Rare disease

Indirect carotid-cavernous fistula following minor head trauma treated with incomplete radiographic endovascular occlusion

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Summary

We report the unusual case of a patient with an indirect carotid-cavernous fistula treated with only partial angiographic embolisation of the fistula; however, by 8 weeks postembolisation, she was found to have both symptomatic and angiographic cure. This case highlights the potential for treating indirect carotid-cavernous fistulas with partial embolisation as a means of achieving angiographic and symptomatic cure by altering flow dynamics to attain spontaneous thrombosis.

BACKGROUND

Carotid-cavernous fistulas (CCFs) are an abnormal communication between the cavernous carotid artery and the cavernous sinus venous system. Fistulas may be spontaneous or traumatic, high-flow or low-flow and direct or indirect with respect to arterial feeding vasculature.¹ Trauma is a common cause of direct high-flow CCFs, resulting from a traumatic tear in the cavernous portion of the internal cerebral artery (ICA), hypothesised to result from shear injury to the ICA at its dural attachment to the cavernous sinus.^{2 3} Indirect fistulas occur when dural branches of the internal/external carotid system drain indirectly into the cavernous system and are more often spontaneous in nature.¹ We report the unusual case of a patient who presented with an indirect fistula secondary to trauma. Presentation occurred in a delayed manner and was successfully treated with only partial embolisation of the fistula.

CASE PRESENTATION

A 58-year-old woman was involved in a motor vehicle collision with minor closed head injury 3 weeks prior to presentation. Upon presentation, she was noted to have proptosis, periorbital oedema, chemosis, restriction of horizontal gaze and moderately elevated intraocular pressures (22–30 mm Hg) of the left eye. MRI of the brain showed dilatation of the intraorbital venous structures including the left ophthalmic vein, and diagnostic cerebral angiography confirmed CCF between the frontoparietal branch of the left middle meningeal artery and left deep temporal artery with drainage into the ipsilateral sphenoparietal sinus. There was drainage from the sphenoparietal sinus into the cavernous sinus with subsequent retrograde flow into the left superior ophthalmic vein causing dilatation of these venous structures without evidence of cortical venous reflux (figure 1).

TREATMENT

The frontal branch of the middle meningeal artery and the deep temporal branch were embolised successfully

using the Onyx Liquid Embolic system (ev3, Irvine, California, USA). Despite seemingly good arterial pedicle embolisation, the fistula persisted via small frontal branches of the middle meningeal artery, although with a slower filling rate following embolisation (figure 2). Given the low-flow state of the fistula, further embolisation was abandoned and the decision was made to follow the patient clinically.

OUTCOME AND FOLLOW-UP

One day after embolisation, the patient was noted to have subjective and objective improvement of proptosis, chemosis and periorbital swelling. Approximately 1 week after embolisation, the patient had lower intraocular pressures (22 mm Hg) and by 6 weeks after embolisation repeat angiography showed complete occlusion of the dural arteriovenous fistula (figure 3). At her 2-month

Figure 1 Photograph taken at the time of initial presentation.

Figure 2 Early (left) and late (right) arterial phases of initial cerebral angiogram showing indirect carotid-cavernous fistula.

Figure 3 Cerebral angiogram after partial Onyx embolisation demonstrating fistulous connection in low-flow state.

Figure 4 Cerebral angiogram at 8-week follow-up visit showing complete obliteration of carotid-cavernous fistula.

Figure 5 Photograph taken at 8-week clinical follow-up visit.

clinical follow-up visit, the patient was asymptomatic and neurologically intact with normal intraocular pressures (figures 4 and 5).

DISCUSSION

CCFs can be classified based on their pathogenicity, rate of flow and the direct or indirect vascular course of the fistula. Barrow *et al*¹ classified fistulas into four types based on anatomical features of the feeding pedicles. Type A fistulas are direct fistulas between the cavernous portion of the ICA and the cavernous sinus. These fistulas are most commonly caused by trauma¹ from a tear in the ICA via shearing forces or laceration from adjacent bone fractures.^{3–4} Types B, C and D fistulas are most commonly spontaneous in nature and have been associated with hypertension, pregnancy and atherosclerotic arterial disease.¹

In this report, we describe an unusual case of delayed presentation of an indirect CCF that presented secondary to minor trauma. While rare, trauma has been implicated in indirect CCF formation previously.^{5–9} Traumatic indirect CCFs (TICCFs) differ from spontaneous indirect CCFs (SICCFs) in that they often involve a higher flow state, are larger and often present more acutely than SICCFs. Subsequently, these TICCFs cause more pronounced symptomatology than SICCFs⁵; however, the time point for presentation of symptoms can be highly variable.¹⁰

Indirect CCFs often resolve spontaneously and do not require treatment. Indications for intervention include visual decline, diplopia, intractable headache and severe proptosis.¹ In our case, we chose to treat the patient secondary to her ophthalmoparesis and elevated intraocular pressures. Sakata *et al*¹¹ studied the dynamic changes of a traumatic middle meningeal arteriovenous fistula over the course of 15 years and found a significant dilatation of draining venous structures over this time period that eventually lead to a devastating haemorrhage. Given that the flow characteristics of TICCFs differ from SICCFs, we advocate treatment of these fistulas as they

usually present with either severe ocular symptoms or cortical reflux.

Usually complete occlusion of the fistula by angiography is necessary to relieve the presenting symptoms. However, with newer embolic agents such as Onyx, complete occlusion of the fistula may be possible without complete angiographic embolisation of all arterial feeders, as evidenced in our case. This may be due to the more complete occlusion of arterial feeders embolised with Onyx and better penetration into the venous outflow, thereby altering flow dynamics of the fistula. In our patient, complete obliteration of her fistula was not achieved initially; however, at her 6-week angiographic follow-up visit, it was evident that spontaneous thrombosis of the remainder of the fistula had occurred.

In summary, it seems that TICCF has a different pathophysiology from SICCF, and often requires treatment secondary to higher flow rates and the tendency to enlarge and produce more symptoms over time. While we advocate complete occlusion at the time of intervention, if complete angiographic occlusion of the TICCF is not achieved but the flow of the fistula is slowed down, it is possible for some of these fistulas to spontaneously thrombose.

Learning points

- Indirect carotid-cavernous fistulas are connections between the cavernous sinus and meningeal branches of the external carotid, internal carotid or a combination of both. Indirect carotid-cavernous fistulas most commonly occur spontaneously.
- Indirect carotid-cavernous fistulas often present with subtle symptoms rather than the dramatic symptoms typically associated with carotid-cavernous fistulas including pain, pulsatile proptosis and chemosis.
- Low-flow indirect carotid-cavernous fistulas can resolve spontaneously without treatment. When these fistulas do require treatment, complete occlusion is the goal. However, partial embolisation can result in eventual spontaneous resolution of the fistula.

Competing interest None.

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