Craniopharyngioma resection and aneurysm clipping performed simultaneously by pure endoscopic endonasal approach: A case report and literature review

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Abstract. Craniopharyngioma is one of the most challenging issues for neurosurgeons as a brain tumor. Among the approaches of neurosurgery, in comparison to craniotomy, the endoscopic endonasal approach (EEA) has risen in popularity over the last two decades; unruptured intracranial aneurysms are relatively commonly found in the general population. The EEA as a new paradigm in the treatment of aneurysm has been reported to successfully clip dozens of cases of intracranial aneurysm. However, when reviewing the domestic and foreign literature, it appeared that cases of craniopharyngioma complicated with intracranial aneurysm purely treated by EEA have not been reported so far. In the present study, the published literature regarding endoscopic endonasal surgery for craniopharyngioma and intracranial aneurysms was reviewed, accompanied with a case of craniopharyngioma complicated with intracranial aneurysm, both of which were simultaneously treated by EEA.

Introduction

Craniopharyngioma is a common benign tumor type of the central nervous system. It accounts for 3% of total tumors and 4% of cranial tumors in children (1); however, the co-occurrence of craniopharyngioma and intracranial aneurysm is rarely reported. Fusiform dilation of the internal carotid artery (ICA) following resection of craniopharyngioma has been reported previously, but it has been suggested to occur as a result of surgical manipulation (2). For the first time, intracranial aneurysms were described to be associated to a certain extent with craniopharyngioma by Shida *et al* (3). In 2013, Takeuchi *et al* (4) present a case of anterior cerebral artery dissecting aneurysm associated with craniopharyngioma and discussed the relationship between these two lesions, but they did not perform any treatment of the craniopharyngioma. However, to the best of our knowledge, the treatment of both of the co-occurrence lesions purely by endoscopic endonasal approach (EEA) simultaneously has not been reported so far (search strategy shown as Data S1).

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Case report

A 62-year-old female was admitted to Huanhu Hospital (Tianjin, China) in August 2021. The patient had been diagnosed with intracranial space-occupying lesions 2 years previously with occasional double vision and had recently developed a headache, which was getting worse. Neurological examination of the patient revealed partial temporal visual field defects in both eyes. CT scan indicated a space-occupying lesion in the anterior tri-ventricle (Fig. 1), and together with the MRI findings (Fig. 2), a diagnosis of craniopharyngioma was determined. Specifically, on MRI, the tumor was irregularly defined, with a cyst-solid intra- and suprasellar cystic lesion expansively growing into the third ventricle, and indicated to be isointense on T1 and hyperintense on T2. The optic nerve, optic chiasma and pituitary stalk were obviously displaced under compression. Enhancement was inhomogeneous and the imaging specialist diagnosed craniopharyngioma empirically. CT angiography (CTA) was routinely scheduled for the patient, which revealed a 6.0x6.0x5.0 mm aneurysm originating at the C5 segment of the right internal carotid artery (ICA) (Fig. 3A). For further confirmation, digital subtraction angiography was performed, based on which the presence of the aneurysm was confirmed (Fig. 3B-D). After sufficient preoperative preparation, endoscopic endonasal transsphenoidal tumor resection and aneurysm clipping were carried out. Informed consent was obtained from the patient for the publication of the study.

After the general anesthesia accompanied by intravenous infusion of third-generation cephalosporins, the patient was placed in the supine position with the head elevated at 20˚ and slightly tilted to the operator's side. Following routine facial disinfection with chlorhexidine, the nasal cavity was disinfected with iodophor cotton balls and then filled with absorbent cotton soaked with epinephrine to facilitate its mucosal vasoconstriction, thus reducing intraoperative bleeding. A Karl Storz endoscope was adopted with 0 and 30˚ lenses, equipped with an image 1HUB HD camera (Karl Storz SE & Co. KG). A bilateral nasal access was applied but the surgical approach was with a predominantly right-sided nasal access. The endoscope with a 0˚ lens was inserted into the right nostril to determine the middle turbinate and middle nasal meatus, and followed the middle nasal meatus inward to localize the aperture of the sphenoidal sinus above the root of the middle turbinate at the junction with the nasal septum, and then a standard nasoseptal flap was prepared (the upper edge was kept 1 cm away from the olfactory fissure, avoiding to affect the sense of smell).

Special attention should be paid to the sphenopalatine artery or its branches in order to preserve the irrigation of the nasoseptal flap. The anterior wall of the sphenoid sinus was removed by a grinding drill or biting forceps. For a smooth operation, the posterior wall of the pterygoid sinus, also known as the saddle base, was required to be adequately exposed. The following anatomical landmarks were identified: The planum sphenoidale, tuberculum sellae, both optic nerve protuberance, medial and lateral optic-carotid recess, floor of the sella turcica, clivus and inferior margin of the sphenoid sinus, cavernous segment of ICA and clinoidal carotid artery protuberance. The enlarged posterior wall of the sphenoid sinus was obviously exposed and then removed. The dura was then prepared for incision and the dura was then incised around the optic chiasm in an I-beam pattern after satisfactory dural hemostasis by bipolar electrocoagulation. After dissection of the arachnoid, the tumor process was fully removed (Fig. 4).

The next step was to examine the aneurysm (Fig. 5), for which the dura at the right optic-carotid recess was initially gradually incised and the distal and proximal rings of the right ICA were then further opened, so as to clearly expose the C5 segment of the right ICA and the C4-C5 transition, thus obtaining manipulation of the ICA proximal to the aneurysm. It was further indicated that the apex of the aneurysm was medially oriented and was in close proximity to the optic nerve; the adherent arachnoid was gradually separated by a microdebrider and the aneurysm dome and the aneurysm neck were then carefully freed. The visualization and control of the aneurysm were robust and a simulation of the application of a test clip confirmed the possibility of safe clipping. The aneurysm was initially clipped with one straight FT 720 T Yaşargil clip (B. Braun Melsungen AG); however, a small amount of bleeding from the aneurysm appeared during the clamping process. The C4 segment of the ICA was temporarily blocked with an aneurysm clip applied for additional clamping, then the aneurysm was perfectly clamped without bleeding (Fig. 6). Finally, the fascia lata of the thigh with adipose tissue was excised and the skull base was repaired with a combined artificial dural patch and nasoseptal flap (5).

During the postoperative treatment, the patient did not report any symptoms of intracranial infection, cerebrospinal fluid leakage, serious water and electrolyte metabolic disorders or ischemic symptoms of cerebral tissue. A transient urinary collapse was reported, which was relieved after symptomatic treatment with desmopressin tablets. Finally, the diagnosis of craniopharyngioma was confirmed by pathology (Fig. 7) and a postoperative enhanced MRI indicated the complete tumor resection (Fig. 8). Furthermore, CTA indicated complete aneurysm clamping without any residual aneurysm (Fig. 9). Nasal endoscopic examination 2 weeks after the surgery indicated a stable saddle base restoration with abundant blood flow in the nasal mucosal flap. The patient was discharged from the hospital without any neurological deficiency. The patient did not receive any radiotherapy or chemotherapy after surgery, and no tumor recurrence was observed at the follow-up until now.

Figure 1. Coronal CT scan of sellar region: The supratentorial ventricle system was dilated, most of the tumors were in the suprasellar cisterna and parts of them protruded into the third ventricle with calcification. Localized bone defect was observed at the base of the sella region.

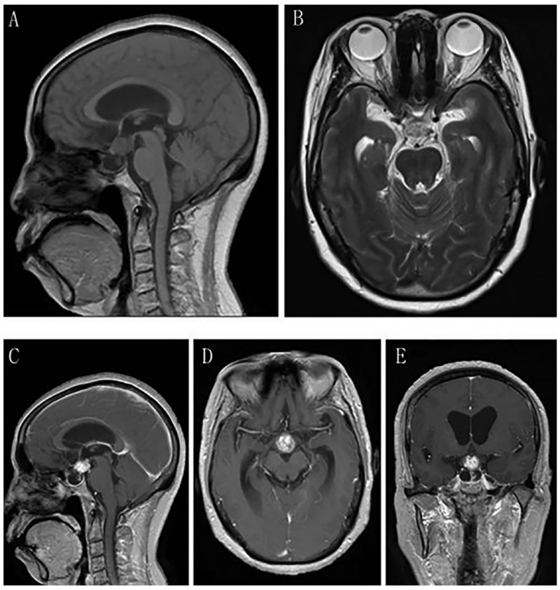

Figure 2. (A and B) On MRI, the mass was irregularly defined, with a cyst-solid intra- and suprasellar cystic lesion expansively growing into the third ventricle, and indicated to be (A) isointense on T1 and (B) hyperintense on T2. The optic nerve, optic chiasma and pituitary stalk were obviously displaced under compression. (C) Sagittal, (D) axial and (E) coronal images of enhanced MRI; the enhancement was inhomogeneous.

Discussion

Craniopharyngioma as a benign tumor type arising from the epithelial remnants of Rathke's pouch, commonly presenting in children with non-specific symptoms, accounting for 3% of total tumors and 4% of cranial tumors in children, with an

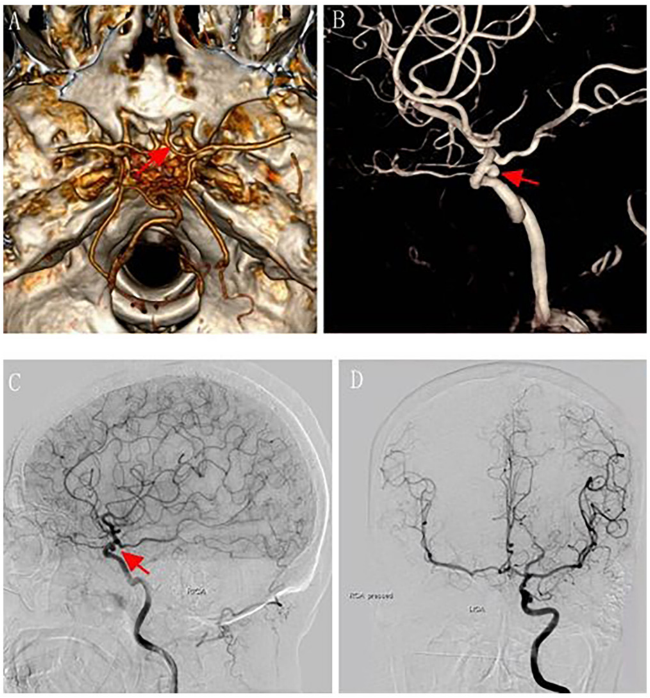

Figure 3. (A) On CT angiography, the size of the aneurysm was determined to be ~6.0x6.0x5.0 mm and observed to originate at the C5 segment of the right ICA. (B) 3D reconstruction of digital subtraction angiography. (C) The original lateral image; the results confirmed the presence of the aneurysm, of which the apex was projecting infero-medially; the red arrow points to the aneurysm. (D) Temporary balloon occlusion indicating that the anterior communicating artery was opened with the left ICA providing a good supply to the right middle cerebral artery, further confirming the relative safety of temporary occlusion of the right ICA during the operation. ICA, internal carotid artery.

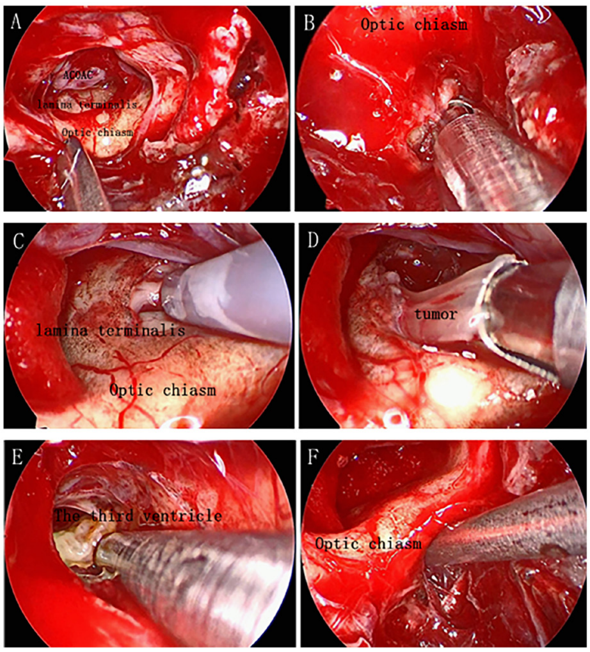

Figure 4. Intraoperative images at the ACOAC. (A) A cyst-solid tumor tissue was found in the suprasellar cistern, expansively growing into the third ventricle. (B) The suprasellar cisterna was accessed from below the optic chiasm. The internal tumor was resected with suction cutting pliers; (C) the lamina terminalis was then incised through the optic chiasm. (D) The tumor was removed gradually in sections. (E) Accessing the tricuspid ventricle for further resection of the residual tumor. (F) The tumor was completely removed. ACOAC, anterior communicating artery complex.

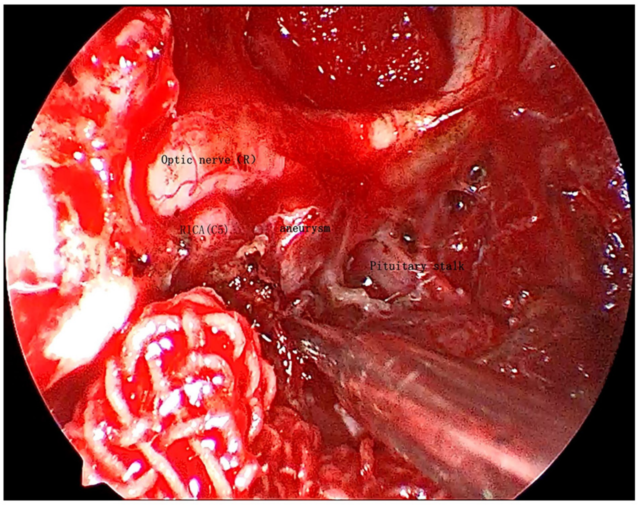

Figure 5. Aneurysm, ICA, optic nerve and pituitary stalk were fully exposed on endoscopy. RICA, right internal carotid artery.

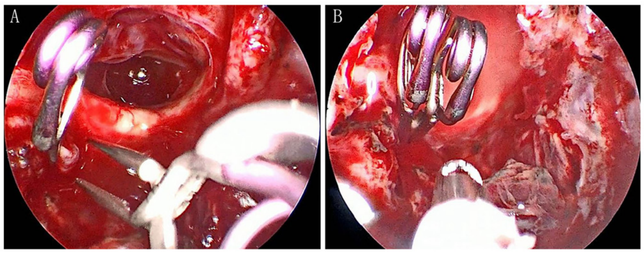

Figure 6. (A) Clipping the aneurysm with one straight FT 720 T Yaşargil clip. (B) A small amount of bleeding was found from the aneurysm and an aneurysm clip was added for additional clamping.

annual incidence of 0.05-0.20 per 100,000 individuals (1,6,7). Unruptured intracranial aneurysms are relatively common in the general population, with a prevalence of 3.2% (8). With the popularity and application of non-invasive imaging technology for intracranial vessels, the diagnosis of unruptured intracranial aneurysms is made with increasing frequency. However, to the best of our knowledge, the probability of simultaneous occurrence of craniopharyngioma and intracranial aneurysm is rather low, and there is a lack of related literature reporting whether craniopharyngioma has a role in the occurrence of intracranial aneurysm. Shida *et al* (3) argued that craniopharyngioma typically invades the brain, eliciting anaplastic transformations and an intense glial reaction in subjacent brain and vascular structures, thus contributing to the development of aneurysm; however, the hypothesis has also been put forward that the craniopharyngioma itself or cyst rupture produces chemicals that induce asymptomatic chemical meningitis, resulting in degeneration of the vessel wall and development of the dissecting aneurysm (4). However, in any case, scientific and uniform evidence is still lacking to sufficiently explain the relationship between craniopharyngioma and aneurysm occurrence. For selecting the optimal surgical approach, craniopharyngiomas may be generally classified into the following four subtypes: Intrasellar, intra-suprasellar, suprasellar and intra-third ventricle (9). Due to its close reach to hypothalamus, pituitary gland, optic nerve and

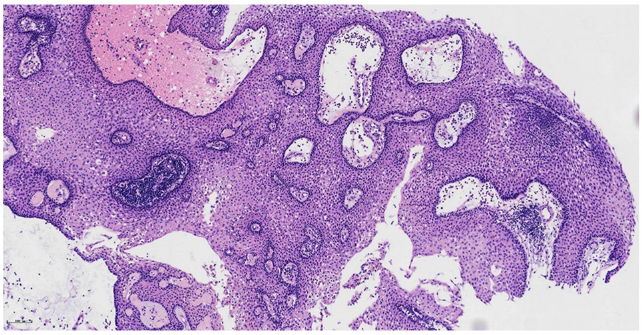

Figure 7. Histopathological evaluation revealed that craniopharyngioma (papillary type), peripheral glial proliferation with Rosenthal fiber formation and focal neuronal disorder (hematoxylin and eosin; magnification, x100).

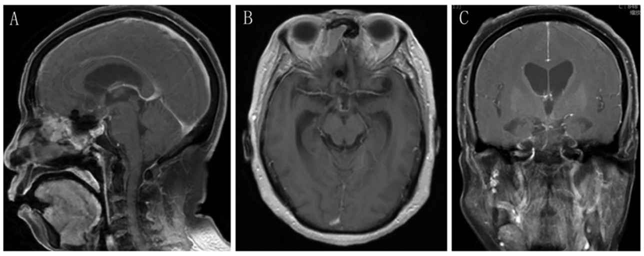

Figure 8. (A) Sagittal, (B) axial and (C) coronal images of enhanced MRI at the postoperative review. The result indicated complete tumor resection.

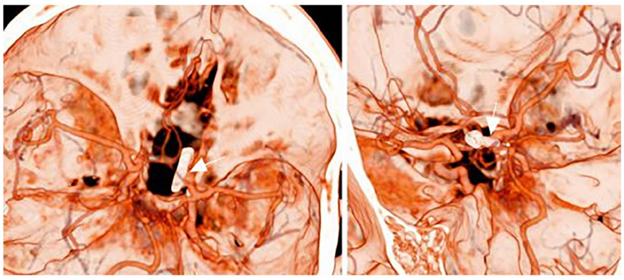

Figure 9. Postoperative review by CT angiography. The left is the view from the top downward angle and the right view is from the left back top side angle. The result indicated complete aneurysm clamping without residual aneurysm. The white arrow points to the aneurysm clips.

carotid artery, or other crucial nerve vessels, it is difficult to be completely removed, carrying a significant risk for recurrence and postoperative morbidity despite the general postoperative radiotherapy. The patient's tumor was completely removed. The team of clinicians suggested that radiotherapy should be decided at the follow-up of 3 months according to the results of the review. and in consideration of the wishes of the patient and the patient's family, the patient did not receive radiotherapy or chemotherapy. Transcranial approaches have always served as the main treatment for craniopharyngioma. In recent years, with the development of endoscope equipment and the enhancement of surgical techniques, the EEA has been widely adopted in the treatment of craniopharyngioma (10,11), despite its unique disadvantages, including cerebrospinal fluid (CSF) leakage and high incidence of intracranial infection, as well as the requirement of a nasal septum mucosal flap to repair the skull base and lumbar cisterna drainage to reduce the postoperative CSF leak, it has increased in popularity all over the world (12). Microsurgical clipping and endovascular coiling have always been adopted as the two main treatments for aneurysms (13). The pace of the progress in neurosurgeons' treatment and technological innovation for aneurysms have never stopped. EEA is a contributing technique for midline lesions of the skull base; however, to the best of our knowledge, only dozens of cases of intracranial aneurysm clipped by intranasal endoscopy have been reported so far (14,15) (Table I). As early as 2006, Kassam *et al* (16) successfully treated an intracranial aneurysm using an expanded EEA. Back in 2011, Fischer *et al* (17) reported that endoscopic enhancement of the visual field provided by the endoscope prior to, during and after microsurgical aneurysm occlusion may serve as a safe and effective application to increase the quality of treatment. In 2015, Gardner *et al* (18) reported a series of patients undergoing EEA for microsurgical clipping of intracranial aneurysm. In 2018, EEA clipping in 7 patients with 12 anterior circulatory aneurysms was reported by the group of Professor Hong Tao of the First Affiliated Hospital of Nanchang University (Nanchang, China) (15). However, craniopharyngioma complicated with aneurysm has been rather rarely reported and no previous case of craniopharyngioma complicated with intracranial aneurysm purely treated by EEA has been reported at once so far, to the best of our knowledge. The present case was the first in which the patient was admitted for a craniopharyngioma and prepared for endoscopic resection according to the established treatment plan; however, the presence of aneurysm was incidentally discovered during the preoperative examination. The considerations for simultaneous treatment of the two lesions under pure endoscopy were as follows: First, the aneurysm was located in the deep C5 segment of the ICA and preoperative cerebral angiography indicated that the tip of the aneurysm was facing medially; thus, it was difficult to maximize the exposure of the lesion, which, however, was resolved by adopting an endoscopic approach, which provided excellent access. Furthermore, the craniopharyngioma is mostly located in the anterior tri-ventricle, which is not contraindicated for the EEA according to the 'Kassam type' (19). Finally, it was a wide-necked aneurysm with the size of 6.0x6.0x5.0 mm; if adopting an interventional method, endovascular stent placement may be required for auxiliary embolization, which means the patient requires to take oral antiplatelet drugs after the interventional procedure, resulting in the impossibility to perform tumor resection in a short time, thus missing the best time for tumor treatment. The anatomical exposure technique of the cavernous sinus segment of the ICA under endoscopy is sophisticated but is feasible to perform by a skilled surgeon, so it was possible to confidently perform proximal flow control of the aneurysm without any significant risk of serious adverse effects such as intraoperative hemorrhage and postoperative cerebral infarction. Ultimately, in the present study, two lesions were successfully managed in one operation, which also further demonstrated that skull base lesions (tumor or vascular disorders) located in the midline area may be completely resolved by transnasal endoscopic surgery, depending on an adequate preoperative evaluation. However, this requires skilled neuro-endoscopic surgical experience and extensive knowledge of endoscopic anatomy, as well as advanced endoscopic surgical equipment and perfect teamwork. There is still

Table I. Summary of previously published cases of aneurysm clipped via the endonasal approach.

Author (year)	Patient age, sex	Clinical presentation	Location/size, mm	Complications	Outcome	(Refs.)
Kassam (2006)	51, F	Focal deficit	Verteb/11	None	Complete recovery	16
Kassam (2007)	56, F	Incidental finding	Sup Hyp/5	None	Complete recovery	20
Masahiko (2007)	58, F	Incidental finding	Acom/n.a	None	Complete recovery	21
Ensenat (2015)	74, F	SAH	PICA/1.2	CSF Leak	Complete recovery	22
Froelich S (2011)	55, M	Incidental finding	Acom/7	None	Complete recovery	23
Germanwala (2011)	42, F	SAH	Ophth/5Paracl/10	None	Complete recovery	24
Drazin (2012)	59, F	SAH	Bas.Tr/4	None	Repeat surgery for reclipping	25
Dehdashti A R (2015)	42, F	SAH	Bas.Ap/10	None	Endovascular coiling for residual neck	26
Dehdashti A R (2015)	70, F	SAH	Bas.Ap/5	Lacunar stroke	Neurological disability	26
Dehdashti A R (2015)	35, M	Focal deficits	PCA/9.4	Stroke CSF leak Meningitis	Neurological disability	26
Dehdashti A R (2015)	50, M	SAH	Bas.Tr/9	None	Complete recovery	26
Gardner (2015)	42, F	Incidental finding	Ophth/3.5	None	Complete recovery	18
Gardner (2015)	74, M	CN palsy	PCA/19	CSF leak Meningitis Lacunar stroke	Mild disability	18
Gardner (2015)	43, F	Incidental finding	Sup Hyp/5	CSF leak	Complete recovery	18
Gardner (2015)	47, F	Incidental finding	Bas.Ap/9	Lacunar stroke	Complete recovery	18
Gardner (2015)	45, M	Vision loss hypopituitarism	Ophth/giant Ophth/5	None	Complete recovery	18
Gardner (2015)	73, F	Incidental finding	Ophth/6	CSF leak Meningitis	Complete recovery	18
Gardner (2015)	45, F	SAH	Ophth/7	None	Complete recovery	18
Gardner (2015)	34, F	Incidental finding	Ophth/4	None	Complete recovery	18
Gardner (2015)	55, F	Incidental finding	Sup.Hyp/NA	None	Complete recovery	18
Gardner (2015)	42, F	Incidental finding	Sup.Hyp/NA	None	Complete recovery	18
Yildirim (2015)	72, F	Incidental finding	Acom/NA	None	Complete recovery	27
Xiao (2018)	42, M	SAH	Acom/7.2	None	Complete recovery	15
Xiao (2018)	63, F	Incidental finding	R.para/13.3[a] L.para/7.2 Acom/3.1	None	Endovascular coiling of the right large paraclinoid aneurysm	15
Xiao (2018)	61, F	Incidental finding	L.cav-ICA/7.9[a] R.par/10	None	Complete recovery	15
Xiao (2018)	52, M	Incidental finding	Acom.an/3.5	None	Complete recovery	15
Xiao (2018)	50, M	Incidental finding	Acom/5.7	None	Complete recovery	15
Xiao (2018)	45, F	Incidental finding	Acom/2.8	None	Complete recovery	15
Xiao (2018)	47, F	Incidental finding	L.para/4.2 L.oph/2.2L. cav-ICA/2.4[a]	None	Complete recovery	15
Present case	62, F	Double vision; headache	R.C5-ICA (R.para)	Transient urinary collapse	Complete recovery	/

[a]Indicated aneurysm was not clipped. F, female; M, male; R, right; L, left; SAH, subarachnoid hemorrhage; Acom, anterior communicating aneurysm; para, paraclinoid aneurysm; oph, ophthalmic aneurysm; cav-ICA, cavernous segment of ICA; Verteb, vertebral artery; BS, brain stem; Endovasc, endovascular; Sup Hyp, superior hypophyseal artery; S.medial, supero-medial; P.medial, postero-medial; Bas. Tr, basilar trunk; Bas. Ap, basilar apex; CN, cranial nerve; CSF, cerebrospinal fluid; I.medial, infero-medial; NA, not available; ICA, internal carotid artery; PICA, posterior inferior cerebellar artery.

a long way to go before the indications and contraindications of the procedure are fully demonstrated and the technique is widely used.

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Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Authors' contributions

The patient's surgery was performed by JYC and JMK. MCZ, JWL, JYC, HY and JM were involved in manuscript preparation and in data interpretation. All authors approved the final version of the manuscript. JWL and HY confirm the authenticity of all the raw data.

Ethics approval and consent to participate

Not applicable.

Patient consent for publication

Informed consent was obtained from the patient for the publication of the study.

Competing interests

The authors declare that they have no competing interests.

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