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CASE REPORT

Application of Continuous Intraoperative Neuromonitoring (cIONM) in the Endovascular Treatment of Pediatric Spinal Arteriovenous Malformation - A Case Report

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Abstract

Spinal arteriovenous malformations (AVM) are rare vascular occurrences that, if left untreated, can result in devastating neurologic sequelae. Prevalence, especially in the pediatric population, is low but requires prompt diagnosis and management to avoid neurologic impairment. Embolization has been advocated as the treatment of choice for spinal AVMs. We present a rare case of a spinal AVM in an 11-year-old male managed with endovascular embolization during continuous intraoperative monitoring to ensure good functional outcome.

Introduction

Spinal arteriovenous malformations (AVM), accounting for 2-4% of intraspinal lesions and are of even rarer occurrences in children. Different angiographic and clinical classifications exist to determine appropriate choice of treatment. Referring to the classification proposed by Anson and Spetzler in 1992, type I to IV lesions has been established to further classify anatomic presentation. If left untreated, they can result in chronic myelopathy and permanent neurologic deficits. However, treatment attempts can possibly result in devastating functional outcome given the vulnerable vascular anatomy.

In the present report we describe the rare case of a complex Type II intramedullary arteriovenous malformation successfully managed endovascularly and discuss the role of continuous neurophysiological monitoring (IONM) in monitoring the integrity and function of the motor pathway and preventing new onset of neurological deficits.

Case Description

An 11-year-old male initially presented with pain and redness of his right arm after playing sport. The symptoms also included complete paresis and hypoesthesia of the right leg. The symptoms were self-limiting and ceased after a few hours. An MRI of the spine was performed to identify the cause of the symptoms. MRI scans revealed numerous flow voids along the surface of the cord, predominantly on the dorsal surface, as well as hematoma and swelling. There was increased signal and expansion of the lower cervical cord, due to venous congestion, as observed in the T2-weighted images. Additionally, the post-contrast T1-weighted images showed enhancement within these prominent flow voids. These findings extend over approximately 6 cm, ranging from the intraspinal space from mid C3 vertebra to the C7 vertebra.

Digital subtraction angiography (DSA) was performed which confirmed the presence of a complex arteriovenous malformation arising from the V3 segment of the right posterior spinal artery and a caudal inflow originating from the right thyrocervical trunk, measuring 7 mm horizontally and 15 mm vertically.

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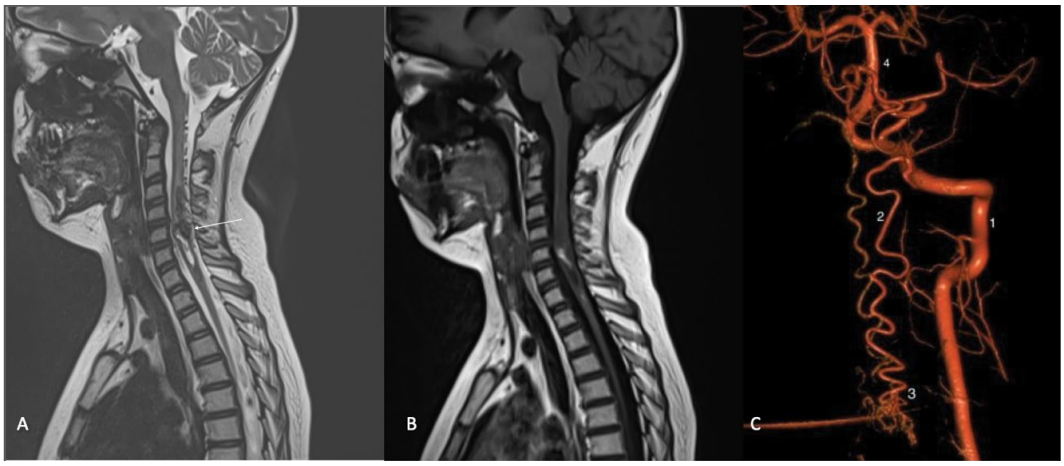

Figure 1: Preoperative MRI scan of the cervical and thoracic spine. (A) T2-weighted sagittal image shows hyper-intense cord signal as well as cord edema on level C4-C6 (white arrow); (B) Corresponding T1 weighted sagittal image; (C) DSA showing the central nidus of the AVM (3) with feeding vessel (2) originating from V3 segment of the right vertebral artery (1).

The main nidus of the AVM was located at the C5 level. While the primary venous drainage of the AVM occurred through the superior bulb of the internal jugular vein, a minor portion drained via epidural veins (Figure 1).

Given the complex anatomy of the AVM the embolization was performed under general anesthesia in two separate sessions with continuous intraoperative neuromonitoring. Transcranial electrical stimulation of the motor cortex and cortical spinal tract (CTS) was used. Stimuli were delivered through subdermal needle electrodes placed from C1 to C4. Subdermal needle electrodes were used for recording compound muscle action potential (CMAPs) in the target muscles of the limbs bilaterally. During the procedure MEPs were continuously recorded and analyzed in real time regarding latency and amplitude. A decline in amplitude of > 50% was considered as alarm criteria and was immediately reported to the neuro-radiologist. In this case, the procedure was halted and, after stabilization of the potentials, embolization was continued.

Cannulations of the feeding vessels were performed using a flow-directed microcatheter (Synchro-10, Marathon) and liquid embolic agent (20% Magic glue) was injected closed to the nidus. Complete obliteration was achieved after second intervention (Figure 2).

During the interventions the patient was continuously monitored with base monitoring (heart rate, oxygenation, blood pressure and electrocardiogram) by the anesthetist as well as IONM by a neurosurgeon to detect possible alterations in motor evoked potentials (MEP). During the first intervention no decline in MEP´s amplitude was detected. During the second intervention a 50% decline in MEP amplitude was observed and immediately reported to the interventional neuroradiologist. The procedure was halted till the stabilization of the potentials (Figure 3). After the intervention the child initially presented with temporary motor deficit of the right leg, which completely resolved within few hours. No complications associated with the use of IONM were observed.

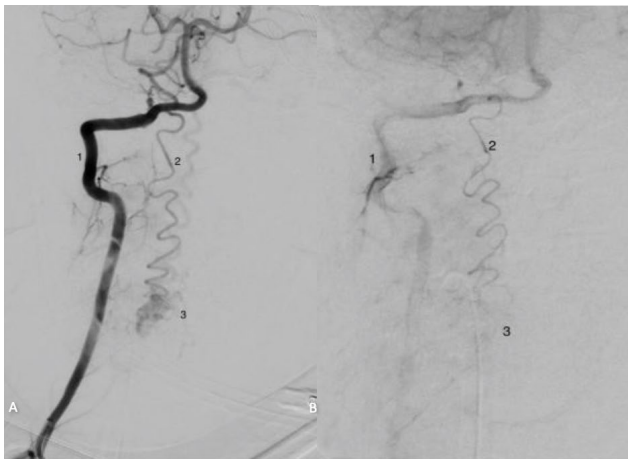

Figure 2: Intra-procedural DSA. (A) Before treatment; (B) Post-glue embolisation shows the obliteration of AVM (3).

Discussion

Vascular malformations of the spine account for 2-4% of the entirety of spinal lesions in adults [1-3], in the pediatric populations are even rare, making them an exceptional entity that require prompt diagnosis and optimized treatment regimes. This report refers to the classification proposed by Anson and Spetzler [4-6]. There are only a few series available that describe clinical features and treatment of these lesions in children. Our case illustrates a Type II, intramedullary arteriovenous malformation, fed by anterior and posterior spinal artery. MRI is a sensitive diagnostic tool to confirm the presence of hemorrhage and intramedullary AVM. Classic representation of myelopathy correlates with possible MRI findings such as hyper-intense T2 cord signal or edema, presenting over multiple spinal levels [4-8].

Computer tomography (CT) and computed tomography angiography (CTA) scans may be a viable

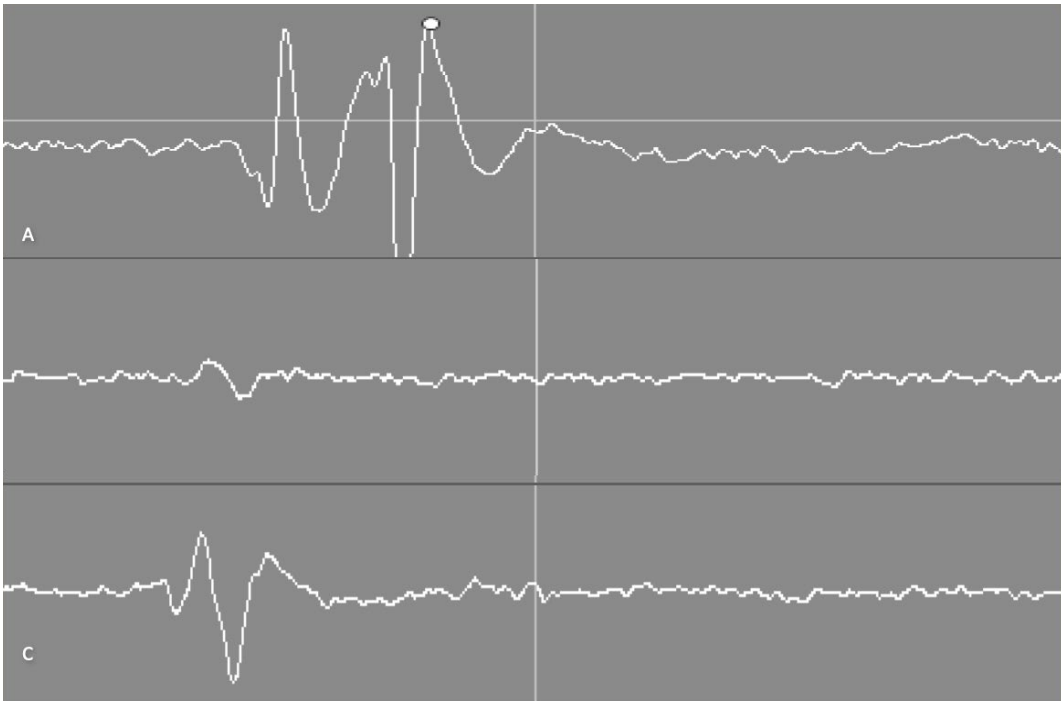

Figure 3: Intraoprocedural MEPs. (A) Shows MEPs baseline before the start of procedure; (B) Iintraprocedural MEP Amplitude loss > 50%; (C) MEPs at the end of the procedure.

option in acute situation, but better tissue resolution and especially in the pediatric population, the radiation exposure calls for alternative imaging provided by MRI [7]. If spinal AVM is suspected, DSA is the gold standard for sensitive detection of involved segments, feedings arteries and fistulous components, proving superior to both CT and MRI [5-7].

Although pediatric spinal AVMs are a heterogeneous group with a wide spectrum of symptoms, treatment options do not differ from those of adult AVMs [1]. Successful treatment depends on the degree of neurological impairment before the diagnose as well as the timespan of treatment after the diagnosis [1]. Treatment options for spinal AVM include surgery, endovascular embolization and radiotherapy, with embolization used alone or before resection [9]. In Type II Spinal AVM, embolization is the first-line treatment with open surgery carrying a significant intraoperative risk [1,2,9].

To ensure favorable functional outcome, IONM has been established, mainly during surgery [3-7,9,10]. Monitoring via somatosensory evoked potentials (SEP) is more accessible but MEPs should be included whenever possible to monitor sensory and motor pathways [11]. Niimi, et al. in 2001 suggest the use of provocative chemical testing in 41 embolization procedures for the treatment of spinal cord AVM using amytal and lidocaine injections to test for changes in SEPs and MEPs before applying a liquid embolic agent [12]. This could prove a viable option in the future considering that MEPs reflect on the current intraprocedural spinal integrity, working as a monitoring device but ineffective at preventing changes in the vascular territories, e.g. spasm or vessel occlusion through catheter placement.

Conclusion

IONM is a reliable and safe technique during endovascular treatment of spinal AVMs in children. The implementation of IONM can be useful in the decision-making process to evaluate the risk/benefit ratio between achieving a complete AVM embolization and a patient's short and long-term neurological status.

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