



Flow diverter stents for pediatric traumatic carotid cavernous fistula: a case report and literature review

Kyoji Tsuda¹ · Takahiro Ota² · Maya Kono¹ · Satoshi Ihara¹

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Abstract

A 10-year-old, male patient with a head injury caused by a fall presented with chemosis, exophthalmos, right orbital bruit, and intracranial venous reflux, based on which posttraumatic carotid cavernous fistula (CCF) was diagnosed. Coil embolization was semi-urgently performed for the dangerous venous drainage. After the treatment, right abducens nerve palsy newly appeared. To treat the neurological symptoms and preserve the parent artery, curative endovascular treatment using a pipeline embolization device (PED) with coil embolization was performed after starting dual antiplatelet therapy (DAPT). The CCF and abducens nerve palsy finally resolved, and the internal carotid artery (ICA) was remodeled. Use of the PED with adjunctive coil embolization was effective and safe in the present case of pediatric traumatic direct CCF.

Keywords Pediatrics · Posttraumatic · Carotid cavernous fistula · Flow diverter stent

Abbreviations

CCF	Carotid cavernous fistula
ICA	Internal carotid artery
ICH	Intracerebral hemorrhage
MCA	Middle cerebral artery
ACA	Anterior cerebral artery
TVE	Transvenous embolization
TAE	Transarterial embolization
PED	Pipeline embolization device
DAPT	Dual antiplatelet therapy

vision loss, oculomotor or abducens nerve palsy, headache, and, occasionally, convulsions or intracerebral hemorrhage (ICH) [1–3]. The ideal treatment involves occluding the arteriovenous shunt while maintaining ICA patency [4]. Detachable coils, n-butyl-2-cyanoacrylate, and Onyx are normally used in the standard treatment. However, numerous studies have recently examined the use of flow diverter stents in CCF treatment [1, 4–11] although most of the patients were adults or adolescents. We herein reported a pediatric case of traumatic direct CCF successfully treated using a flow diverter stent with detachable coils.

Introduction

Posttraumatic carotid cavernous fistula (CCF) usually presents high-flow shunt via direct fistulas between the internal carotid artery (ICA) and cavernous sinus. Symptoms include chemosis, exophthalmos, orbital bruit, pulsating tinnitus,

Case presentation

The patient was a 10-year-old male who fell from a height of two meters. He presented with bruising of the right eye and vision loss (0.02). Head CT revealed right orbital fractures (Fig. 1A). On day 28, chemosis and exophthalmos of the right eye (Fig. 1B) and pulsatile bruit were observed. Ophthalmological examination revealed temporal hemianopia and visual loss (0.15) in the right eye. Brain MRI showed engorgement of the right superior orbital vein and superficial middle cerebral veins. Based on these findings, posttraumatic direct CCF was diagnosed. To ameliorate the symptoms, semi-emergent endovascular treatment was performed. A right internal carotid angiogram performed under general anesthesia revealed the direct CCF

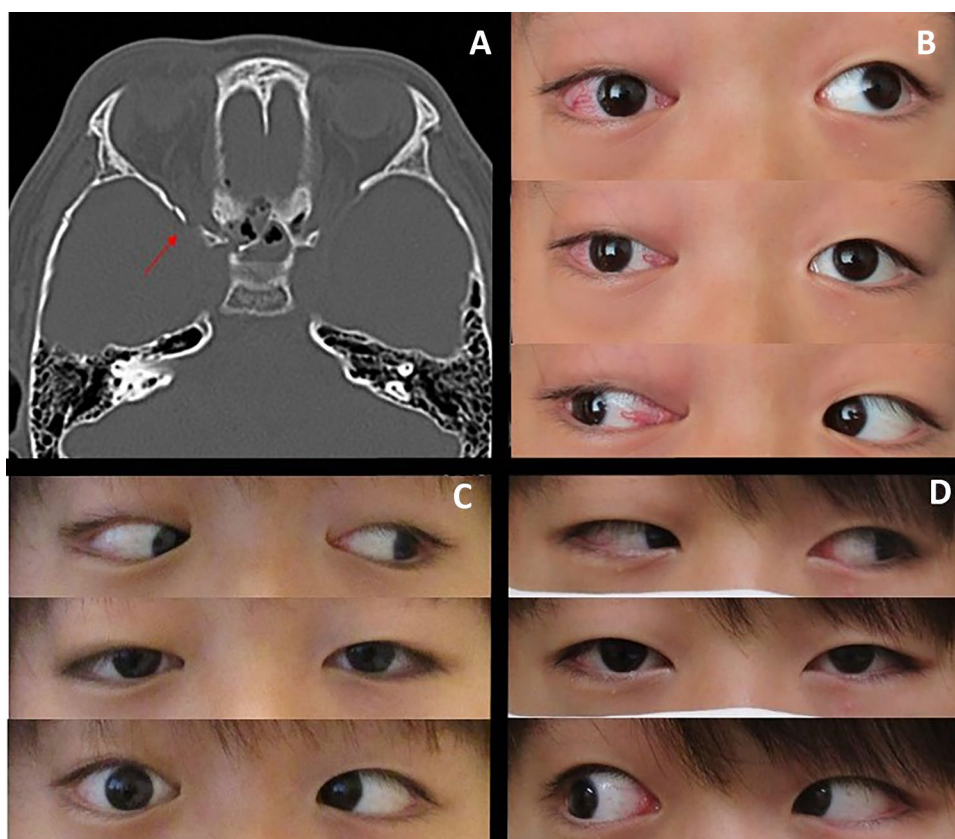
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✉ Kyoji Tsuda
kyoji.tsuda0412@gmail.com

¹ Department of Neurosurgery, Tokyo Metropolitan Children's Medical Center, 2-8-29 Musashidai, Fuchu, Tokyo 183-8561, Japan

² Department of Neurosurgery, Tokyo Metropolitan Tama Medical Center, Tokyo, Japan

Fig. 1 Head CT at the first visit (A) and photographs of eye positions at diagnosis (B) 3 days after the first treatment (C) and 2 weeks after final treatment (D). The head CT revealed lateral right orbital bone fractures (arrow) (A). Chemosis and exophthalmos of the right eye were observed at posttraumatic CCF diagnosis (B). Although the chemosis and exophthalmos improved, right abducens nerve palsy appeared 3 days after the first TVE for the venous drainage (C). The right abducens nerve palsy resolved after the last treatment (D)



between the right ICA and cavernous sinus, venous reflux into the right superior orbital vein, superficial middle cerebral veins, and uncal vein, and poor opacification of the right middle cerebral artery (MCA) and anterior cerebral artery (ACA) (Fig. 2A). A left internal carotid angiogram revealed opacification of the right ACA and MCA via the anterior communicating artery. Under systemic heparinization, a 5-Fr guiding sheath and 3.4-Fr intermediate catheter were advanced into the right internal jugular vein and the cavernous sinus, respectively. The microcatheter was advanced into the uncal vein, superior orbital vein, and superficial middle cerebral veins, then, transvenous embolization (TVE) with coils was performed. The post-treatment angiogram showed resolution of the venous reflux (Fig. 2C). Postoperatively, the chemosis and exophthalmos improved, but on postoperative day 3, right abducens nerve palsy appeared (Fig. 1C). To treat the neurological symptoms and preserve the ICA, curative endovascular treatment using a pipeline embolization device (PED) with TAE was scheduled for postoperative day 21. The patient was pretreated with dual antiplatelet therapy (DAPT) consisting of aspirin (81 mg/day) and clopidogrel (37.5 mg/day). Under general anesthesia, a triaxial system, including a microwire, microcatheter, and 5-Fr intermediate catheter, was navigated into the proximal region of the CCF. A 4 × 18-mm PED (Pipeline shield, Medtronic, Ireland)

was navigated distal to the posterior communicating artery and deployed to cover the CCF (Fig. 2D). Subsequently, an adjunctive TAE was performed with coils using the jailing technique (Fig. 2F). Since the TAE failed to occlude the fistula, and natural thrombosis had not occurred, TVE with coils was performed in the next session six weeks after the second treatment. The final angiogram revealed almost complete disappearance of the shunt flow and restoration of the antegrade ICA flow (Fig. 2G). By postoperative day 14, the right abducens nerve palsy had completely resolved (Fig. 1D). However, visual acuity in the right eye returned only to 0.20, and the temporal hemianopia remained. Four months later, a cerebral angiogram visualized complete occlusion of the CCF and ICA remodeling (Fig. 2H). DAPT was then terminated. The patient was followed up for ten months but experienced no recurrence.

Discussion

Three sessions of endovascular treatment were performed in this case. These staged therapies ensured that the dose of contrast medium per session could be kept to a minimum, DAPT could be safely started without dangerous venous reflux, and neurological symptoms could be improved prior to radical treatment. Unexpectedly, the

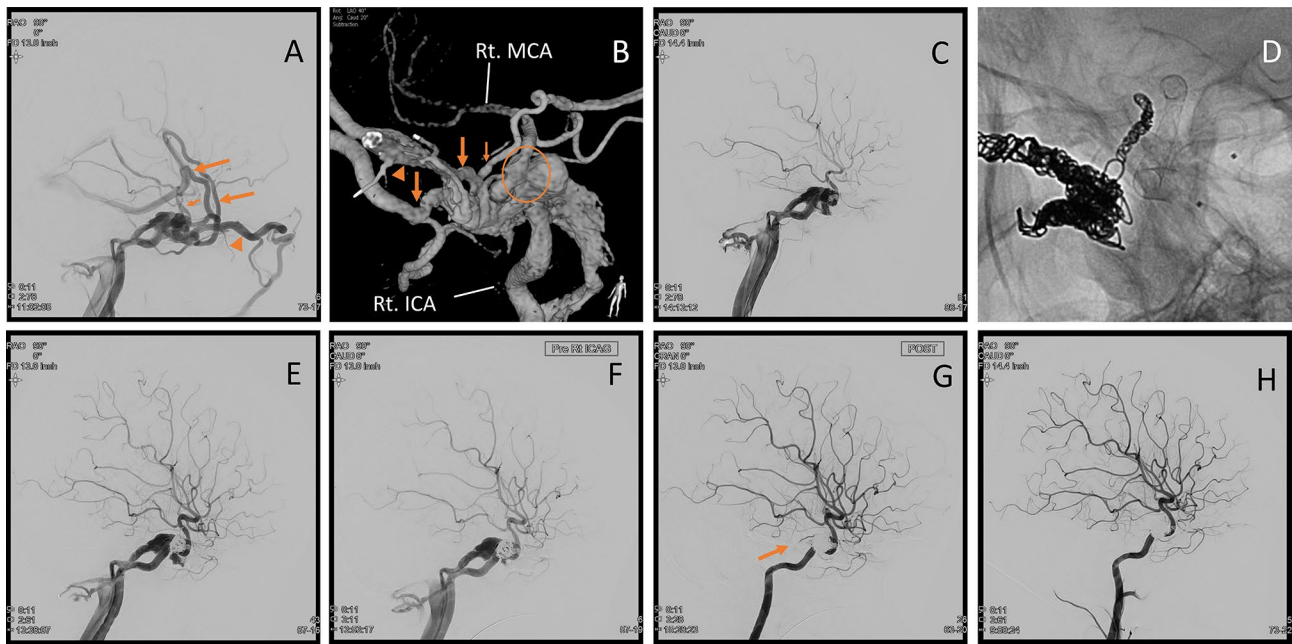


Fig. 2 Right internal carotid angiograms. Lateral view (A) and 3-dimensional rotational image (B) of preoperative internal carotid angiogram revealed the direct CCF, venous reflux into the right superior orbital vein (arrow head), superficial middle cerebral veins (arrow), and uncal vein (small arrow), and the shunt point (circle). Lateral view of the internal carotid angiogram after the first treatment demonstrated resolution of the venous reflux (C). PED placement

across the fistula can be seen (D). Deployment of PED with adjunctive TAE using coils was performed, resulting in incomplete fistula occlusion (E). Lateral view of the right internal carotid angiogram before (F) and after (G) the third treatment demonstrated nearly complete resolution of the shunt (arrow). Four months later, the CCF was completely occluded (H)

right abducens nerve palsy appeared after the first session, possibly due to increased pressure in the cavernous sinus after the drainage of the CCF decreased. The right abducens nerve palsy resolved completely in two weeks after the final treatment. In contrast, limited improvement was observed in the vision of the right eye, apparently due to the initial, traumatic damage to the optic nerve.

Several case series have reported the safety and efficacy of flow diverter stents [1, 4–11]. However, most of the patients enrolled in these studies were adults or adolescents [1, 4, 11]. We reviewed the literature on posttraumatic CCF in pediatric patients aged 10 years or younger (Table 1) and found that Barburoglu et al. reported two pediatric cases of CCF treated with flow diverter stents but

Table 1 Reported cases of pediatric posttraumatic carotid cavernous fistula aged 10 years or younger

Author	N	Sex	Age*	Treatment	Preservation of parent artery	Complete occlusion of CCF
Debrun [13]	1	F	5	Detachable balloon	No	Yes
Barrow [12]	1	F	10	Detachable balloon	Yes	Yes
Yang [18]	1	M	6	TVE with coil	Yes	Yes
Paiva [15]	1	M	10	TVE with coil	Yes	Yes
Pawer [16]	1	F	6	TVE and TAE with coil	N/A	Yes
Wajima [17]	1	M	8	Aneurysm coil embolization with PAO	No	Yes
Barburoglu [1]	2	N/A	N/A [†]	Flow diverters	N/A	N/A
Morais [14]	1	M	8	Detachable balloon	Yes	Yes
Present case	1	M	10	TVE and TAE with coil using flow diverter stent	Yes	Yes

*Age at treatment

[†]Their ages are between 3 and 16 years, but the details are not available

gave no details about their age [1]. Only seven traumatic CCF cases in patients aged 10 years or younger who were treated with other methods have been reported [12–18].

Treatment is generally required for posttraumatic CCF. However, packing too many coils into the cavernous sinus may increase the internal pressure, exacerbating the neurological symptoms [19]. Parent artery occlusion may be required if the fistula cannot be occluded by other means. Hemodynamic stress caused by non-physiological conditions may increase the risk of de-novo aneurysm formation later [20]. Sacrificing the parent artery should be considered carefully in children.

Progress in flow diverter stent technology has significantly impacted the treatment of direct CCF by allowing (1) preservation of the parent artery during fistula occlusion; (2) reduction in the amount of coil used with multiple flow diverter stents; and (3) thrombosis induction even in the presence of residual shunt flow after treatment. These effects can reduce the stenosis and thrombosis risk in the parent artery and prevent neurological complications [4]. Flow diverter stents may be used as a scaffold and treated with coils or liquid embolic substances [4, 5]. In stand-alone treatments, multiple stents will usually be required to occlude the fistula completely [5].

Flow diverter stents have come to be used commonly for most cases of CCF in recent years. However, the indications for its use should be carefully considered especially in children because long-term follow-up is required, and the cost may be high in many countries.

Conclusion

Deployment of PED with adjunctive coil embolization for pediatric direct CCF was effective in occluding the fistula while reducing the total amount of coil placed in the cavernous sinus and preserving the parent artery.

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Author contribution Kyoji Tsuda wrote the first draft. Takahiro Ota, Maya Kono, and Satoshi Ihara reviewed and edited the manuscript.

Availability of data and material Not applicable.

Code availability Not applicable.

Declarations

Ethics approval Not applicable.

Consent to participate Not applicable.

Consent for publication The patient and his parents consented to the submission of this case report to the journal. Patients signed informed consent regarding publishing their data and photographs.

Conflict of interest The authors report no conflicts of interest regarding the content of this study.

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