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Endovascular treatment of a supraclinoid internal carotid artery fenestration aneurysm: A case report and literature review

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ABSTRACT

Supraclinoid internal carotid artery (ICA) fenestration aneurysm is rare. Except for open surgery, endovascular treatment (EVT) is considered an alternative for such an aneurysm. However, experience with this procedure is lacking. Therefore, we reported such a case. A 61-year-old woman suffered subarachnoid hemorrhage. Digital subtracted angiography (DSA) showed bilateral middle cerebral artery (MCA) aneurysms and a saccular aneurysm associated with fenestration of the supraclinoid ICA. Two MCA aneurysms were treated with single coiling, and the supraclinoid ICA fenestration aneurysm was coiled under stent assistance. The postoperative recovery was uneventful. At this time, a literature review was performed on the role of EVT in supraclinoid ICA fenestration aneurysms. A total of 13 supraclinoid ICA fenestration aneurysms treated by EVT in 11 cases, including our case, were obtained. After EVT, good outcomes were obtained in all cases. To our knowledge, this is the first study to review the role of EVT for supraclinoid ICA fenestration aneurysms. Our case report and literature review indicated that EVT for such aneurysms may be feasible and act as a therapeutic alternative.

1. Introduction

Supraclinoid internal carotid artery (ICA) fenestration is a rare condition [1–4]. Although the fenestration itself is asymptomatic, any associated aneurysms may rupture or become symptomatic [5–8]. At this time, treatment is necessary [9,10]. Clipping supraclinoid ICA fenestration aneurysms is a choice. In addition, endovascular treatment (EVT) can act as an alternative. Due to insufficient experience resulting from the small number of cases and the inherent complexities of the procedure, EVT for such aneurysms remains challenging. Here, we report a case of supraclinoid ICA fenestration aneurysm treated by EVT. Additionally, cases of supraclinoid ICA fenestration aneurysms treated by EVT were collected from PubMed, and a literature review was performed.

2. Case presentation and literature review

2.1. Case presentation

A 61-year-old woman presented with sudden headache, nausea and vomiting for 6 h. She was of Han Chinese nationality and was healthy. On physical examination, her condition was classified as grade I on the Hunt-Hess scale. Her limbs had grade V muscle

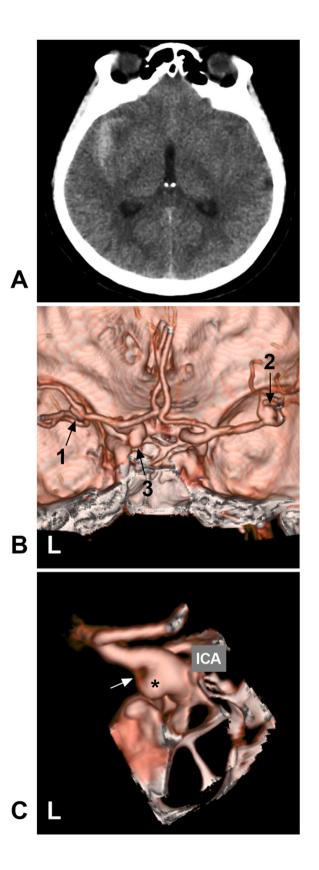
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Fig. 1. Preoperative CT and CTA images. A: CT showing subarachnoid hemorrhage, primarily in the right sylvian fissure. B: CTA showing three aneurysms at the bilateral middle artery bifurcations (arrows with numbers 1 and 2) and the left supraclinoid ICA (arrow with number 3). C: CTA showing a fenestration (arrow) at the supraclinoid ICA with an aneurysm (asterisk) in it.

Abbreviations: CT: computed tomography, CTA: computed tomography angiography, ICA: internal carotid artery.

strength. The Babinski sign was positive in both lower limbs. Computed tomography (CT) showed subarachnoid hemorrhage (SAH) focusing on the right sylvian fissure (Fig. 1A). CT angiography (CTA) showed bilateral middle cerebral artery (MCA) bifurcation mirror aneurysms and a saccular aneurysm associated with fenestration of the supraclinoid ICA (Fig. 1B and C). Based on the SAH primary location, the right MCA aneurysm was considered ruptured.

EVT of the three aneurysms was planned and performed under general anesthesia. Antiplatelet agents (clopidogrel 300 mg and aspirin 300 mg) were administered 3 h before EVT. First, EVT was performed to coil the ruptured right MCA aneurysm using an Echelon-10 microcatheter (Medtronic, Irvine, CA, USA). After packing the aneurysm with three Axium Prime coils (5 mm \times 15 cm, 2 mm \times 8 cm, and 1.5 mm \times 4 cm (Medtronic, Irvine, CA, USA)), EVT was considered complete (Fig. 2A). Then, the left MCA aneurysm was coiled using two Axium Prime coils (5 mm \times 15 cm and 2 mm \times 8 cm) (Fig. 2B). Finally, the saccular aneurysm in fenestration was coiled by using two Axium Prime coils (5 mm \times 20 cm, 2 mm \times 8 cm) under the assistance of a Neuroform Atlas stent (Stryker Neurovascular, Fremont, California, USA) (4.5 mm \times 21 mm) (Fig. 3A and B). The postoperative recovery was uneventful.

Preprocedural antiplatelet agents (clopidogrel 75 mg and aspirin 100 mg) were administered for three months. Then, follow-up digital subtracted angiography was performed and showed complete coiling of the three aneurysms (Fig. 4A–C). After follow-up, clopidogrel was suggested to be stopped, and aspirin was prescribed for half a year and then discontinued.

2.2. Literature review

Eligible English language literature was searched in the PubMed database (last search date was May 10, 2023). The key words "supraclinoid internal carotid artery", "fenestration" and "aneurysm" were combined for use. The reference lists of the identified articles were also screened. The flowchart of article collection is shown in Fig. 5. We collected 9 articles [6,11–18], including 10 cases

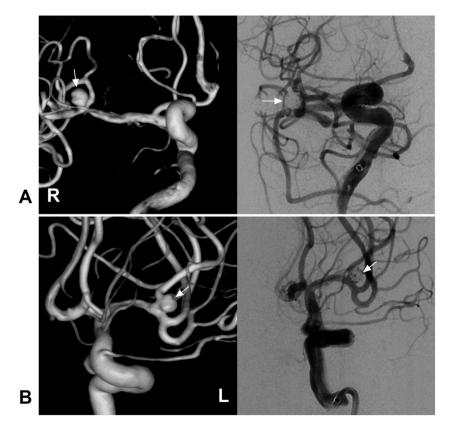


Fig. 2. EVT of bilateral MCA aneurysms. A: Left panel: DSA of the right ICA showing the aneurysm (arrow) at the right MCA bifurcation; Right panel: Postoperative DSA showing coiling of the aneurysm (arrow). B: Left panel: DSA of the left ICA showing the aneurysm (arrow) at the left MCA bifurcation; Right panel: Postoperative DSA showing coiling of the aneurysm (arrow).

Abbreviations: DSA: digital subtraction angiography, EVT: endovascular treatment, ICA: internal carotid artery, L: left, MCA: middle cerebral artery, R: right.

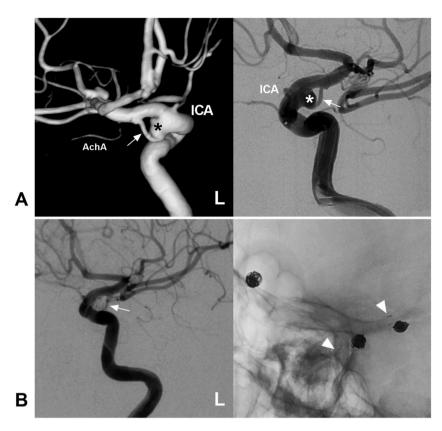


Fig. 3. EVT of supraclinoid ICA fenestration aneurysm. A: Three-dimensional DSA (left panel) and two-dimensional DSA (right panel) of the left ICA showing fenestration (arrows) at a supraclinoid ICA with an aneurysm (asterisks). The upper limb is thick, and the lower limb is thin. B: Left panel: DSA of the left ICA showing coiling of the aneurysm and preservation of the lower limb (arrow); Right panel: X-ray image showing the markers (arrowheads) of the stent to assist coiling.

Abbreviations: DSA: digital subtraction angiography, EVT: endovascular treatment, ICA: internal carotid artery, L: left, R: right.

with supraclinoid ICA fenestration aneurysm, including our case, and a total of 11 cases were reviewed. These 11 cases contained 14 aneurysms of supraclinoid ICA fenestration, 12 of which arose from the proximal fenestration and were in or beyond the fenestration, and 2 of which arose from the limb. Of 14 aneurysms, except for 1 aneurysm clipped, the other 13 aneurysms in 11 cases were treated by single coiling in 3 aneurysms, coiling with balloon/stent assistance in 7 aneurysms, and flow diversion deployment in 3 aneurysms. Of 13 aneurysms, the thinner limb of the fenestration was obstructed during EVT in 3 aneurysms. After EVT, all 11 patients achieved good outcomes without complications (Table 1).

3. Discussion

Artery fenestration is a congenital variant defined as segmental duplication, presenting with a vessel into two distinct endotheliumlined channels; it consists of a proximal portion, fenestrated limb and distal end [19,20]. Fenestrations of the intracranial arteries are observed in the anterior cerebral artery (ACA) in approximately 1% of autopsies and in the vertebrobasilar system (especially at the proximal segment of the basilar artery) in 1%–5% of autopsies [8,21]. Supraclinoid ICA fenestration is rare [6,7].

Failure of division or persistence of the plexiform may cause supraclinoid ICA fenestration. At the 4- to 5-mm embryological stage, the primitive ICA divides into the cranial and caudal divisions just distal to the ophthalmic artery (OphA), which are often connected with a small plexiform. The cranial division gives rise to the ACA, MCA and anterior choroidal artery (AchA), while the caudal division gives rise to the posterior communicating artery [22]. Embryology explains why supraclinoid ICA fenestration occurs between the OphA and AchA [3,7]. Based on the developmental condition of the fenestration, a supraclinoid ICA fenestration can be divided into large or small and symmetric with duplicate limbs or asymmetric with thick upper and thin lower limbs [19,23]. In our case, the supraclinoid ICA fenestration was large and asymmetric (Fig. 1C and 3A).

When the fenestration is large, an aneurysm may occur. In Haryu et al.'s review, 25 supraclinoid ICA fenestrations were collected, and 71% had lesion-associated aneurysms [7]. In a review by Filep et al., of 23 supraclinoid ICA fenestrations, 78% were associated with at least one aneurysm at the proximal and/or distal part [6]. These aneurysms often arise from the proximal fenestration and can grow very large beyond the fenestration [21]. Rarely, these aneurysms can also occur on the limb and grow inside or outside. Most supraclinoid ICA fenestration aneurysms are saccular. The formation may be due to arterial weakness or unusual hemodynamic

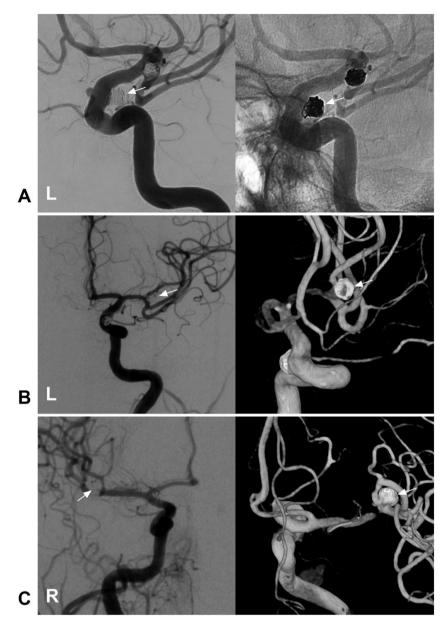


Fig. 4. Follow-up angiographic outcome of three aneurysms. A: DSA (left panel) and unsubtracted DSA (right panel) of the left ICA showing no recurrence of the supraclinoid ICA fenestration aneurysm. B: DSA (left panel) and three-dimensional DSA (right panel) of the left ICA showing no recurrence of the left MCA aneurysm. C: DSA (left panel) and three-dimensional DSA (right panel) of the right ICA showing no recurrence of the left MCA aneurysm.

Abbreviations: DSA: digital subtraction angiography, ICA: internal carotid artery, L: left, R: right.

stresses, similar to the morphology of the circle of Willis branch points [3]. Congenital factors may also play an important role because supraclinoid ICA fenestration aneurysms may be associated with other intracranial aneurysms, duplicate MCAs, persistent primitive trigeminal artery, arteriovenous malformation, etc. [20,24–26].

In our case, both factors can be considered because the saccular aneurysm was at the proximal portion of the supraclinoid ICA fenestration, likely reflecting arterial wall defects or unusual hemodynamic stresses, and the association with bilateral MCA aneurysms likely reflected a congenital factor.

For ruptured or symptomatic aneurysms with supraclinoid ICA fenestrations, treatment, including open surgery and EVT, is necessary [7]. Previously, open surgery was the mainstream method to resolve these aneurysms. In a review by Filep et al., a total of 23 cases with supraclinoid ICA fenestration were published between 1983 and 2018, and 18 cases had at least one associated aneurysm. EVT was undertaken in 8 aneurysms, and the rest were treated surgically [6].

Recently, EVT has been increasingly utilized for supraclinoid ICA fenestration aneurysms [6,19]. However, fenestration introduced

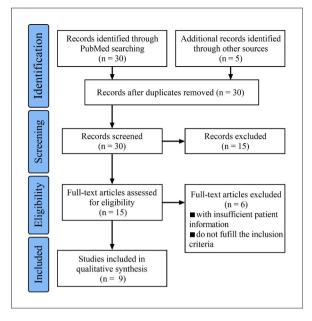


Fig. 5. Flowchart of collected articles.

additional complexity to EVT. During EVT, consideration should be given as to whether the fenestration limb should be preserved or sacrificed [6]. Because no notable perforators originate from the lower limbs, generally, compromise of a thin lower limb appears safe [6,19]. Flow-diverting stents with/without coil embolization can be a promising therapeutic option, especially for large or wide-necked aneurysms with difficult remolding, to avoid recurrence [11].

In our case, the supraclinoid ICA fenestration aneurysm was 5 mm in diameter and was a medium unruptured aneurysm. Due to the available EVT technique in our institute and previous reported experience, we coiled this supraclinoid ICA fenestration aneurysm under the assistance of the Neuroform Atlas stent [27]. Similar to other supraclinoid ICA fenestration aneurysms treated by EVT in previous reports, good outcomes were obtained.

However, due to stent assistance, the disadvantage of postoperative platelet antiaggregatory medication must be considered. For the Neuroform Atlas stent deployed in large vessels, such as in the ICA, due to the low metal volume and good apposition for conformation to the vessel wall, this stent may require antiplatelet therapy at a lower dose and over a shorter period than other stents. Dual antiplatelet therapy can be administered for 1–3 months, followed by single antiplatelet therapy for 6 months and then stopped, which can avoid the disadvantage of long-term platelet antiaggregatory medication [28]. Therefore, in our case, we chose the Neuroform Atlas stent to assist in coiling supraclinoid ICA fenestration aneurysms.

In addition, we had an additional consideration: the associated MCA mirror bifurcation aneurysms can be coiled together to solve all the aneurysms in this case. For MCA aneurysms, based on a systematic review and meta-analysis in 2015 comparing the efficacy and safety of coiling versus clipping for unruptured MCA aneurysms, surgical clipping is still recommended first and remains a better option [29]. However, with the development of techniques and equipment, current EVT is becoming a safe and effective therapy for unruptured MCA aneurysm management and results in a stable aneurysm occlusion rate during follow-up, especially for small and medium MCA aneurysms [28,30–33]. In some selective cases, such as MCA aneurysms in our case, they can be embolized by single coiling, and EVT is favored.

3.1. Limitations

Until now, including our case, there have been very few cases that can be collected in the literature, making it difficult to draw a clear conclusion for EVT of a supraclinoid ICA fenestration aneurysm. However, to our knowledge, this is the first study to review the role of EVT for supraclinoid ICA fenestration aneurysms, and it is still very valuable.

4. Conclusion

Our case report and literature review indicated that EVT for supraclinoid ICA fenestration aneurysms may be feasible and act as a therapeutic alternative.

Ethics approval and consent to participate

Ethics approval was not required by the authors' institution, as the present study is a case report. All methods were performed in

Table 1
Literature review data of cases with EVT for fenestration aneurysm.

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No.	Author/ year	Age/ sex	Onset	Preoperative status	Fenestration type	Fenestration aneurysm location	Fenestration aneurysm size	EVT for fenestration aneurysm	Management of other aneurysms	Complication	Follow up
1	Ichikawa et al., /2011 [12]	47/F	Headache	Good	Thick and thin limbs	One: arising from the proximal fenestration and beyond the fenestration	7 mm diameter	Balloon-assisted coiling with MRR class I; Sacrifice of lower limb	AchA aneurysm/ Balloon-assisted coiling	No	Good
2	Park et al., /2012 [13]	44/F	SAH	Good	Thick and thin limbs	One: arising from the proximal fenestration and beyond the fenestration	$3.5 \times 4.5 \text{ mm}$	Balloon-assisted coiling with MRR class I; Preservation of both limbs	MCA/Clipping at an early stage	No	Good
3	Nakiri et al., /2012 [14]	47/F	Headache	Good	Symmetric limbs	One: arising from the proximal fenestration and beyond the fenestration	$6.5 \times 12 \text{ mm}$	Coiling with MRR class II; Preservation of both limbs	No	No	Good
4	Nakiri et al., /2012 [14]	44/ M	SAH and IVH	Good	Symmetric limbs	Two: One arose from the proximal fenestration and beyond the fenestration; The other arose from the upper limb outside the fenestration	$5.5 \times 6 \text{ mm}$ and <2.5 mm diameter	Balloon and stent- assisted coiling the two aneurysms with MRR class I; Preservation of both limbs	No	No	Improved and no new symptoms
5	Ng et al., /2016 [15]	34/F	Headache and oculomotor paralysis	Good	Thick and thin limbs	Two: One arose from the proximal fenestration and beyond the fenestration; The other arose from the small limb above the fenestration	small and medium size	Clipping small aneurysm with sacrifice of thin limb; Stent-assisted coiling large aneurysm with MRR class I	No	No	3 months; Good
6	Sgreccia et al., /2018 [16]	55/ M	Acute ischemic stroke	Hemiparesis	Thick and thin limbs	One: arising from the proximal fenestration and beyond the fenestration	$10\times 11 \text{ mm}$	FD-assisted coiling; Preservation of both limbs	PcomA/FD coverage	No	6 months; mRS 0; complete occlusion of two aneurysms
7	Lee et al., /2018 [17]	65/F	Headache and dizziness	Good	Thick and very thin limbs	One: arising from the proximal fenestration and beyond the fenestration	medium size	Stent-assisted coiling with MRR class I; Preservation of both limbs	No	No	Good
8	Jha et al., /2018 [11]	60/F	Headache	Good	Symmetric limbs	Two: One arose from the proximal fenestration and in the fenestration; The other arose from the lower limb and in the fenestration	5- and 2-mm diameter	FD-assisted coiling of the large aneurysm; Sacrifice of the lower limb with small aneurysm by coiling	No	No	Good
9	Kasper et al., /2021 [18]	53/F	Migraine	Good	Symmetric limbs	One: arising from the proximal fenestration and in the fenestration	medium size	FD deployment in upper limb; Preservation of both limbs	No	No	2 years; Good; Regression of the aneurysm, lower limb occlusion
10	Filep et al., /2022 [6]	61/F	SAH and IVH	Drowsy	Thick and thin limbs	One: arising from the proximal fenestration and beyond the fenestration	$12\times 20 \text{ mm}$	Coiling with MRR class IIIb; Preservation of both limbs	MCA/Clipping at a later stage	No	15 days; mRS 1 with a slight disorientation
11	Present case/2023	61/F	SAH	Good	Thick and thin limbs	One: arising from the proximal fenestration and in the fenestration	5 mm diameter	Stent-assisted coiling with MRR class I; Preservation of both limbs	Bilateral MCAs/ Coiling at the same time	No	3 months; Good, No recurrence of three aneurysms, lower limb occlusion

Abbreviations: AchA: anterior choroidal artery, F: female, FD: flow diversion, IVH: intraventricular hemorrhage, M: male, MCA: middle cerebral artery, MRR: Modified Raymond-Roy, mRS: modified Rankin Scale, PcomA: posterior communicating artery, SAH: subarachnoid hemorrhage.

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accordance with the relevant guidelines and regulations. Informed signed consent to participate was obtained from the patient.

Consent for publication

The patient provided consent and agreed for all images and clinical data and other data included in the manuscript to be published.

Author contribution statement

All authors listed have significantly contributed to the investigation, development and writing of this article.

Data availability statement

Data will be made available on request.

Additional information

No additional information is available for this paper.

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Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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