

An Unruptured Aneurysm Coexisting with an Infundibular Dilatation: A Case Report

Takao Kitamura, Yasuo Murai, Kazutaka Shirokane,
Fumihiro Matano, Takayuki Kitamura and Akio Morita

Department of Neurological Surgery, Nippon Medical School, Tokyo, Japan

Background: Infundibular dilatation (ID) is a funnel-shaped enlargement of the origin of cerebral arteries. The coexistence of an aneurysm and ID is relatively rare. Patients with IDs are rarely followed up. However, some IDs have been reported to develop into aneurysms with subsequent rupture. Here we report on a case of an aneurysm that coexisted with ID of the posterior communicating artery.

Case Presentation: A 51-year-old woman underwent magnetic resonance imaging (MRI) to check for aneurysms and other problems. MRI revealed an unruptured aneurysm of the right internal carotid artery, for which the patient was admitted to our hospital. Three-dimensional computed tomographic angiography revealed an aneurysm, which protruded outward, and ID of the posterior communicating artery, which protruded inward. A right pterional craniotomy was performed with aneurysm clipping. The postoperative course was uneventful. In this report, we demonstrate operative views of the aneurysm and ID with the use of neuroendoscopy.

Conclusion: ID can develop into a true arterial aneurysm and potentially rupture. Therefore, we need to observe the patients with IDs carefully, particularly in young women.

(J Nippon Med Sch 2016; 83: 268–271)

Key words: carotid artery, cerebral aneurysm, infundibular dilatation, neuroendoscopy

Introduction

A funnel-shaped dilatation of the posterior communicating artery occurs at its origin from the internal carotid artery is called an infundibular dilatation (ID). While the incidence of ID slightly differs among different populations, it has been reported to be observed in 6%–8% of cerebral angiography or autopsy cases, and this percentage increases with age^{1–3}. In actual clinical practice, few cases of ID undergo follow-up. However, it has been reported that in some cases, ID can progress into an aneurysm, which ruptures and leads to subarachnoid hemorrhage^{4–17}. We experienced a case of an unruptured cerebral aneurysm, which was believed to have coexisted with ID on the basis of preoperative imaging and intraoperative findings. Here we report on this case along with a literature review regarding the relationship between IDs and aneurysms and the associated changes.

Case Presentation

The patient was a 51-year-old woman with a history of hypertension and dyslipidemia. Her family history revealed that her mother had died from a subarachnoid hemorrhage. An aneurysm of the right internal carotid artery was identified on brain screening, and the patient was referred to our hospital for surgery. When the diagnosis of a right internal carotid posterior communicating (IC-PC) cerebral aneurysm was made, the patient was admitted for surgery.

The patient's consciousness level was 15 on the Glasgow Coma Scale. No neurological deficits were observed. Three-dimensional computed tomographic angiography (3D-CTA) revealed a cerebral aneurysm in the right IC-PC, which was 5 mm in size, and ID protruding posteriorly (**Fig. 1A, B**).

Clipping was performed through a right pterional craniotomy. After opening the sylvian fissure and verifying

Correspondence to Takao Kitamura, MD, Department of Neurological Surgery, Nippon Medical School, 1–1–5 Sendagi, Bunkyo-ku, Tokyo 113–8602, Japan

E-mail: taka-kitamu@nms.ac.jp

Journal Website (<http://www.nms.ac.jp/jnms/>)

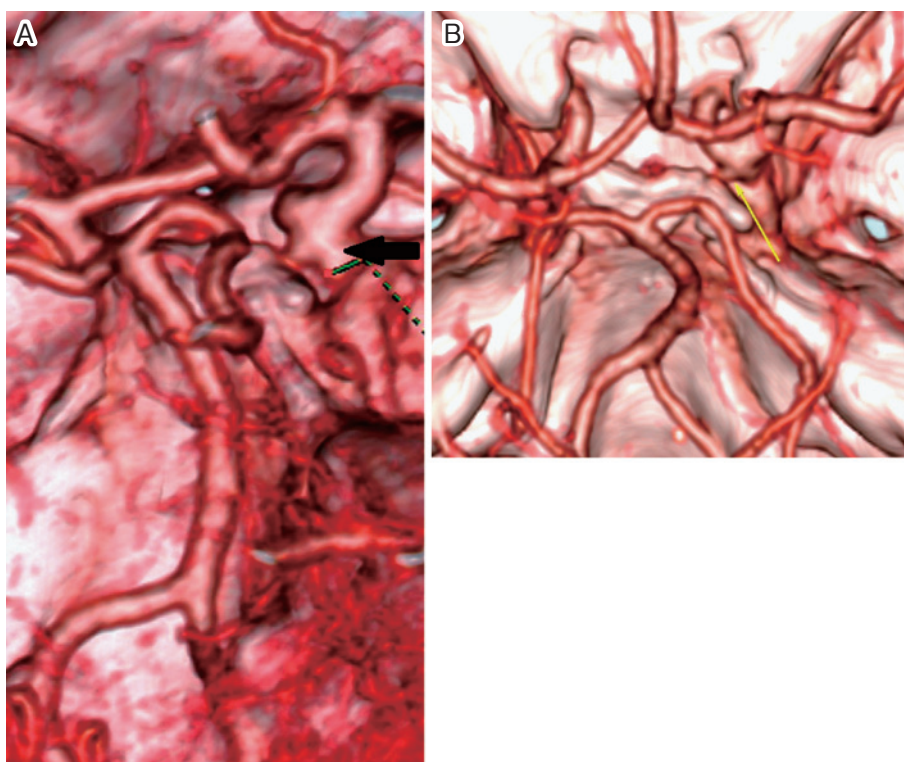


Fig. 1 Preoperative three-dimensional computed tomographic angiography (3D-CTA) on admission

(A) Right internal carotid posterior communicating (IC-PC) artery aneurysm protruding outward (**arrow**)

(B) Infundibular dilatation (ID) of the posterior communicating artery protruding inward (**arrow**). Arrow head indicates posterior communicating artery.

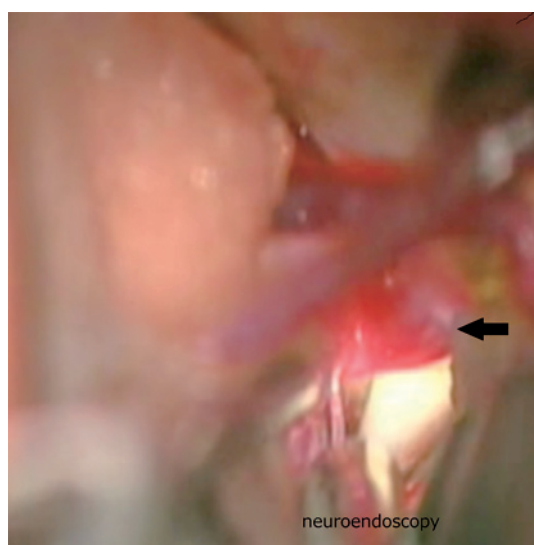


Fig. 2 Operative view

Operative view showing an aneurysm at the origin of the internal carotid posterior communicating (IC-PC) artery (**arrow**)

the internal carotid artery from behind with a microscope, the aneurysm in the internal carotid artery, which had been observed on preoperative imaging, was seen to be attached to the lower region of the dura mater (Fig. 2). The neck of the aneurysm was observed using a neuroendoscope, which revealed a funnel-shaped reddish enlargement without thinning or irregularity of the vascular wall at the tip of which was the posterior communicating artery; therefore, the presence of ID was confirmed (Fig. 3). Neck clipping was performed only for the aneurysm, and ID was avoided. Surgery was completed without coating of ID.

Postoperative diffusion-weighted magnetic resonance imaging performed on the day following surgery revealed no intracranial hemorrhage or findings suggestive of infarction. There were no neurological deficits, and the mini-mental state examination score was 30 points. On day 16 of hospitalization, the patient was discharged and returned home.

Discussion

We treated a case of an IC-PC aneurysm with ID. The vascular wall was thinner in the aneurysm than in ID. The border between the aneurysm and ID was clearly confirmed using neuroendoscopy, and aneurysmal clipping was performed. In addition, we reviewed the literature regarding unruptured IC-PC aneurysms with concurrent IDs.

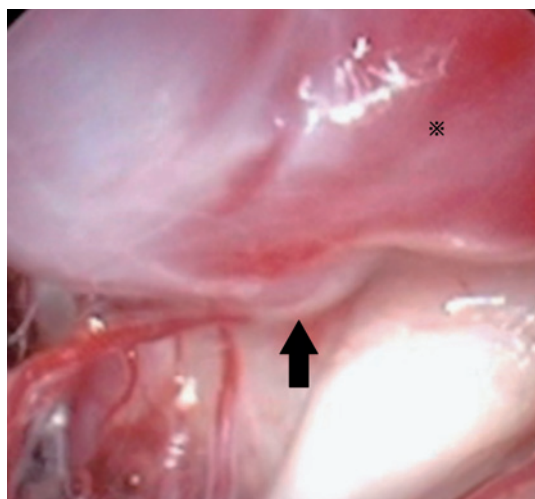


Fig. 3 Neuroendoscopic view
Enlarged infundibular dilatation (ID) (**arrow**) contiguous with the internal carotid posterior communicating (IC-PC) artery aneurysm (*)

The definition of ID on cerebral angiography, as used by Pool et al.¹⁸, includes the following diagnostic criteria: 1) the posterior communicating artery is found at the tip of ID; 2) the maximum diameter is 3 mm or less; 3) there is no aneurysm-like neck present; and 4) the shape is round or conical with no irregularity. Kubota et al.¹⁹ reported that 3D-CTA, which has high spatial resolution, is a useful means of distinguishing aneurysms from IC-PC vascular protrusions, which are difficult to be differentiated by digital subtraction angiography.

Various reports have discussed whether or not ID is a preaneurysmal state^{12,15,20}. It is believed that IDs can progress into cerebral aneurysms because the decreased blood flow velocity in ID due to its shape increases pressure on the vascular wall, which then ruptures the internal elastic membrane, damaging the intima at ID and developing into an aneurysm.

In summary, in past reports regarding concurrent ID and cerebral aneurysms (Table 1), the mean age at the time of diagnosis was 42.1 years⁴⁻¹⁷. Regarding gender, 12 out of 14 patients reported were female. Furthermore, 9 patients presented with multiple aneurysms, including 6 with symmetrical IC-PC aneurysms, all of which developed from IDs. Based on these results, we believe that a more rigorous follow-up is required for IDs in young women, with multiple aneurysms, and particularly, in

Table 1 Summary of previously reported cases with the progression of infundibular dilatation (ID) to aneurysm

Year	Author	Sex/Age	Time to AN ^{a)} Formation (year)	Site of ID	SAH ^{b)}	Site of Associated AN	HT ^{c)}	
1	1962	Bjorksten ⁴	F/40	11	Lt	+	-	-
2	1966	Drake ⁵	F/34	4	Lt	+	Rt IC-PC ^{d)}	-
3	1970	Stuntz ⁶	M/36	9	Lt	+	Rt IC-PC	-
4	1971	Young ⁷	F/29	4	Lt	+	Rt IC-PC	+
5	1974	Yoshimoto ⁸	F/56	8	Lt	-	-	-
6	1979	Waga ⁹	F/36	9	Lt	-	Lt ACA ^{e)}	-
7	1981	Trasi ¹⁰	F/26	0.5	Lt	-	-	-
8	1983	Itakura ¹¹	F/26	7	Rt, Lt	+	ICA Bif ^{f)}	-
9	1983	Patrick ¹²	F/35	9	Rt	+	Lt IC-PC	-
10	1986	Kamiya ¹³	F/49	3	Lt	-	-	-
11	1998	Marshman ¹⁴	F/49	5	Lt	+	Rt MCA ^{g)}	+
12	2002	Carolina ¹⁵	F/53	1.1	Rt	+	Lt IC-PC, Rt MCA	-
13	2005	Chokyu ¹⁶	F/57	5	Rt	+	Lt IC-PC, MCA, Rt MCA	+
14	2009	Nashimoto ¹⁷	M/63	4	Lt	+	-	+
15	2014	Present case	F/51	-	Rt	-	-	+

^{a)} AN: aneurysm

^{b)} SAH: subarachnoid hemorrhage

^{c)} HT: hypertension

^{d)} IC-PC: internal carotid posterior communicating artery

^{e)} ACA: anterior cerebral artery

^{f)} ICA Bif: internal carotid artery bifurcation

^{g)} MCA: middle cerebral artery

patients with symmetrical IC-PC aneurysms. History of hypertension did not show any relationship.

In our patient, the use of neuroendoscopy revealed the posterior communicating artery at the tip of ID, and we were able to clearly confirm the absence of thinning and irregularity of the ID wall. This enabled a more adequate observation of the boundary between the aneurysm and ID, leading to the safe completion of the clipping. Although intraoperative observation by neuroendoscopy has revealed a relationship between the aneurysm and ID in only a few cases, some reports have shown that the use of neuroendoscopy in cerebral aneurysmal clipping enabled a more precise examination of the cerebral aneurysm and surrounding structures. Furthermore, it has been reported that with neuroendoscopy, the state of the blood vessels before and after the aneurysm can be easily ascertained, which is useful in distinguishing aneurysms of the internal carotid artery from IDs, which are difficult to be distinguished by preoperative cerebral angiography^{21,22}.

Conclusion

We reported a case of an unruptured cerebral aneurysm, which was believed to have coexisted with ID. It is possible that ID is a preaneurysmal state. Taking into consideration our case and past reported cases, we believe that we need to observe patients with IDs carefully, particularly in young women.

Conflict of Interest: The authors declare no conflict of interest.

References

1. Bisaria KK: Anomalies of the posterior communicating artery and their potential clinical significance. *J Neurosurg* 1984; 60: 572-576.
2. Saltzman GF: Infundibular widening of the posterior communicating artery studied by carotid angiography. *Acta Radiol* 1959; 51: 415-421.
3. Watanabe M, In S, Kuramoto S: Junctional dilatation originating from posterior communicating artery (in Japanese). *Neurological Surgery* 1975; 3: 917-924.
4. Bjorksten G, Troupp H: Changes in the size of intracranial arterial aneurysms. *J Neurosurg* 1962; 19: 583-588.
5. Drake CG: On the surgical treatment of ruptured intracranial aneurysms. *Clin Neurosurg* 1966; 13: 122-155.
6. Stuntz JT, Ojemann GA, Alvord EC: Radiographic and histological demonstration of an aneurysm developing on the infundibulum of the posterior communicating artery. *J Neurosurg* 1970; 33: 591-595.
7. Young B, Mecham F, Allen JH: Documented enlargement and rupture of a small arterial sacculatation: Case report. *J*

8. Yoshimoto T, Suzuki J: Surgical treatment of an aneurysm on the funnel-shaped bulge of the posterior communicating artery. *J Neurosurg* 1974; 41: 377-379.
9. Waga S, Morikawa A: Aneurysm developing on the infundibular widening of the posterior communicating artery. *Surg Neurol* 1979; 11: 125-127.
10. Trasi S, Vincent LM, Zingesser LH: Development of aneurysm from infundibulum of posterior communicating artery with documentation of prior hemorrhage. *AJNR Am J Neuroradiol* 1981; 2: 368-370.
11. Itakura T, Ozaki F, Nakai E, Fujii T, Hayashi S, Komai N: Bilateral aneurysms formation developing from junctional dilatation (infundibulum) of the posterior communicating arteries. *J Neurosurg* 1983; 58: 117-119.
12. Patrick D, Appleby A: Infundibular widening of the posterior communicating artery progressing to true aneurysm. *Br J Radiol* 1983; 56: 59-60.
13. Kamiya K, Inagawa T, Ogasawara H, Yano T: Aneurysmal development and enlargement from the infundibular dilatation documented by cerebral angiography: A case report (in Japanese). *Med J Shimane Pref Cent Hosp* 1987; 15: 86-90.
14. Marshman LA, Ward PJ, Walter PH, Dossetor RS: The progression of an infundibulum to aneurysm formation and rupture: case report and literature review. *Neurosurgery* 1998; 43: 1445-1449.
15. Martins C, Macanovic M, Silva I, Griz F, Azevedo HR: Progression of an arterial infundibulum to aneurysm. *Arq neuropsiquiatr* 2002; 60: 478-480.
16. Chokyu I, Okawa T, Masuo O, Ogura M, Nakamura Y: Rupture of an internal carotid-posterior communicating arterial aneurysm originating from infundibular dilatation after five years follow-up: a case report (in Japanese). *Practical Currently* 2005; 15: 964-969.
17. Nashimoto T, Saito T, Kurashima A, Seki Y: Rupture of small aneurysm originating from an infundibular dilatation—Case report. *Brain and Nerve (Tokyo)*; 2009; 61: 1177-1181.
18. Pool JL, Potts GD: In *Aneurysms and Arteriovenous Anomalies of the Brain: Diagnosis and Treatment*, 1965; pp 132-133, Harper and Row, New York.
19. Kubota T, Niwa J, Tanigawara T, Chiba M, Akiyama M, Inamura S: Differential diagnosis between aneurysm and infundibular dilatation in the IC-PC region with 3D-CTA (in Japanese). *Neurological Surgery* 2000; 28: 31-39.
20. Ohkuma H, Ebina K, Iwabuchi T: Angiographic study on infundibular dilatation of the posterior communicating artery. *Neurol Med Chir (Tokyo)* 1985; 25: 907-914.
21. Fujimoto S, Kusaka N, Adachi Y, Terai Y, Takasugi N, Yoshino K, Nishimoto A: Endoscopy-assisted microscopic operation for cerebral aneurysm (in Japanese). *Surgery for Cerebral Stroke* 1997; 25: 293-299.
22. Katayama M, Ohira T, Onozuka S, Mayanagi K, Fukunaga A, Akaji K, Kawase T: A case of enlarged infundibular dilatation diagnosed by vertebral angiograms with carotid compression and neuroendoscope. *Brain and Nerve (Tokyo)* 1999; 51: 253-257.

(Received, June 15, 2016)

(Accepted, October 19, 2016)