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Case Report

Trigeminal neuralgia secondary to onyx embolization of a right occipital arteriovenous malformation

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ABSTRACT

Background: Trigeminal neuralgia is a debilitating chronic condition characterized by severe recurrent hemifacial pain which is often caused by compression of the trigeminal nerve by an adjacent vessel loop. Microvascular decompression (MVD) surgery is an effective procedure that can lead to full symptomatic relief. Intracranial arteriovenous malformations (AVMs) are primarily congenital abnormalities that may be asymptomatic or manifest as seizures or focal neurologic deficits. They may cause intracranial bleeding and hence are promptly treated, often by endovascular embolization. This procedure is safe but may have a multitude of unpredictable complications.

Case Description: A 33-year-old female presented with medically refractory trigeminal neuralgia secondary to Onyx embolization of a right occipital AVM 3 years prior. She underwent surgical exploration and MVD of the trigeminal nerve root which was found to be compressed by the previously embolized superior cerebellar artery. The procedure was successful and full symptomatic resolution was immediately achieved.

Conclusion: Postprocedural trigeminal neuralgia is a procedural complication of Onyx endovascular embolization. It may be treated by MVD surgery regardless of the presence or absence of a compressive vascular loop on imaging.

Keywords: Adverse events, Arteriovenous malformation, Embolization, Microvascular decompression, Onyx, Trigeminal neuralgia

INTRODUCTION

Trigeminal neuralgia is a chronic condition characterized by severe, often unilateral, lancinating pain in the trigeminal nerve distribution of the face that is brief and recurrent. It is estimated to affect approximately 4/100,000 people/year in the United States.[6] A common cause of trigeminal neuralgia is thought to be compression of the trigeminal nerve root, most often by the superior cerebellar artery (SCA),[16] making microvascular decompression (MVD) surgery a valid treatment option for medically refractory cases.[5]

Intracranial arteriovenous malformations (AVMs), on the other hand, are rare primarily congenital vascular abnormalities estimated to affect <0.01% of the general

population.[1] Intracranial hemorrhage is one of the most common and feared complications, affecting approximately 53% of patients with AVMs,[4] thus highlighting the importance of treatment. AVM management is often multimodal, involving microvascular surgery, stereotactic radiosurgery, and endovascular embolization.[3]

Onyx liquid endovascular embolization has recently emerged as a versatile therapeutic choice. Often, it is employed perioperatively to reduce AVM nidus size; however, it is also a valid standalone curative modality in select cases.[10] Adverse events of this procedure are not uncommon, and this case report will serve as an example of a rare complication and how to resolve it.

We present the case of a 33-year-old female with trigeminal neuralgia secondary to Onyx endovascular embolization of a right occipital AVM 3 years prior which we successfully treated by MVD surgery despite no evidence of a compressive vessel loop on imaging.

CASE DESCRIPTION

Initial presentation

A 33-year old female, known to have a history of right occipital AVM treated by Onyx endovascular embolization 3 years prior, presented with severe right-sided trigeminal neuralgia and occipital lobe visual seizures. Embolization was done at an outside hospital and right hemifacial pain began after it. She failed three lines of medical treatment since then and has been maintained on carbamazepine 800 mg twice daily with no improvement in pain.

She underwent a diagnostic cerebral angiography which showed total occlusion of the previously embolized AVM along with distal occlusion of the right posterior cerebellar artery and SCA [Figure 1]. No other abnormalities were detected in the posterior circulation.

Subsequent brain MRI showed the previously embolized AVM in the right occipital lobe [Figure 2], along with no evidence of a vascular loop impinging on the right trigeminal nerve that may explain her trigeminal neuralgia [Figure 3]. Nevertheless, the decision was made to surgically intervene for exploration and MVD of the trigeminal nerve after thorough consultation with the patient.

Operative procedure

One week after the angiogram, the patient underwent successful Jannetta MVD of the trigeminal nerve. She was positioned in lateral position and a retroauricular lazy-S skin incision was made two finger-widths away from the ear for a retrosigmoid approach. The flap was reflected from medial to lateral and the paravertebral muscles

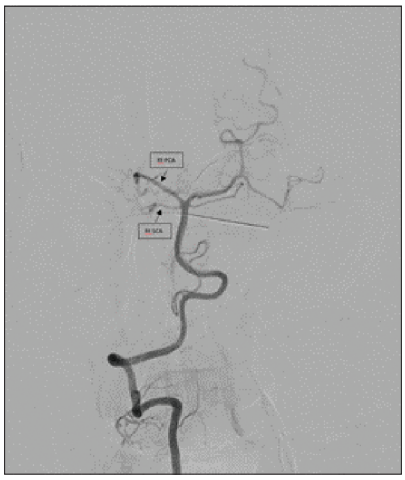

Figure 1: AP cerebral angiogram showing a right vertebral artery injection. Indicated by the arrows are the right SCA and the right PCA, both occluded postembolization. SCA: Superior cerebellar artery, PCA: Posterior cerebellar artery.

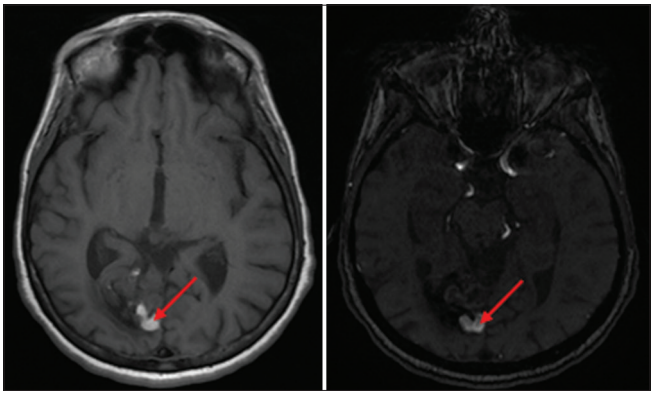

Figure 2: Axial sections of T1-weighted brain MRI with and without contrast showing the embolized right occipital intracranial arteriovenous malformation (red arrows) with the embolization material appearing as hyperintense.

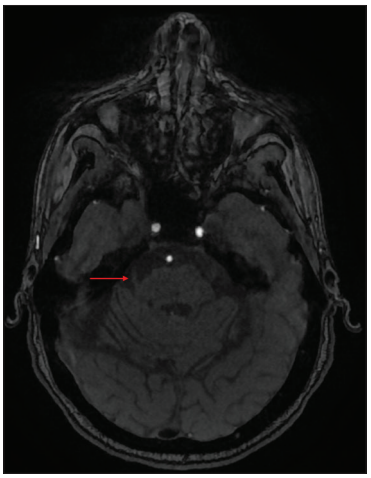

Figure 3: Axial section of T1-weighted brain MRI showing the right trigeminal nerve (red arrow) with no impinging vascular loop.

were elevated subperiosteally. A burr hole was made at the asterion just below the junction of the transverse and the sigmoid sinuses and a retrosigmoid craniotomy was done using a footplate. The craniotomy was enlarged by drilling the bone over the transverse and sigmoid sinuses. The mastoid air cells were covered with bone wax to avoid postoperative CSF leakage. The dura was then opened and reflected over the transverse and the sigmoid sinuses, and CSF was drained from the lateral cerebellomedullary cistern to relax the cerebellum.

Initial inspection of the cerebellopontine angle showed the seventh and eighth nerve complex. More CSF drainage was attempted to provide additional working space and the trigeminal nerve was subsequently localized and freed from the surrounding arachnoid adhesions. A whitish tubular structure that resembled an empty vessel was noticed crossing and abutting the trigeminal nerve root entry zone [Figure 4]. After thorough inspection of the area, it was identified as the previously embolized SCA. As no other obvious vessel loop was noted, we considered this empty vessel as the offending structure causing the trigeminal neuralgia.

The SCA was hence carefully dissected off the trigeminal nerve root entry zone and the brain stem, creating space for two small pieces of Teflon sponges which were inserted to separate the vessel from the mentioned structures it was compressing [Figure 5]. The dura and the muscle fascia were thereafter closed tightly and the procedure was completed.

The patient did well postoperatively and left the hospital 2 days later with no reported hemifacial pain. She was seen 2 weeks after the surgery in clinic with full resolution of trigeminal neuralgia and her medication dosage was halved.

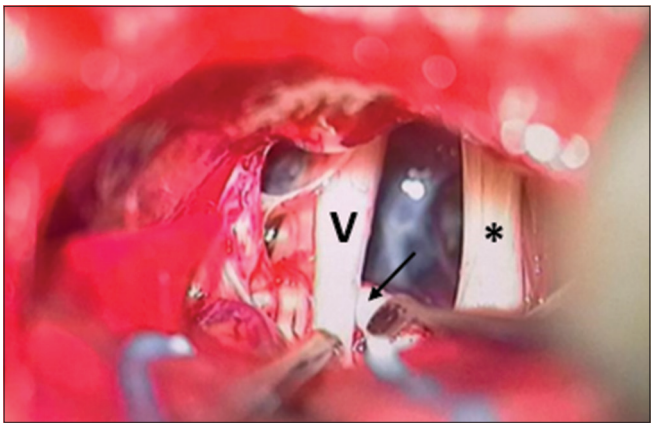

Figure 4: Operative view showing the right trigeminal nerve (v) with the embolized superior cerebellar artery (black arrow) under the instrument. Note the seventh-eighth cranial nerve complex on the right (*).

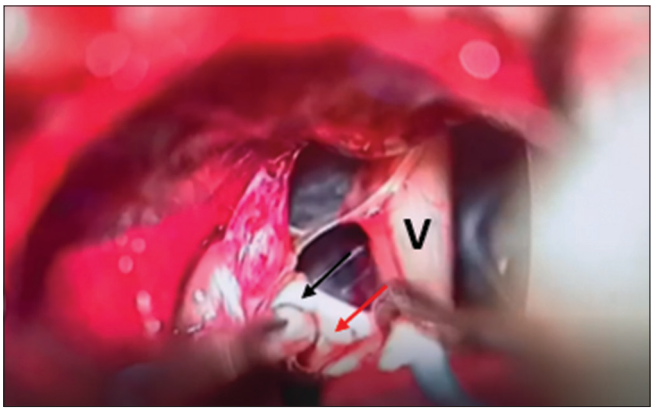

Figure 5: Operative view showing the insertion of a Teflon sponge (red arrow) between the trigeminal nerve (v) and the embolized superior cerebellar artery (black arrow).

DISCUSSION

Onyx endovascular embolization is routinely performed as part of the management plan of intracerebral AVMs. It is deemed safe and effective, but it is in fact not without risk, and many adverse events have been described in the literature. A large retrospective study by Sato *et al.* found that complications arose in 13.1% of procedures,[12] most common of which were hemorrhage (5.7%)[12] and ischemia (5.5%).[12] Other less common adverse events are reported, such as Onyx extravasation,[14] varix or abscess formation,[7,13] and cranial nerve (CN) palsies.[8,9] Nyberg *et al.* detailed transient self-limited CN V and VII palsy following endovascular embolization,[8] while Ozluoglu *et al.* described the development of persistent facial paralysis and CN VII palsy which was only relieved by nerve decompression surgery.[9] Interestingly, there is one reported case of iatrogenic trigeminal neuralgia secondary to AVM embolization which was unsuccessfully treated with MVD surgery.[15]

How such CN palsies arise after endovascular embolization is not clear, but many mechanisms have been proposed to explain this phenomenon, which is most likely due to downstream vascular effects of the procedure. As Nyberg *et al.* hypothesized, the transient CN dysfunctions they encountered may have been due to axonotmetic traction injuries during microcatheter removal from the Onyx cast.[8] Buell *et al.* showed that multiple factors promote angiogenesis following embolization.[2] Such new vessels might compress existing structures including CNs as they exit the brainstem. Finally, Quinn *et al.* described findings of eosinophilic vasculitis with vessel wall thickening and mural calcifications in thrombosed AVM vascular channels following n-BCA embolization.[11] Such a reaction, if applicable to Onyx, could also theoretically lead to local compression of CNs.

Any of these mechanisms, alone or in combination, may have contributed to the presentation we have encountered. The trigeminal neuralgia in our patient can be safely attributed to

the embolization procedure as it began immediately after it. Another supporting piece of evidence is the fact that during the exploratory surgery, the previously embolized SCA, now empty, was found abutting the trigeminal nerve, and decompression lead to prompt resolution of symptoms.

CONCLUSION

By reporting this case, we hope to add to the repertoire of known adverse events attributable to Onyx embolization. We also aim to shed light on possible mechanisms of this event, which, if generalizable to other procedures involving different intracranial arteries, may provide an explanation to some of the more obscure complications and CN palsies. We also show that MVD is a valid and highly effective treatment of trigeminal neuralgia even when no compressing vessel loop is identifiable on imaging, lending credence to the mostly vascular etiology of the condition.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

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