Revascularization and pediatric aneurysm surgery

Clinical article

*M. Yashar S. Kalani, M.D., Ph.D., Ali M. Elhadi, M.D., Wyatt Ramey, B.S., Peter Nakaji, M.D., Felipe C. Albuquerque, M.D., Cameron G. McDougal, M.D., Joseph M. Zabramski, M.D., and Robert F. Spetzler, M.D.*

Division of Neurological Surgery, Barrow Neurological Institute, Saint Joseph’s Hospital and Medical Center, Phoenix, Arizona

Object. Aneurysms are relatively rare in the pediatric population and tend to include a greater proportion of large and giant lesions. A subset of these large and giant aneurysms are not amenable to direct surgical clipping and require complex treatment strategies and revascularization techniques. There are limited data available on the management of these lesions in the pediatric population. This study was undertaken to evaluate the outcome of treatment of large and giant aneurysms that required microsurgical revascularization and vessel sacrifice in this population.

Methods. The authors retrospectively identified all cases in which pediatric patients (age < 18 years) with aneurysms were treated using cerebral revascularization in combination with other treatment modalities at their institution between 1989 and 2013.

Results. The authors identified 27 consecutive patients (19 male and 8 female) with 29 aneurysms. The mean age of the patients at the time of treatment was 11.5 years (median 13 years, range 1–17 years). Five patients presented with subarachnoid hemorrhage, 11 with symptoms related to mass effect, 2 with stroke, and 3 with seizures; in 6 cases, the aneurysms were incidental findings. Aneurysms were located along the internal carotid artery (n = 7), posterior cerebral artery (PCA) (n = 2), anterior cerebral artery (n = 2), middle cerebral artery (MCA) (n = 14), basilar artery (n = 2), vertebral artery (n = 1), and at the vertebrobasilar junction (n = 1). Thirteen were giant aneurysms (45%). The majority of the aneurysms were fusiform (n = 19, 66%), followed by saccular (n = 10, 34%). Three cases were previously treated using microsurgery (n = 2) or an endovascular procedure (n = 1). A total of 28 revascularization procedures were performed, including superficial temporal artery (STA) to MCA (n = 6), STA to PCA (n = 1), occipital artery to PCA (n = 1), extracranial-intracranial (EC-IC) bypass using radial artery graft (n = 3), EC-IC using a saphenous vein graft (n = 7), STA onlay (n = 3), end-to-end anastomosis (n = 1), and in situ bypasses (n = 6). Perioperative stroke occurred in 4 patients, but only one remained dependent (Glasgow Outcome Scale [GOS] score 3). At a mean clinical follow-up of 46 months (median 14 months, range 1–232 months), 26 patients had a good outcome (GOS score 4 or 5). There were no deaths. Five patients had documented occlusion of the bypass graft. The majority of aneurysms (n = 24) were obliterated at last follow-up. There was a single case of a residual aneurysm and one case of recurrence. Angiographic follow-up was unavailable in 3 cases.


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**Key Words**
- aneurysm
- clipping
- endovascular
- complex
- extracranial-to-intracranial
- intracranial-to-intracranial
- bypass
- cerebral revascularization
- vascular disorders

The incidence of aneurysms in the pediatric population is not well established, but several studies suggest that between 1% and 5% of all aneurysms occur in children. It is speculated that the etiology of aneurysm formation in children may be different than in adults, as a disproportionate number of pediatric aneurysms are large to giant and fusiform or dissecting. These characteristics of pediatric aneurysms make them challenging for standard microsurgical treatment options. Although endovascular techniques are a well-established option for the management of aneurysms in the adult population, the long-term risks and durability of endovascular treatment in pediatric patients has not been established. Given the average life expectancy of 80 years, it is our institutional view that microvascular surgery should be
the treatment of choice for aneurysms in the pediatric population, although we have used combined endovascular/microsurgical procedures in complex cases. In the subset of aneurysms not amenable to direct microsurgical treatment or current endovascular therapies, cerebral revascularization and vessel sacrifice or trapping is a robust treatment alternative.9–12,17 We sought to evaluate the outcomes of pediatric patients with complex aneurysms treated using this treatment strategy.

**Methods**

**Patient Population**

Between 1989 and 2013, we treated 27 pediatric patients (age < 18 years) with 29 aneurysms using cerebral revascularization either alone or in conjunction with endovascular or microsurgery at the Barrow Neurological Institute (Tables 1 and 2). Nineteen (70%) of the patients were male and 8 (30%) were female; their average age was 11.5 years (median 13; range 1–17 years). Medical records, results of neurological examinations, and radiographic studies were retrospectively reviewed. Preoperative and postoperative neurological function was evaluated using the Glasgow Outcome Scale (GOS).

**Presentation**

The most common presentation was that of symptoms related to mass effect, and these were the presenting symptoms in 11 (41%) of 27 patients. Other presentations included subarachnoid hemorrhage (SAH) in 5 patients (18%), transient ischemic attacks in 2 (7%), and seizures in 3 (11%). In 6 patients (22%), the aneurysms were incidental findings. Of patients with SAH, the Hunt and Hess grade was II in 2 patients and IV in 1 patient. Hunt and Hess grades were not available in 2 cases, including a 1-year-old child in whom a score could not be assigned. The Fisher classification was used to grade the degree of bleeding noted on the CT scan. The Fisher grade was 2 in 2 cases, 3 in 1 case, and 4 in 1 case. Three patients had undergone previous treatment for their aneurysms prior to referral, including endovascular coil embolization (1 case) and microsurgical treatment (2 cases).

**Aneurysm Location and Characteristics**

The majority of the aneurysms were on the left side (21 [72.4%] of 29). There were 2 basilar artery (BA) aneurysms (7% of the 29 lesions). There were 14 giant (48%) and 5 large aneurysms. Nineteen aneurysms (66%) were fusiform and 10 (34%) were saccular. Fourteen (48%) of the aneurysms were located along the middle cerebral artery (MCA), 7 (24%) along the internal carotid artery (ICA), 2 (7%) on the posterior cerebral artery (PCA), 2 (7%) on the anterior cerebral artery (ACA), 2 (7%) on the basilar artery (BA), 1 (3%) at the vertebrobasilar junction (VBJ), and 1 (3%) on the vertebral artery (VA).
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Diagnostic Evaluation

All patients were evaluated using digital subtraction angiography, CT angiography, or MR angiography. Balloon test occlusion was not routinely performed.

Treatment Paradigm

All patients underwent microsurgical exploration with the goal of direct clipping or clip reconstruction of their aneurysms. If at the time of intraoperative evaluation the surgeon concluded that direct clipping was not possible, a microvascular bypass was performed, followed by occlusion of the parent vessel proximal or distal to the aneurysm, trapping, or trapping and excision of the aneurysm. Before the procedure, patients were treated with aspirin (81 mg/day), with the dosage continued postoperatively. Some of our patients received intravenous administration of heparin postoperatively to prevent sudden thrombosis of aneurysms. Electroencephalographic monitoring was conducted intraoperatively. We perform revascularization procedures under mild hypothermia (33°–34°C) and barbiturate protection. Evaluation of the bypass patency was done using intraoperative angiography or, more recently, using indocyanine green videoangiography. Postoperatively, digital subtraction angiography or CT angiography was used to evaluate the aneurysm and bypass patency.

Aneurysms were treated by proximal vessel clip occlusion (n = 10), clipping of the aneurysm (n = 4), distal clip occlusion (n = 1), trapping (n = 2), excision (n = 4), coil embolization (n = 2), and other interventions in an additional 3 patients. In 3 patients the aneurysms thrombosed after the bypass procedure and additional interventions were not necessary.

Results

Bypass Procedures and Patency

A total of 28 revascularization procedures were performed. Revascularization was performed with superficial temporal artery (STA) to MCA bypass (n = 6), STA to PCA bypass (n = 1), occipital artery (OA) to PCA bypass (n = 1), extracranial-intracranial (EC-IC) bypass using a radial artery graft (n = 3), EC-IC bypass using a saphenous vein graft (n = 7), STA onlay (n = 3), end-to-end anastomosis (n = 1), or in situ bypasses (n = 6). The in situ bypasses included an A3-A3, a posterior inferior cerebellar artery–posterior inferior cerebellar artery and 3 anterior temporal artery to MCA bypasses. The mean length of angiographic follow-up for patients with available follow-up (n = 13) was 49.6 months (range 1–232). During this period, graft occlusion was identified in 5 patients. Although intraoperative bypass patency was observed and documented in every case, data on long-term bypass patency were not available in 3 cases.

Aneurysm Outcomes

The majority of aneurysms (n = 24) were obliterated at follow-up (Table 3). There was 1 case of residual aneurysm and 1 case of aneurysm recurrence. Follow-up angiography demonstrating aneurysm fate was not available in 3 cases. The case of residual aneurysm involved a fusiform BA lesion. The patient had a history of a fusiform ICA aneurysm that was successfully treated 2 years earlier. The patient with the aneurysm recurrence had a small but complex MCA aneurysm that was recalcitrant to multiple treatments including direct microsurgical repair, multiple endovascular coil embolizations, and revascularization and proximal vessel occlusion.

Complications and Outcomes

There were no cases of perioperative mortality. Long-term clinical follow-up (mean 61.9 months, range 1–232 months) was available in 20 cases and immediate postoperative follow-up was available in every case. Postoperatively, 25 patients (92%) had a good outcome (GOS score 4 or 5). There were 4 cases of postoperative ischemic stroke (Table 4). Of these patients, 1 made a complete recovery (GOS score 5), 2 improved to be functionally independent (GOS score 4), and 1 patient remains dependent (GOS score 3) with hemiplegia and a facial droop. There were 5 cases of postoperative graft occlusion. Although none of these patients exhibited a cerebrovascular accident (CVA), 1 patient did experience permanent blindness. One patient suffered a postprocedural iliac pseudoaneurysm after endovascular treatment.

TABLE 3: Summary of patient outcomes in this series, including bypass patency and aneurysm fate

<table>
<thead>
<tr>
<th>Outcome</th>
<th>Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>patient outcomes (n = 27)</td>
<td></td>
</tr>
<tr>
<td>mean clinical follow-up (mos)</td>
<td>46</td>
</tr>
<tr>
<td>median clinical follow-up (mos)</td>
<td>14</td>
</tr>
<tr>
<td>GOS 4–5</td>
<td>26 (96.3)</td>
</tr>
<tr>
<td>GOS &lt;4</td>
<td>1 (3.7)</td>
</tr>
<tr>
<td>aneurysm outcomes (n = 29)</td>
<td></td>
</tr>
<tr>
<td>obliterated</td>
<td>24 (82.7)</td>
</tr>
<tr>
<td>residual</td>
<td>1 (3.4)</td>
</tr>
<tr>
<td>recurrent</td>
<td>1 (3.4)</td>
</tr>
<tr>
<td>unknown</td>
<td>3 (10.3)</td>
</tr>
</tbody>
</table>

* Values represent numbers of patients or aneurysms (with percentages in parentheses) except as otherwise indicated.

TABLE 4: Summary of morbidity and mortality noted with revascularization as a treatment strategy in this series

<table>
<thead>
<tr>
<th>Variable</th>
<th>No. of Cases (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>perioperative mortality</td>
<td>0</td>
</tr>
<tr>
<td>perioperative morbidity*</td>
<td></td>
</tr>
<tr>
<td>CVA</td>
<td>4 (14.3)</td>
</tr>
<tr>
<td>infection</td>
<td>1 (3.6)</td>
</tr>
<tr>
<td>iliac pseudoaneurysm†</td>
<td>1 (3.6)</td>
</tr>
<tr>
<td>unilateral blindness</td>
<td>1 (3.6)</td>
</tr>
<tr>
<td>neck hematoma</td>
<td>1 (3.6)</td>
</tr>
</tbody>
</table>

* Calculated based on 28 revascularization procedures.
† After endovascular treatment.
Discussion

Pediatric aneurysms are relatively rare lesions. Available studies suggest that no more than 1% to 5% of aneurysms occur in children. As a result, data on the long-term outcome of various treatment modalities are limited. Although the results of microsurgery for pediatric aneurysms have generally been good, there remains a subset of cases in which the aneurysms are not amenable to direct clipping, or clip reconstruction, and require complex techniques for management (Fig. 1).

Recently several groups have described their experience with endovascular or combined endovascular and microsurgical repair of complex aneurysms in the pediatric population. These reports suggest that these treatment options are generally well tolerated and result in acceptable outcomes in the majority of patients, but the studies suffer from the effects of small sample size and relatively short follow-up.

While endovascular techniques are a well-accepted option for the treatment of aneurysms in adults, their use in pediatric populations poses additional concerns. Despite continued advances in endovascular techniques, the rate of retreatment for large and giant aneurysms remains high, and the long-term risks of new devices, such as flow-diverting stents, remains to be defined in the pediatric population, where the projected life expectancy is in excess of 80 years.

In the current study, we review the clinical and angiographic outcome in a series of 27 consecutive pediatric patients with large or giant complex aneurysms treated at our institution. Our results indicate that bypass surgery combined with various methods of aneurysm exclusion is well tolerated and effective in this population. This treatment strategy was associated with an 83% rate of aneurysm obliteration and a good to excellent clinical outcome in 26 (96%) of 27 patients. At a mean follow-up of 46 months, we identified one case of recurrence requiring additional treatments and one case of a residual aneurysm. The case of recurrence requiring retreatment has been previously published. This case highlights the importance of close follow-up in this patient population (Fig. 2) and underscores the likelihood that pediatric and adult aneurysms are associated with different pathological mechanisms.

Perioperative complications occurred in 7 of 28 operative procedures, but the majority of these resolved completely. Only 2 complications resulted in permanent deficits—one case of unilateral blindness and one case of a CVA with a permanent deficit. Other complications included 3 cases of CVA with temporary deficits, 1 case of neck hematoma, and 1 case of an iliac artery pseudoaneurysm after endovascular intervention. Our morbidity profile is comparable to those described in other reports of cerebral revascularization for complex aneurysms in the literature.

Fig. 1. An otherwise healthy 17-year-old male presents with the worst headache of his life. Axial CT scan of the head (A) revealed SAH. Formal anteroposterior ICA angiography (B) revealed the presence of a proximal complex fusiform MCA aneurysm. A 3D reconstruction of the angiogram (C) reveals the complex morphology of the aneurysm and the incorporation of distal branches of the MCA by the fusiform aneurysm. The patient was taken to the operative theater for an attempt at clip ligation, but the aneurysm was not amenable to direct clipping. The patient underwent aneurysmorrhaphy with a radial artery interposition graft. Postoperatively the patient was neurologically intact. Selective MCA angiography (D) revealed an excellent reconstruction of the parent vessel with intact distal flow. 3D reconstruction of the angiogram (E) better illustrates the results of the aneurysmorrhaphy with a radial artery interposition graft. The patient continued to smoke tobacco. At the 18-month follow-up, the selective MCA angiography (F) revealed proximal stenosis. The patient underwent angioplasty of the stenosed segment, with no evidence of hemodynamically significant stenosis on follow-up angiography (G), and he was neurologically intact 36 months posttreatment. Figures 1A–E used with permission from the Barrow Neurological Institute. Previously published in Spetzler RF, Nakaji P, Rhoton AL Jr, Kawashima M (eds): Color Atlas of Cerebral Revascularization. New York: Thieme Medical Publishers, 2013.
Conclusions

Vessel occlusion and microsurgical revascularization remains a reliable treatment option for the management of selected large and giant cerebral aneurysms in children. Regardless of the method of treatment, children with cerebrovascular aneurysms require close follow-up for recurrence and the possibility of de novo aneurysm formation.

Disclosure

The authors have no personal financial or institutional interest in any of the materials or devices described in this article.

Author contributions to the study and manuscript preparation include the following. Conception and design: Zabramski, Kalani, Nakaji, Albuquerque, McDougall, Spetzler. Acquisition of data: Kalani, Elhadi, Ramey, McDougall. Analysis and interpretation of data: Kalani, Elhadi, Ramey, Spetzler. Drafting the article: Kalani, Elhadi. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Study supervision: Zabramski, Nakaji, Spetzler.

References


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Address correspondence to: Joseph M. Zabramski, M.D., c/o Neuroscience Publications, Barrow Neurological Institute, St. Joseph’s Hospital and Medical Center, 350 W. Thomas Rd., Phoenix, AZ 85013. email: neuropub@dignityhealth.org.