



# Endovascular embolization of dural arteriovenous fistula in a child presented with slight conjunctival hyperemia

Lei Guo<sup>1</sup> · Ting Liu<sup>1</sup> · Xianli Lv<sup>2</sup>

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## Abstract

**Background** Dural arteriovenous fistulas (DAVFs) are rare in pediatrics. A case of DAVF diagnosed because of a slight conjunctival hyperemia and endovascular coil embolization at 2 years old is reported.

**Case description** The 2-year-old boy presented with a slight conjunctival hyperemia of the left eye for 1 month. Magnetic resonance imaging (MRI) examination of the head showed abnormal blood flow in the left middle cranial fossa. On digital subtraction angiography, a DAVF with a dural feeder shunt and a venous varix at the middle cranial fossa was confirmed. After transarterial coil embolization, shunt blood flow disappeared.

**Conclusions** This report describes a case of DAVF with a slight conjunctival hyperemia treated by coil embolization in a child.

**Keywords** Dural, Arteriovenous fistula · Coil · Child · Embolization

## Introduction

Neurointervention in pediatrics, especially neonates, can be more challenging than analogous procedures in old children or adults [1–3]. It needs a multidisciplinary team involved in the treatment of these children [1–3]. Intracranial dural arteriovenous fistulas (DAVFs) are rare in children and constituted 5.7% of pediatric intracranial arteriovenous shunting lesions [4]. Management strategies for these lesions have undergone considerable evolution in the last decade with the advent of new endovascular, surgical, and radiosurgical technologies. Endovascular treatment is currently the first choice of

treatment for most pediatric DAVFs [4]. Embolization of DAVFs in these patients prevented adverse cardiac effects, hydrovenous disorders, and rebleeding. In this study, we present a case of DAVF diagnosed because of a slight conjunctival hyperemia and endovascular coil embolization at 2 years old.

## Case presentation

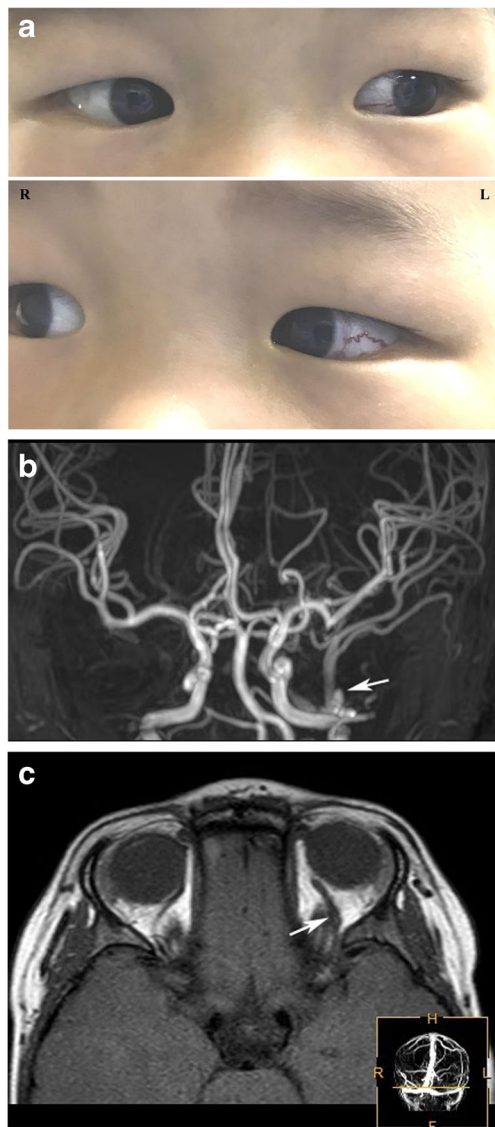
A 2-year-old boy presented with slight conjunctival hyperemia of the left eye for 1 month. Magnetic resonance imaging (MRI) of the head and magnetic resonance angiography (MRA) confirmed that the left middle meningeal artery flowed into the dilated pial veins and the left ophthalmic vein was dilated than that of the right side (Fig. 1a–c).

It was decided that it would be difficult to wait any longer, and endovascular treatment was needed. First, diagnostic digital subtraction angiography was performed with the patient under general anesthesia. The right femoral artery was punctured and a 4F arterial sheath was inserted. The left middle meningeal artery connected directly into the superficial middle cerebral

✉ Xianli Lv  
lvxianli000@163.com

<sup>1</sup> Department of Vascular Anomalies and Interventional Radiology, QILU Children's Hospital of Shandong University, Jinan, Shandong, China

<sup>2</sup> Neurosurgical Department, Beijing Tsinghua Changgung Hospital, School of Clinical Medicine, Tsinghua University, Litang Road 168, Changping, Beijing 102218, China



**Fig. 1** **a** The pictures showed a slight conjunctival hyperemia of the left eye. R, right; L, left. **b** There is arteriovenous shunt of the left middle cerebral artery and the enlargement of the left superficial middle cerebral veins (arrow) on magnetic resonance angiography. **c** T1-weighted magnetic resonance imaging (MRI) showed the enlargement of the left ophthalmic vein (arrow)

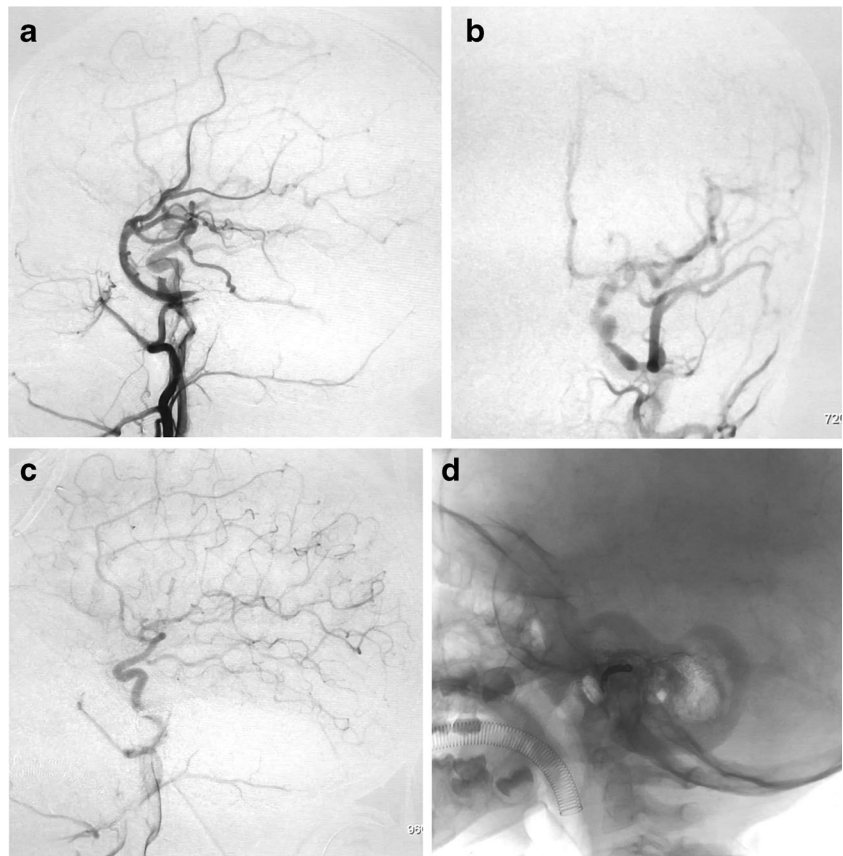
vein and drained into the superior sagittal sinus (Fig. 2a, b). An Echelon-10 microcatheter was introduced through a 4F diagnostic catheter into the left middle meningeal artery. Transarterial obliteration of the shunt was performed using coils and the shunt blood flow disappeared completely after treatment (Fig. 2c, d). After the procedure, there were no new ischemic complications and the patient's course was good. Follow-up MRI and MRA examination was scheduled at 6 months.

## Discussion

DAVFs not directly shunting into the cavernous sinus can also present conjunctival hyperemia as a result of rerouting venous drainage promoting the intracavernous pressure [5]. DAVFs in young children are characterized by frequent high-flow fistulas and the physiological condition of the developing brain and heart; each DAVF type tends to present at a certain age with unique symptoms [4]. Dural sinus malformation with arteriovenous (AV) shunt tends to present in the neonate with high-output cardiac failure. In infancy, infantile DAVF tends to present with hydrodynamic disorder such as macrocephaly, ventriculomegaly, prominent facial veins, developmental delay, or focal neurological signs such as seizure or hemorrhage at older ages [1–4]. In a series of 43 cases of Galen malformation treated with a combined transarterial and transvenous approach, the child did not develop DAVF as long as we used coils solely and the child developed DAVF after the transarterial use of Onyx [6].

Endovascular treatment is currently the first choice of treatment for most pediatric ICAVS. The treatment goal should be defined on a patient-by-patient basis, according to the unique physiological condition of the child. Endovascular embolysates included Onyx, N-butyl cyanoacrylate, or coil embolization [1]. Hettis et al. retrospectively reviewed 22 DAVFs in children and found that good clinical outcome (modified Rankin Scale score 0–2) was documented in 77% of patients > 1 year old at presentation compared with 57% of patients ≤ 1 year old at presentation [4]. Six patients (27%) died. They concluded that compared with other pediatric vascular shunts, DAVFs had lower rates of angiographic obliteration and poorer clinical outcomes. Puccinelli et al. described endovascular treatment of consecutive 52 children weighing less than 5 kg with neurovascular arteriovenous malformations, including 38 vein of Galen aneurysmal malformations, 3 pial AVM, 6 pial arteriovenous fistulas, and 5 dural sinus malformations [7]. Their treatment goals were control of cardiac failure or hydrocephalus in cases of nonhemorrhagic malformations and prevention of new bleeding in cases of previous hemorrhage. A hemorrhagic complication occurred in 12 procedures and an ischemic complication in 2. Both complication types were correlated with the age of the infant (age cutoff at 3 months). No correlation was found with the weight of the infant or the duration of the procedure. The risk of major cerebral complications seems mainly correlated with age, with a threshold at 3 months.

**Fig. 2** **a** Left carotid artery angiogram, lateral view. **b** Left carotid artery angiogram, frontal view. Showed the left middle meningeal artery flow into the left superficial middle cerebral veins and out into the superior sagittal sinus. After coil embolization of the arteriovenous fistula, the middle meningeal artery is plugged with coils. **c** Left common carotid artery angiogram showed blood flowing into the veins has disappeared. **d** Fluoroscopic image showed coils, lateral view



## Conclusion

This report describes a case of DAVF with a slight chemosis treated by coil embolization in a child.

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## Compliance with ethical standards

**Conflict of interest** The authors declare that they have no conflict of interest.

## References

- Zaidi HA, Kalani MY, Spetzler RF, McDougall CG, Albuquerque FC (2015) Multimodal treatment strategies for complex pediatric cerebral arteriovenous fistulas: contemporary case series at Barrow Neurological Institute. *J Neurosurg Pediatr* 15(6):615–624
- Okazaki T, Sakamoto S, Ishii D, Oshita J, Matsushige T, Shinagawa K, Ichinose N, Matsuda S, Kurisu K (2019) A pial arteriovenous fistula in infancy as the presenting manifestation of hereditary hemorrhagic telangiectasia. *World Neurosurg* 122:322–325
- Niimi Y (2017) Endovascular treatment of pediatric intracranial arteriovenous shunt. *Pediatr Int* 59(3):247–257
- Hetts SW, Mofitakhar P, Maluste N, Fullerton HJ, Cooke DL, Amans MR, Dowd CF, Higashida RT, Halbach VV (2016) Pediatric intracranial dural arteriovenous fistulas: age-related differences in clinical features, angioarchitecture, and treatment outcomes. *J Neurosurg Pediatr* 18(5):602–610
- Lv X, Zhang J, Li Y, Jiang C, Wu Z (2008) Dural arteriovenous fistula involving the transverse sigmoid sinus presenting as chemosis. A case report. *Neuroradiol J* 21(3):428–432
- Meila D, Schmidt C, Melber K, Grieb D, Jacobs C, Jacobs C, Lanfermann H, Brassel F (2018) Delayed and incomplete treatment may result in dural fistula development in children with vein of Galen malformation. *Interv Neuroradiol* 24(1):82–87
- Puccinelli F, Tran Dong MNTK, Iacobucci M, Mazoit JX, Durand P, Tissieres P, Saliou G (2019) Embolization of cerebral arteriovenous shunts in infants weighing less than 5 kg. *J Neurosurg Pediatr* 22:1–9

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