Giant vertebral artery aneurysm in a child treated with endovascular parent artery occlusion and coil embolization

Hun-Soo Park, Ichiro Nakagawa, Takeshi Wada, Hiroyuki Nakagawa, Yasuo Hironaka, Kimihiko Kichikawa, Hiroyuki Nakase

Departments of Neurosurgery, and Radiology, Nara Medical University, Nara, Japan

E-mail: Hun-Soo Park - memepakusan@hotmail.com; Ichiro Nakagawa - nakagawa@naramed-u.ac.jp; Takeshi Wada - twneuro@m5.kcn.ne.jp; Hiroyuki Nakagawa - h_naka@naramed-u.ac.jp; Yasuo Hironaka - hironaka@naramed-u.ac.jp; Kimihiko Kichikawa - kkichika2005@yahoo.co.jp; Hiroyuki Nakase - nakasehi@naramed-u.ac.jp

*Corresponding author

INTRODUCTION

Intracranial aneurysms in the pediatric age group account for 1-2% of all intracranial aneurysms,[7,17] and vertebral artery (VA) giant aneurysms are even more rare in the pediatric population. The usual clinical presentation of giant aneurysms in children includes neurological symptoms following transient ischemic attacks or brainstem, thalamic, cerebellar, or cerebral infarctions. However, these lesions sometime progress insidiously and may be difficult to detect.[4] These lesions are associated with significant morbidity and mortality because of the associated mass effect caused by compression of the brainstem or lower cranial nerves rather than subarachnoid hemorrhage.[12] In general, the standard treatment for VA aneurysms, especially giant aneurysms, is to exclude
the lesion from the circulation while simultaneous preserving effective blood flow to the brain. This can be achieved by surgery, VA trapping with/without bypass, endovascular treatment, coil embolization, or parent artery occlusion (PAO). However, surgery for giant VA aneurysm may be very difficult to perform because of the risk of damage to the vertebral perforating arteries. Endovascular techniques are an effective therapeutic option, but complete occlusion is rarely achieved, and aneurysms often recur. Furthermore, coil embolization of giant VA aneurysm may result in permanent complications due to potentiation of the mass effect on the brainstem or due to hemorrhage. We report a case of pediatric patient with giant VA aneurysm who was successfully treated using staged endovascular treatment.

CASE REPORT

A 7-year-old girl presented with a 1-month history of gait disturbance, dysarthria, dysphagia, and hoarseness and with a 1-day history of muscular weakness of the extremities. Neurological examination revealed oculomotor, trochlear, abducens, glossopharyngeal, vagus, and accessory nerve palsy and tetraparesis. Computerized tomography revealed a giant right VA aneurysm expanding at the vertebrobasilar junction (diameter, 40 mm) without calcification. Magnetic resonance (MR) imaging demonstrated a giant aneurysm without intraaneurysmal thrombosis that was compressing the brainstem [Figure 1]. Digital subtraction angiography (DSA) showed the aneurysm arising 2 cm distal to the posterior inferior cerebellar artery (PICA) orifice to vertebrobasilar junction, fetal-type left posterior communicating artery, and bilateral VAs of similar caliber [Figure 2a and b]. A 6-Fr guiding catheter (Shuttle; Cook Medical, Inc., Bloomington, IN) was inserted into the right femoral artery and guided into the right VA. An Excelsior 1018 microcatheter (Boston Scientific, Watertown, MA) and a balloon catheter (HyperForm 7mm; 7mm; Micro Therapeutics) were introduced into the parent artery (right VA) through each guiding catheter. Parent artery coil embolization distal to the PICA was performed under flow control by balloon inflation at V4 portion of right VA, and complete PAO with preservation of the right PICA patency was achieved by the end of the procedure [Figure 2c and d]. However, one-third of the aneurysm was not thrombosed [Figure 3a]. Coil embolization was subsequently performed 14 days after PAO after the patient presented with respiratory depression due to mass effect. A 6-Fr guiding catheter (Shuttle) was inserted into the right femoral artery and guided into the left VA. An Excelsior 1018 microcatheter (Boston Scientific) was introduced into the left VA through the guiding catheter. Thirteen bare platinum coils were introduced into the aneurysm through the microcatheter, and total occlusion was confirmed at the end of this procedure [Figure 3b]. However, 1.5 months after the first coil embolization, follow-up DSA showed recurrent dome filling at the distal side of the aneurysm because of sinking of the coils into the intraaneurysmal thrombus due to pulsation effect from the VA union [Figure 3c]. A second coil embolization procedure was then performed using the balloon (HyperForm 4mm; 7mm; Micro Therapeutics) remodeling technique for tight packing and avoiding coil protrusion to the union of VA. An Excelsior 1018 microcatheter (Boston Scientific) was introduced into

Figure 2: Right vertebral angiography demonstrates a giant vertebral artery aneurysm (dome size: 42×30×30 mm). The aneurysm arises from 2 cm distal to the posterior inferior cerebellar artery orifice to vertebrobasilar junction (a,b). Left vertebral angiography shows the lumen of the contralateral left vertebral artery had a similar caliber to ipsilateral one (c). Rt. Vertebral angiography after parent artery occlusion shows complete occlusion with preservation of PICA patency (d)
the aneurysm and a balloon catheter (HyperForm 4 mm; 7 mm; Micro Therapeutics) was placed at the union of VA through the guiding catheter introduced into the left VA. Seventeen bioactive coils were introduced into the aneurysm. The aneurysm was totally occluded [Figure 3d], but reappearance of dome filling was noted 1 month after the second embolization [Figure 3e]. A third coil embolization procedure was performed, and eight bioactive coils were introduced into the aneurysm using the balloon-assist technique. Postembolization DSA confirmed total occlusion of the aneurysm [Figure 3f]. All procedures were performed with systemic heparinization to maintain the activated clotting time at levels above 2- to 3-fold the baseline value and oral aspirin (100 mg) was started after introducing guiding catheter through a gastric tube. Follow-up DSA at 1 month [Figure 4a] after the last fourth embolization did not show recurrence of the aneurysm aside from a small neck remnant. The patient’s postoperative course was uneventful. Although no ischemic or edematous changes of the brainstem were depicted in the course of the treatment, mass effect to the brainstem gradually decreased, and her cranial nerve palsy completely resolved [Figure 4b]. No new neurological deficits developed, and the patient was able to resume her normal life. Two years later, follow-up MR angiography did not show any recurrence of aneurysm [Figure 4c].

**DISCUSSION**

Giant aneurysm are defined as aneurysms greater than 25 mm in diameter, represent approximately 5% of all intracranial aneurysms, and are often located at the posterior circulation. Outcomes for patients with giant aneurysms are very poor, mainly due to associated hemorrhage, re-bleeding, mass effect, or thromboembolism. The natural history of untreated giant aneurysm has been extensively described; these lesions are associated with a mortality rate of about 60% at a year and over 80% at 5 years. Stroke is more frequent in affected pediatric patients when compared with adult patients. Subarachnoid hemorrhage is uncommon in affected pediatric patients when compared with adult patients. In general, the most likely cerebrovascular etiologies include hemodynamic stress and structural change in the vessel wall, which results in congenital, traumatic, and infectious disease. Trauma, such as minor craniocervical blunt injury during sports or other physical activities, represents the most common predisposing factor for giant aneurysm during childhood. However, factors predisposing to VA giant aneurysm are not yet well elucidated. The patient in the present case did not have congenital or acquired risk factors, such as minor and major trauma. The mechanism of the persistent growth of giant aneurysms might be related to recurrence of intramural/intraaneurysmal hemorrhage. However, some...
authors reported that pediatric posterior circulation aneurysms might be pathologically distinct from adult saccular aneurysms because fusiform shape, giant size, de novo aneurysm formation, and rapid progression are common in pediatric aneurysms. Developmental vessel wall defects or dissection may be considered the cause of enlarging aneurysms. In the present case, radiological findings did not reveal findings of dissection (e.g. “pearl and string sign” or “double lumen sign”). However, aneurysmal growth may occur secondary to repeated intimal dissection caused by hemodynamic stress.

Although advances in surgical and endovascular therapy have enabled the treatment of some cases of giant VA aneurysm, the optimal treatment strategy has not yet been determined. Some reports recommended that VA aneurysms should be treated with aneurysmal trapping and aneurysmectomy after bypass surgery because microsurgical therapy leads to higher rates of complete obliteration and lower rates of recurrence, indicating an advantage over endovascular therapy. From a flow reductive point of view, uni/bilateral VA occlusion may be a superior treatment options, but such treatment is still associated with a significant risk of acute ischemic events. Aneurysmal trapping and aneurysmectomy was also considered in the present case, however, we gave up the microsurgical approach because the remaining right VA distal to the aneurysm was too short to obtain distal VA trapping, anatomic deep location, and tight posterior fossa. Endovascular treatments can also be effective. Coil embolization of the aneurysm without PAO can be used for unclippable lesions, but this strategy can be associated with incomplete occlusion, rebleeding, regrowth, recanalization of aneurysms, coil compaction, and mass effect. Recent reports suggest that endovascular PAO resulted in favorable outcome, however, persistent enlargement of giant aneurysm even after complete endovascular occlusion may occur because endovascular PAO, unlike surgical PAO, cannot block a blood flow to the aneurysm neck beyond the occluded arterial segment through vasa vasorum on the adventitia. However, although the use of neck-remodeling devices, such as stent-assisted embolization and flow diveters, has therapeutic utility, these strategies can also be associated with thromboembolic complications, such as in-stent thrombosis and occlusion/stenosis. Furthermore, use of an intracranial stent and continuous antiplatelet medication can be problematic for young women who can become pregnant. In the present case, several recurrences of distal part of the aneurysm were expected because of sinking of the coils into the intraneurysmal thrombus, even if complete coil embolization of the aneurysm was performed. However, we initially utilized endovascular PAO alone prior to aneurysmal coil embolization in an effort to avoid the coiling mass effect, not PAO with rough or tight aneurysmal coil embolization. However, we initially utilized endovascular PAO alone prior to aneurysmal coil embolization in an effort to avoid the coiling mass effect to the brainstem, not PAO with rough or tight aneurysmal coil embolization. Endovascular trapping without inserting coils into the aneurysm is better treatment option in terms of avoiding coil mass effect, however, the remaining right VA distal to the aneurysm was too short to obtain distal PAO. We did not use stent-assist techniques or a flow diverter device. Outcomes were favorable, and the patient had no neurological deficits or aneurysm recurrence at the 2-year follow-up time point after the last fourth coil embolization. Furthermore, the mass effect to the brainstem was markedly reduced. This case suggests that staged endovascular treatment can be a viable treatment option for giant VA aneurysm in pediatric patients, although further long-term follow up is needed.

REFERENCES

1. Artmann H, Vonofakos D, Müller H, Grau H. Neuroradiologic and neuropathologic findings with growing giant intracranial aneurysm. Review of the literature. Surg Neurol 1984;21:391-401.
2. Chalouhi N, Tjoumakaris S, Starke RM, Gonzalez LF, Randazzo C, Hasan D, et al. Comparison of flow diversion and coiling in large unruptured intracranial saccular aneurysms. Stroke 2013;44:2150-4.
3. Cohen JE, Gomori JM, Moscovici S, Bala M, Itshayek E. The use of an intracranial stent and continuous antiplatelet medication can be problematic for young women who can become pregnant. In the present case, several recurrences of distal part of the aneurysm were expected because of sinking of the coils into the intraneurysmal thrombus, even if complete coil embolization of the aneurysm was performed. However, we initially utilized endovascular PAO alone prior to aneurysmal coil embolization in an effort to avoid the coiling mass effect, not PAO with rough or tight aneurysmal coil embolization. However, we initially utilized endovascular PAO alone prior to aneurysmal coil embolization in an effort to avoid the coiling mass effect to the brainstem, not PAO with rough or tight aneurysmal coil embolization. Endovascular trapping without inserting coils into the aneurysm is better treatment option in terms of avoiding coil mass effect, however, the remaining right VA distal to the aneurysm was too short to obtain distal PAO. We did not use stent-assist techniques or a flow diverter device. Outcomes were favorable, and the patient had no neurological deficits or aneurysm recurrence at the 2-year follow-up time point after the last fourth coil embolization. Furthermore, the mass effect to the brainstem was markedly reduced. This case suggests that staged endovascular treatment can be a viable treatment option for giant VA aneurysm in pediatric patients, although further long-term follow up is needed.

REFERENCES

1. Artmann H, Vonofakos D, Müller H, Grau H. Neuroradiologic and neuropathologic findings with growing giant intracranial aneurysm. Review of the literature. Surg Neurol 1984;21:391-401.
2. Chalouhi N, Tjoumakaris S, Starke RM, Gonzalez LF, Randazzo C, Hasan D, et al. Comparison of flow diversion and coiling in large unruptured intracranial saccular aneurysms. Stroke 2013;44:2150-4.
3. Cohen JE, Gomori JM, Moscovici S, Bala M, Itshayek E. The use of an intracranial stent and continuous antiplatelet medication can be problematic for young women who can become pregnant. In the present case, several recurrences of distal part of the aneurysm were expected because of sinking of the coils into the intraneurysmal thrombus, even if complete coil embolization of the aneurysm was performed. However, we initially utilized endovascular PAO alone prior to aneurysmal coil embolization in an effort to avoid the coiling mass effect, not PAO with rough or tight aneurysmal coil embolization. However, we initially utilized endovascular PAO alone prior to aneurysmal coil embolization in an effort to avoid the coiling mass effect to the brainstem, not PAO with rough or tight aneurysmal coil embolization. Endovascular trapping without inserting coils into the aneurysm is better treatment option in terms of avoiding coil mass effect, however, the remaining right VA distal to the aneurysm was too short to obtain distal PAO. We did not use stent-assist techniques or a flow diverter device. Outcomes were favorable, and the patient had no neurological deficits or aneurysm recurrence at the 2-year follow-up time point after the last fourth coil embolization. Furthermore, the mass effect to the brainstem was markedly reduced. This case suggests that staged endovascular treatment can be a viable treatment option for giant VA aneurysm in pediatric patients, although further long-term follow up is needed.

REFERENCES

1. Artmann H, Vonofakos D, Müller H, Grau H. Neuroradiologic and neuropathologic findings with growing giant intracranial aneurysm. Review of the literature. Surg Neurol 1984;21:391-401.
2. Chalouhi N, Tjoumakaris S, Starke RM, Gonzalez LF, Randazzo C, Hasan D, et al. Comparison of flow diversion and coiling in large unruptured intracranial saccular aneurysms. Stroke 2013;44:2150-4.
3. Cohen JE, Gomori JM, Moscovici S, Bala M, Itshayek E. The use of an intracranial stent and continuous antiplatelet medication can be problematic for young women who can become pregnant. In the present case, several recurrences of distal part of the aneurysm were expected because of sinking of the coils into the intraneurysmal thrombus, even if complete coil embolization of the aneurysm was performed. However, we initially utilized endovascular PAO alone prior to aneurysmal coil embolization in an effort to avoid the coiling mass effect, not PAO with rough or tight aneurysmal coil embolization. However, we initially utilized endovascular PAO alone prior to aneurysmal coil embolization in an effort to avoid the coiling mass effect to the brainstem, not PAO with rough or tight aneurysmal coil embolization. Endovascular trapping without inserting coils into the aneurysm is better treatment option in terms of avoiding coil mass effect, however, the remaining right VA distal to the aneurysm was too short to obtain distal PAO. We did not use stent-assist techniques or a flow diverter device. Outcomes were favorable, and the patient had no neurological deficits or aneurysm recurrence at the 2-year follow-up time point after the last fourth coil embolization. Furthermore, the mass effect to the brainstem was markedly reduced. This case suggests that staged endovascular treatment can be a viable treatment option for giant VA aneurysm in pediatric patients, although further long-term follow up is needed.

REFERENCES

1. Artmann H, Vonofakos D, Müller H, Grau H. Neuroradiologic and neuropathologic findings with growing giant intracranial aneurysm. Review of the literature. Surg Neurol 1984;21:391-401.
2. Chalouhi N, Tjoumakaris S, Starke RM, Gonzalez LF, Randazzo C, Hasan D, et al. Comparison of flow diversion and coiling in large unruptured intracranial saccular aneurysms. Stroke 2013;44:2150-4.
3. Cohen JE, Gomori JM, Moscovici S, Bala M, Itshayek E. The use of an intracranial stent and continuous antiplatelet medication can be problematic for young women who can become pregnant. In the present case, several recurrences of distal part of the aneurysm were expected because of sinking of the coils into the intraneurysmal thrombus, even if complete coil embolization of the aneurysm was performed. However, we initially utilized endovascular PAO alone prior to aneurysmal coil embolization in an effort to avoid the coiling mass effect, not PAO with rough or tight aneurysmal coil embolization. However, we initially utilized endovascular PAO alone prior to aneurysmal coil embolization in an effort to avoid the coiling mass effect to the brainstem, not PAO with rough or tight aneurysmal coil embolization. Endovascular trapping without inserting coils into the aneurysm is better treatment option in terms of avoiding coil mass effect, however, the remaining right VA distal to the aneurysm was too short to obtain distal PAO. We did not use stent-assist techniques or a flow diverter device. Outcomes were favorable, and the patient had no neurological deficits or aneurysm recurrence at the 2-year follow-up time point after the last fourth coil embolization. Furthermore, the mass effect to the brainstem was markedly reduced. This case suggests that staged endovascular treatment can be a viable treatment option for giant VA aneurysm in pediatric patients, although further long-term follow up is needed.
Intracranial aneurysm in childhood: 27-year single-institution experience. AJNR Am J Neuroradiol 2009;30:1315-24.

8. Iihara K, Murao K, Sakai N, Soeda A, Ishibashi-Ueda H, Yutani C, et al. Continued growth of and increased symptoms from a thrombosed giant aneurysm of the vertebral artery after complete endovascular occlusion and trapping: The role of vasa vasorum. J Neurosurg 2003;98:407-13.

9. Kanaan H, Jankowitz B, Aleu A, Kostov D, Lin R, Lee K, et al. In-stent thrombosis and stenosis after neck remodeling device-assisted coil embolization of intracranial aneurysms. Neurosurgery 2010;67:1523-33.

10. Klisch J, Eger C, Sychra V, Strasilla C, Bashe S, Weber J. Stent-assisted coil embolization of posterior circulation aneurysms using solitaire ab: Preliminary experience. Neurosurgery 2009;65:258-66.

11. Lasjaunias PL, Campi A, Rodesch G, Alvarez H, Kanaan I, Taylor W. Aneurysmal disease in children. Review of 20 cases with intracranial arterial localisations. Interv Neuroradiol 1997;3:215-29.

12. Massimi L, Moret J, Tamburrini G, Di Rocco C. Dissecting giant vertebro-basilar aneurysm. Childs Nerv Syst 2003;19:204-10.

13. Michael WF. Posterior fossa aneurysms simulating tumours. J Neurol Neurosurg Psychiatry 1974;37:218-23.

14. Mizutani T. A fatal, chronically growing basilar artery: A new type of dissecting aneurysm. J Neurosurg 1996;84:962-71.

15. Mocco J, Snyder KV, Albuquerque FC, Bendok BR, Alan S B, Carpenter JS, et al. Treatment of intracranial aneurysms with the Enterprise stent: A multicenter registry. J Neurosurg 2009;110:35-9.

16. Nagahiro S, Takada A, Goto S, Kai Y, Ushio Y. Thrombosed growing giant aneurysms of the vertebral artery: Growth mechanism and management. J Neurosurg 1995;82:796-801.

17. Sanai N, Quinones-Hinojosa A, Gupta NM, Perry V, Sun PP, Wilson CB, et al. Pediatric intracranial aneurysms: Durability of treatment following microsurgical and endovascular management. J Neurosurg 2006;104 (2 Suppl): S82-9.

18. Santillan A, Greenberg E, Patsalides A, Salvaggio K, Riina HA, Gobin YP. Long-term clinical and angiographic results of Neuroform stent-assisted coil embolization in wide-necked intracranial aneurysms. Neurosurgery 2012;70:1232-7.

19. Schubiger O, Valavanis A, Wichmann W. Growth-mechanism of giant intracranial aneurysms; demonstration by CT and MR imaging. Neuroradiology 1987;29:266-71.

20. Sluzewski M, Brilstra EH, van Rooij WJ, Wijnalda D, Tulleken CA, Rinkel GJ. Bilateral vertebral artery balloon occlusion for giant vertebrobasilar aneurysms. Neuroradiology 2001;43:336-41.

21. Steinberg GK, Drake CG, Peerless SJ. Deliberate basilar or vertebral artery occlusion in the treatment of intracranial aneurysms. J Neurosurg 1993;79:161-73.