CASE REPORT

4D flow preliminary investigation of a direct carotid cavernous fistula due to a ruptured intracavernous aneurysm

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SUMMARY

Inadequate information is available about the cerebral blood flow and surgical strategies of a direct aneurysmal carotid cavernous fistula (daCCF). We report a quantitative analysis of flow velocity and volume using preoperative time-resolved phase-contrast MRI (four-dimensional (4D) flow MRI) in a daCCF. This is the first report of 4D flow findings with a daCCF. A 55-year-old woman developed a sudden headache and bruit of the right orbit, and MRI suggested the presence of a daCCF. Quantitative analysis using preoperative 4D flow MRI revealed the flow volume of the right internal carotid artery. The daCCF was successfully treated by high-flow bypass using a radial artery graft and internal carotid artery trapping. Postoperative angiography showed a complete obliteration of the daCCF. Studies to collect data from additional cases are required so that 4D flow findings can be further used in the management of daCCFs.

BACKGROUND

Intracavernous aneurysms are a cause of direct carotid cavernous fistula (CCF) and have an incidence rate of 2.3% of direct and indirect CCFs.¹ Lewis *et al*² reported that direct CCF caused by a ruptured cavernous sinus aneurysm (direct aneurysmal CCF, daCCF) accounted for 18% of direct CCFs. Therefore, only a few large series of daCCFs have been reported.

Most CCF cases are treated by intravascular surgery.³ Intravascular surgery with parent artery sacrifice is not infrequent in daCCFs in cases of a large fistula or high-flow shunt.⁴ However, no

studies have considered arteriovenous shunt volume because no method has been available to confirm flow volume or flow velocity in daCCF. In the present study, preoperative, time-resolved four-dimensional (4D) phase-contrast^{5 6} MRI (4D flow MRI) was used to quantify flow in a daCCF. In this case, a quantitative analysis of flow velocity, volume and streamlines using preoperative and post-operative time-resolved phase-contrast 4D flow MRI was also performed. This is the first report concerning 4D flow MRI findings of CCF or radial artery graft (RAG) for complex carotid disease. Therapeutic processes including blood flow reconstruction are also discussed.

CASE PRESENTATION

A 55-year-old woman with no history of head injury developed a sudden headache and continuous bruit at the circumference of her right orbit. MR angiography (MRA) revealed a right CCF. The patient was admitted to another hospital for endovascular treatment. Cerebral angiography ([figure 1](#)) confirmed the diagnosis of daCCF caused by a ruptured right cavernous aneurysm and internal carotid artery (ICA)-anterior choroidal artery unruptured aneurysm. CCF cortical reflux through a superficial sylvian vein was performed as Barrow type A ([figure 2](#)). The right ophthalmic artery was not detected using angiography because of the shunt flow around the cavernous sinus. Although endovascular treatment is normally preferred for

Figure 1 Three-dimensional digital subtraction angiography showing the aneurysm causing the direct aneurysmal carotid cavernous fistula. The red arrow indicates the aneurysm.

Figure 2 Right internal carotid angiogram showing a carotid cavernous sinus fistula and cortical reflux.

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Table 1 Results of a quantitative analysis using perioperative 4D flow MRI

	Flow volume (mL/min)	Endodiastolic Flow velocity (cm/s)	Systolic Flow velocity (cm/s)
Preoperative right ICA	798	10.4	132
Preoperative left ICA	294	16.3	45.4
Postoperative right RA	166.8	13.2	33.2
Postoperative left ICA	288	54.4	25.3

4D, four-dimensional; ICA, internal carotid artery; RA, radial artery.

CCF, in this case, high-flow, cortical reflux and an ipsilateral intracranial aneurysm were all present. The authors discussed and concluded that for this 55-year-old woman, the daCCF required immediate treatment because three points would be required in the near future. However, we agreed it would be difficult to treat an ICA-anterior choroidal artery aneurysm and daCCF with endovascular treatment in one session. Moreover, the reported rate of ICA occlusion with endovascular treatment is 17–47%. Sacrificing the ICA via interventional radiology would make surgical clipping of the cerebral aneurysm difficult. In this case, the radiological findings suggested that performing RAG with trapping of the ICA and clipping of the ICA aneurysm simultaneously would be more reliable.

After the surgery, the patient's symptoms improved, and there was no new ischaemia on diffusion-weighted MRI. There were no differences in the right and left cerebral blood flow mean transit times, as measured by perfusion CT. The results of postoperative 4D flow MRI are shown in [table 1](#) and indicated a decrease in the right-side and left-side differences in flow volume and flow velocity. Streamline images indicated a normalisation of the flow direction of the right A1 segment of the anterior cerebral artery ([figure 3](#)). Furthermore, blood flow from the RAG into the ICA terminal segment was confirmed. The patient was discharged without any neurological deficits.

INVESTIGATIONS

The methodology of 4D flow MRI^{5–7} is presented in [box 1](#). The prominent shunt flow from the cavernous ICA was confirmed by preoperative 4D flow MRI. The results of quantitative analysis using perioperative 4D flow MRI are shown in [table 1](#). The preoperative 4D flow MRI showed a marked increase in the flow volume and systolic flow velocity of the right ICA.

The magnitude data and the flow velocity data were encoded on the x-axis and y-axis, respectively, and Z shaft orientations were obtained using 3D cine phase contrast, which is a phase-contrast MRI with ECG synchronisation.

Evaluation of the MRI data was performed using 'GT Flow'.

Regions of interest were defined manually through the respective vessels for measurements of mean flow volume (mL/min) and maximum systolic, mean and minimum end diastolic flow velocity (cm/s).

The streamline data express the flow velocity by the coloured line and the curvilinear rate vector.

TREATMENT

The operation was performed; the Dolenc approach was used to remove the anterior clinoid process and the ICA was exposed to the proximal ophthalmic artery bifurcation. Then, we clipped the ICA-anterior choroidal artery aneurysm. RA anastomosis was performed at the M2 segment of the MCA and ECA in order. The patency of an chor A and RAG was checked using indocyanine green videoangiography.

OUTCOME AND FOLLOW-UP

After the surgery, the patient's symptoms improved, and there was no new ischaemia on diffusion-weighted MRI. There were no differences in the right and left cerebral blood flow mean transit times, as measured by perfusion CT. The results of the postoperative 4D flow MRI are shown in [table 1](#). This showed a decrease in right-side and left-side differences in flow volume and flow velocity. Streamline images indicated a normalisation

Figure 3 Four-dimensional flow MRI showing the radial artery retrograde flow supplying the M1 segment and covering a wide range. The shunt flow has disappeared. The orange arrow indicates the radial artery graft on the right side.

Box 1 The methodology of four-dimensional flow MRI

Philips Medical system; 3.0 T-Achieva
 TR: 8.7 ms
 TE: 5.4 ms
 FA: 13°
 VENC: 70 cm/s
 Voxel size: 1.19×1.36×1.4 mm

of the flow direction of the right A1 segment of the anterior cerebral artery (figure 3). Furthermore, blood flow from the RAG into the ICA terminal segment was confirmed. The patient was discharged without any neurological deficits.

DISCUSSION

We encountered a case of daCCF that was successfully treated by high-flow bypass, ICA trapping and clipping of an aneurysm at the same time. We used 4D flow MRI to assess an arteriovenous shunt. This is the first report of 4D flow findings with CCF. The high-flow shunt of CCF and retrograde flow of the ophthalmic artery were confirmed on preoperative 4D flow MRI. The blood flow volume of the RAG, which had adequate flow comparable to that of the contralateral ICA, was confirmed on postoperative 4D flow MRI. In this study, the 4D flow findings included abnormal increased systolic flow velocity and markedly increased flow volume in the carotid artery. Sufficient blood flow volume from a RAG to the proximal M1 segment of the MCA was also shown by postoperative 4D flow images. This case provides an important example of preoperative 4D flow imaging of CCF and could have a potential benefit for assisting in preoperative treatment strategy planning. The collection of data from a greater number of similar cases is required to clarify the benefits.

Intravascular treatment with parent artery preservation has been used to treat many CCFs. Gupta *et al*^{8,9} reported treatment results from intravascular surgeries for 91 direct CCFs (85 traumatic and 6 aneurysmal); their rate of complications was 3.4%. Out of 91 cases, only 89% had a complete filling of the fistula. However, in the daCCFs, the difficulties inherent in the complete occlusion of the fistula with parent artery preservation in endovascular treatment have been reported, and some reports even state that parent artery occlusion is necessary.^{4,10} Kobayashi *et al* reported the treatment results of 23 cases for direct CCF and experienced a rate of 83% ICA preservation

with interventional treatment for daCCF. Of course, some reports exist on CCFs treated by parent artery occlusion and vascular reconstructive surgery of the ICA (table 2).^{10–13} In the present case, high-flow, symptomatic daCCF and the ipsilateral intracranial aneurysm were all present. After discussion, the authors concluded that treating an intracranial aneurysm and daCCF with endovascular treatment in a single session would be extremely difficult. Therefore, the authors elected to perform reconstructive surgery with parent artery occlusion and clipping in the single session.

There are many treatment strategies for complex carotid disease, including ICA intentional occlusion, with or without high-flow bypass, as based on the results of balloon test occlusion (BTO) (so-called selective bypass^{12,14}), and ICA intentional occlusion with high-flow bypass in all cases without BTO (so-called universal bypass). Some case reports have described non-traumatic direct CCF treated with vascular reconstructive surgery.^{2,15,16} For complex carotid disease, universal bypass strategies are safe to prevent ischaemic complications.¹⁷

Learning points

- Four-dimensional flow MRI is useful when assessing an arteriovenous shunt.
- For complex carotid disease, universal bypass strategies are safe to prevent ischaemic complications.
- There are some cases of direct aneurysmal CCF that are difficult to treat using endovascular techniques.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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Table 2 Reported cases^{10–13} of CCFs treated with reconstructive surgery

Case	Author	Year	Age	Sex	Graft	Cause
1	Eguchi	1982	37	F	SV, STA	Spontaneous
2	Ogawa	1986	72	F	SV	Spontaneous
3	Sekhar	2001			RA	Aneurysm
4	Inoue	2005	48	M	STA	Aneurysm

F, female; M, male; RA, radial artery; STA, superficial temporal artery; SV, saphenous vein.

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