Childs Nerv Syst (2012) 28:169–173
DOI 10.1007/s00381-011-1602-y

CASE REPORT

Three-year-old patient with giant MCA aneurysm treated by trapping–resection plus STA–MCA bypass. Case report

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Received: 11 September 2011 / Accepted: 26 September 2011 / Published online: 8 October 2011

Introduction

Intracranial aneurysm in the pediatric population is very rare; the prevalence has been reported from 0.5% to 4.6% [1–3]. This pathology is completely different from aneurysms in adults: gender predominance; location of aneurysm; incidence of spontaneous thrombosis; incidence of giant, dissecting, and fusiform aneurysm; and rate of subarachnoid hemorrhage, among others [4]. Different multidisciplinary approaches have been used, ranging from conservative follow-up to the most complex microsurgical and endovascular treatments. In the microsurgical group, there are different alternatives, direct clipping and reconstruction or trapping with bypass from extracranial–intracranial (EC–IC) or intracranial–intracranial (IC–IC) types. For the middle cerebral artery aneurysms, the revascularization is an excellent option, especially for the giant lesions in which the reconstruction is not an option. Even more, if we analyze this subject, the overall complication rate of EC–IC procedures is very low, and the 10-year patency rate is as high as 73% [5]. Different types of graft used in children have been reported for EC–IC bypasses with vein graft [5], with radial artery graft [2], and superficial temporal artery (STA) [2, 6]. In the pediatric group, the youngest patient previously reported with an EC–IC bypass with STA–middle cerebral artery (MCA) for a MCA aneurysm had 11 years [6]. We present our case report as the youngest patient ever treated with a combination of trapping with a successful STA–MCA bypass in a giant MCA aneurysm.

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Case report

A 3-year-old boy presented with nausea, vomiting, and generalized seizures. The neurologic examination was normal. In the emergency room, a CT scan (Fig. 1) was taken and showed a hyperdense large mass related with the left proximal middle cerebral artery. The study was completed with a CT-angiography (Figs. 2 and 3) and 3-D conventional angiography (Fig. 4) combined with a balloon test occlusion. The study concluded a giant partially thrombosed fusiform aneurysm of the left M1, measuring 55×36×29.5 mm, with a significant mass effect. The MRI showed sequelar lesions in the left lenticulostriate territory. A small anterior temporal artery and most of the lenticulostriate arteries had their origin proximal to the aneurysm. The balloon test occlusion concluded that it had opening of pial anastomoses depending on the anterior and posterior left cerebral arteries that supply in a retrograde fashion the M2 segments. The case was comprehensively evaluated by

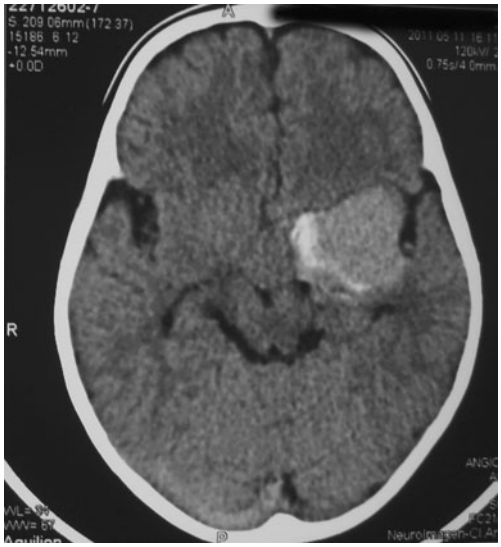

Fig. 1 Preoperative non-contrasted CT scan of our patient showing a giant left MCA territory hyperdensity secondary to intraaneurysmal thrombosis

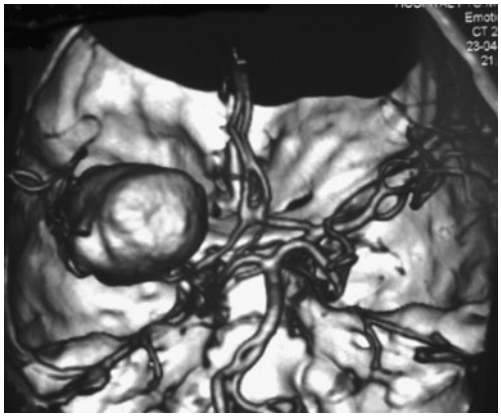

Fig. 3 AngioCT tridimensional reconstruction. The giant aneurysm can be seen occupying more than half of the middle fossa

a multidisciplinary team with interventional neuroradiologists, vascular neurosurgeons, and peripheral vascular surgeon. The treatment pre-designed for the patient was microsurgery with trapping and resection of the aneurysm to eliminate the mass effect, combined with a left ECA–ICA bypass with radial artery graft as a flow restoration procedure. The microsurgery was performed with a peripheral vascular surgeon who exposed the radial artery. The left carotid artery was exposed in the neck, and then a left pterional interfascial approach was performed with preservation of the superficial temporary artery. Because of the sufficient size of the STA, we preferred to use it instead of the radial artery graft. The STA-M2 bypass was made followed by the trapping and resection of the aneurysm. The lenticulostriate arteries and the anterior temporal artery were preserved in the proximal segment of the M1. The distal portion flow was restored with the STA–MCA bypass as we mentioned before (Fig. 5). An immediate CT scan and a CT-angiography were performed showing the total exclusion of the lesion and confirmed the patency of the bypass (Figs. 6 and 7). The patient had an excellent and uneventful postoperative clinical condition, and the neurological examination remained normal (Fig. 8). The patient was discharged 2 weeks after the microsurgery with a second normal CT-angiography and CT scan that showed no new ischemic lesion and restoration of the brain position secondary to aneurysm resection.

Discussion

Intracranial aneurysms in the pediatric population are rare. According to the literature, they occur in less than 5% of all aneurysms [1–3]. It is well known that the clinical features and presentation of pediatric aneurysm differ from adults:

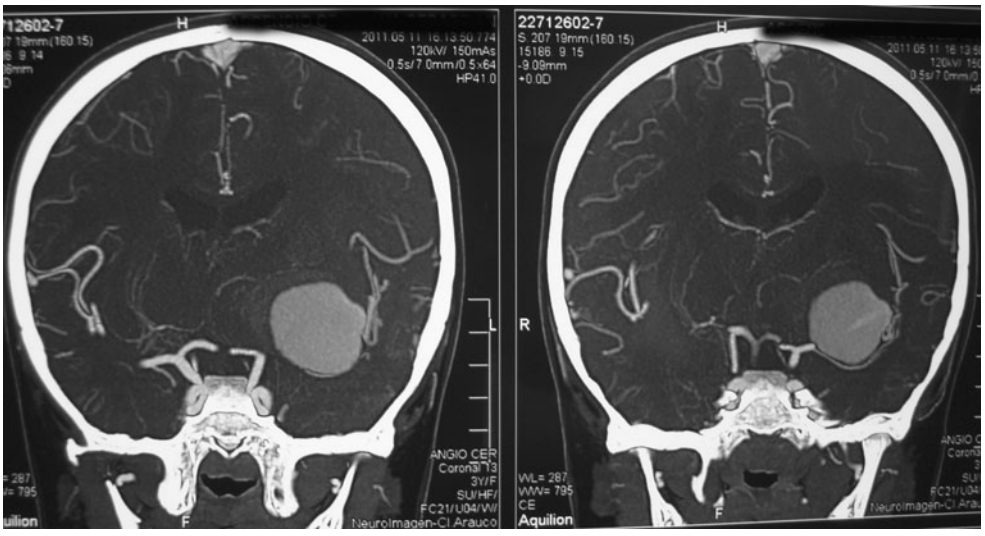

Fig. 2 Coronal angioCT showing the giant MCA aneurysm, with the anterior temporal artery arising proximal to the aneurysm and the Sylvian bifurcation distal to the aneurysm

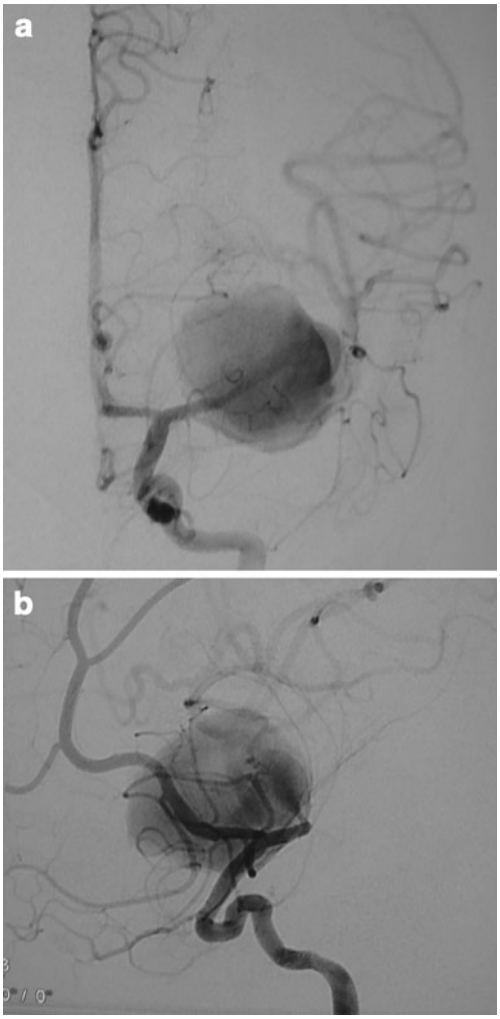

Fig. 4 Left carotid angiography, anteroposterior (**a**) and lateral (**b**) views

pediatric patients have male predominance; high incidence of posterior circulation aneurysm; higher rate of giant, dissecting, and fusiform aneurysms; and a high incidence of spontaneous thrombosis [4, 7], although there are recent publications of series that show a predominance of anterior circulation aneurysm up to 72% [2, 8]. Aneurysm in the MCA has similar proportion in children and adults [7]. In the demographic aspect, it has been suggested that they could follow a bimodal presentation from birth to 6 years and another peak from 8 years to adolescence, but other reports have shown most of the aneurysms in the second decade of life [2]. The rate of aneurysm rupture is 22% [8], but it is more often in saccular aneurysms [7]. If we analyze why it is more often fusiform or dissecting aneurysm in the MCA, specifically in the M1 segment, there is not a clear answer. There are some hypotheses that try to explain this, based on the fact that in the earlier brain development, the MCA supplies more blood flow than other vessels; it would be exposed to more hemodynamic stress, and in the presence of an embryological deficit or in a structural abnormality, it can be the reason of the aneurysm formation [3]. Nevertheless, we do not know if the proposed mechanism of formation in adults can be used in children [9].

A variety of modalities have been published for microsurgical and endovascular treatments. In the microsurgical group, the options range from direct clipping, trapping, bypass EC–IC or IC–IC, proximal parent artery occlusion, aneurysm excision–reanastomosis, cotton reinforcement, muslin wrap, or any combination between them [2, 5–8]. If we analyze the overall results of microsurgical and endovascular treatments, Sanai showed no deaths for either group and similar neurological morbidity for both. But he showed a 14% of aneurysm recurrence in the

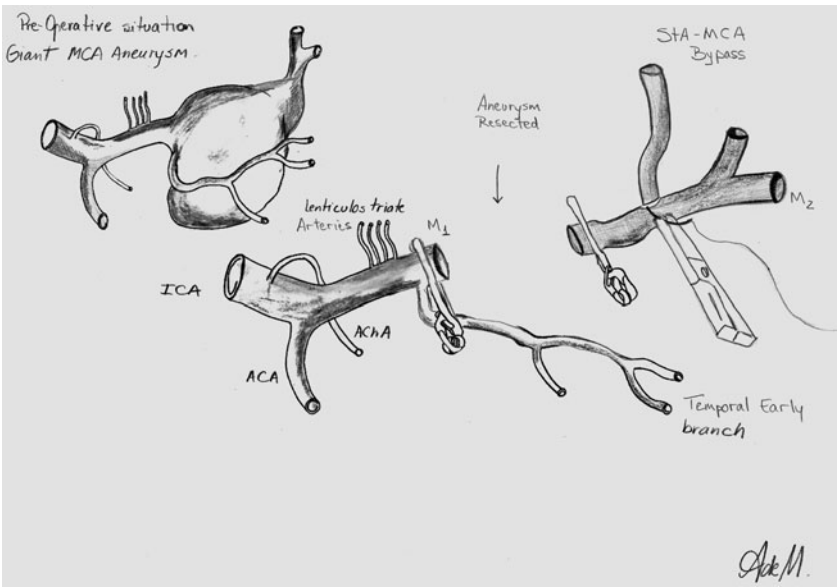

Fig. 5 Schematic view of the microsurgery performed in the patient

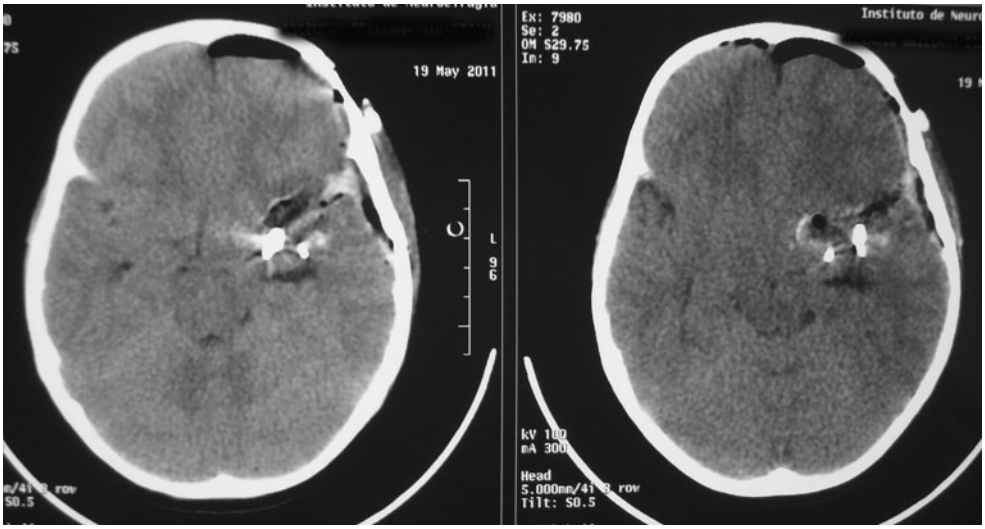

Fig. 6 Postoperative CT scan showing, the miniclips used for the trapping and the partial excision of the aneurysm

endovascular group, with no recurrence in the microsurgical group. In addition, "de novo" aneurysm formation was higher for the endovascular compared with the microsurgical group, with 19% and 6%, respectively, with a mean of 5.7 years of follow-up [10]. The data from Kakarla showed an annual recurrence rate of 2.6% and a novo formation in 7.8% in 53 months of angiographic follow-up. Their numbers for the long-term morbidity were 14% with 3% mortality with 59 months of follow-up [2]. Recently, Sanai showed a 93% obliteration rate in the microsurgical group against 79% in the endovascular group, with functional results and a morbidity similar for both treatments, concluding that the need for an additional aneurysm treatment was over four times higher in children receiving the endovascular therapy [8].

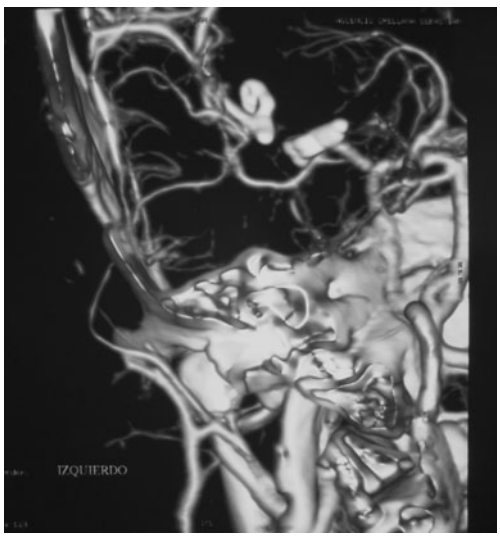

Fig. 7 AngioCT 3D reconstruction showing the clipping and trapping of the lesion, the preservation of the anterior temporal artery, the STA–MCA bypass just distal to the miniclips, and the filling of the arteries distal to the bypass

In the bypass group, there are reports from IC–IC and EC–IC bypass with vein graft [5], with radial artery graft [2], and superficial temporal artery [2, 6]. The STA–MCA bypass was published only in two pediatric patients before, the first by Goedee in an 11-year-old girl with a giant intracranial M2 aneurysm [6] and the other by Kakarla in a 12-year-old patient [2].

In our knowledge, our case may be the first STA–MCA in a 3-year-old patient. We decided on the microsurgical option, because we think that even if the endovascular option is available, if the aneurysm is giant and thrombosed, the mass effect would not disappear after this kind of treatment. In our case, the aneurysm was in very close relation with the anterior temporal artery and most of the lateral lenticulostriate arteries; therefore, the endovascular treatment carries the risk of sacrificing them. Moreover, we think that a balloon occlusion test that showed that pial anastomosis or the opening of leptomeningeal channels that

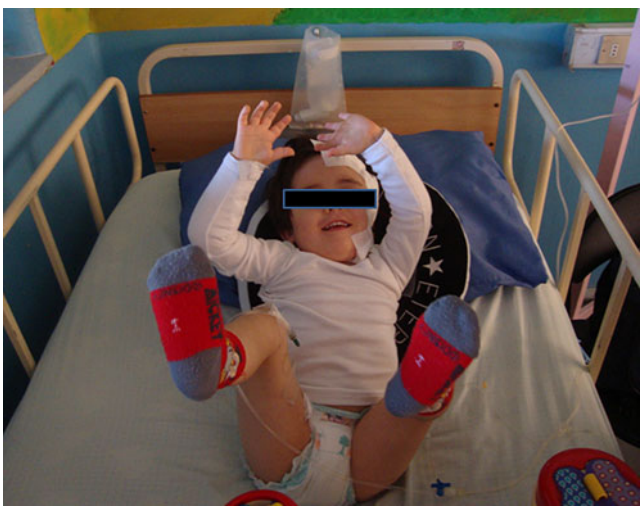

Fig. 8 Postoperative photography of our patient in very good condition and without any deficit

supply in a retrograde fashion the M2 segment in a controlled scenario is not completely reliable to predict long-term ischemic complications. The so-called "collateral blood flow compensation" can be insufficient if the children do sports or any exercise that increases the hemodynamic stress, and it can lead them to a delayed cerebral ischemia or infarction. We strongly believe that microsurgical pure trapping of the aneurysm will have the same effect that endovascular treatment has. However, if the aneurysmal decompression can be achieved combined with a revascularization flow restoration procedure, the final clinical result can be extremely different, with a reduction of the long-term risks secondary to the MCA occlusion. Finally, we think that even if there are no randomized clinical trials evaluating the extracranial–intracranial bypass surgery in intracranial aneurysms, there is a long list of case series that support our conduct and show that these revascularization procedures can reduce the risk of late ischemic stroke [11]. There are still some questions that need answers like which is the best choice of graft in each patient and what will happen in 20 years of follow-up with that graft. We believe that a maturation of the bypass will take place as we have seen in our series of young adult patients treated with STA–MCA bypasses in cases of acute arterial sacrifice of the ICA or the MCA.

Conclusion

For giant fusiform or dissecting M1 aneurysm, literature is very clear showing that microsurgery, with trapping and decompression, combined with a revascularization flow restoration procedure is the best choice for having better, complete, and permanent aneurysm exclusion; lesser recurrence rate; de novo aneurysm formation; and four times less need for an additional treatment over the endovascular therapy. Moreover, the most important effect is the prevention of delayed ischemic events in a developing normal child brain. Endovascular treatment is still a good option for some aneurysms in children that do not have mass effect, with severe co-morbidity. We acknowledge that both treatment therapies have similar morbimortality risks in these particular cases. Nevertheless, the microsurgical approach combined with flow restoration revascularization is the best available technique considering the long-term outcome in the pediatric population.

Acknowledgments We thank Mabel Amores for providing the artwork and John Jane, Jr. for critical review and assistance in the preparation of the paper.

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