gait model. This, through its programs, brings several benefits, namely: increased independence, improved gait and muscle tone, increased mobility, and gait (33).

Another means of recovery used to achieve the proposed objectives was the Virtual Reality Assistance Device used for upper and lower train training (34). Creating goals in the virtual world that reproduce real-life ensures the re-learning and regaining of lost physical skills and functions and increases the success rate of rehabilitation programs that use the complexity of virtual reality. The peculiarity of the case consists in myelopathy suddenly installed, at the age of 49, this mode of onset being specific to patients under 30 years. Another peculiarity is the slow recovery, with the paraparesis type

sequelae. After 2 years from the onset of the disease and intensive recovery treatment, the ASIA C classification is maintained, although studies mention neurological independence in the percentage between 89 and 97.7% (27). Also, the patient returns to the previous activity, following the adaptation of the vehicle.

CONCLUSIONS

Spinal EAVFs are rare lesions with a low risk of bleeding; the clinical manifestations are determined by the compression of the bone marrow these being significantly improved after the endovascular and surgical treatment (15). Studies show a good long-term prognosis, which is determined by the absence of recurrences (16). An essential role in the integration of patients with disabilities in the socio-professional life is the identification of their deficiencies and their reorientation according to the outstanding abilities, the stimulation of the preserved skills. This is possible with the support of competent institutions, depending on occupational requirements, health status and previous cognitive impairment (2), in close relation to the education, skills, motivation of the patient (35). Currently, the possibilities of complex recovery, together with occupational therapy, occupational therapy, psychotherapy, manage to obtain outstanding results, which lead to the reinsertion of a significant number of patients. A systematic review of the PATHWAYS literature has shown that multidisciplinary involvement, ergonomic interventions, part-time work, active labour market policies to promote employment will promote the integration into the work of patients with chronic diseases (35).

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