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CASE REPORT

Incidental occlusion of anterior spinal artery due to Onyx reflux in embolization of spinal type II arteriovenous malformation

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Abstract
Purpose Onyx embolization is one of the standard treatments for brain arteriovenous malformations (AVMs) and is a promising method for spinal AVMs as well. Its advantages have been emphasized, and few complications have been reported with Onyx embolization in spinal AVMs. Here, we report an incidental anterior spinal artery (ASA) occlusion due to Onyx reflux during embolization of a spinal type II AVM.
Methods A 15-year-old boy presented with weakness in both upper and lower extremities. Magnetic resonance imaging and spinal angiogram revealed a spinal type II AVM with two feeders including the right vertebral artery (VA) and the right deep cervical artery.
Results Onyx embolization was performed gradually from the VA to the deep cervical artery and an unexpected Onyx reflux to the ASA was observed during the latter stage deep cervical artery embolization. Post-operative quadriplegia and low cranial nerves (CN) dysfunction were observed. Rehabilitation treatment was performed and the patient showed marked improvement of neurologic deterioration at 1-year follow-up.
Conclusions Onyx is an effective treatment choice for spinal AVMs. However, due to the small vasculature of the spine compared to the brain, the nidus is rapidly packed with a small amount of Onyx, which allows Onyx reflux to unexpected vessels. Extreme caution is required and dual-lumen balloon catheter could be considered for Onyx embolization in spinal AVMs treatment.

Keywords Anterior spinal artery · Arteriovenous malformation · Onyx · Embolization · Reflux

Introduction

Spinal glomus arteriovenous malformation (AVM), which is classified as a spinal type II AVM in the classification scheme of DiChiro and Wener, is a rare disease and a particularly challenging type of spinal cord vascular malformation [1]. Endovascular embolization is preferred for most spinal AVMs because of the high risk associated with surgical resection [2]. Onyx (ev3 Neurovascular, Irvine, CA, US) is a liquid embolic agent that has become one of the standard treatments for brain AVMs; it has also shown promise in in spinal AVM treatment [3, 4]. As with brain AVMs, Onyx treatment of spinal AVMs was previously reported to yield good clinical overall outcomes with few complications [2, 5, 6]. However, we report an unusual complication associated with Onyx embolization of spinal type II AVM in which the non-targeted anterior spinal artery (ASA) was clogged incidentally. Details of the case and outcomes are presented below.

Case report

A 15-year-old boy was admitted via the emergency room (ER) on a weekend with a 1-day history of progressive weakness and paresthesia in his bilateral upper and lower extremities. Neurologic examination revealed motor grade 4 for all extremities, American Spinal Injury Association

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(ASIA) grade D, and a Functional Independence Measure (FIM) score of 6 [7, 8]. Cervical magnetic resonance imaging (MRI) was performed immediately and the result suggested a spinal AVM with extensive medullary swelling (Fig. 1). The patient showed rapid deterioration of neurology within an hour after MRI was taken (motor grade 3, ASIA grade C, FIM score 5). Progression of spinal cord compression due to medullary swelling was suspected, therefore, the emergency multilevel laminoplasty was performed. Intra-operative surgical resection of the AVM suspected lesion was not considered because of the absence of vascular information. The patient was stable after surgery without further neurologic deterioration.

Spinal angiography was performed subsequent to the surgery. The angiogram demonstrated a spinal type II AVM with two feeders, the right VA and right deep cervical artery. The nidus of the AVM and congested medullary draining veins were also observed. The ASA was visualized on a selective deep cervical artery angiogram with the microcatheter placed proximal (before passing) to the ASA branch orifice. After the microcatheter was advanced closer to the nidus and passed the ASA branch orifice, the ASA was not visualized with contrast agent (Fig. 2). Further endovascular embolization was performed under general anesthesia after heparinization (3000 IU). Successful embolization of the nidus supplied from the VA was observed with Onyx 0.2 ml. The microcatheter was subsequently repositioned to the deep cervical artery and advanced close to the nidus, passing the ASA branch orifice. Less than 0.2 ml of Onyx was carefully injected; however, unexpected retrograde reflux occurred and Onyx instantly occluded the ASA branch (Fig. 3). Immediate anticoagulation with heparin (1000 IU) was performed and no further embolization was attempted. Post-operative quadriplegia and impairments of cranial nerves (CN) IX, X, XI, and XII were observed (ASIA grade A, FIM score 1). The patient remained in the intensive care unit (ICU) for a month after the procedure because of respiratory insufficiency. He underwent rehabilitation therapy after transfer to a general ward, where he showed gradual improvement of motor deficits and low CN dysfunction. Follow-up neurologic examination 1 year postoperatively showed marked improvement of both motor and low CN function (motor grade 3, ASIA grade C, and FIM score 5). A 1-year follow-up MRI revealed significantly improved cord swelling compared with the initial postoperative MRI (Fig. 4).

Fig. 1 T2 sagittal MRI demonstrates the nidus formation at the C4–5 level (*arrow*). Engorged vascular signal voids and associated massive medullary swelling from level of C2 to C7 are noted

Discussion

Spinal type II AVM is a rare lesion that causes neurological morbidity as a result of hemorrhage and/or venous hypertension, arterial steal, or mass effect [9]. Treatment of the malformations remains challenging because of their complex microanatomy. Spinal vascular malformations have been treated surgically and by endovascular embolization. Endovascular treatment is preferred for most of these complex vascular malformations because of the high degree of morbidity associated with surgery [2]. Endovascular embolization of spinal AVMs has been traditionally performed with polyvinyl alcohol (PVA), N-butyl cyanoacrylate (NBCA) and Onyx [3]. PVA particles can be injected directly through metameric arteries without super-selection because of its size, which ranges from 10 to 30 μm. In spite of the advantages of PVA in terms of the safety of particulate embolization, recanalization and clinical recurrence after embolization have emerged as its main drawbacks. Compared to PVA, NBCA has the advantage of a lower recurrence rate. However, NBCA occasionally adheres to the catheter and has high risk of spread into unexpected spinal vessels through anastomoses, even though the microcatheter is advanced deeply into the nidus of a spinal cord AVM [10].

Onyx, the latest material to be utilized for spinal embolization, has slower polymerization kinetics and higher viscosity compared to NBCA; this results in superior delivery characteristics and enables increased penetration of the AVM nidus, potentially leading to decreased surgical blood loss and less catheter adherence than NBCA

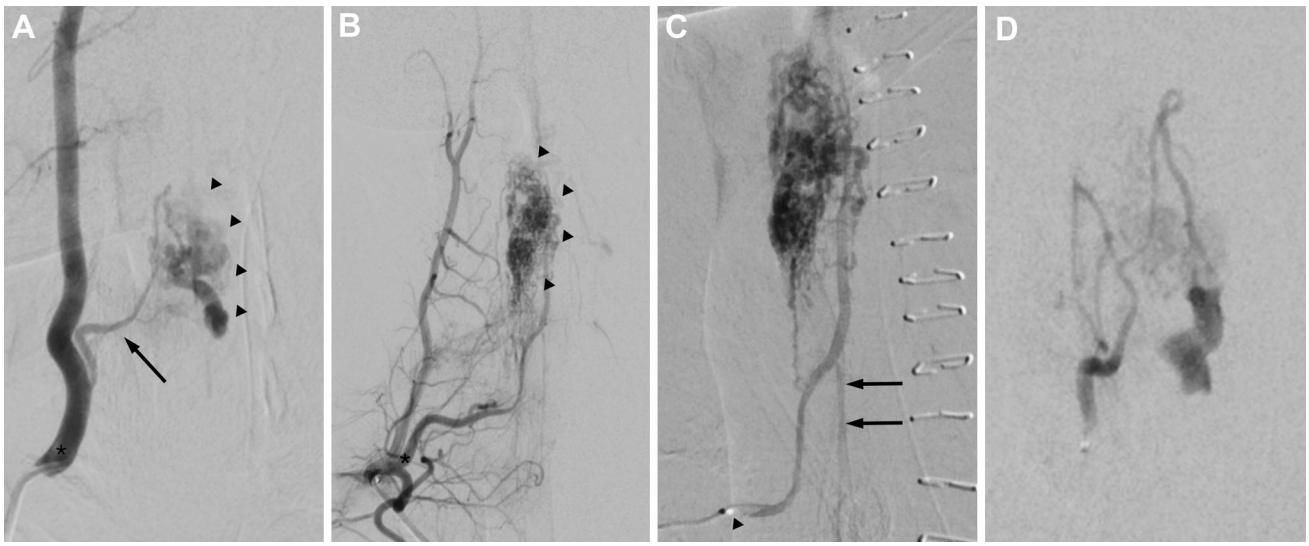

Fig. 2 **a** Lateral (LAT) angiogram via the right VA (*asterisk*) reveals the nidus (*arrow heads*) and feeding artery (*arrow*). **b** Anteroposterior (AP) angiogram via the right deep cervical artery (*asterisk*). The nidus (*arrow heads*) measured 13 × 35 mm. **c** AP angiogram through the right deep cervical artery reveals the nidus and the ASA (*arrows*). The microcatheter distal tip (*arrow head*) is placed proximal to the ASA branch orifice. **d** The ASA is not visualized with contrast agent after the advance of microcatheter distal to the ASA branch orifice

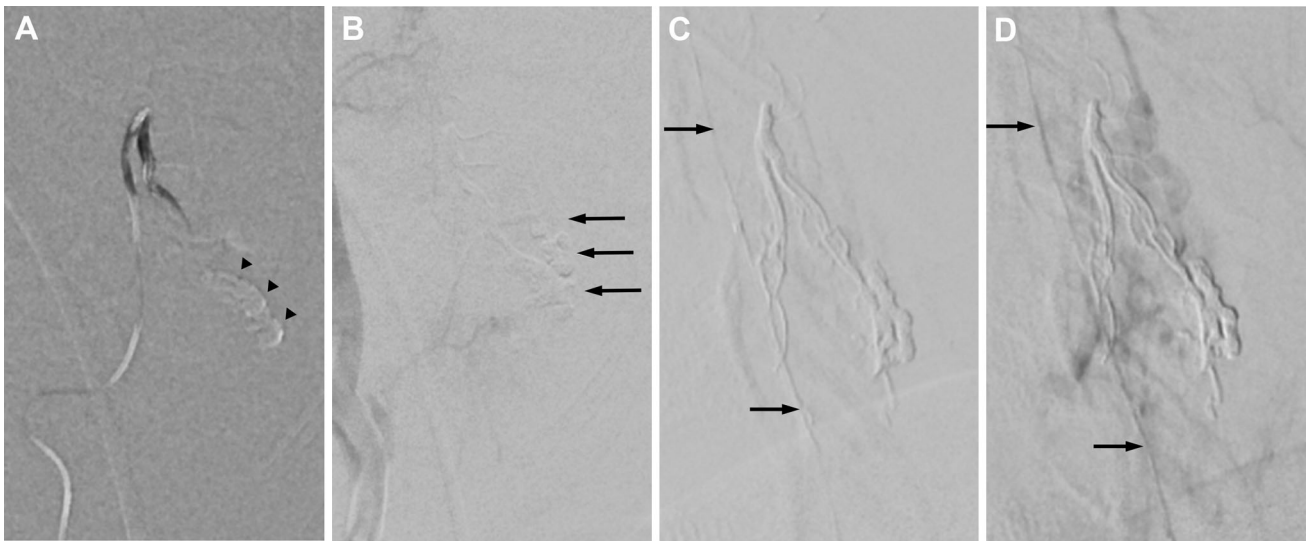

Fig. 3 **a** Stored fluoroscopic roadmap image during Onyx injection toward the nidus (*arrow heads*) via the right VA. **b** Successful obliteration of the nidus (*arrows*) supplied from the VA was observed. **c** The microcatheter was positioned distal to the ASA branch orifice via the deep cervical artery. However, off-target Onyx reflux occurred and ASA occlusion (*arrows*) is noted. **d** Onyx cast (*arrows*) is still visible after heparinization

[2, 5, 6, 11]. Despite its advantages, Onyx has some drawbacks that have rarely been reported. The biological toxicity of ethyl alcohol-dimethyl sulfoxide (DMSO, the chemical formula of Onyx) is well known in general chemistry and biology; however, in the case of endovascular treatment, the detailed toxicity of the compound has not been clear [12]. Hemorrhage has been reported as one potential complication of Onyx [2, 6]. Small intra-procedural subarachnoid hemorrhages have been reported, with subsequent neurological decline observed in the affected patients [6]. An unusual case of hemorrhage from a radicular artery tear due to withdrawal of the microcatheter has also been reported [5]. However, complications such as the one observed in our case have not been reported with regard to spinal AVM treatment with Onyx. The microcatheter was advanced close enough to the nidus, which passed the ASA branch orifice via the deep cervical artery, to prevent an off-target effect. We injected a total volume of no more than 0.4 ml of Onyx, which is well within the recommended range of practical use (0.2–1.0 ml)

Fig. 4 T2 sagittal MRI 1 year after Onyx embolization. The nidus cast is prominent (*arrow*), and cord swelling has markedly improved

[3–5, 12]. Nonetheless, retrograde Onyx reflux occurred, resulting in incidental occlusion of the ASA.

Retrograde embolization of patent arterial vessels with Onyx is infrequently reported in brain AVMs [13–15]. Ashour et al. published three cases of inadvertent cerebral artery-to-artery Onyx embolization out of a total of 51 pediatric brain AVM embolization procedures (5.9 %) [15]. They argued that retrograde embolization was more likely to occur during an aggressive attempt or in the final stages of embolization after a significant portion of the AVM has already been embolized. To interpret our case, we referred to the references; the retrograde reflux might have occurred in the final stage of embolization after a significant portion of the nidus supplied from the VA had been obliterated. The affected vasculature in our patient was smaller than the brain vasculature of adults. This might have caused the retrograde reflux from the distal tip of the microcatheter, even though only a small amount of Onyx was packed.

Corkill et al. published a series of spinal type II AVM in 17 patients [2]. Long-term clinical follow-up demonstrated that approximately 82 % of the patients showed improved neurological functioning. Three complications, all associated with hemorrhage, were reported without subsequent neurological sequelae. Our patient, however, had severe neurologic deterioration due to an unexpected complication. He also was ventilator-dependent for 1 month. Fortunately, he showed marked improvement of neurological deterioration 1 year after rehabilitation treatment.

Onyx embolization is a satisfactory treatment option for spinal AVMs. However, the spinal vasculature is smaller than that of the brain, and this size difference could lead to an incidental retrograde reflux of Onyx, resulting in off-target vessel occlusion. In recent studies, the Scepter catheter (MicroVention, Tustin, California, USA), a dual-lumen balloon catheter, has been applied to Onyx embolization of tumors and AVMs [11, 16–19]. An initial proximal plug is often necessary prior to Onyx injection for preventing reflux with conventional catheter-based techniques. However, the inflation of a balloon proximal to the Onyx injection site may avoid the need for a plug and its associated risks [16]. In conclusion, meticulous handling and extreme caution are required during Onyx embolization procedures for spinal AVMs and the balloon-assisted Onyx embolization could be also considered to reduce the complication.

Compliance with ethical standards

Conflict of interest None of the authors has any potential conflict of interest.

Ethical approval For this type of study formal consent is not required.

Informed consent Informed consent was obtained from the participant included in the study.

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