Autologous repair of an internal carotid artery aneurysm by resection, caliber reduction, and external mesh tube reinforcement in a 9-year-old boy

Nestoras Papadopoulos, MD,[a] **Cornelius Klein, MD,**[b] **Koray Ak, MD,**[c] and **Anton Moritz, PhD,**[a] *Frankfurt, Germany; and Istanbul, Turkey*

Extracranial internal carotid artery aneurysms in children are rare, with a reported incidence of 0.5% to 1.9% in internal carotid artery aneurysm operations compared with all carotid operations in adult patients. We report a case of surgical reconstruction of an extracranial internal carotid artery aneurysm in a 9-year-old boy. Our patient complained of episodic neck pain on the left site under the mastoid process for the last year. The child was otherwise healthy. Autologous reconstruction without graft interposition was planned. Surgical repair was performed by resection of the main body of the aneurysm and restoration of the arterial continuity with end-to-end anastomosis. Because nondilated proximal and distal vessels could not be approximated, the most distal end of the aneurysm was tapered over a mandril. To prevent redilation, a tubular polyester external stent was fitted around the diseased segment. (J Vasc Surg 2007;46:1280-2.)

Extracranial internal carotid artery aneurysms (EICAA) in children are rare, with a reported incidence of 0.5% to 1.9% in ICAA operations compared with all carotid operations in adult patients. We report a case of surgical reconstruction of an EICAA by resection, caliber reduction, and external mesh tube reinforcement in a 9-year-old boy.

CASE REPORT

A 9-year-old boy was admitted to the hospital with the finding of a pulsating mass on the left side of his neck. His only complaint was episodic neck pain on the left side under the mastoid process for 1 year. The child was otherwise healthy. He had no history of trauma or any coexisting conditions.

The examination found a pulsatile cervical mass with an audible bruit located behind the left-sided sternocleidomastoid muscle. Angiography and magnetic resonance tomography (MRT) scans detected a large aneurysm of the left ICA, 33 × 35 mm in diameter and 88 mm in length, located between the bifurcation and the base of the skull (Fig 1). The amount of remaining normal artery at the distal end remained unclear. To achieve optimal distal exposure, we planned the surgery with the hospital's head and neck surgeons. No intraluminal thrombus was present in the aneurysm, and there was no evidence of significant disease on the contralateral side.

Under general endotracheal anesthesia, a neck incision was performed anterior to the left sternocleidomastoid muscle. The aneurysm was carefully dissected and exposed. The common, external, and proximal internal carotid artery segments were encircled with Silastic (Duecker, Melsungen, Germany) vascular loops. Moderate general and topical hypothermia by head cooling was begun, and the proximal ICA was clamped after systemic heparinization. Blood pressure was kept at 140 mm Hg with vasopressors.

After complete clamping of the ICA, a slight collapse of the aneurysm occurred and thus it was possible to free its distal end. A short segment of regularly sized distal ICA could be dissected free. The distal end was clamped and a shunt (Carotid Artery Shunts, Kendall, Argylle, Wollerau, Switzerland) was inserted but could not be stabilized in place because of the short distance available between the remaining vessel and the skull. Two attempts to stabilize the shunt in place were not successful. Because the backpressure was very good, we proceeded without shunt.

The aneurysm was resected between the proximal remnant of the ICA and the distal end at a distance that would allow the two stumps to be approximated. Excess distal aneurysm wall was resected, and a regular vessel diameter was reconstructed by using a longitudinal suture line over a 6-mm mandril. A tubular polyester mesh external stent (vein graft support, B/BRAUN, Aesculap AG & Co. KG, Tuttlingen, Germany) was flipped over the distal ICA before end-to-end anastomosis. The arterial continuity was restored by an end-to-end anastomosis with running 6-0 polypropylene suture. Then, the stent was pulled over the anastomosis and fixed on both ends to prevent redilation (Fig 2). The time of carotid clamping was 54 minutes. The rest of the operation was completed in a standard fashion.

The patient's postoperative course was uneventful. Control postoperative magnetic resonance angiography showed normal ICA morphology and flow after reconstruction (Fig 3). The patient was discharged on postoperative day 4 without complications.

Pathologic evaluation of the aneurysm revealed fibromuscular dysplasia. Alternating areas of thinned media and thickened fibro-

From the Departments of Thoracic and Cardiovascular Surgery,[a] and Oral, Maxillofacial and Plastic Facial Surgery,[b] Johann Wolfgang Goethe University Hospital; and Marmara University Faculty of Medicine Department of Cardiovascular Surgery.[c]

Competition of interest: none.

Correspondence: Dr Papadopoulos, Johann Wolfgang Goethe University Hospital, Theodor-Stern-Kai 7, 23A, 60590 Frankfurt/Main Germany (e-mail: nestoras.papadopoulos@gmail.com).

0741-5214/$32.00

doi:10.1016/j.jvs.2007.07.036

Fig 1. Angiography shows a large aneurysm of the left internal carotid artery located between its origin and the base of the skull. Parts of the aneurysm wall appear to be irregular. Most probably this is due to the rough inner surface and may be aggravated by flow phenomena in the preoperative diagnostics. During surgery, no thrombus was found adherent to the aneurysm wall.

muscular ridges containing collagen were evident microscopically. This coincides with medial fibroplasia according to the Harrison and McCormack classification.

DISCUSSION

Aneurysms of the EICA are extremely rare vascular lesions in childhood. The pathogenesis of these aneurysms can be inflammatory, traumatic, congenital, or previous operations such as tonsillectomy. EICAA in childhood have been found to have different features than those of adults. Atherosclerosis and traumatic cause has been shown to be a leading pathology in adults, but congenital and inflammatory causes predominate in children.[1,2] Also, there is a male preponderance and a tendency for large or even giant aneurysms in childhood.[3]

An EICAA can present with a variety of symptoms, depending on the size, etiology, and location. A common

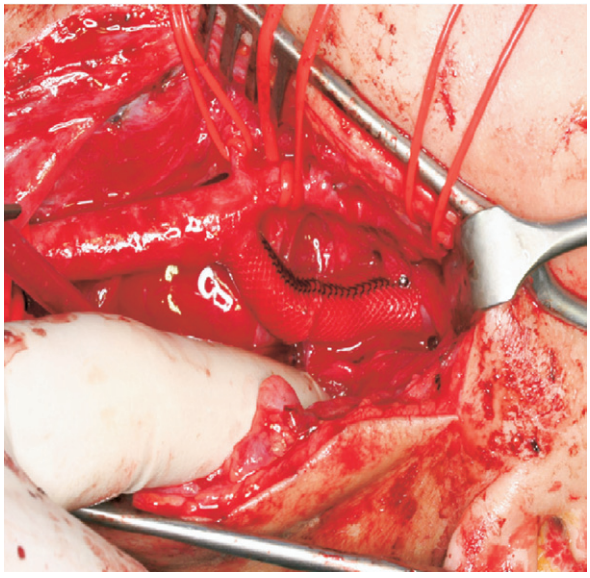

Fig 2. Intraoperative view shows end-to-end anastomosis of the proximal and distal segments of the left internal carotid artery and reposition of the polyester mesh tube over the anastomosis to prevent later redilation.

Fig 3. Follow-up magnetic resonance angiography shows normal internal carotid artery morphology and flow after reconstruction.

symptom is reported to be a pulsatile neck mass and pain, as in our case. Other nonspecific clinical manifestations can be related to cranial nerve palsy due to the compression, such as Horner syndrome, tinnitus, dizziness, hoarseness, and dysphagia.[4,5] Epistaxis, otorrhagia, hematemesis, and embolic stroke have also been reported.[6,7]

Zwolak et al[8] reported a stroke risk of 50% in nonoperated EICAAs in adult patients. Owing to the high rate of fatal neurologic complications and high risk of rupture, surgical treatment is strongly advocated in the treatment of patients with EICAA whenever the diagnosis is made. The outcome of surgery in children is reported to be much more favorable compared with adult patients, partly because young patients tolerate carotid clamping well due to sufficient collateral flow.[9]

Historically, ligation of the aneurysmatic ICA was performed to treat these patients. This technique has been totally abandoned owing to the high risk of cerebral morbidity and mortality after carotid artery ligation.[10,11] Vascular reconstruction with resection of the aneurysm and end-to-end anastomosis is now a well-accepted surgical procedure. In addition, vascular reconstruction with a synthetic or autologous saphenous vein graft is an alternative in the treatment of these cases.[12] Resection of the aneurysm, followed by primary closure of the artery, can be applied in particular cases with a sacciform EICCA.[13]

In our patient, we used resection and size reduction of the remaining distal aneurysm to achieve an autologous reconstruction. To prevent redilation of the remaining aneurysm wall, the repair was supported by an external Dacron (DuPont, Wilmington, Del) mesh tube. With this technique, the interposition of a synthetic or autologous graft, which is prone to further stenosis, intimal hyperplasia, or redilation[14] can be avoided. The 6 to 7 mm inner diameter gives sufficient space for normal development of the ICA.

The pathologic examination revealed fibromuscular dysplasia (FMD) involving the wall of the aneurysm. It has been shown that FMD is rarely associated with stroke, aneurysm formation, and intracranial hemorrhage in childhood.[15] Deficiency of α-1 antitrypsin and enhanced lipid peroxidation has been proposed in the pathogenesis of FMD.[16] The inherent FMD pathology of the vessel wall mandated an external support. External synthetic stents or sheaths have been used to taper vein grafts clinically or to reduce intimal hyperplasia caused by the arterialization process.[17] Jeremy et al[18] showed that external synthetic stents inhibit neointima formation and promote angiogenesis in vein grafts. They also prevent thickening of vein grafts and by this possibly prevent late failure.[18] These data indicate that external synthetic stents may provide increased long-term patency by inhibiting neointimal proliferation or dilatation.

CONCLUSION

EICAAs are rare pathologies in children, and prompt diagnosis and surgical treatment are essential to avoid complications. To achieve an autologous repair in this patient, a small portion of the distal aneurysm was tapered to allow direct end-to-end anastomosis. A commercially available external polyester mesh tube, which had been designed for external support of vein grafts, was used to prevent later redilation.

REFERENCES

1. Pourhassan S, Grotemeyer D, Heinen W, Ramp U, Heusch A, Sandmann W, et al. Extracranial aneurysm of the carotid artery in a 12-year-old girl. Chirurg 2006;77:173-8.
2. Cinar B, Fazliogullari O, Goksel O. True aneurysm of extracranial internal carotid artery in a 10-year-old. Eur J Vasc Endovasc Surg 2006;32:386-8.
3. Swamy NK, Pope FM, Coakham HB. Giant aneurysm of internal carotid artery in a four-year-old child: a case report. Surg Neurol 1993;40:138-41.
4. DeFatta RJ, Verret DJ, Bauer P. Extracranial internal carotid artery pseudoaneurysm Int J Pediatr Otorhinolaryngol 2005;69:1135-9.
5. Kawada T, Oki A, Iyano K, Bitou A, Okada Y, Matsuo Y, et al. Surgical treatment of atherosclerotic and dysplastic aneurysms of the extracranial internal carotid artery. Ann Thorac Cardiovasc Surg 2002;3:183-7.
6. Lueg EA, Awerbuck D, Forte V. Ligation of the common carotid artery for the management of a mycotic pseudoaneurysm of an extracranial internal carotid artery. A case report and review of the literature. Int J Pediatr Otorhinolaryngol 1995;33:67-74.
7. Moreau P, Albat B, Thevenet A. Surgical treatment of extracranial internal carotid artery aneurysm. Ann Vasc Surg 1994;8:409-16.
8. Zwolak RM, Whitehouse WM, Knake JE, Bernfeld BD, Zelenock GB, Cronenwett JL, et al. Atherosclerotic extracranial carotid artery aneurysms. J Vasc Surg 1984;1:415-22.
9. El-Sabrout R, Cooley DA. Extracranial carotid artery aneurysms: Texas Heart Institute experience. J Vasc Surg 2000;31:702-12.
10. McCann RL. Basic data related to peripheral artery aneurysms. Ann Vasc Surg 1990;4:411-4.
11. Rhodes EL, Stanley JC, Hoffman GL, Cronenwett JL, Fry WJ. Aneurysms of extracranial carotid arteries. Arch Surg 1976;111:339-43.
12. Bakoyiannis CN, Georgopoulos SE, Tsekouras NS, Klonaris CN, Skrapari IC, Papalambros EL, et al. Surgical management of extracranial internal carotid aneurysms by cervical approach. ANZ J Surg 2006;76:612-7.
13. Redaelli C, Carrel T, Turina M. Surgery of extracranial aneurysms of the carotid artery. Analysis of 8 cases. Chirurg 1991;62:620-4.
14. Fagioli GL, Freyrie A, Stella A, Pedrini L, Gargiulo M, Tarantini S, et al. Extracranial internal carotid artery aneurysms: results of a surgical series with long-term follow-up. J Vasc Surg 1996;23:587-95.
15. DiFazio M, Hinds SR 2nd, Depper M, Tom B, Davis R. Intracranial fibromuscular dysplasia in a six-year-old child: a rare cause of childhood stroke. J Child Neurol 2000;15:559-62.
16. Solder B, Streif W, Ellemunter H, Mayr U, Jaschke W. Fibromuscular dysplasia of the internal carotid artery in a child with alpha-1-antitrypsin deficiency. Dev Med Child Neurol 1997;39:827-9.
17. Moritz A, Grabenwoger F, Raderer F, Ptakovsky H, Staudacher M, Magometschnigg H, et al. Mesh tube-constricted varicose veins used as bypass grafts for infrainguinal arterial reconstruction. Arch Surg 1992;127:416-20.
18. Jeremy JY, Gadsdon P, Shukla N, Vijayan V, Wyatt M, Newby AC, et al. On the biology of saphenous vein grafts fitted with external synthetic sheaths and stents. Biomaterials 2007;28:895-908.

Submitted May 13, 2005; accepted Jul 29, 2007.