Delayed Spontaneous Resolution of a Traumatic Middle Meningeal Artery Pseudoaneurysm

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Patient: Male, 42-year-old
Final Diagnosis: Middle meningeal artery pseudoaneurysm
Symptoms: Migraine
Medication: —
Clinical Procedure: —
Specialty: Radiology

Objective: Unusual clinical course
Background: Middle meningeal artery (MMA) aneurysms are a very rare entity, comprising less than 1% of all intracranial aneurysms. In particular, traumatic MMA pseudoaneurysms (MMAP) are reported in the literature to have a poor outcome in about 20% of cases. Moreover, in extremely rare cases, MMAPs can spontaneously thrombose.

Case Report: We present the case of a 42-year-old Hispanic man with multiple craniofacial hemangiomas and history of chronic migraines that increased in frequency after blunt head trauma 1 month prior to initial evaluation. CTA and brain MRI showed a right-sided MMAP adjacent to the foramen spinosum with a pan-hemispheric subdural hematoma and no associated skull fractures. The MMAP was not visualized 2 days later on digital subtraction angiography (DSA) and was therefore presumed to be thrombosed. CTA at 3 months showed interval progression of the MMAP with subsequent spontaneous resolution on CTA at 10 months.

Conclusions: Knowledge regarding MMAPs is limited since it is based on a small number of cases and literature reviews. Additional studies are needed to elucidate the true incidence and natural course of this entity and produce adequate treatment guidelines.

Keywords: Aneurysm, False • Angiography, Digital Subtraction • Craniocerebral Trauma • Intracranial Aneurysm • Meningeal Arteries

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Background

Middle meningeal artery (MMA) aneurysms are a very rare entity, with about 70 cases described in the literature [1]. They account for less than 1% of all intracranial aneurysms [2]. The natural course of this entity is not well known given that in most cases prompt treatment is employed in order to avoid the high mortality risk of 20% reported in the literature [3]. MMA pseudoaneurysms (MMAP) are associated with temporal skull fractures in up to 92% of cases [4]. Only 4 prior case reports have described spontaneous resolution of MMAPs, usually within the first month after diagnosis [5-8]. We describe a fifth rare case of spontaneous resolution of a traumatic MMAP. This case is of particular interest because the patient presented no associated skull fractures, the MMAP was angiographically occult, and it demonstrated growth at 4 months after initial trauma, with subsequent delayed spontaneous resolution at some point before the 11-month follow-up.

Case Report

A 42-year-old Hispanic man with past medical history of chronic migraines and multiple right-sided facial hemangiomas came to the emergency room with a 3-week history of increased migraine frequency, usually beginning in the right coronal region with subsequent irradiation to the right side, with associated phonophobia, photophobia, nausea, and multiple vomiting episodes. The patient referred blunt head trauma as a result of physical aggression by repeated blows to the head 4 weeks prior to the emergency room (ER) visit. Prior to the traumatic event, migraines occurred every 3 to 6 months, increasing to daily episodes after the blunt head trauma. Additional past medical history was remarkable for prior heroin abuse, and he was currently on methadone. On physical examination, the patient showed no motor or sensory deficits, with a Glasgow Coma Scale score of 15 out of 15.

An outside institution brain MRI report (images were not available) performed approximately 3 weeks after the blunt head trauma described a possible aneurysm at the middle cranial fossa “adjacent to the petrous portion of the internal carotid artery”. Unfortunately, multiple attempts to obtain the images for this study were unsuccessful. The patient arrived at our institution’s ER 28 days after the inciting traumatic event, at which time a contrast-enhanced brain MRI was performed (Figure 1). It showed a round mildly lobulated T2 flow void at the right middle cranial fossa lateral to Meckel’s cave with corresponding contrast enhancement, peripheral rim-like susceptibility artifact, and a thin flow-void stalk at its inferior aspect, which appeared to connect with the foramen spinosum. There was associated right temporal lobe vasogenic edema as well as a small right pan-hemispheric subdural hematoma.

In view of these findings suggesting an MMAP, a computed tomography angiography (CTA) with delayed views (75 s after the standard arterial phase) was performed 3 days later (31 days after initial trauma) (Figure 2). The lesion showed early arterial enhancement, with delayed peripheral contrast pooling, in keeping with a posttraumatic intracranial pseudoaneurysm, likely from the right MMA. No skull fractures were identified. Additionally, a craniofacial hemangioma along the right masticator space and superficial to the temporal bone was identified with multiple feeders from the right external carotid artery.

Figure 1. (A) Post-contrast 3D T1 in the coronal plane shows a hyper-enhancing lesion at the right middle cranial fossa (long arrow) adjacent to the right foramen spinosum. In this image, the MMA can be seen traversing the foramen spinosum (dashed arrow). (B) Gradient echo axial image shows a ring-like blooming artifact (short arrow) associated to this lesion. (C) Coronal T2W image demonstrates a tubular flow void (arrowhead) connecting the vascular lesion with the foramen spinosum.
Figure 2. (A–C) CTA in axial, coronal, and sagittal planes shows a hyper-enhancing lesion (solid arrows) at the right middle cranial fossa. (D–F) Delayed CTA images (75 s after the standard CTA phase) show subsequent peripheral contrast pooling (short arrows) in keeping with a middle meningeal artery pseudoaneurysm. A craniofacial hemangioma is also seen superficial to the right temporal bone (dashed arrows in A, B, D and E). (G) Sagittal CTA in the bone window better demonstrates the relationship between the right middle cranial fossa pseudoaneurysm and the foramen spinosum (arrowhead).

The Endovascular Neurosurgery team was consulted after the CTA was performed, and 2 days later (33 days after trauma), the patient underwent digital subtraction angiography (DSA) including selective catheterization of the right ECA (Figure 3). The study showed the craniofacial hemangioma with feeders from the right ECA and right ophthalmic artery previously seen on CTA; however, the MMAP was not visualized in this study and was therefore presumed by the performing neurosurgeon to be thrombosed. Of note, a super-selective catheterization of the right MMA was not performed, which...
could have contributed to non-visualization of the MMAP. The 2-day delay in performing the DSA was due to non-availability of the endovascular neurosurgery team after the MMAP diagnosis was made by CTA. Given the neurologic stability during the hospital admission and taking into account the chronicity of his traumatic event, the patient was discharged home by the Neurosurgery service after completion of the DSA study with no additional imaging performed.

Approximately 4 months after the initial traumatic event, a follow-up contrast-enhanced brain MRI was performed at an outside institution, which confirmed the presence of the MMAP with improved depiction of an associated pulsation artifact (Figure 4C). Unfortunately, the lesion was misinterpreted in the MRI by the outside institution radiologist as a meningioma. A follow-up head CTA also performed at an outside institution around the same time as the aforementioned brain MRI showed an interval progression in size of the MMAP (Figure 4A, 4B). The patient had a follow-up visit with the Endovascular Neurosurgery service in charge of his care 7 months after the initial trauma. Unfortunately, based on chart review, at this 7-month visit the clinician only evaluated the brain MRI report obtained at 4 months, which wrongly suggested a middle cranial fossa meningoic; the results of the head CTA that confirmed the presence of the MMAP were not provided to the evaluating clinician at that time.

Upon case review by the Radiology service, the patient was contacted 11 months after the initial traumatic event and was asked to return to our institution for further evaluation. In view of the interval increase in size of the MMAP when comparing the studies at 1 month and 4 months after the head trauma, a follow-up CTA was performed at this 11-month evaluation (Figure 5). At this time, no vascular lesion was identified at the site of the previously identified MMAP, likely due to interval thrombosis. The previously described craniofacial hemangiomas remained stable.

**Discussion**

Aneurysms of the MMA are a rare entity first described by Schultze in 1957 [9]. Prior studies have found about 40 traumatic and 28 non-traumatic MMA cases in the literature [1]. Aneurysms can be true vs false or pseudoaneurysms, with the distinction based on aneurysmal wall histology. The wall of true aneurysms includes all 3 of the normal vessel layers, while pseudoaneurysms have fibroconnective tissue covering the vessel wall with none of the normal 3 layers. Pseudoaneurysms occur when there is a tear through the vessel wall where a blood clot forms, which subsequently converts to fibrous tissue. This fibrous tissue of pseudoaneurysms is more fragile when compared to a true aneurysm wall, and therefore carries a higher risk for size progression and/or delayed bleeding.
Figure 4. (A, B) Three-month follow-up CTA in axial and coronal planes shows interval progression in size and change in configuration of the right middle cranial fossa MMAP (solid arrows). (C) Post-contrast T1 in the sagittal plane shows the right-sided MMAP (solid arrowhead) with a subtle pulsation artifact in the phase-encoding direction (empty arrowheads).

Figure 5. (A, B) Ten-month follow-up CTA in axial and coronal planes shows complete resolution of the previously identified right-sided MMAP at the right middle cranial fossa (solid arrow). The craniofacial hemangioma is still seen superficial to the temporal bone (dashed arrow).

Traumatic MMAPs are associated with both intraparenchymal and extra-axial hematomas, with epidural hematomas being the most prevalent [10].

Various mechanisms have been suggested, including arterial wall tear by a skull fracture or by separation of the dura from the bone [12]. When the MMAP is not associated with a skull fracture, as in our case, or when they occur at locations distant from the fracture site, the postulated mechanism is traction injury to the vessel [13,14].

[10]. MMAPs have been associated with both intraparenchymal and extra-axial hematomas, with epidural hematomas being the most prevalent [10].
The true incidence of MMAPs is unknown, with only a few cases reported in the literature. It is possible that MMAPs are underdiagnosed given the lack of clear guidelines as to when vascular studies (such as CTA and DSA) are to be obtained in traumatic cases. At least 2 prior studies have suggested temporal rim fractures and adjacent small intracranial bleeds as the main predictors of MMAPs, and if both are present, a CTA is strongly recommended [10,11].

Both CTA and DSA have comparable accuracy in the detection of small intracranial aneurysms [15-17]. For MMAPs specifically, in a small prospective study by Paiva et al of 11 patients with epidural hematomas, both CTA and DSA detected 3 MMAPs in this group [18]. To the best of our knowledge, a larger study comparing the accuracy of these modalities for detection of MMAPs has not been published. The typical angiographic appearance of MMAPs has been described, which include an irregular aneurysmal sac without a neck, peripheral location distant from branching points, and a slow delayed contrast filling, which sometimes make them only visible in late injection stages [3]. In our case, the MMAP was not visualized on DSA by the performing Endovascular Neurosurgeon or by a Neuroradiologist who evaluated this DSA retrospectively. We hypothesize that this could have been due to a variety of reasons, including vasospasm of the feeding vessel, intermittent thrombosis and recanalization of the MMA wall tear, steal phenomenon from the craniofacial hemangioma shunting contrast away from the MMA during injection, technical factors such as not performing supra-selective catheterization of the MMA during the catheter angiogram, or a combination of the above. Only a selective catheterization of the right ECA was performed during the study and, in retrospect, a super-selective catheterization of the MMA might have helped in excluding the craniofacial hemangioma as a potential culprit of non-visualization of the MMAP via steal phenomenon. We did not find studies in our literature search regarding missed intracranial aneurysms on conventional angiography due to steal phenomenon by hemangiomas or other vascular malformations in the same vascular distribution. Nevertheless, prior studies comparing catheter angiography and CTA in the evaluation of hemodialysis fistulas have described cases of partially thrombosed aneurysms that were very well characterized on CTA and missed on angiography, in part due to a similar vascular steal mechanism [19]. Due to one or a combination of the above-mentioned reasons, we feel that the MMAP could have been present yet missed on initial catheter angiography. It is unlikely that the MMAP had completely thrombosed at the time of DSA evaluation given the lack of intraluminal filling defects on follow-up CTA at 4 months, which would have suggested some residual thrombus. It has been previously reported that about 6-30% of non-traumatic intracranial aneurysms are occult on initial angiography [20]. Similar data corresponding to angiographically occult MMAPs are not available due to the rarity of this entity.

Among the 4 prior case reports describing spontaneous resolution of a MMAP, 3 are part of the English literature [5-8]. Among these 3 English language articles, only Srinivasan et al and Shah et al disclosed the timeframe in which spontaneous resolution was documented (2 weeks and 1 month, respectively) [5,6]. This makes our case of particular interest, given that the pseudoaneurysm had progressed in size and configuration at its 4-month CTA and subsequently resolved at some point before the 11-month CTA. Both the interval progression and delayed subsequent resolution are unique findings among spontaneously resolving MMAP cases and contributes to the idea that at least some MMAPs could have a benign course, even after interval progression.

We recognize that in our case the patient unknowingly received conservative treatment given that the MMAP was presumed thrombosed by the Endovascular Neurosurgeon after a negative DSA. If the Endovascular Neurosurgery service had been made aware of the MMAP interval progression on the 4-month outside institution follow-up CTA, the patient would have probably undergone either endovascular or open surgery, mainly based on the high mortality rate that the literature reports in these cases. However, this speaks to the lack of treatment guidelines for MMAPs given that knowledge of this entity is based on a limited number of case reports and literature reviews. It is possible that MMAPs are being underdiagnosed given that CTA/DSA studies are not routinely performed in traumatic cases. Similarly, the subset of MMAPs that can spontaneously thrombose could be underrepresented given that CTA/DSA studies are usually only done after patient deterioration.

Conclusions

In conclusion, although rare, MMAPs are an important entity to be aware of in traumatic cases, even if no skull fractures are identified. The majority of the literature regarding this entity supports a poor outcome if untreated; however, our case speaks to the contrary in that some cases can resolve spontaneously even after progression. Nevertheless, it is impossible to generalize as to which MMAPs will spontaneously resolve based on our case and the current literature. Additional studies are needed to determine the true incidence of this entity, its natural history, and treatment guidelines.
References:

1. Gerosa A, Fanti A, Del Sette B, et al. Posttraumatic middle meningeal artery pseudoaneurysm: Case report and review of the literature. World Neurosurg. 2019;128:225-29

2. Bozzetto-Ambrosi P, Andrade G, Azevedo-Filho H. Traumatic pseudoaneurysm of the middle meningeal artery and cerebral intraparenchymal hematoma: Case report. Surg Neurol. 2006;66:529-31

3. Bruneau M, Gustin T, Zekhnini K, Gilliard C. Traumatic false aneurysm of the middle meningeal artery causing an intracerebral hemorrhage: Case report and literature review. Surg Neurol. 2002;57(3):174-78

4. Bezircioǧlu H, Erşahin Y, Demirciɔ F, et al. Nonoperative treatment of acute extradural hematomas: Analysis of 80 cases. J Trauma. 1996;41(4):696-98

5. Shah Q, Friedman J, Mamourian A. Spontaneous resolution of traumatic pseudoaneurysm of the middle meningeal artery. Am J Neuroradiol. 2005;26(10):2530-32

6. Srinivasan A, Lesiuɔ H, Goyal M. Spontaneous resolution of posttraumatic middle meningeal artery pseudoaneurysm. Am J Neuroradiol. 2006;27(4):882-83

7. Lillequist B. Roentgenologic appearances of traumatic lesions of middle meningeal artery: Report of two cases. Acta Radiol. 1967;6(6):513-18

8. Namba K, Ban S, Oda Y, et al. [Case of traumatic false aneurysm of the middle meningeal artery with epidural hematoma – comparison with a case resulting in spontaneous thrombosis]. Rinsho Shinkeigaku. 1972;12(4):165-70 [in Japanese]

9. Schulze A. [Unusual forms of epidural hematoma.] Zentralbl Neurochir. 1957;17(1):40-47 [in German]

10. Montanari E, Polonara G, Montalti R, et al. Delayed intracerebral hemorrhage after pseudoaneurysm of middle meningeal artery rupture: Case report, literature review, and forensic issues. World Neurosurg. 2018;117:394-410

11. Jussen D, Wiener E, Vajkoczy P, Horn P. Traumatic middle meningeal artery pseudoaneurysms: Diagnosis and endovascular treatment of two cases and review of the literature. Neuroradiology. 2012;54(10):1133-36

12. Tsutsumi M, Kazekawa K, Tanaka A, et al. Traumatic middle meningeal artery pseudoaneurysm and subsequent fistula formation with the cavernous sinus – Case report. Surg Neurol. 2002;58(5):325-28

13. Lim D-H, Kim T-S, Joo S-P, Kim SH. Intracerebral hematoma caused by ruptured traumatic pseudoaneurysm of the middle meningeal artery: A case report. J Korean Neurosurg Soc. 2007;42(5):416

14. Wu X, Jin Y, Zhang X. Intraparenchymal hematoma caused by rupture of the traumatic pseudoaneurysm of middle meningeal artery. J Craniofac Surg. 2014;25(2):111-13

15. Lu X, Zhang X. Onyx™ embolization for an angiographically progressive traumatic pseudoaneurysm of the middle meningeal artery: A case report and literature review. Exp Ther Med. 2019;17(5):4144-48

16. Heit JJ, Pastena GT, Nogueira RG, et al. Cerebral angiography for evaluation of patients with CT. Am J Neuroradiol. 2016;37:297-304

17. Yang ZL, Ni QQ, Schoepf UJ, et al. Small intracranial aneurysms: Diagnostic accuracy of CT angiography. Radiology. 2017;285(3):941-52

18. Paiva WS, Andrade AF, Amorim RLO De, et al. Computed tomography angiography for detection of middle meningeal artery lesions associated with acute epidural hematomas. Biomed Res Int. 2014;2014:413916

19. Kο SF, Huang CC, Ng SH, et al. MDCT angiography for evaluation of the complete vascular tree of hemodialysis fistulas. Am J Roentgenol. 2005;185(5):1268-74

20. Inamasu J, Nakamura Y, Saito R, et al. “Occult” ruptured cerebral aneurysms revealed by repeat angiography: Result from a large retrospective study. Clin Neurol Neurosurg. 2003;105(1):33-37