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Case Report

A Case of a Perianeurysmal Cyst Following Stent-Assisted Coil Embolization of an Unruptured Vertebral Artery Aneurysm

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Objective: To report the rare case of a patient with a perianeurysmal cyst following stent-assisted coil embolization of an unruptured vertebral artery aneurysm.

Case Presentation: A 63-year-old woman underwent stent-assisted coil embolization for an unruptured vertebral artery aneurysm embedded in the brainstem (pons). Complete occlusion of the aneurysm was successfully achieved. However, subsequent magnetic resonance imaging (MRI) conducted 8 months after the procedure showed perilesional edematous changes surrounding the aneurysm, and at 20 months, cyst formation was observed in the vicinity of the aneurysm. Progressive enlargement of the cyst eventually led to the development of paralysis and dysphagia, necessitating cyst fenestration surgery. Although postoperative reduction in the cyst size was achieved, the patient experienced complications in the form of aspiration pneumonia and bacterial meningitis, which resulted in a life-threatening condition.

Conclusion: Aneurysms embedded in the brain parenchyma should be carefully followed up, recognizing the risk of perianeurysmal cyst formation after coil embolization.

Keywords ▶ vertebral aneurysm, stent-assisted coil embolization, perianeurysmal cyst formation

Introduction

Intracranial perianeurysmal cysts are rare. Although there have been several reports of them, their pathogenesis has not been fully elucidated. There are reports of intracranial cystic lesions occurring in association with vascular malformations such as cavernous hemangiomas and cerebral arteriovenous malformations[1–3] and case reports of perianeurysmal cysts with findings that could seemingly be mistaken for brain tumors such as gliomas and hemangioblastomas.[4–6] Perianeurysmal cyst formation after coil embolization of aneurysms has been reported, although rarely,[7–9] and it is important to recognize the disease as a complication of treatment.

A case of perianeurysmal edematous changes after coil embolization of an unruptured vertebral artery aneurysm located within the brainstem, which subsequently became a perianeurysmal cyst and presented with symptoms of brainstem compression is presented. Consent for this submission was obtained from the patient's family.

Case Presentation

A 63-year-old woman with a history of hypertension presented with a chief complaint of visual impairment in her right eye. Magnetic resonance imaging (MRI)/magnetic resonance angiography (MRA) was performed to evaluate the chief complaint, and an unruptured aneurysm of the right vertebral artery was incidentally detected. The patient was then referred to our hospital. There were no abnormal neurological findings on the initial clinical examination. MRA showed a right vertebral artery aneurysm of a sidewall type having a broad neck, with no thrombosis or calcification within the aneurysm, and the aneurysm was

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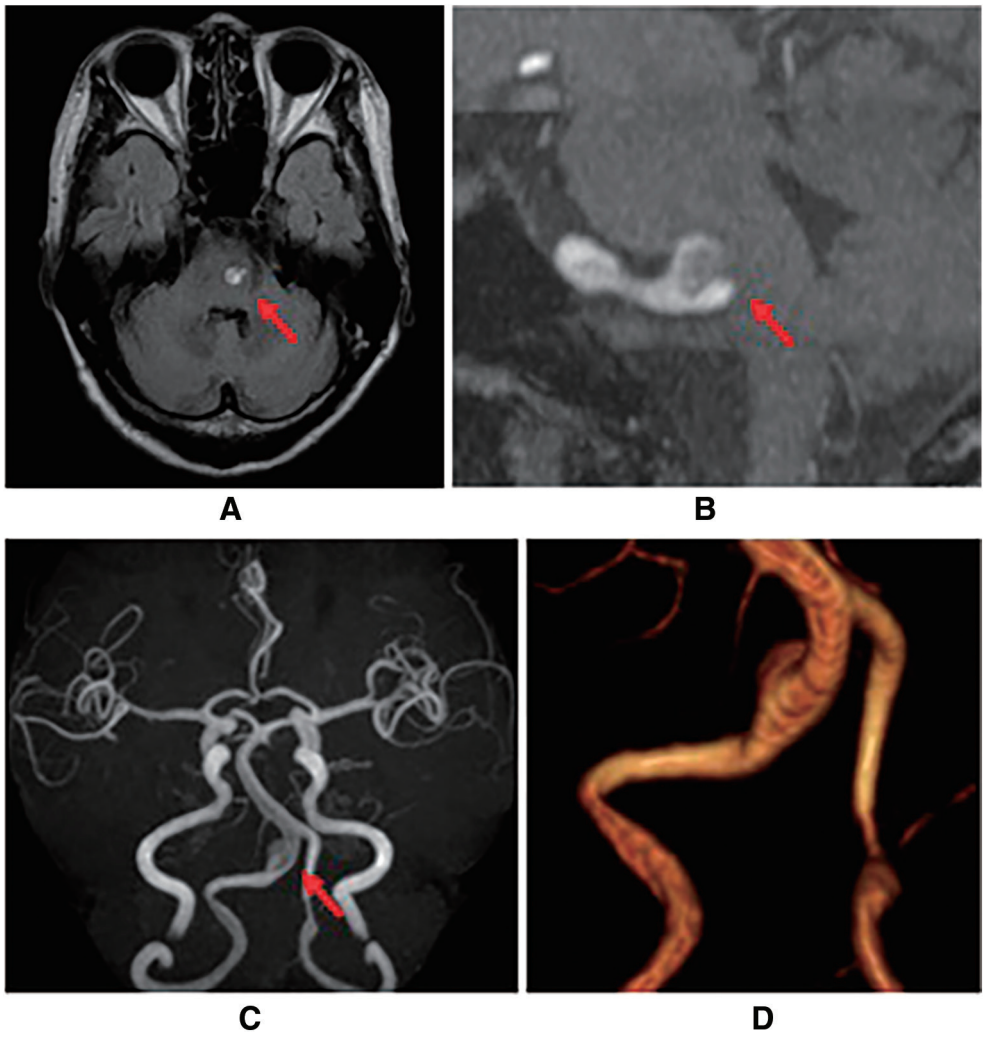


Fig. 1 Axial FLAIR (**A**) showing the aneurysm embedded in the pons without hemorrhage and edema (arrow). Sagittal time-of-flight MRA (**B**) showing the aneurysm embedded in the pons from below (arrow). MRA (**C** and **D**) showing the aneurysm in the right vertebral artery (arrow).

embedded in the brainstem from below (**Fig. 1**). Digital subtraction angiography (DSA) showed that the aneurysm had a maximum diameter of 7.9 mm and a neck diameter of 9 mm, with a small bleb (**Fig. 2**). Considering the size and irregular shape of the aneurysm, treatment suggestions were offered to the patient to prevent its rupture. Since the aneurysm was a posterior circulation lesion close to the brainstem, coil embolization was scheduled.

Two weeks prior to coil embolization, the patient was started on aspirin 100 mg and clopidogrel 75 mg. An LVIS 4.5 mm × 18 mm (Microvention TERUMO, Tustin, CA, USA) was deployed over the neck of the aneurysm. It was framed with a Target360 Soft 8 mm × 30 cm (Stryker, Kalamazoo, MI, USA) using the semi-jail technique, filled with Axium Prime (Medtronic, Minneapolis, MN, USA). Then Hydro Soft 3D (TERUMO) and ED coil Extra Soft (Kaneka Medix Corporation, Osaka, Japan) were inserted. A total of 11 coils were inserted for embolization, of which Hydro Soft 3D 3 mm × 6 cm and 1.5 mm × 3 cm coils were embolized as finishing coils at the end of embolization. The procedure was completed without intraoperative complications, and DSA performed 6 months after treatment showed that the aneurysm was completely occluded (**Fig. 3**). However, MRA performed 8 months after treatment showed no aneurysm recurrence or abnormal finding in the implanted stent. Still, a high-intensity signal appeared around the aneurysm embedded in the brainstem on FLAIR imaging (**Fig. 4**). Although the patient continued to have no symptoms, the high-intensity signal on FLAIR enlarged gradually. Suspecting edematous changes due to an allergic reaction to the coils and stents implanted in the aneurysm and the parent artery, the patient was started on

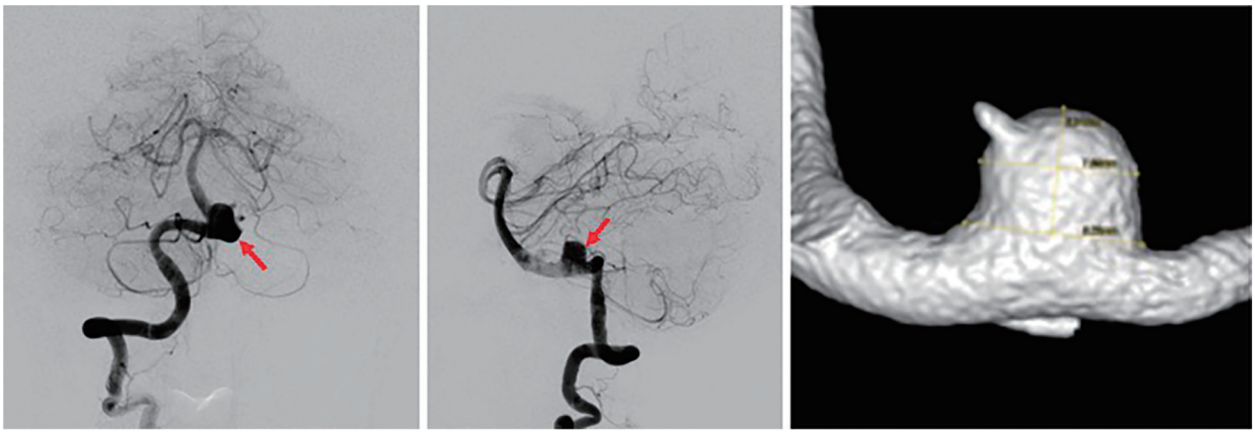


Fig. 2 The right vertebrobasilar artery angiogram, anteroposterior view (**A**) and lateral view (**B**) showing a wide-neck aneurysm (arrow). Three-dimensional reconstruction (**C**) showing the aneurysm with a maximum diameter of 7.9 mm and a bleb.

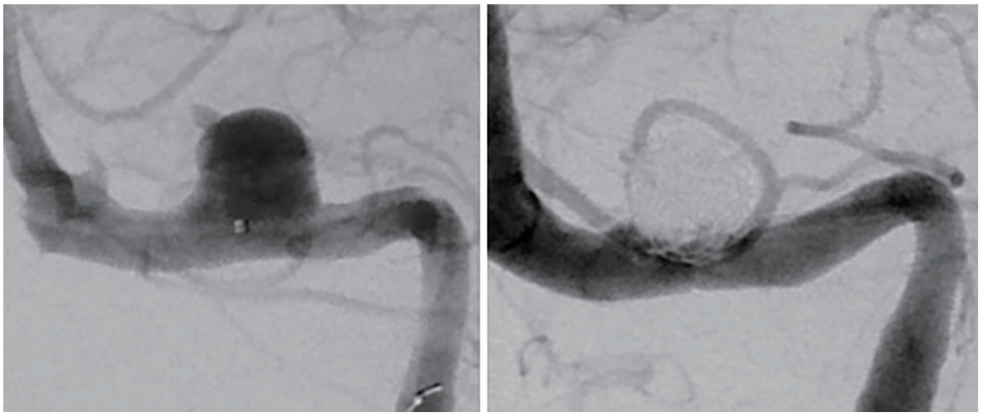
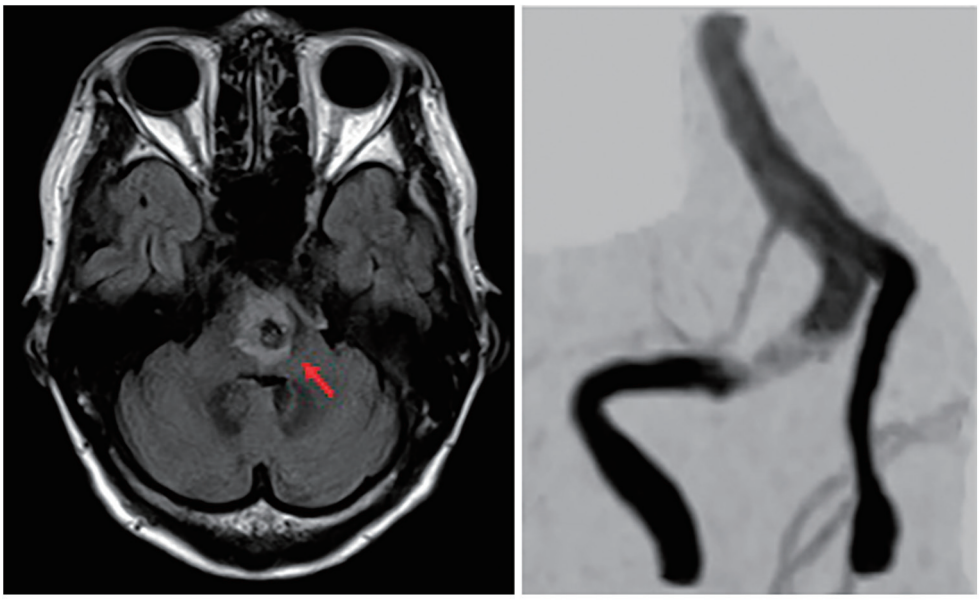


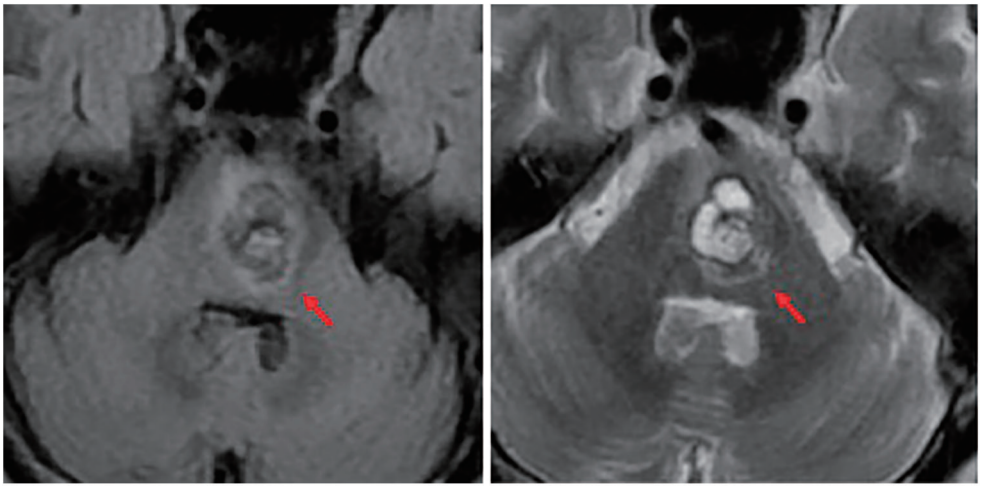
Fig. 3 The right vertebrobasilar angiogram before embolization (**A**) showing the wide-neck aneurysm. The angiogram six months after embolization (**B**) showing complete occlusion of the aneurysm.

oral prednisolone, but the results of allergic patch tests on the coils and stents used were negative. The edematous changes continued to progress, and MRI 15 months after treatment showed a cystic lesion with a high-intensity signal on FLAIR and iso-intensity with cerebrospinal fluid (CSF) components on T2WI (**Fig. 5**). The cyst continued to enlarge, and about 2 years after treatment, she developed right upper and lower extremity paralysis. Three years after treatment, the patient had difficulty walking independently and left facial paralysis, dysphagia, dysarthria, and an ocular motility disorder appeared. MRI showed significant enlargement of the cyst (**Fig. 6**). Since the cyst formation led to obstructive hydrocephalus, a ventriculoperitoneal shunt was placed, and then surgery to remove the cyst was scheduled. Prior to surgery, the patient underwent internal trapping of the vertebral artery, which was the parent artery of the aneurysm. Blood flow disappeared immediately after the parent artery occlusion, but 14 days later, angiography showed recanalization of the parent artery; another coil embolization procedure was performed to achieve complete occlusion and succeeded. Three days later, a transcondylar approach was performed for cyst fenestration. The aneurysm was embedded in the brainstem and showed yellowish degeneration. Indocyanine green fluorescence angiography confirmed no blood flow within the aneurysm. The cyst did not communicate with the ventricles or the subarachnoid space, and an incision through the cyst wall showed leakage of yellow fluid. After the aneurysm wall was incised, bleeding persisted, the parent artery was clipped distal to the lesion and occluded, and then intra-aneurysmal thrombus and the coils were removed as much as possible. Postoperatively, the cyst was seen to



A B

Fig. 4 Eight months after embolization, FLAIR (**A**) showing a high-intensity signal around the aneurysm (arrow). MRA (**B**) showing no apparent recurrence of the aneurysm.



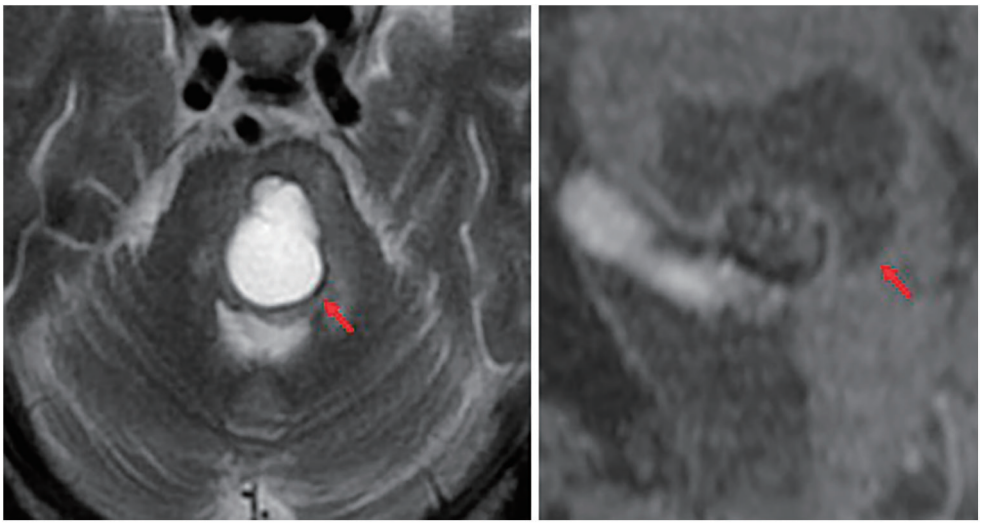
A B

Fig. 5 Fifteen months after embolization, FLAIR (**A**) showing edema and cyst formation around the aneurysm (arrow). The T2-weighted image (**B**) showing the iso-intensity cyst with the cerebrospinal fluid (arrow).

shrink, and the neurological symptoms did not worsen, but aspiration pneumonia and bacterial meningitis developed, and the patient died 7 months after surgery.

Discussion

Perianeurysmal cysts have been reported since 1999 when Hirota et al. described a case of cyst formation around the middle cerebral artery in a patient with a history of subarachnoid hemorrhage,[10] but the number of reports is limited, and they are rare. According to previous reports, their occurrence has been reported in both ruptured and unruptured cases of intracranial cerebral aneurysms, endovascularly treated and untreated cases. In both anterior and posterior circulations,[4,11,12] and no established factors for their pathogenesis are known. Liang et al. published a case series of 19 cases of perianeurysmal cysts[11] and noted that many of them had features of being embedded in the brain parenchyma, which is also consistent with the present case. Some have reported that thrombosis or calcification within



A B

Fig. 6 Three years after embolization, T2-weighted image (**A**) showing significant enlargement of the cyst (arrow). Sagittal time-of-flight MRA (**B**) showing the cyst around the aneurysm embedded in the brainstem (arrow).

the aneurysm is suspected as a causal feature,[12] but this was not seen in the present case.

In the present case, the patient underwent stent-assisted coil embolization of an unruptured vertebral artery aneurysm. During the postoperative period, edematous brain parenchymal changes appeared around the aneurysm, followed by cyst formation. However, no report has shown the change process from parenchymal edema to perianeurysmal cyst that occurs after coil embolization over time. When the perianeurysmal cyst appeared, a metal allergy to the device was suspected, and oral steroids were started. However, allergic patch tests on the coils and stents used were negative for all devices; therefore, a local allergic reaction was unlikely to be the cause. Some reports have shown edematous changes around aneurysms after coil embolization,[13,14] and though the mechanism is not clear, it has been reported to be related to the pulsation of the aneurysm, intramural bleeding, and local inflammation or cytokine release in the aneurysm wall. There is a report[15] of perianeurysmal edema that appeared after coil embolization was improved by administration of steroids to suppress local inflammation, suggesting that postoperative inflammatory changes may have been a contributing factor. However, in the present case, the findings progressed without improvement. Wallaert et al. reported that perianeurysmal edema occurs in about 6.4% of patients at 3-month follow-up after endovascular treatment and that factors related to its occurrence include aneurysm size, adhesion to the brain parenchyma, and aneurysm recurrence.[16] In the present case, complete occlusion was achieved by stent-assisted coil embolization, but the aneurysm was relatively large and was found to be adherent and embedded in the parenchyma, which may have contributed significantly to perianeurysmal edema and subsequent cyst formation.

Friedman et al. reported a case of perianeurysmal cyst formation after stent-assisted coil embolization of an unruptured aneurysm in the main trunk of the basilar artery,[7] similar to the present case. In this case, the aneurysm was embedded in the brainstem, and neurological symptoms due to postoperative cyst formation appeared. The authors performed ventriculoperitoneal shunting and cystotomy for hydrocephalus, and subsequently, the cyst regrew, and a cystic-abdominal shunt was added, resulting in an improvement of the neurological symptoms. They considered that the cyst formation was caused by the pulsatile pressure on the brainstem resulting from the hardening of the aneurysm itself as a result of the embolization of the coil into the aneurysm. It is known that the pulsation of blood flow from aneurysms attached to the brain affects the brain parenchyma (water-hammer effect),[4,11] and aneurysms embedded in the brain parenchyma as in the present case, are expected to be strongly affected by this effect, and coil embolization may have stiffened the aneurysmal sac after treatment, which may have contributed to cyst formation. It is also possible that the effect of the aneurysmal sac embedded in the brain parenchyma may have induced local inflammation and ischemic changes in the surrounding brain parenchyma. In past reports, the following mechanisms have been proposed to form perianeurysmal cysts.[4,11]

Direct exudation theory: Aneurysmal wall inflammatory mediator levels are elevated, resulting in exudate leakage from the aneurysmal wall.

Hemorrhage theory: Blood components leaking from the aneurysm due to hemorrhage or rebleeding of the aneurysm decompose and form a cyst.

Pulsatile blood flow theory: The pulsatile effect of blood flow in an aneurysm affects the brain parenchyma, resulting in the degeneration of parenchymal cells and the formation of cysts.

CSF entrapment theory: CSF is entrapped locally around the aneurysm and forms a cyst.

In the present case, the cystic component was equal in signal intensity to CSF on MRI, but the cyst contained a yellow exudate, confirmed by direct surgery. Moreover, the finding of high signal intensity on FLAIR preceded the cyst formation. These findings might suggest that the cystic component was not simply a finding of entrapment of spinal fluid. Since none of the above theories has been fully substantiated in any of the previous reports and there is no evidence that any one theory is superior to the others, it is likely that a combination of these factors is involved in the pathogenesis of the disease, and it is expected to be a complex condition.

Bioactive coils such as the Hydro coil induce inflammatory changes during the healing process of aneurysms, and it has been reported that embolization using Hydro coils might cause edema, hydrocephalus around aneurysms,[17] and aseptic meningitis.[18] Although the majority of embolized coils in these reports were bioactive, it is unknown how much bioactive coil volume is needed for the effect. In the present case, the length of the Hydro coil was only 9 cm, which accounted for only 10% of the total coil length of 90 cm, but the possibility that the edematous changes were caused by the bioactive coil material cannot be ruled out.

Perianeurysmal cysts often cause neurological symptoms by compressing the brain parenchyma, and symptoms vary depending on the site occupied. As in the present case, cyst formation can also cause a spinal fluid circulation disorder, resulting in secondary hydrocephalus.[19] Since perianeurysmal cysts have been reported to expand over time,[20] treatment should be considered when symptoms due to a cyst develop. The aim of treatment is to relieve pressure in the brain parenchyma caused by cysts; cyst drainage for decompression seems to be the most effective. In the present case, it was decided to remove the cyst, and internal trapping was performed prior to craniotomy to facilitate hemostatic control during surgery. There are reports of cyst fenestration craniotomy and endoscopic drainage.[21] On the other hand, though there are many reports of treatment by removal or fenestration of the cyst, there are also reports of good outcomes with endovascular treatment. Galdamez et al. reported a case of coil embolization of an unruptured aneurysm with a perianeurysmal cyst and subsequent increase in cyst size with recurrence of the aneurysm, in which additional coil embolization resulted in cyst reduction.[20]

In another case series, a perianeurysmal cyst formed and enlarged after coil embolization for the ruptured aneurysm, but the cyst stopped enlarging after the placement of a flow diverter stent for the residual neck after coil embolization. The authors suggested that the cyst expansion was caused by the persistent blood flow in the aneurysm.[11] In the present case, internal trapping with coils was performed before craniotomy, and the thrombus and coils in the aneurysm were removed, but intraoperative findings showed residual blood flow in the aneurysm. Even though the aneurysm appeared to be completely occluded on imaging, blood flow into the aneurysm actually remained. Considering these processes, the embolized coil might induce local inflammation of the wall of the embedded aneurysm, or the aneurysm might affect the brain parenchyma by a water-hammer effect on the brain parenchyma, resulting in perianeurysmal edema and cyst formation, since aneurysms embedded in the brain parenchyma, as in the present case, are thought to be at risk of perianeurysmal cyst formation.

Unlike inserting materials into aneurysms, flow diverter stent placement does not cause local inflammation due to the materials themselves within the aneurysms. Therefore, flow diverter stents may be a superior treatment. However, there are few reports of using a flow diverter stent for treating aneurysms embedded in the parenchyma, and its effectiveness is unknown.

There is still no clear consensus on the appropriate treatment, and the treatment must be considered depending on the location and findings of the aneurysm and cyst. The specimen obtained at cyst removal from the present patient was not examined, but it should have been because closer examination of the cyst contents might have provided more information about its etiology.

It is important to recognize that an aneurysm embedded in the parenchyma, as in the present case, may form a perianeurysmal cyst after coil embolization when determining the indications for treatment, surgical selection, and postoperative follow-up. Meticulous imaging follow-up is required when cyst formation is confirmed, and treatment should be considered when symptoms due to cyst formation are observed. Although there is no consensus on

treatment, the most appropriate therapy must be selected based on the patient and the lesion.

Conclusion

A rare case of edematous changes and cyst formation in the brainstem around an aneurysm after stent-assisted coil embolization of an unruptured vertebral artery aneurysm was reported. Aneurysms embedded in the brain parenchyma should be carefully followed up, recognizing the risk of perianeurysmal cyst formation after coil embolization.

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Disclosure Statement

The first author and all co-authors have no conflicts of interest.

References

1) Diyora B, Patil S, Bhende B, et al. Cerebral arteriovenous malformation associated with a cyst. *J Neurosci Rural Pract* 2018; 9: 420–422.
2) Hatashita S, Tajima A, Takeuchi K, et al. Arteriovenous malformation associated with a large cyst - case report. *Neurol Med Chir (Tokyo)* 1995; 35: 377–379.
3) Huang H, Xu K, Qu L, et al. Cystic cavernous malformation of the cerebellopontine angle: case report and literature review. *World J Surg Oncol*. 2011 23;9:36.
4) Pedro KM, Marie I, Sih IMY. Perianeurysmal parenchymal cyst of the pons: a case report and review of literature. *Interdiscip Neurosurg* 2020; 21: 100731.
5) Birua GJS, Tyagi G, Beniwal M, et al. Large parenchymal perianeurysmal cyst: a case report. *J Neurosci Rural Pract* 2021; 12: 800–803.
6) Benvenuti L, Gagliardi R, Scazzeri F, et al. Parenchymal perianeurysmal cyst in the brain: case report. *Neurosurgery* 2006; 58: E788; discussion, E788.
7) Friedman JA, Mclver JI, Collignon FP, et al. Development of a pontine cyst after endovascular coil occlusion of a basilar artery trunk aneurysm: case report. *Neurosurgery* 2003; 52: 694–699; discussion, 698–699.
8) Grandhi R, Miller RA, Zwagerman NT, et al. Perianeurysmal cyst development after endovascular treatment of a ruptured giant aneurysm. *J Neuroimaging* 2014; 24: 515–517.
9) Marcoux J, Roy MSD, Bojanowski MW. Acquired arachnoid cyst after a coil-ruptured aneurysm. *J Neurosurg* 2002; 97: 722.
10) Hirota N, Ueno J, Naitoh N, et al. Giant aneurysm associated with a large cyst. *Case illustration. J Neurosurg* 1999; 91: 160.
11) Liang ES, Efendy M, Winter C, et al. Intracranial perianeurysmal cysts: case series and review of the literature. *J Neurointerv Surg* 2022; 14: 837–841.
12) Sato N, Sze G, Awad IA, et al. Parenchymal perianeurysmal cystic changes in the brain: report of five cases. *Radiology* 2000; 215: 229–233.
13) Craven I, Patel UJ, Gibson A, et al. Symptomatic perianeurysmal edema following bare platinum embolization of a small unruptured cerebral aneurysm. *AJNR Am J Neuroradiol* 2009; 30: 1998–2000.
14) Fanning NF, Willinsky RA, ter Brugge KG, et al. Wall enhancement, edema, and hydrocephalus after endovascular coil occlusion of intradural cerebral aneurysms. *J Neurosurg* 2008; 108: 1074–1086.
15) Son JH, Fujii N, Matsuno A. A case of perianeurysmal edema after coil embolization. *Jpn J Stroke* 2016; 38: 120–124. (in Japanese)
16) Wallaert L, Soize S, De Beule T, et al. Perianeurysmal edema: prevalence, risk factors and clinical significance. *J Neuroradiol* 2017; 44: 135–142.
17) Stracke CP, Krings T, Möller-Hartmann W, et al. Severe inflammatory reaction of the optic system after endovascular treatment of a supraophthalmic aneurysm with bioactive coils. *AJNR Am J Neuroradiol* 2007; 28: 1401–1402.
18) Im SH, Han MH, Kwon BJ, et al. Aseptic meningitis after embolization of cerebral aneurysms using hydrogel-coated coils: report of three cases. *AJNR Am J Neuroradiol* 2007; 28: 511–512.
19) Jayakumar N, Ughratdar I, White E. Obstructive hydrocephalus secondary to a perianeurysmal cyst: a case report. *Br J Neurosurg* 2022; 36: 658–660.
20) Galdamez MM, Lorente PS, Rico AP, et al. Intracranial perianeurysmal cyst: still a dilemma - a case report with endovascular management. *Neuroradiol J* 2011; 24: 743–748.
21) Takai K, Nishihara T, Nemoto S, et al. Multilocular cystic lesion associated with a giant aneurysm. *J Neurosurg* 2001; 95: 1081.