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Case report

Endovascular treatment of cervical intramedullary arteriovenous malformation

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ABSTRACT

Intramedullary arteriovenous malformations (AVMs) in the cervical region are a rare clinical condition. They represent a therapeutic challenge, as the lesions may cause serious functional disorders due to their location within or immediately adjacent to the critical ascending and descending sensorimotor pathways. In this case report, we present a patient with a cervical intramedullary AVM that was treated with endovascular therapy. Our experience suggests that endovascular treatment is an effective and safe method for treating AVMs located in the cervical region of the spinal cord. More studies are needed to establish appropriate treatment protocols depending on the clinical course, the anatomy of the lesion, and the region in which it is found.

1. Introduction

Spinal arteriovenous malformations (AVMs) are rare clinical entities that can lead to myelopathy, progressive neurological symptoms, and even death if not properly treated [1,2]. Symptomatic spinal AVMs are generally thought to require treatment [3].

Two AVM treatment modalities are available: surgery and endovascular therapy. Endovascular occlusion is considered to be the first-line treatment for most spinal vascular malformations [2–6], because surgical obliteration is accompanied by a high level of intraprocedural risk, especially if the lesion lies on or within the ventral portion of the spinal cord [4–7].

Intramedullary AVMs in the cervical spinal cord require special attention; they occur more rarely than AVMs in other regions of the spinal cord [8] and can be dangerous, due to their location within or immediately adjacent to critical ascending and descending sensorimotor pathways. The clinical presentation, course, and results of endovascular treatment of

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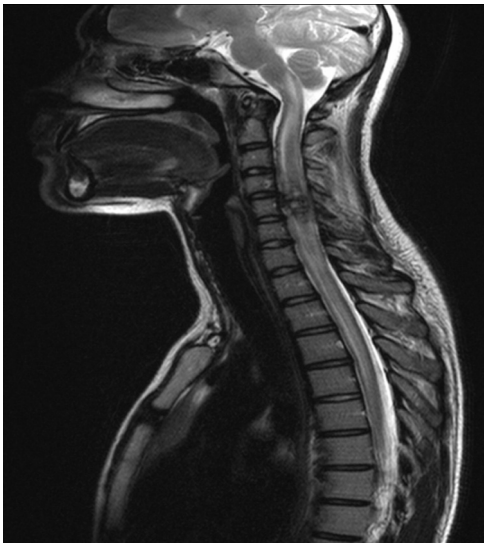

Fig. 1 – Sagittal T2-weighted MR image showing intramedullary spinal AVM at the level of C5. Extension of the spinal cord C3–C7 and edema extending to the medulla oblongata are visible.

intramedullary AVMs are rarely described in the literature. Here, we present a patient with a cervical intramedullary AVM that was treated with endovascular therapy.

2. Case report

A 17-year old girl suddenly experienced neck pain radiating to her right upper limb, weakness of the right upper limb, and right-sided paresthesia that progressed over several weeks. Similar symptoms had occurred 2 years earlier: sudden pain and paresthesia of the right upper limb that resolved spontaneously within a month. In addition, the patient had been under the care of endocrine and orthopedic specialists for several years, because of growth deficiency and curvature of the spine, respectively. A neurological examination at admission revealed right-sided hyperesthesia, muscle weakness in the right hand (4/5 strength), and neck pain upon palpation. Brain magnetic resonance imaging (MRI) was normal. MRI of the cervical spine demonstrated a centrally located, predominantly on the ventral side lesion 10 mm in diameter at the level of C5, which suggested hemorrhage or aneurysm. Anterior to the lesion, we observed a network of small vessels associated with the AVM, which were surrounded by edema. Administration of contrast agent revealed small, punctate enhancement lesions within the AVM (Fig. 1). Digital subtraction angiography was performed to define the type of vascular malformation and consequently decide upon the appropriate therapy.

Digital subtraction angiography was performed under local anesthesia. After transfemoral access and sheath placement, the patient received 3000 units of intravenous heparin. Bilateral vertebral angiography showed a glomus-type intramedullary AVM supplied by the arterial branches from the left and right vertebral arteries. Within the AVM nidus, we observed a large aneurysm that was filling from the left side (Fig. 2). The lesion was believed to carry an extremely high surgical risk given its location; therefore, we decided to perform endovascular treatment.

The first therapeutic goal was to exclude the nidal aneurysm from circulation to prevent possible bleeding. After the diagnostic angiography, a 6-F catheter was placed in the left vertebral artery. A 1.5-F flow-guide microcatheter (Magic; Balt Extrusion, Montmorency, France) was subsequently navigated over a 0.007″ microwire (Sorceror; Balt Extrusion)

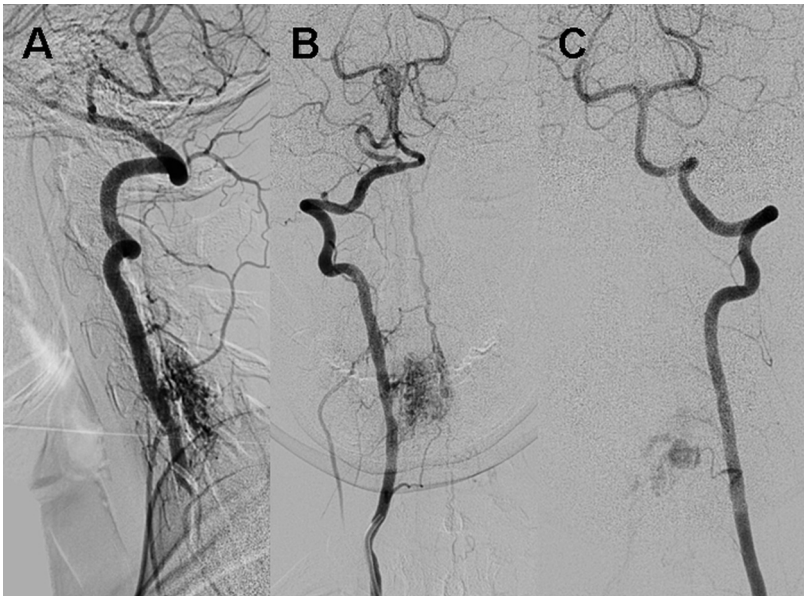

Fig. 2 – Right vertebral angiography (side and anteroposterior) showing the nidus of the glomus-type AVM supplied by the two arterial branches from the right vertebral artery (A, B). Frontal angiogram of the left vertebral artery showing aneurysm within the AVM nidus filling from the left side from one branch of the left vertebral artery (C).

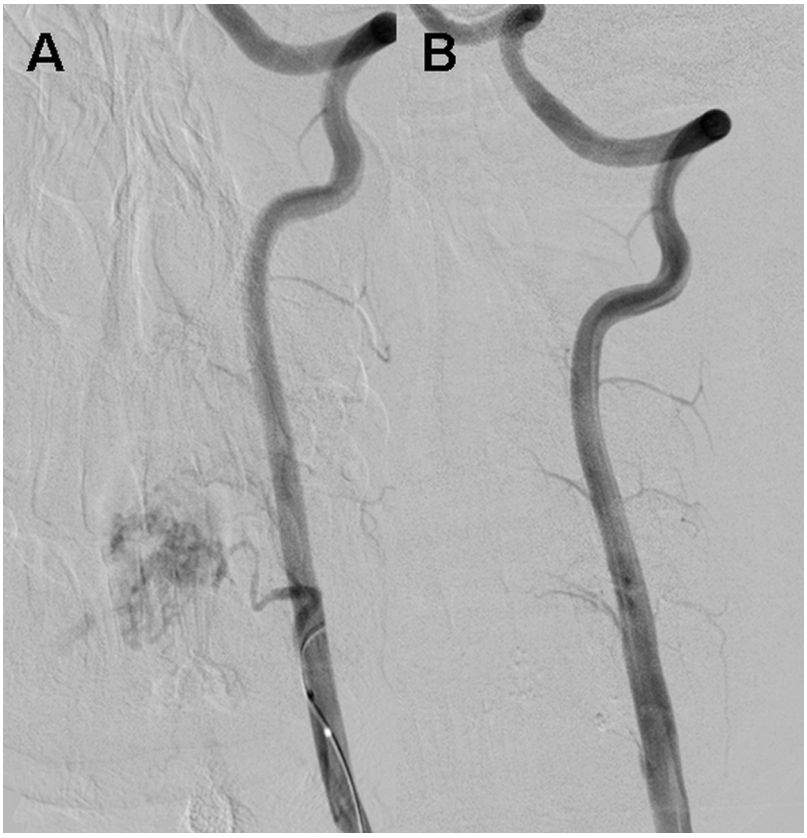

Fig. 3 – Frontal angiogram of the left vertebral artery demonstrating the position of the microcatheter prior to NBCA injection (A). Left vertebral artery angiogram following NBCA injection. Contrast material did not enter the nidus from the left side (B).

to reach the pedicle outgoing from the left vertebral artery and supplying the AVM and aneurysm. We decided to embolize the feeding arteries rather than the AVM to prevent distal migration of embolic material into the veins of the AVM and the spinal cord as much as possible. Blood flow to the AVM from the left vertebral artery was prevented with a 25% mixture of N-butyl cyanoacrylate (NBCA; B. Braun, Aesculap AG, Tuttlingen, Germany) in Lipiodol (Guerbet AG, Zurich, Switzerland) under fluoroscopic control (Fig. 3). No complications occurred. After the procedure, we observed clinical improvement; the patient's pain had disappeared, and the mobility of her right upper limb had improved. However, the patient still reported paresthesia of the right upper limb. After several days, the patient was discharged home and the next stage of the procedure was scheduled for 1 month later.

During the follow-up hospitalization, we performed right vertebral angiography, which showed a residual cervical AVM fed by two arterial branches from the right vertebral artery. Both supplying vessels were embolized with NBCA, which almost completely excluded the AVM from circulation; residual blood flow into the AVM was observed (Fig. 4). No complications occurred, and the patient's general and neurological condition was the same as before the procedure. MRI revealed that there was a reduction in the blood vessel network and less edema. We discharged the patient home and scheduled a follow-up angiography for 8 months later.

3. Discussion

Data from spinal angiography, clinical evaluations, and surgical explorations have revealed that spinal AVMs are a relatively heterogeneous group of vascular lesions, and that each type requires a different management paradigm. The most widely used classification for AVM is based on both the location and the vascular characteristics; four types of AVM are described [2,3]. In our patient, the AVM was relatively small in size (10 mm) and characterized by a compact intramedullary nidus interposed between multiple feeding arteries and their respective draining veins, with angioarchitecture similar to that of brain AVMs. These characteristics, along with the young age of the patient, are consistent with those of Type II (glomus) AVM. Rather than a slow, progressive neurological decline, glomus-type AVMs typically lead to sudden deterioration secondary to rupture of the nidal aneurysm [3].

Endovascular occlusion is considered to be the first-line treatment for most spinal vascular malformations [2–6]. The morphological target (i.e., partial or complete obliteration of the AVM), the sequence of sessions used, the therapeutic goals, and the technique used should be chosen after careful analysis of the patient's history and neurological status with correlation to the angioarchitectural features of the AVM. If complete obliteration is not possible, partial targeted embolization should be performed, with the aim of neutralizing

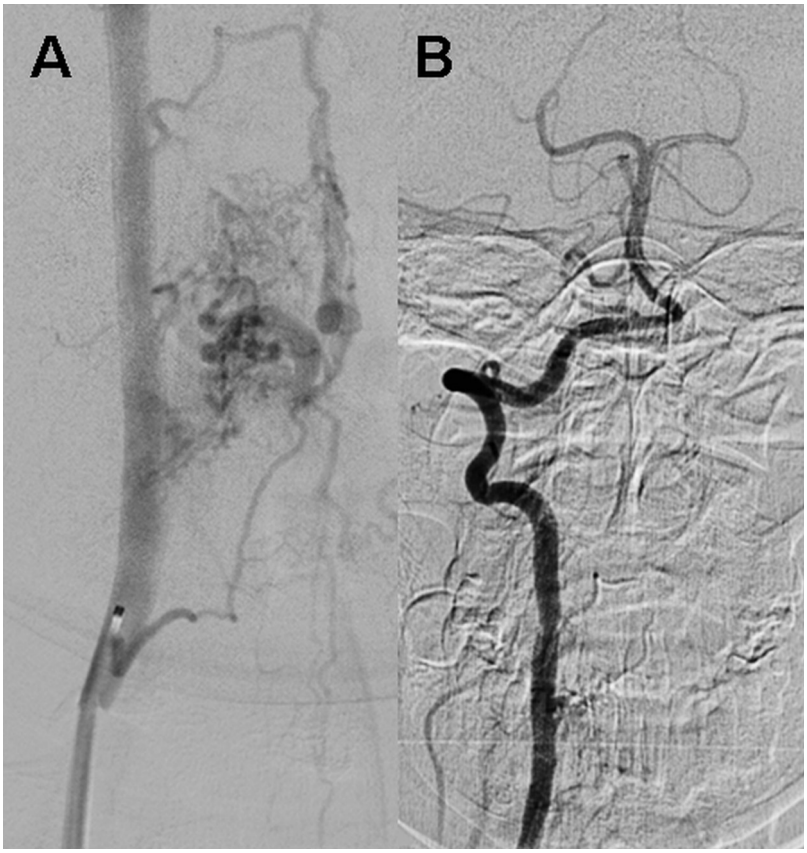

Fig. 4 – Right frontal vertebral angiogram demonstrating residual AVM fed by two arterial branches from the right vertebral artery (A). Right frontal vertebral angiogram after NBCA injection showing almost complete exclusion of the AVM from circulation (B).

dangerous anatomic structures such as aneurysms. In contrast to brain AVM, in most cases partial treatment of cervical AVM appears to be sufficient to dramatically improve the prognosis [9].

Endovascular therapy has traditionally been performed with particulate agents such as polyvinyl alcohol (PVA) and liquid adhesives such as NBCA. We used NBCA in our patient. This may be the best therapeutic option for embolization of spinal AVMs in regions where special caution is required. The use of this liquid embolic agent is clearly established in the literature and in clinical practice, and it is associated with good anatomical and clinical results. In most cases, there is improvement or stabilization of patient clinical status and protection from hemorrhagic events with good long-term effects, a low rate of operative morbidity, a reduction in the number of therapeutic sessions, and a low rate of recanalizations [7,10]. However, acrylic glue should be used only by experienced teams, particularly for spinal AVMs in dangerous regions like the cervical region, since it requires considerable skill to prevent inadvertent proximal reflux of the embolic material, excessive premature venous penetration, and gluing of the catheter within the vascular pedicle during slow injections.

There is a growing body of evidence regarding the successful use of Onyx to treat spinal AVMs endovascularly [9,11,12]. As an alternative liquid embolic agent, Onyx appears to have several advantages: it can be delivered at a significantly slower injection speed over several minutes, and there is a lower risk of premature occlusion of the venous side and a reduced possibility of gluing the catheter [9]. However, limitations remain with regard to technique and iatrogenic episodes associated with the delivery of Onyx in small perimedullary veins involved in drainage in the normal spinal cord. Although Onyx has been used many times in the treatment of brain AVMs [13], there is a need for more studies on the use of Onyx in the embolization of spinal AVMs.

Glomus AVMs in cervical regions represent a therapeutic challenge, as the lesions may cause serious functional disorders due to their location within or immediately adjacent to critical ascending and descending sensorimotor pathways. To the best of our knowledge, relatively few cases in the literature have described embolization of spinal intramedullary AVMs in the cervical region; they have mostly been reported as part of larger series of cases [9,11,12,14]. However, these studies did report that endovascular treatment of AVMs in the cervical region seems to be effective and safe (no change or transitory worsening in immediate outcome, with improvement upon clinical follow-up after a few months), few details about these patients, the procedures, and the clinical courses have been reported. More-detailed series and cases need to be described before appropriate and precise guidelines for treatment can be set forth.

In conclusion, we present a patient with a glomus-type intramedullary AVM in the cervical region of the spinal cord

that we treated endovascularly. Endovascular treatment seems to be an effective and safe method for the treatment of lesions located in this region of the spinal cord; however, this method requires considerable experience and caution. More studies are needed to help establish appropriate algorithms of treatment depending upon the clinical course, the anatomy of the lesion, and the region in which the AVM occurs.

Conflict of interest

None declared.

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Ethics

The work described in this article has been carried out in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki) for experiments involving humans; Uniform Requirements for manuscripts submitted to Biomedical journals.

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