CASE REPORT

Reversed ocular ischaemic syndrome secondary to carotid cavernous sinus embolisation

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SUMMARY

Ocular ischaemic syndrome is a progressive and serious vision-threatening condition that is usually associated with carotid artery disease, and poor effective therapeutic options are available. In the present case, a 65-year-old woman was submitted to embolisation of spontaneous carotid-cavernous fistula with complete resolution of initial ophthalmoplaegia and ocular hypertension. Later, ocular ischaemia was detected. Cerebral angiography showed delayed choroidal flush and the patient was admitted for hypervolaemic hypertension therapy and medicated with nimodipine. The ischaemia was reversed and visual function improved.

BACKGROUND

Ocular ischaemic syndrome (OIS) is caused by a reduction of arterial blood flow to the eye and is usually associated with carotid artery disease; it affects individuals in their late 60s and rarely occurs in those under 50 years of age. The incidence is estimated at only seven cases per million, as this condition usually only develops in cases with poor collateral circulation between internal carotid artery (ICA) and external carotid artery or ICA-ICA. In fact, ocular vascularisation is highly susceptible to circulation imbalances between ICA and collateral circulation. In this case, the patient developed an ocular ischaemia 3 weeks after embolisation of the spontaneous carotid-cavernous fistula (SCCF), possibly due to short posterior ciliary artery insufficiency. The patient was medicated with nimodipine via intravenous infusion, and was submitted to hypervolaemic hypertension therapy. The ischaemia was fully reversed, visual acuity (VA) improved, intraocular pressure (IOP) was raised back to normal, the retinal haemorrhages were reabsorbed and the anterior chamber assumed its anterior architecture. This is an unusual case as the acute ischaemia responded to medical treatment, and therefore was probably related to short posterior ciliary artery spasm.

CASE PRESENTATION

A 65-year-old woman with an otherwise unremarkable medical history and blood pressure values around 120/60 mm Hg was submitted to left SCCF embolisation, at the time associated with right cavernous sinus thrombosis and ipsilateral full ophthalmoplaegia, involving III without ptosis nor pupil, IV and VI cranial nerves, and raised IOP and dilated episcleral vessels. IOP lowered to 20 mm Hg the day after the procedure, and in the next 2 weeks the ophthalmoplaegia reversed completely.

In the third week after the procedure, the patient's vision decreased in the right eye (RE).

Examination revealed a best corrected VA (BCVA) 1/10 (Snellen) in the RE and 10/10 in the left eye (LE); Ishihara plates tested 17/17 in both eyes, in spite of lower reading speed and red desaturation on the RE, anisocoria (RE>LE, the pupil was fixed on medium mydriasis presumably due to iris ischaemia and, furthermore, did not constrict under near reflex, which it would have done if III cranial nerve pupillary fibres were involved and no relative afferent papillary deficit was present) and no oculomotor palsies. On biomicroscopy, the RE showed conjunctival and episcleral dilated vessels, corneal oedema, Descemet folds, flair 1+, and low anterior chamber depth with iridotrabecular contact 360°, no rubeosis iridis, anterior capsule opalescence and anterior subcapsular cataract (figure 1). The fundus observation revealed deep, superficial and round retinal haemorrhages, especially in the posterior pole (figure 2), with no significant optic disc alterations; the IOP was 16 mm Hg on both eyes.

New cerebral angiography (CA) revealed delayed choroidal flush in spite of the normal ophthalmic artery (OA) perfusion that was already present on first examination of the RE.

The patient was treated with reinforced hydration and intravenous nimodipine. Control CA showed improvement of the choroidal flush and the patient was discharged maintaining hydric reinforcement and oral nimodipine 30 mg/day.

Blood pressure during the treatment at the hospital was around 120/60 mm Hg.

DIFFERENTIAL DIAGNOSIS

- Retinal veins show tortuosity, most likely caused by compromised venous drainage associated with right cavernous sinus thrombosis.

Figure 1 Retinal haemorrhages in the posterior pole.

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Figure 2 Delayed choroidal flush despite normal ophthalmic artery perfusion.

- Fluorescein angiography as well as *spectral domain* optical coherence tomography were performed, which showed the presence of deep, superficial and round retinal haemorrhages in the posterior pole plus delayed choroidal and retinal perfusion followed by staining of the retinal arteries as expected in acute ischaemic ocular syndrome.
- At this point, the differential diagnosis included diabetic retinopathy and central retinal vein occlusion; nevertheless, the patient was not diabetic and the retinal haemorrhages were round rather than flame-shaped.
- CA showed delayed choroidal flush, which strongly supported ocular ischaemic syndrome.
- Carotid Doppler study was not performed in this particular patient because the OIS diagnosis was suspected after SCCF embolisation, and useful information was present in CA.

TREATMENT

At the time CA was performed, normal OA perfusion but delayed choroidal perfusion was identified. The neuro-ophthalmologist, neuroradiologist and neurosurgeon decided to admit the patient to reinforce hydration and medicate with nimodipine. The patient was observed daily and the ocular ischaemia started to reverse in 1 week. The following week, CA showed choroidal perfusion improvement and normal ocular blood perfusion.

No ocular treatment was prescribed.

OUTCOME AND FOLLOW-UP

The patient was monitored closely and at 7-month follow-up, there was a significant improvement in the chief visual complaints.

BCVA was 10/10 RE and 10/10 LE, Ishihara plates were 17/17 bilaterally, right IOP 14 mm Hg, left IOP 18 mm Hg, with no iridotrabecular contact 360°, no rubeosis iridis, appropriate anterior chamber depth and complete resorption of the retinal haemorrhages (figure 3). The iris ischaemia reversed, and RE direct pupillary reflex was present and identical to the LE.

As after effects on the RE, an anterior subcapsular cataract was evident.

Currently, the patient is being medicated with nimodipine.

Figure 3 Complete reabsorption of retinal haemorrhages.

DISCUSSION

OIS is an uncommon condition, and usually secondary to atherosclerotic carotid stenosis of more than 90%,¹ nevertheless, this was not the case in our patient.

OIS on the RE was diagnosed in the third week after left transvenous SCCF embolisation with platinum coils, and CA showed delayed right choroidal flush that justified OIS on the RE.

In spite of a well documented delayed choroidal perfusion, no anterior ischaemic optic neuropathy was detected, probably due to vascularisation from pial and central retinal arterial branches, justifying the absence of a relative afferent pupillary deficit.

Several complications of CCF embolisation have been reported,^{2 3} such as aggravation of symptoms, including increased proptosis, elevation of IOP, choroidal detachment, venous stasis retinopathy and central retinal vein obstruction. Ophthalmic artery obstruction and cerebral infarction have also also described.

In our patient, the ischaemia was presumably due to posterior ciliary artery insufficiency, probably caused by an arterial spasm. CCF embolisation was transvenous and, therefore, did not support the hypothesis of arterial spasm. Nevertheless, the ischaemia was progressing and reversed after reinforced hydration and nimodipine, which is also a drug of choice for cerebral artery vasospasm. The goal was to maintain arterial perfusion and counter arterial spasm as well as to employ the blood pressure-lowering effect of nimodipine.

Furthermore, on the last CA, the choroidal flush occurred sooner than on the first examination, thus we believe delayed choroidal perfusion was actually caused by generalised spasm of posterior ciliary arteries. After arterial circulation was improved, both in short and long ciliary arteries, the retinal and anterior segment ischaemia reversed, except for an anterior subcapsular cataract, possibly caused by metabolic insufficiency in the acute stage.

A possible explanation for ischaemia onset occurring after SCCF embolisation and not before, is that choroidal perfusion was already compromised as shown on CA due to right cavernous sinus thrombosis, which caused arterial perfusion on the RE. When SCCF embolisation was performed, the collateral arterial circulation was further disturbed and OIS developed.

During the course of the disease, IOP was always above 10 mm Hg, in spite of ciliary shutdown, but iridotrabecular contact caused by reduction of the anterior chamber depth prevented fluid from flowing out of the eye, thus maintaining IOP.

Learning points

- ▶ Ocular ischaemic syndrome (OIS) can present after successful carotid-cavernous fistula embolisation.
- ▶ Close monitoring by an ophthalmologist is necessary to prevent complications from occurring.
- ▶ OIS can be reversed entirely if diagnosed soon enough and treated with hypervolaemic hypertension therapy and nimodipine.

To our knowledge, this is the first successfully treated case of presumed posterior ciliary artery insufficiency after CCF embolisation.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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