Mechanical thrombectomy performed in thrombosed fusiform aneurysm after surgery for craniopharyngioma in adult

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Introduction. Fusiform dilatation of the internal carotid artery (ICA) is reported as a possible complication of craniopharyngioma resection in childhood. Here, the authors describe such a complication in an adult patient who presented with acute symptomatic thrombosis 7 months after surgery.

Materials and Methods. A 45-year-old woman presented with left hemispheric stroke due to a thrombotic supraclinoid occlusion of the terminal ICA (so called "T" occlusion). Successful revascularisation was achieved with mechanical thrombectomy. Beside recanalization of the M1 middle cerebral artery segment and ICA, an irregular filling of the fusiform aneurysm of the communicating segment of the left ICA was observed. The patient recovered after mechanical thrombectomy with no clinical sequelae. Due to the persistent filling of the aneurysm sac, a flow diverter stent was deployed across the diseased vessel segment two weeks later. The patient underwent resection of the craniopharyngioma from ipsilateral pterional craniotomy 7 months ago. Five years later the patient works full time as a nurse with no regrowth of the craniopharyngioma and no aneurysm reperfusion.

Results. This case, together with four other previously reported cases, documents that fusiform aneurysm as a complication of the craniopharygioma resection is not restricted to the childhood population but may also rarely occur in adults. As the patient suffered from acute symptomatic thrombosis which required treatment under the protocol for acute large vessel occlusions, we decided to treat the aneurysm with the flow diverter stent.

Key words: dissecting aneurysm, acute ischemic stroke, endovascular therapy, flow diverter stent, craniopharyngioma

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INTRODUCTION

Fusiform dilatation of the internal carotid artery (ICA) after resection of the suprasellar craniopharyngioma is a known possible complication in children and has been reported previously[1-4]. This vascular complication is extremely rare in adults[5]. There is a general opinion that these fusiform dilatations should be managed conservatively since few of them become symptomatic. Besides partial arterial wall injury during tumor resection, excessive radiation was observed to be another pathogenetic mechanism in several cases[6].

We present a case of iatrogenic, suspiciously dissecting, fusiform aneurysm of the communicating ICA segment in a woman who underwent resection of the craniopharyngioma from ipsilateral pterional craniotomy.

CASE REPORT

A 45-year old female patient was admitted to the primary stroke center with sudden onset of motor weakness of the right sided extremities, expressive aphasia and central paresis of the facial nerve. Her neurological deficit was assessed based on the National Institutes of Health Stroke Scale (NIHSS) score as 5. The non-contrast CT (NCCT) scan showed no early signs of brain ischemia, CT angiogram (CTA) revealed "T" occlusion of the left intracranial ICA (Fig. 1.A,B). The perfusion CT showed extensive penumbra with no ischemic core.

The patient had a previous history of partial transcranial resection of the craniopharyngioma from pterional craniotomy 7 months prior. Before the surgery, she underwent local radiation therapy using an implanted yt-

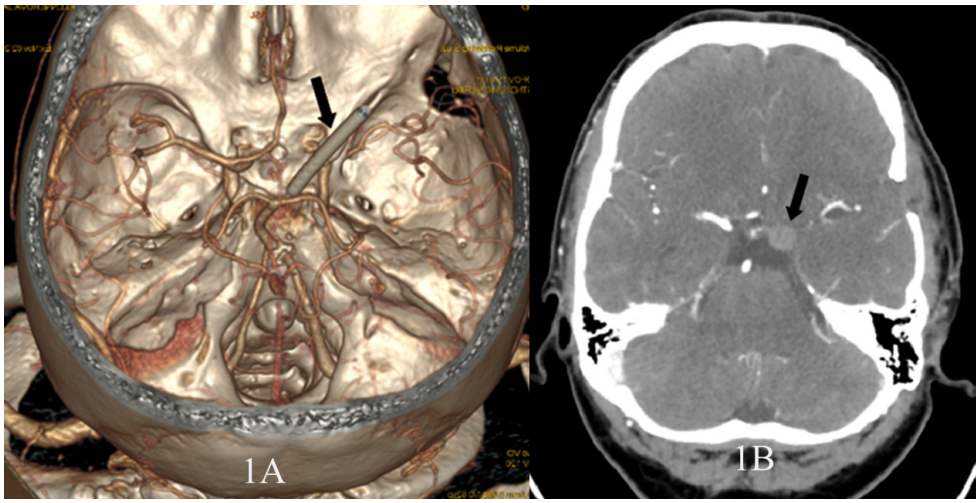

Fig. 1A. The volume CT angiography reconstruction shows occlusion of the communicating segment of the left ICA, M1 and A1.There is an Ommaya reservoir (arrow), which is in vicinity of the occluded artery**. B.** The CT angiogram reveales fusiform thrombosed aneurysm of the ICA (arrow).

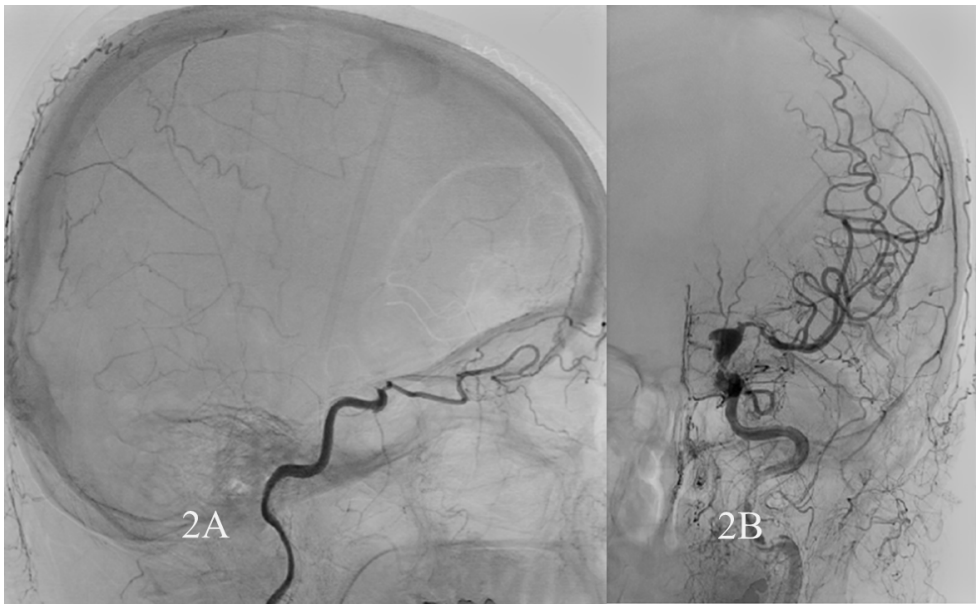

Fig. 2A. Angiogram of the left ICA proved its occlusion. **B.** Angiogram after 3 passes of the stent retriever. The flow was restored and an irregular fusiform aneurysm was revealed. There was also a stenosis of the M1 segment of the MCA and vasospasm on extracranial ICA.

trium Ommaya reservoir. The last MR scan, which was performed 3 months after surgery, demonstrated subtotal reduction of the tumorous volume and no pathological changes of the left ICA.

The patient had a stable neurological deficit from the time of symptom onset, and based on suitable findings on CT perfusion, she was transferred to a comprehensive stroke center, where the digital subtracted angiogram (DSA) was performed 11.5 h from symptom onset. The DSA revealed a "T" occlusion of the left terminal ICA (Fig. 2A) and no cross filling from the contralateral carotid artery. Retrospective assessment of the baseline NCCT and CTA revealed the presence of hyperdense fusiform ectasia of the left supraclinoid ICA segment, which was occluded (Fig. 1A).

Mechanical thrombectomy was performed with a stent retriever 4 x 20 mm (Trevo XP ProVue Retriever, Stryker Neurovascular, Fremont, USA) using a 9F balloon guide catheter (Concentric – Stryker Neurovascular, Salt Lake City, USA) (Fig. 2B). The stent retriever was deployed in the M1 segment of the middle cerebral artery (MCA). Mechanical thrombectomy yielded a significant amount of thrombotic material with TICI 2B reperfusion of the M1 MCA segment and the ICA after 3 passes. Aresidual stenosis of the M1 segment and vasospasm of the extracranial ICA were observed. There was an irregular filling of the fusiform aneurysm of the left communicating ICA segment. The aneurysm had a diameter of 8 mm and was extended to a 10 mm long segment of the terminal ICA (Fig. 2B). The left anterior cerebral artery was occluded

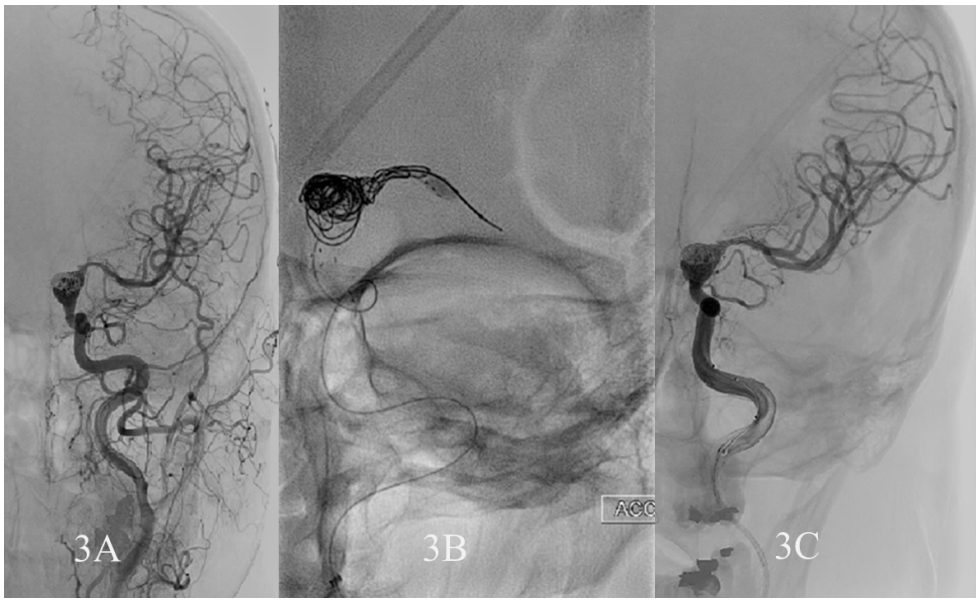

Fig. 3A. Angiogram of the left ICA after stent-assisted coiling. The most prominent part of the aneurysm was partially coiled. **B.** Balloon angioplasty of the distal part of the flow diverter stent immediately after its implantation. There were dislodged coils, which were placed into the aneurysm 3 weeks ago. **C.** Final angiogram which proved a brisk flow into the MCA. The A1 had already been occluded during acute ischemic stroke. There was no cross filling via the anterior communicating artery. The left anterior cerebral artery had retrograde flow through the pial collaterals from the left posterior cerebral artery (not shown).

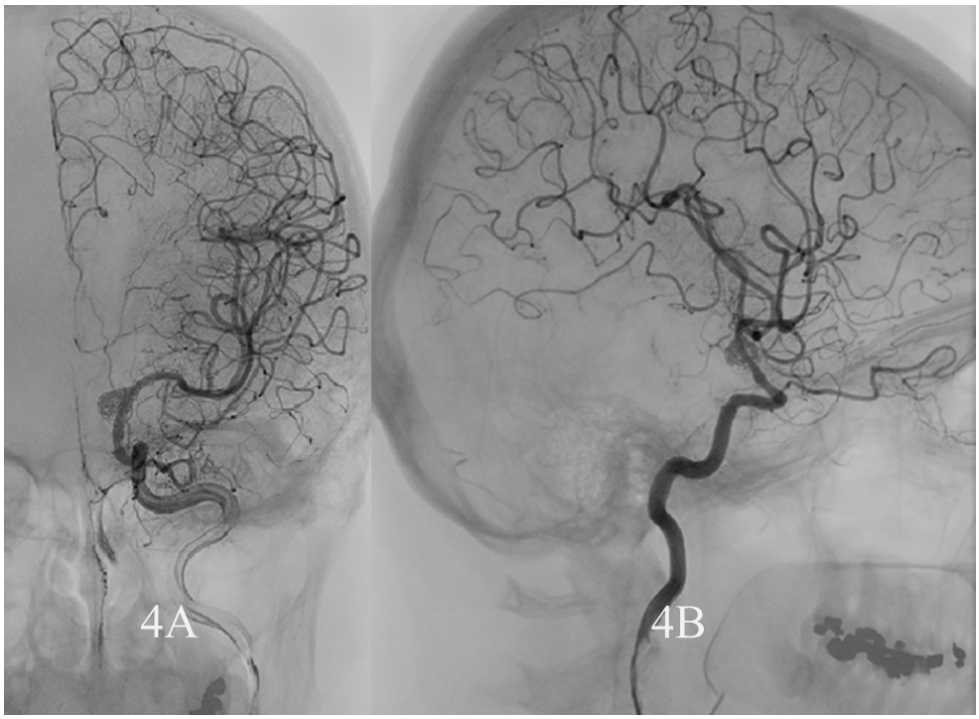

Fig. 4A,B. The digital subtraction angiogram in lateral and anteroposterior view 7 months later proved complete remodelling of the ICA. The patient made a full recovery.

at the level of the A1 segment, and the whole territory was filled via collaterals from the left posterior cerebral artery.

The patient recovered with no neurological deficit. The follow-up MRI showed small, patchy cortical infarcts. A stent-assisted coiling was performed three days later using Neuroform EZ 3.5 x 30 mm (Stryker Neurovascular, Cork, Ireland) with loose coiling in the most prominent part of the fusiform aneurysm (Fig. 3A). The patient was on dual antiplatelet therapy (acetylsalicylic acid 100 mg and clopidogrel 75 mg per day). Due to the persistent filling of the aneurysm sac, a flow diverter stent 3.3x22mm (Fred 3516, MicroVention, Tustin, USA) was deployed across the diseased vessel segment two weeks later. The catheterisation of the previously deployed Neuroform

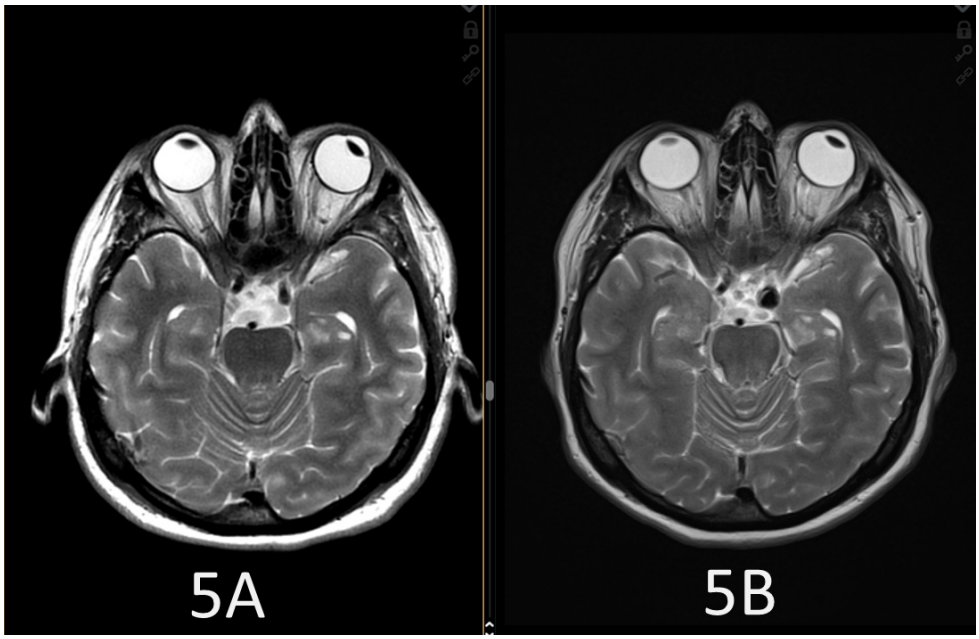

Fig. 5A,B. A Axial T2W MR image demonstrates normal calibre of the internal carotid artery in 3 months after surgery. **B** There is aneurysma at the same level on follow up MR after ischemic stroke in 7 months after surgery.

stent was technically challenging and the introduction of the delivery microcatheter (Headway 27, MicroVention) was complicated by the coils' dislodgment downstream into the M1 MCA segment (Fig. 3B). After the successful deployment of the flow diverter stent, angioplasty of the distal end of the implanted flow diverter stent was required to improve blood flow through M1 MCA using the Scepter C balloon (MicroVention) (Fig. 3C). During the procedure, a bolus of eptifibatide glycoprotein blocker (Integrilin, Glaxo Group Ltd, Middlesex, UK), was administered intravenously and followed by continuous infusion due to the formation of distal embolus in the left parietooccipital branch of the MCA. The patient was extubated with no neurological deficit. She was kept on dual antiplatelet therapy for the next 7 months.

A 7-month follow-up DSA demonstrated complete remodeling of the dissected fusiform aneurysm with no residual filling (Fig. 4A,B). Five years later the patient works full time as a nurse with no regrowth of the craniopharyngioma and no aneurysm reperfusion confirmed on the follow-up MRI in April 2020.

DISCUSSION

We present the case of a patient who experienced development and subsequent acute thrombosis of a large fusiform aneurysm of the communicating ICA segment which clinically manifested as left hemispheric stroke. The patient was treated with mechanical thrombectomy under the protocol for acute ischemic stroke. The most probable etiology of the aneurysm's development was iatrogenic injury of the vessel wall during neurosurgical resection of craniopharyngioma 7 months prior to the ischemic event. Beside mechanical injury, we should consider effect of the ultrasonic aspiration, which is widely used in tumor resection[7]. However, there was not such injury mentioned in the surgery protocol, moreover injury had to be minimal since 3 months after it, there was no ectasia seen on the MR follow up images (Fig. 5A,B).

The location and fusiform morphology of the aneurysm was almost identical to numerous pediatric cases described in the literature. In most cases, the ICA was diffusely enlarged from the distal dural ring of the cavernous sinus to the carotid bifurcation with no evidence of focal vessel wall injury. Some reported cases showed less extended fusiform aneurysms. The dilatation of the vessel segment was probably caused by prior injury during the surgical peeling of the tumor mass from the arterial adventitia of the exposed artery which resulted in adventitial thinning.

Another pathogenetic mechanism leading to aneurysm formation is radiation exposure during locally applied radiotherapy. However, ionising radiation from an yttrium solution via Ommaya reservoir is absorbed in the 1 mm layer of the craniopharyngioma cyst wall, so it does not reach nearby anatomical structures[8].

Participation of the vasa vasorum was also taken into consideration based on histological findings from the resected wall of aneurysms in patients who had undergone microneurosurgical remodeling[1]. Whether an injury of the sympathetic plexus around the ICA, resulting in neurogenic paralysis of the arterial wall, is a possible explanation of aneurysm formation, remains unclear. There is a documented case of a child with full regression of postoperative ICA dilatation after 6 months, which supports this particular mechanism[4].

Fusiform dilatation is broadly reported in the child population. In a series of 31 iatrogenic intracranial aneurysms related to radical craniopharyngioma surgery,

Table 1. Clinical data of adult patients with postsurgical fusiform dilatation of the supraclinoid segment of the ICA.

Age	Gender	Therapy	Diagnosis after surg	Suplement. radiotherapy	Course	Ref.
24	F	vascular surgery	4 months	none	progression in 3 years	5
19	NR	none	17 months	none	7years/alive	13
21	F	none	13 months	none	NR	8
39	F	FD placement	1 year	none	progression in 21years	14

note: F = female, surg = surgery, NR = not reported, FD = flow diverter

fusiform aneurysm of the supraclinoid ICA segment developed in 9 children[2]. Only 3 of those children received additional radiation therapy. These fusiform aneurysms occurred in a period of 4 to 17 months after surgery and were apparently fully developed by 18 months. The eight children experienced no clinical symptoms caused by the aneurysms at the mean follow-up of 3.7 years after diagnosis. In the German Childhood Craniopharyngioma Registry, which included patients from 2001 to 2015, fusiform dilatation of the ICA was observed in 14 cases out of 583, demonstrating that this vascular complication occurred in only 2.4% (ref.[4]). In a series of 62 children who were followed after surgery of suprasellar tumors, 7 children (11.3%) with fusiform dilatation of the ICA were identified. In all observed cases the fusiform aneurysm developed within 15 months following the surgery. Its occurrence was not related to radiotherapy or distinct histology findings. Complete regression was observed in one child, the aneurysm remained stable in 3 children and aneurysm progression occurred in the remaining 3 children. However, none of them were treated[4].

In another series[8] of 8 patients with iatrogenic intracranial aneurysms, 3 patients with fusiform aneurysms were reported. One patient was diagnosed with an aneurysm 6 months after surgery for hypothalamic gangliocytoma, a second was operated for hamartoma of tuber cinereum and diagnosed 6 months later, and in the last case, the aneurysm was observed 13 months after craniopharyngioma resection. None of these three patients were treated and their fusiform aneurysms remained unchanged and asymptomatic. There are several other cases described in case report publications[9-12].

The vast majority of craniopharyngiomas are treated in childhood, therefore, the occurrence of this arterial complication is quite rare in adults. We identified only 4 other adult patients in the available literature (Table 1). Three patients were women, the gender was not reported in one case, and their age ranged from 19 to 39 years. All fusiform aneurysms were observed within 4 to 17 months after surgery for craniopharyngioma[5,8,13,14,15]. The presented case was a 45 year old woman with an aneurysm which presented clinically with acute stroke 7 months after surgery.

As previously described[16], intracranial artery dissection can present with aneurysmal dilatation, segmental stenosis, or occlusion, with a widely varying distribution of these radiological subtypes. Our patient presented with acute thrombosis of the fusiform aneurysm and stenosis of the M1, which are both typical signs of arterial dissection. Underlying occlusion and thus outflow obstruction of A1 ACA could play a significant role in acute thrombosis of the ICA. The occlusion of the A1 ACA was most likely also due to surgery, since on follow-up MRI there was infarction in the territory of the medial lenticulostriate arteries. Relatively mild neurological symptoms with very slow or even no progression in time, despite the "T" occlusion which usually presents with fast progression of the brain ischemia[16], as well as large penumbra-core mismatch on CT perfusion, could be explained by a good collateral network due to pre-existing M1 stenosis and A1 occlusion.

Mechanical thrombectomy was indicated based on persisting, mild but disabling neurological deficit and suitable baseline imaging findings (absent early ischemic changes on NCCT and no ischemic core on perfusion CT). The interventional neuroradiologist was aware of the ectatic segment of the ICA. The artery was recanalized after 3 passes with a stent retriever, including a majority of the aneurysm cavity. There was a risk of aneurysm rupture and hemorrhage following the mechanical thrombectomy, as reported previously[17-20]. Approaching the thrombus in an optimal working projection is crucial for a proper navigation of the microcatheter during catheterization of the middle cerebral artery. In general, mechanical thrombectomy performed by an experienced operator has a low rate of procedure related complications. The prevalence of incidental brain aneurysms is approximately 3% in the overall population[18], which is in concordance with an incidence rate of 2.9% in a population of 240 endovascularly treated patients with large vessel occlusion[19]. In that study, no periprocedural aneurysm rupture was observed. Based on the available data, there is a general recommendation to opt for contact aspiration thrombectomy over stent retriever thrombectomy in the presence of suspected intracranial aneurysm[17,18].

To prevent acute aneurysm rupture, we aimed to place a regular intracranial stent with additional coiling in the most prominent part of the aneurysm 3 days after thrombectomy. Since there was no tendency of the aneurysm sac to thrombose, we decided to implant a flow diverter stent across the diseased vessel segment. In spite of technical problems during flow diverter stent delivery and stenosis of the distal part of the flow diverter stent, which required additional angioplasty, the artery was completely remodeled on the follow up angiogram 7 months later and blood flow in the MCA territory was preserved.

In general, the therapeutic approach is dependent on local possibilities. There are reported childhood cases of fusiform dilatation of the ICA treated surgically by wrapping and clipping[1-3,5,13], or by extra-intracranial bypass[11]. In

the endovascular approach, either stent assisted coiling[9] or flow diverter stent placement[12,14] was performed.

The endovascular therapy became a dominant treatment of intracranial aneurysms in the past decade[13,15]. A recent meta-analysis by reported complete occlusion in 48/60 (80%) fusiform/dissecting distally located aneurysms treated with flow diverter stents, confirming the safety and efficacy of this endovascular treatment[21].

CONCLUSION

The presented case demonstrates that fusiform dilatation of the ICA as a rare complication of the ipsilateral resection of the craniopharyngioma can occur in adult age. Acute thrombosis of fusiform aneurysm resulted in left hemisphere neurological deficit. Acute thrombosis and stenosis of the M1 MCA segment are typical signs of arterial dissection, which is the most likely etiology of the fusiform ICA dilatation in this case. This case also demonstrates that implantation of the flow diverter stent is a minimally invasive therapy with durable effect in symptomatic patients, or as reported in the literature, in those who are asymptomatic but show aneurysm progression during follow up.

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