Middle meningeal artery pseudoaneurysm and pterygoid plexus fistula following percutaneous radiofrequency rhizotomy: illustrative case

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BACKGROUND Percutaneous treatment for trigeminal neuralgia is a safe and effective therapeutic methodology and can be accomplished in the form of balloon compression, glycerol rhizotomy, and radiofrequency thermocoagulation. These procedures are generally well tolerated and demonstrate minimal associated morbidity. Moreover, vascular complications of these procedures are exceedingly rare.

OBSERVATIONS We present the case of a 64-year-old female with prior microvascular decompression and balloon rhizotomy who presented after symptom recurrence and underwent a second balloon rhizotomy at our institution. Soon thereafter, she presented with pulsatile tinnitus and a right preauricular bruit on physical examination. Subsequent imaging revealed a middle meningeal artery (MMA) to pterygoid plexus fistula and an MMA pseudoaneurysm. Coil and Onyx embolization were used to manage the pseudoaneurysm and fistula.

LESSONS This case illustrates the potential for MMA pseudoaneurysm formation as a complication of percutaneous trigeminal balloon rhizotomy, which has not been seen in the literature. Concurrent MMA-pterigoid plexus fistula is also a rarity demonstrated in this case.

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KEYWORDS trigeminal neuralgia; percutaneous rhizotomy; arteriovenous fistula; pseudoaneurysm

The percutaneous approach to the trigeminal nerve via the foramen ovale originated in the early 20th century from Taptas and Hartel in 1911 and 1913, respectively, with electrocoagulation developed by Reith in 1913. Over the subsequent century, much advancement in the percutaneous techniques has occurred including chemodenervation, radiofrequency ablation, cryoablation, nerve blocks, Botulinum injections, nerve stimulation, and balloon decompression.

Percutaneous treatment for trigeminal neuralgia is generally considered a well-tolerated procedure, with the most common complications of dysesthesia, reduced corneal reflex, paresthesias, and herpes flare. Each therapeutic technique in the percutaneous approach has different risk-benefit profiles. However, vascular complications are rare. If a case is affected, the most common vascular sequelae is a fistula between the cavernous internal carotid artery and the surrounding venous cavernous sinus. Other fistulous connections have been reported but are even more infrequent. Pseudoaneurysm formation is exceptionally rare and, to the best of our knowledge, has not been reported. Here, we report the case of a 64-year-old female who developed a pseudoaneurysm of the middle meningeal artery (MMA) as well as a fistulous connection between the MMA and pterygoid plexus as a complication of percutaneous rhizotomy.

Illustrative Case

Presentation

A 64-year-old female initially developed symptoms of bilateral trigeminal neuralgia 40 years prior. In addition, she was status post-right microvascular decompression 23 years prior, balloon rhizotomy 14 years prior, and oxcarbazepine treatment. She described the recurrence of pain on the right as 3–5/10, needle-like, “lightning bolt,” and stabbing. Precipitating events included talking, eating/chewing, brushing her teeth, washing her face, and wind exposure. An increase in medication was not tolerated because of sedative side effects. She underwent a right-sided percutaneous balloon rhizotomy without immediate complication (Fig. 1A and B). She tolerated the procedure well and went home the same day.

ABBREVIATION MMA = middle meningeal artery.

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Complication

The patient reported symptoms of pulsatile tinnitus 2 weeks after the procedure and demonstrated a right preauricular bruit on a physical exam. Magnetic resonance angiography of the head demonstrated an extracranial fistulous connection of unclear etiology (Fig. 2A and B). Digital subtraction angiography was subsequently performed, which demonstrated an MMA to pterygoid plexus fistula as well as an MMA pseudoaneurysm (Fig. 2C–F).

Management

The patient was taken to the angiography suite for further characterization and definitive management. Catheterization of the MMA with microinjections opacifying the pseudoaneurysm and draining veins was performed (Fig. 2C–F). The distal intracranial MMA and its anastomosis to the ophthalmic artery were evaluated. This was followed by coil placement distal to the pseudoaneurysm (within the proximal skull base MMA and foramen spinosum) and subsequently Onyx-34 penetration casting the fistula and immediately proximal MMA supply until the internal maxillary artery was reached (Fig. 3A and B). Final images showed no residual arteriovenous fistula or pseudoaneurysm filling (Fig. 3C and D).

Follow-Up

The patient was seen 5 years later in 2019 for symptoms of left trigeminal neuralgia pain. Residual right-sided numbness was endorsed. No report of tinnitus was documented.

Patient Informed Consent

The necessary patient informed consent was obtained in this study.
Discussion
Observations

We present the case of MMA pseudoaneurysm formation as a complication of percutaneous trigeminal balloon rhizotomy, which, to our knowledge, has not been seen in the literature. Additionally, the pseudoaneurysm occurred concurrently with an MMA-pterigoid plexus fistula, which has been reported only once. Fistula formation following rhizotomy is a rare complication, with only 19 cases of fistulous formations reported in the literature.5–17 The majority have been abnormal connections between the cavernous carotid and the surrounding venous plexus of the cavernous sinus. Seven cases have described fistulous connections involving segments of the internal maxillary artery.5,9,11,14,15,17 One connected to the venous cavernous sinus, two to the pterygoid plexus, one to the internal jugular vein, and others were not recorded. One additional nonrhizotomy-related fistulous connection during percutaneous foramen ovale manipulation has been reported between the cavernous internal carotid and the inferior petrosal sinus during foramen ovale telemetry.18

Anatomical variants are thought to be specific risk factors for fistula formation complications. The most commonly referenced anatomical variant thought to predispose patients to a fistulous complication is a primitive foramen lacerum medius in which the foramen ovale and lacerum are conjoined (reported in 4% of the population) or a primitive foramen lacerum in which the foramen ovale, lacerum, and spirnous form one large foramen (recorded in 3% of the population). It is thought that this variant causes inadvertent medialization of the percutaneous needle, resulting in injury to the carotid artery. Of the 14 cases reported, only the original case series by Sekhar et al.7 documents a patient with primitive foramen lacerum. Nonetheless, this is a frequent variant of caution reported in subsequent case reports.6–8,10–12

Other case reports have mentioned the foramen of Vesalius; however, this variant is exceedingly rare.20 Additionally, our patient’s prior balloon rhizotomy likely contributed to scar formation resulting in less reliability when using the traditional anatomical markers. A similar conclusion was suggested by Langford et al.14 in a 72-year-old patient with a previous radiofrequency procedure. To our knowledge, no cases of pseudoaneurysm formation after percutaneous rhizotomy for trigeminal neuralgia have been reported. A prior instance of a pseudoaneurysm of the proximal internal maxillary artery was reported with needle injection for masticatory muscle reduction; however, the exact trajectory was not well described. In one rare instance, a pseudoaneurysm was reported as a late complication of Gamma Knife treatment for trigeminal neuralgia.22

HärteI’s anatomical landmarks begin 2.5 cm lateral to the corner of the mouth with the needle directed to intersect an imaginary line along the medial ipsilateral pupil and 2.5 to 3 cm anterior to the external auditory canal.23 Thus, prior to arriving at the foramen ovale, the needle must first transverse the infratemporal fossa, which houses the diffuse pterygoid plexus. This plexus extends from the ramus of the mandible to the lateral pterygoid plate, allowing for connections with the maxillary vein, deep facial vein, and inferior ophthalmic and emissary veins to the cavernous sinus.24 Simultaneously, the infratemoral fossa houses the internal maxillary artery and its branches as the artery dives behind the neck of the mandible until it reaches the pterygopalatine fossa.21 Thus, it is conceivable to have simultaneous injury to these adjacent structures and create an inadvertent fistula, as was seen in our case and twice in the literature, or an isolated injury to an arterial structure resulting in a pseudoaneurysm or both, as described in our patient.

Previously reported treatments for these fistulas have primarily been endovascular, via embolization/casting at the internal maxillary artery or branches, and one instance of open carotid clipping.8 The durability of these treatments is difficult to reliably characterize because of their infrequent use. One report of distal embolization failure for an iatrogenic fistula resulted in the need for carotid embolization. In this instance, the authors noted a very high-flow fistula in which arterial steal occurred from the left internal carotid artery to the fistula. This patient was noted to have filling of the affected hemisphere through the contralateral circulation via the anterior communicating, and after a 15-minute test occlusion demonstrated no deficits, the internal carotid artery was sacrificed proximal to the ophthalmic. In the largest series of cases, Li et al.17 demonstrated no vascular recurrences with a mean follow up of 20 months. Spontaneous closure has been reported, twice within 3 months and once within 3 weeks. Our patient was most recently seen in a pain clinic 5 years after treatment with persistent numbness on her affected side. No symptoms of her prior fistula or pseudoaneurysm were reported.

Lessons

Care should be utilized when conducting a percutaneous rhizotomy for trigeminal neuralgia, and one should be mindful of the vascular structures and anatomical variants that can arise. Knowledge of possible vascular complications is critical for the early diagnosis and management of these rare complications.

References


**Disclosures**

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

**Author Contributions**

Conception and design: Ismail, Kessler. Acquisition of data: Ismail, Kessler. Analysis and interpretation of data: Ismail, Schwartz, Kessler. Drafting the article: Ismail, Van Hoang, Kessler. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Ismail. Administrative/technical/material support: Van Hoang, Kessler. Study supervision: Kessler.

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