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Journal of Neuroscience and Neurological Surgery
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Review Article

Microsurgical Treatment of a Giant Intracavernous Carotid Artery Aneurysm in a Pediatric Patient Case Report and Literature Review

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Received date: January 02, 2022; **Accepted date:** January 24, 2023; **Published date:** February 03, 2023

Citation: Edgar Natha, Javier Degollado-García, Alfredo Bonilla-Suastegui, Héctor A. Rodríguez-Rubio, Bill Roy Ferrufino-Mejia, et all (2023). Microsurgical Treatment of a Giant Intracavernous Carotid Artery Aneurysm in a Pediatric Patient Case Report and Literature Review. *J. Neuroscience and Neurological Surgery.* 13(1); DOI:**10.31579/2578-8868/256**

Abstract

Introduction: Intracranial aneurysms in pediatric age represent 4 - 5% of all cases, and 20% are giant (>25mm). The main sites of occurrence are ICA and MCA; about 55% - 72.5% of cases present rupture and secondary SAH with a 10% - 23% mortality.

Case Description: We report the case of a previously healthy nine-year-old woman that initiated severe right retroocular pain and holocraneal headache. Her relatives detected paresis of the III, IV, and VI cranial nerves three days later. After evaluation, an MRI showed the presence of a giant aneurysm in the cavernous portion of the internal carotid artery (ICA) with a mass effect. The patient was treated surgically through a high-flow bypass using a radial artery graft and trapping of the aneurysm. The patient had an uneventful postoperative course and was discharged three days after the operation to continue in follow-up at the outpatient clinic.

Discussion: The options for treatment are endovascular treatment through flow diverters or stenting and coiling, with the risk of incomplete occlusion or thrombosis. On the other hand, the goal of surgery is the permanent occlusion through the proximal closure of the ICA if a balloon occlusion test shows good collateral circulation or by trapping the aneurysm combined with a high-flow bypass if the collateral circulation is not good or absent.

Conclusion: Even when the surgical option was successful in this case, there is no consensus about the best treatment; the selection of the method to use in these aneurysms depends on the center´s experience when confronting this rare entity.

Key words: giant intracranial aneurysm; bypass; trapping; pediatric neurosurgery

Introduction

Intracranial aneurysms in pediatric age represent 4 - 5% of all cases, and they have an annual "de novo" rate of 0.7% [1,2,4]. Aneurysms of the anterior circulation represent 75% of the cases. The main sites of occurrence are the internal carotid artery (ICA) and the middle cerebral artery (MCA), representing 27% and 26% of the total. In the posterior circulation, the basilar top is the most common site of location [4]; About 55% - 72.5% of cases present rupture and secondary subarachnoid hemorrhage (SAH) with a 10% - 23% mortality [1,2,4]. About 27.5% - 30% of unruptured aneurysms present with thunderclap headache without SAH, partial III nerve palsy, ischemic stroke, and seizures (2). On the other side, saccular aneurysms exceeding 25 mm in maximal diameter have traditionally been classified as "giant" in size. Giant lesions comprise approximately 2 to 5 percent of all cases. Still, in children, they occur with far greater frequency (about 20% in some reported series). Approximately 40% occur in the carotid distribution, 25% in the anterior and middle cerebral artery distribution, and 30% in the vertebrobasilar territory [8]. In this report, we discuss the case of an unruptured giant intracavernous carotid artery aneurysm, presenting as an acute cavernous

sinus syndrome, in a previously healthy 9-year-old female patient. The surgical strategy used to solve the case is also discussed.

Case Report/Case Presentation

A 9-year-old female without important background information presented a pulsatile right retroocular pain with secondary holocraneal headache associated with nausea and vomiting. She received medical attention and was diagnosed with ocular infection. Three days later, her mother observed the absence of ocular movements, and the patient was sent to an ophthalmologist who diagnosed probable right orbital cellulitis. The patient was redirected to a pediatric third-level hospital. The neurological examination showed a right palpebral ptosis, mydriatic pupil not reactive to light stimuli, and complete paralysis of the right III, IV, and VI cranial nerves. An MRI study revealed a giant aneurysm in the cavernous portion of the right ICA (shown in Fig. 1). An angiography with a balloon occlusion test (BOT) was completed showing poor collateral circulation. After analysis of the case, the vascular neurosurgery department decided to perform trapping of the aneurysm with a high flow-bypass from the external carotid artery to the middle cerebral artery with a radial artery graft.

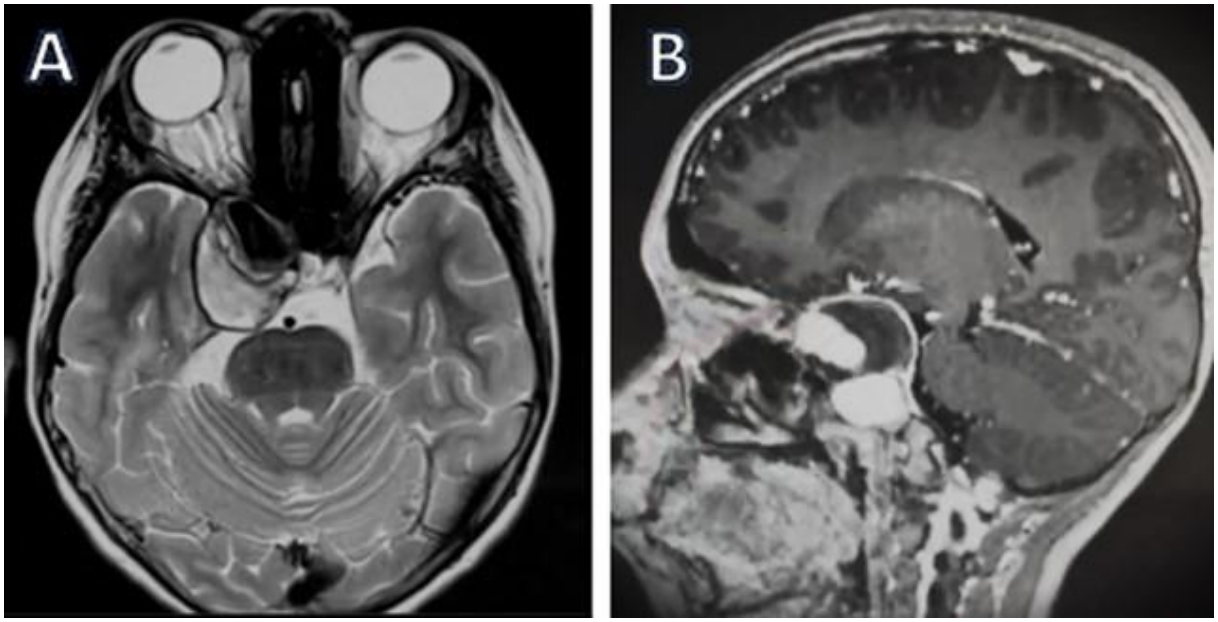

Figure 1: MRI, axial (T2), and sagittal (T1) views show an aneurysm of 54mm in diameter partially thrombosed in the cavernous segment of the right ICA.

Surgical Technique

The patient was positioned supine with a left-side head rotation of 30o and fixed with the three-pin Mayfield head holder. A pterional approach was completed with an additional neck incision following the anterior border of the sternocleidomastoid muscle for carotid exposure. The carotid bifurcation was identified and isolated. Simultaneously, the left forearm was prepared to obtain a radial artery graft. A previous Allen test showed good collateral blood flow at the palmar arch. After obtaining the graft, it was tunneled from the neck to the temporal side of the craniotomy. The inferior trunk of the M2 segment of the MCA was isolated with a dam. A proximal end-to-side anastomosis was completed using nylon 9-0. After, we performed an end-to-side anastomosis between the radial graft and the external carotid artery with an 8-0 suture (shown in Fig. 2). The patency of the bypass was confirmed with fluorescein video angiography (FL-VAG) and intraoperative Doppler. The postoperative course was uneventful, and the patient was discharged three days later to continue with a follow-up at the outpatient clinic (shown in Fig. 3).

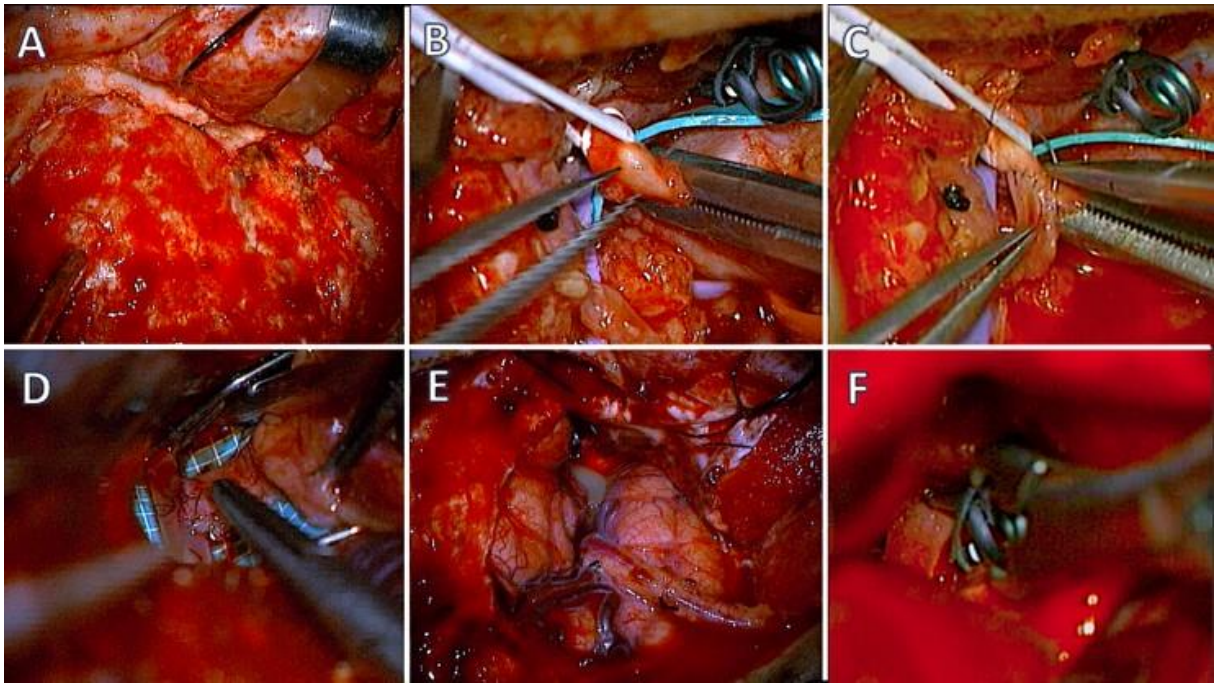

Figure 2. Intraoperative images. A) Right pterional craniotomy. B) The carotid bifurcation is identified, and the external carotid artery is isolated. C) A termino-lateral anastomosis is made with a radial artery graft previously obtained from the left arm with an 8-0 suture. After completion of the anastomosis, the clamp is released, and patency of the proximal bypass is confirmed. D) The recipient artery is prepared at the level of the Sylvian fissure. The inferior trunk of the MCA is selected for the second end-to-side anastomosis. The anastomosis is completed with a 9-0 suture using separate points. E) The anastomosis is completed, and its functioning is verified with the intraoperative Doppler. F) After closing the ICA at the bifurcation with a 0-silk suture, a clip is positioned just proximal to the origin of the ophthalmic artery to trap the cavernous sinus aneurysm permanently.

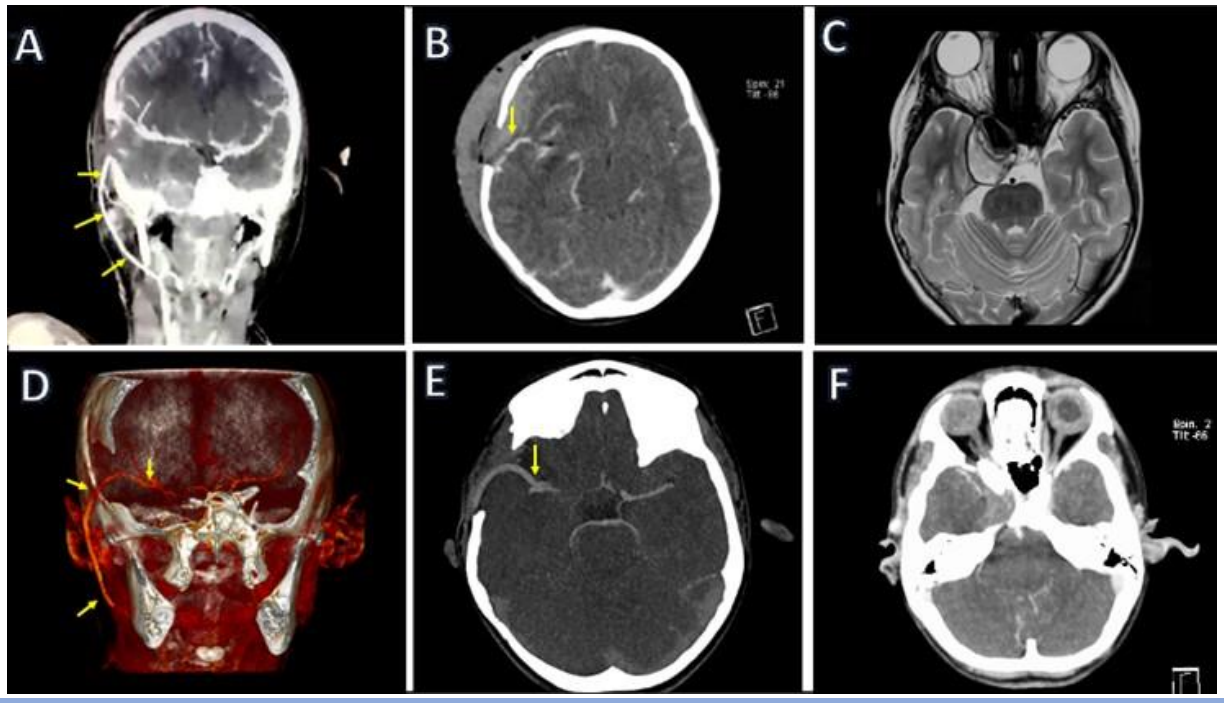

Figure 3. Postoperative images of 2 months (A-C) and 6 months (D-F) after surgery. A and D) Coronal view shows a patent bypass (yellow arrows). B and E) CTA in axial view. The MCA's bypass (arrow) and branches are seen without evidence of low-density areas. C and F) Pre and postop (6 months) images of the aneurysm. The mass effect is reduced with improvement of the cavernous sinus symptomatology.

Figure 4. Postoperative evolution. Superior row (A-C) shows the postop course at 2 months. A) Partial opening of the right eyelid, with an adequate response of the superior rectus muscle and spontaneous pupillary reflex. B) Adequate response of superior oblique muscles. Resolution of the right IV nerve lesion. C) Paralysis of the right abducens muscle (VI nerve palsy) is still present. Inferior row (D-F), postop course at 6 months. D-E) Adequate function of III nerve. F) Complete remission of VI nerve palsy.

Discussion/Conclusion

Clinical presentation

Treatment of giant cavernous aneurysms is still a matter of concern. There exists the possibility of endovascular treatment using flow diverters or some alternative technique such as stenting and coils ("jailing") [3,4,10]. However, the availability of these devices is limited in some countries. In the case of the "jailing" technique, there exists the possibility of perpetuating the mass effect producing permanent pressure over the cranial nerves [13]. Conversely, the surgical technique depends upon the BOT results to demonstrate the grade of collateral blood flow. If the patient shows an excellent flow, the simple proximal ligation of the ICA (Hunterian ligation) is enough to induce aneurysm thrombosis. In this case, some authors recommend completing a low-flow superficial temporal artery to middle cerebral artery (STA-MCA) bypass to avoid "de novo" aneurysms in the long term. In case of insufficient collateral blood flow, trapping the aneurysm together with a high-flow bypass will solve the situation [4,6,11,12]. Our patient presented a cavernous sinus syndrome secondary to a giant aneurysm in the cavernous portion of the ICA. A similar case was reported in an 11 years old child who presented with right eye exotropia associated with a severe headache [7]; in another case, the aneurysm caused a severe retroocular pain diagnosed as a Tolosa–Hunt syndrome [9]. These syndromes are explained by the mass effect caused by the aneurysm over the lateral wall of the cavernous sinus.

Association with other pathologies

Some studies have described the relationship of intracranial aneurysms in children with diverse diseases such as problems in reticular fiber formation, gene mutations, connective tissue diseases (v.gr. Marfan's syndrome, Ehler-Danlos syndrome), coarctation of the aorta, fibromuscular dysplasia, sickle cell anemia, Paget syndrome and tuberous sclerosis [2,4,5,6]. Approximately 15% of the cases can be due to an infectious etiology [7]. Further work did not show any of these conditions in our patient.

Treatment of choice

Intracranial aneurysms in pediatrics are uncommon but have a higher risk of rupture than in the adult population [8]. In the case of cavernous sinus aneurysms, they present with a mass effect, acting like a tumor. In the case of rupture, they produce a carotid-cavernous fistula with ipsilateral ocular signs and symptoms. As was said before, treatment could be accomplished through clipping or endovascular coiling; however, for aneurysms at this location, studies show better results with clipping in terms of durability, recurrence rate, and less need for additional procedures [4,6]. Some authors, like Sania et al., describe a higher recurrence rate in endovascular therapy and a higher rate of "de novo" aneurysm formation in children [11].

Regarding the endovascular treatment for a similar case like ours, there exist reports of successful treatment of a giant aneurysm in a child using a flow diverter stent with a good evolution three years after the procedure [10]. On the other side, surgical treatment was used in a child with a bilateral giant aneurysm that underwent a bilateral STA-MCA bypass, followed by endovascular carotid artery occlusion with a balloon on both sides, tolerating both procedures without postoperative complications [6]. These cases are described in table 1, together with other reports from the literature.

Table 1. Published cases of giant intracavernous aneurysms in pediatric age

Case No	Authors	Year	Age/ Sex	Associated pathology	Clinical presentation	No.	Side	Treatment	Complications	Follow-up
1	Devadiga et al. [14]	1969	1/M	None	Paresis of left side	1	Lt	None	Dead	None
2	Banna et al.[15]	1990	1/F	Chest infection	Proptosis	1	Rt	Embolization	None	4 Mo
3	Linskey et al.[16]	1991	15/M	None	Orbital pain	1	Lt	Proximal occlusion	None	2 Mo
4	Cekirge et al. [3]	1996	4/F	None	Left third nerve palsy	2	Bil	Coiling	None	3 Mo
5	Sanai et al.[11]	2010	15/M	None	Diplopia and headache	1	Lt	Proximal balloon occlusion	None	1 Mo

6	Meyer et al.[12]	1989	NI	None	Mass Effect	1	Rt	Proximal occlusion and Bypass	None	2 yrs
7	Rehman et al.[6]	2010	11/M	Juvenile Paget's disease	Left abducens nerve palsy	2	Lt	Bypass / proximal occlusion	None	2 Mo
8	Kumaria et al.[10]	2018	12/M	Septic artristis	Diplopia	1	Lt	Flow diverter stent	None	3 yrs
9	Buño et al.[7]	2018	11/M	Horner Syndrome	Diplopia	1	Rt	Clipping	None	6 Mo
10	Present case	2022	9/F	None	Proptosis	1	Rt	Bypass/ Trapping	None	2 Mo

Abbreviations: NI, no information; Rt, right-side; Lt, left-side; Bil, bilateral; female gender; M, male gender; Mo, months; yrs, years.

Table 1: Published cases of giant intracavernous aneurysms in pediatric age.

Outcome giant aneurysms in the pediatric population

A retrospective multicentric study documented unfavorable outcomes with aneurysms larger than 5 mm, and this size was also independently associated with aneurysm recurrence. The annual "de novo" aneurysm rate, mainly based on 3 Tesla MRI, was 0.7%. [2]. In the case of cavernous sinus aneurysms, it is expected that the mass effect will improve after treatment, reducing the compression of the aneurysm over the cavernous sinus, and alleviating the symptoms of the patients. In our case, the extraocular motility deficits and proptosis resolved after surgical treatment, as was previously reported in similar cases (shown in Figure. 4) [7,15].

Conclusions

There has yet to be a consensus about the best treatment for giant cavernous aneurysms in the pediatric age. Favorable results depend on comorbidities, clinical presentation, and the centers' experience treating this rare entity, so the published cases still need to permit us to draw a unique conclusion. Regarding the treatment used for this case, we can say that proximal carotid occlusion, based on a BOT showing excellent collateral circulation, is an acceptable method of treatment to induce thrombosis of the aneurysm and further improve the symptoms caused by the mass effect. In case of poor collateral circulation, a high-flow bypass and trapping the aneurysm is the best way of treatment. Direct clipping of cavernous sinus aneurysms has been abandoned because of the high incidence of cranial neuropathies.

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DOI: 10.31579/2578-8868/256