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Case Report

Unruptured Saccular Aneurysm Arising from the Fenestrated A1 Segment of the Anterior Cerebral Artery: Report of 2 Cases

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Keywords

Anterior cerebral artery · Cerebral aneurysm · Fenestration · Clipping

Abstract

Some cases of aneurysms originating from the fenestrated A1 segment of the anterior cerebral artery (ACA) have been reported, but the pitfalls of the surgical procedure have not been well determined. We herein report 2 cases of a saccular aneurysm arising from the fenestrated A1 segment. Case 1 was a 72-year-old man incidentally diagnosed with an unruptured left ACA aneurysm on magnetic resonance imaging (MRI). Cerebral angiography revealed a saccular aneurysm arising from the proximal end of the left A1 segment. He underwent surgical clipping via the left pterional approach. The aneurysm originated from the proximal bifurcation of the fenestrated left A1 segment. A fenestrated ring clip was applied to obliterate the aneurysmal neck and one small fenestrated trunk, preserving the other fenestrated trunk and perforators

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around the fenestration. Case 2 was a 73-year-old man incidentally diagnosed with an unruptured ACA aneurysm on MRI. Cerebral angiography revealed a saccular aneurysm arising from the proximal end of the fenestrated left A1 segment. He underwent surgical clipping via the interhemispheric approach. The aneurysm originated from the proximal bifurcation of the fenestrated left A1 segment. A fenestrated ring clip was applied to obliterate the aneurysmal neck and one hypoplastic fenestrated trunk, preserving the other fenestrated trunk and perforators around the aneurysm. Detailed intraoperative evaluations of the anatomical structure and hemodynamics around the fenestration are important. The intentional obliteration of a fenestrated trunk and application of fenestrated clips need to be considered in difficult cases in order to expose the aneurysmal neck.

Introduction

Arterial fenestration has an angiographical incidence of 0.3–0.9% and is frequently associated with aneurysms [1]. Fenestrated arteries are commonly observed in the posterior circulation, including the basilar artery and vertebral artery [2]. Fenestrated arteries in the anterior circulation are usually observed in the anterior communicating artery but are rare in the others, including the anterior cerebral artery (ACA). Some cases of aneurysms arising from the fenestrated A1 segment of the ACA have been reported, but the pitfalls of the surgical procedure have not been well determined.

We herein report 2 cases of a saccular aneurysm arising from the fenestrated A1 segment and discuss the pitfalls of the surgical procedure in detail.

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Case 1

A 72-year-old man presented with slight dizziness. Magnetic resonance imaging (MRI) showed an unruptured left ACA aneurysm. Cerebral angiography revealed a saccular aneurysm with a maximum diameter of 10 mm arising from the proximal end of the left fenestrated A1 segment and a perforating artery arising from the rostral trunk of the fenestrated A1 segment (Fig. 1a, b). He underwent surgical clipping via the left pterional approach. The aneurysm originated from the proximal bifurcation of the fenestrated left A1 segment. A small perforating artery arising from the aneurysmal neck and 2 small perforating arteries arising from the rostral trunk of the fenestrated A1 segment were detected around the fenestration (Fig. 2a). The perforator arising from the aneurysmal neck and the proximal perforator artery arising from the rostral trunk of the fenestration were not identified using preoperative cerebral angiography. The aneurysmal body strongly adhered to the rostral small fenestrated trunk. A fenestrated ring clip was applied to obliterate the aneurysmal neck and rostral fenestrated trunk, preserving the caudal fenestrated trunk and perforators (Fig. 2b). Using a microvascular Doppler ultrasonography and fluorescein angiography, the patency of the perforating arteries and the obliteration of the aneurysm were confirmed (Fig. 2c, d). The remnant neck was obliterated with an additional clip (Fig. 2e). Postoperative angiography demonstrated complete obliteration of the aneurysm (Fig. 2f). Postoperative computed tomography (CT) did not demonstrate any abnormal findings. The postoperative course was uneventful, and the patient was discharged without any neurological deficits.

Case 2

A 73-year-old man presented with chronic headaches. MRI showed an unruptured left ACA aneurysm. Cerebral angiography revealed a saccular aneurysm with a maximum diameter of 7 mm arising from the proximal end of the fenestrated left A1 segment (Fig. 3a–c). He underwent surgical clipping via the interhemispheric approach. The aneurysm originated from the proximal bifurcation of the fenestrated left A1 segment and strongly adhered to the rostral hypoplastic fenestrated trunk. Two small perforating arteries arising from the rostral fenestrated trunk were detected around the fenestration (Fig. 4a). The proximal perforator was not identified using preoperative cerebral angiography. A fenestrated ring clip was applied to obliterate the aneurysmal neck and rostral fenestrated trunk, preserving the caudal fenestrated trunk and perforators (Fig. 4b). Using a microvascular Doppler ultrasonography and fluorescein angiography, the patency of perforating arteries and the obliteration of the aneurysm were confirmed (Fig. 4c). Postoperative angiography demonstrated complete obliteration of the aneurysm (Fig. 4d). Postoperative CT did not demonstrate any abnormal findings. The postoperative course was uneventful, and the patient was discharged without any neurological deficits.

Discussion

ACA fenestration has been reported in 0.1–7.2% of dissected autopsy specimens [2], which is more frequent than in the distal A1 segment. Fenestration of the A1 segment may occur due to the absence of the fusion of the plexiform anastomosis, which is present in the distal primitive ACA during the 18- to 43-mm stage of the embryo [3, 4]. This failure may increase blood flow in one of the A1 segments because of contralateral A1 hypoplasia, resulting in increased hemodynamic stress on the ipsilateral A1 segment. Increased blood flow and hemodynamic stress may prevent normal fusion of the plexiform anastomosis in the distal primitive ACA, resulting in the formation of A1 fenestration, and may also lead to the formation of aneurysms on the fenestrated A1 segment [5]. However, the pathophysiological mechanisms underlying the development of a fenestrated A1 segment have not yet been elucidated in detail.

Table 1 summarizes the clinical characteristics of 16 cases of aneurysms arising from the fenestrated A1 segment reported in the literature in the last 20 years, including the present cases [2, 3, 5–15]. The patients comprised 7 men and 8 women (no description in 1 case) with a mean age of 57.5 years (range 33–80). Nine aneurysms were located on the right side and 6 on the left side (no description in 1 case). Ten cases (62.5%) were ruptured. Twelve aneurysms were treated by neck clipping and 4 by endovascular treatments. The location of the aneurysm was described in detail in 13 cases, with 10 (76.9%) aneurysms developing on the proximal end of the fenestrated A1 segment. Hemodynamic stress may induce the formation of aneurysms at the bifurcation, and the development of a fenestrated A1 segment may also be strongly associated with hemodynamic stress at the site of the bifurcation.

Small arteries around the fenestrated A1 segment, such as perforating arteries and hypothalamic arteries, may not always be identified in digital subtraction angiograms and 3-dimensional CT angiograms due to the complexity of cerebral blood flow around them. In our cases, the perforating arteries around the fenestrated A1 segment were not preoperatively identified in radiological examinations but were detected during surgical exploration. The complex blood circulation of fenestrations may affect radiological detection. Detailed intraoperative evaluations of anatomical structures and hemodynamics around the fenestrated A1 segment are important for decision-making regarding the sacrifice or preservation of branches.

Aneurysms arising from the fenestrated A1 segment often develop on the proximal end of fenestrated trunks, which branch at an acute angle from the distal A1 segment. The neck and body of aneurysms commonly adhere strongly to fenestrated trunks, and the complete exposure of the aneurysmal neck or securing spaces for clip blades is difficult in some cases. In our cases, the aneurysms originated from the proximal bifurcation of the fenestrated A1 segment associated with strong adhesion to the fenestrated trunk, and the exposure of aneurysmal necks was difficult. Fenestrated ring clips, enclosing one larger fenestrated trunk, were applied to obliterate both the aneurysmal neck and the other fenestrated trunks that had a double blood supply from the fenestrated trunks. After clipping, the patency of perforating arteries was confirmed using a microvascular Doppler ultrasonography and fluorescein angiography. The intentional obliteration of the fenestrated trunk and application of fenestrated clips need to be considered in difficult cases under cautious observation of anatomical structures during surgery. In addition, it may be important that the patency of perforators around fenestrations is checked as much as possible using Doppler ultrasonography and fluorescent angiography after clipping.

Statement of Ethics

The patients consented to the publication.

Disclosure Statement

The authors declare that they have no conflicts of interest.

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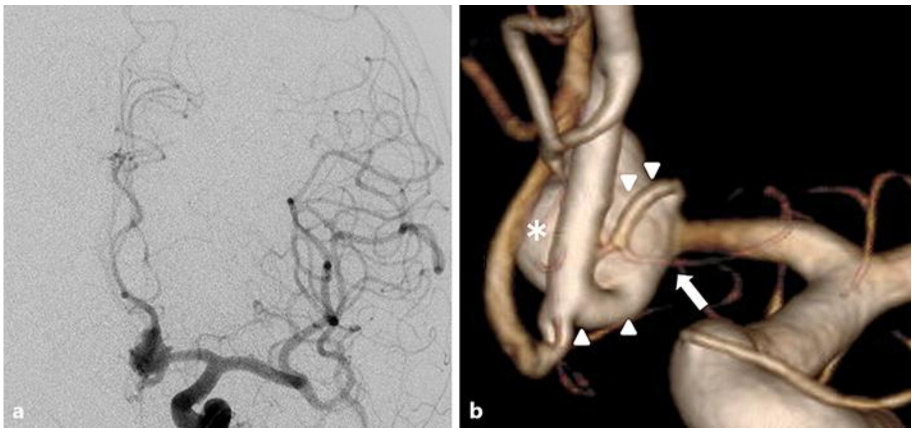

Fig. 1. Left cerebral angiography (**a**) and 3-dimensional rotational angiography (**b**) showing a saccular aneurysm (asterisk) arising from the proximal end of the left fenestrated A1 segment (arrowheads) and a perforating artery (arrow) arising from the rostral trunk of the fenestrated A1 segment.

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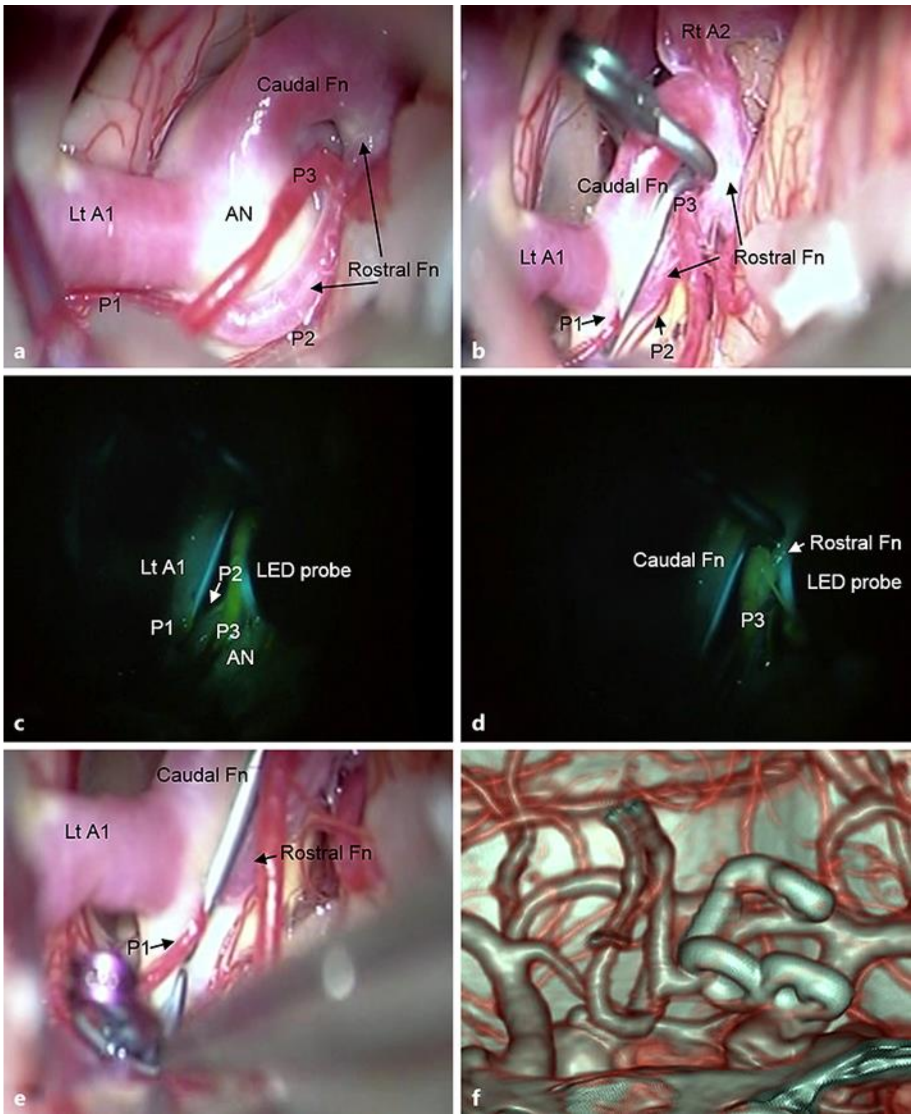

Fig. 2. **a** Intraoperative photograph before clipping showing an aneurysm (AN) originating from the proximal bifurcation of the fenestrated left A1 segment, a perforating artery (P1) arising from the aneurysmal neck, and 2 perforating arteries (P2, P3) arising from the rostral trunk of the fenestration (Rostral Fn). Caudal Fn, caudal trunk of the fenestration; Lt A1, left anterior cerebral artery A1 segment. **b** Intraoperative photograph after clipping showing a fenestrated ring clip attached to obliterate the aneurysmal neck and rostral fenestrated trunk. Rt A2, right anterior cerebral artery A2 segment. **c**, **d** Intraoperative photograph after clipping showing the patency of the parent arteries and of the perforating arteries and the obliteration of the aneurysm, which were confirmed by fluorescein angiography. LED probe, pencil type probe with a blue LED. **e** Intraoperative photograph after additional clipping for a remnant aneurysmal neck. **f** Postoperative 3-dimensional computed tomographic angiography showing complete obliteration of the aneurysm.

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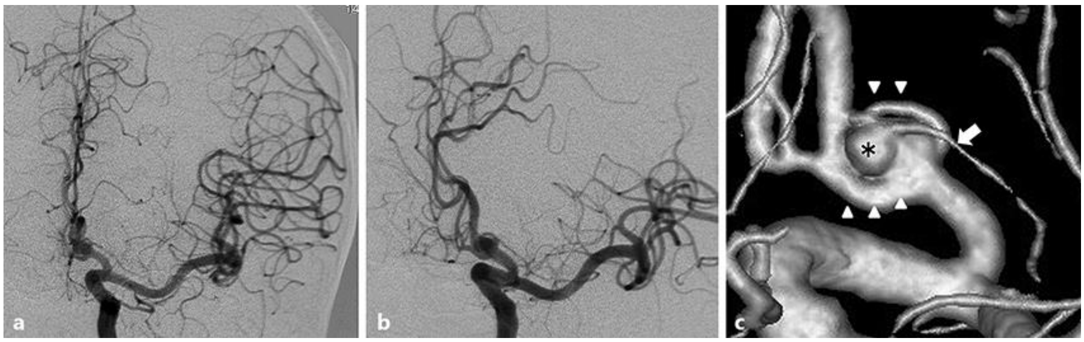

Fig. 3. Left cerebral angiography (**a**, **b**) and 3-dimensional rotational angiography (**c**) showing a saccular aneurysm (asterisk) arising from the proximal end of the left fenestrated A1 segment (arrowheads) and a perforating artery (arrow) arising from the rostral trunk of the fenestrated A1 segment.

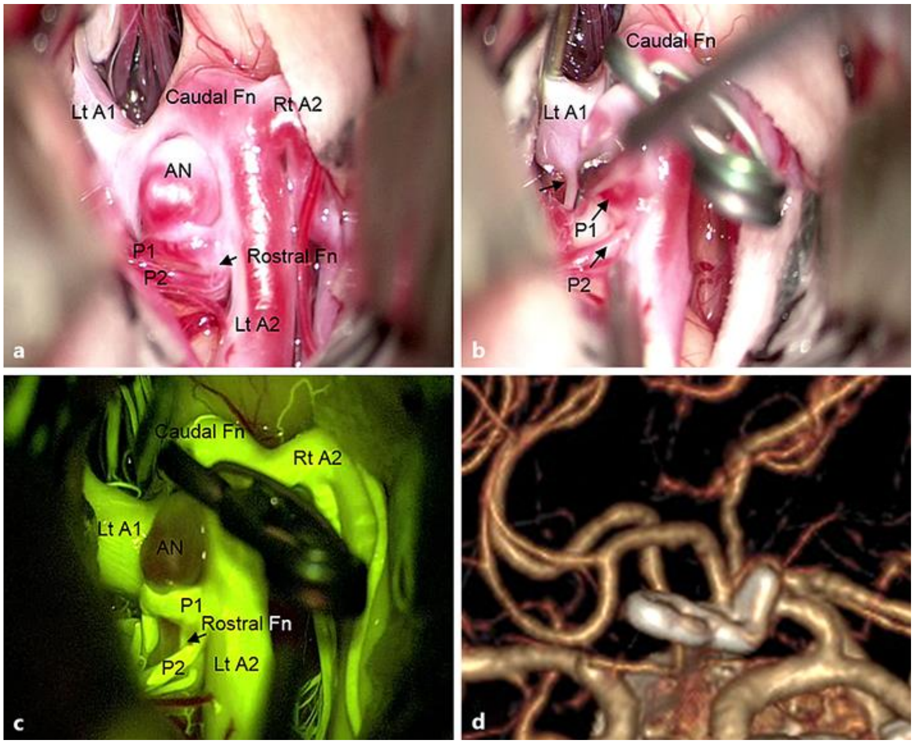

Fig. 4. a Intraoperative photograph before clipping showing an aneurysm (AN) originating from the proximal bifurcation of the fenestrated left A1 segment and 2 perforating arteries (P1, P2) arising from the rostral trunk of the fenestration (Rostral Fn). Caudal Fn, caudal trunk of the fenestration; Lt A1, left anterior cerebral artery A1 segment; Lt A2, left anterior cerebral artery A2 segment; Rt A2, right anterior cerebral artery A2 segment. **b** Intraoperative photograph after clipping showing a fenestrated ring clip attached to obliterate the aneurysmal neck and rostral fenestrated trunk. **c** Intraoperative photograph after clipping showing the patency of the parent arteries and of the perforating arteries and the obliteration of the aneurysm, which were confirmed by fluorescein angiography. **d** Postoperative 3-dimensional computed tomographic angiography showing complete obliteration of the aneurysm.

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Table 1. Literature review of aneurysms arising from A1 fenestrations

Authors [ref.]	Year	Age, years	Sex	Side	Ruptured or unruptured	Location	Treatment	Outcome
Friedlander and Oglivy [3]	1996	33	M	Rt	ruptured	proximal end	clipping	GR
Kachhara et al. [2]	1998	50	F	Rt	ruptured	proximal end	clipping	GR
Taylor et al. [6]	2000	68	M	Lt	ruptured	trunk	clipping	GR
Park et al. [7]	2000	35	F	Lt	ruptured	proximal end	clipping	GR
Wanibuchi et al. [8]	2001	52	F	Lt	unruptured	proximal end	clipping	GR
Ihara et al. [5]	2003	78	F	Rt	ruptured	trunk	clipping	GR
Terui et al. [9]	2010	70	M	Rt	unruptured	NR	clipping	GR
Mitsuhara et al. [10]	2011	71	F	Rt	ruptured	proximal end and trunk	coiling	GR
Mantatzis et al. [11]	2011	52	M	Rt	ruptured	NR	coiling	GR
Mantatzis et al. [11]	2011	39	NR	Lt	unruptured	proximal end	coiling	GR
Aktüre et al. [12]	2012	50	F	Rt	ruptured	proximal end	clipping	GR
Kwon et al. [13]	2013	60	F	Rt	ruptured	proximal end	clipping	GR
Kumar et al. [14]	2013	47	F	Rt	ruptured	trunk	clipping	GR
Eto et al. [15]	2015	80	M	NR	unruptured	NR	coiling	GR
Present case	2018	72	M	Lt	unruptured	proximal end	clipping	GR
Present case	2018	63	M	Lt	unruptured	proximal end	clipping	GR

F, female; M, male; NR, not reported; Rt, right; Lt, left; GR, good recovery.