CASE REPORT

Bilateral atherosclerotic internal carotid artery occlusion and recurrent ischaemic stroke

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SUMMARY

Bilateral internal carotid artery occlusion (BICAO) is a rare disease that carries a gloomy prognosis. We report a case of a 52-year-old man who developed ischaemic infarction at the region of the right middle cerebral artery; he was found to have atherosclerotic occlusion of both internal carotid arteries on Doppler-duplex examination. He received medical treatment only. After 1 year, he developed a new infarction at the region of the left middle cerebral artery. Conventional angiography revealed bilateral occlusion of internal carotid arteries at their origin, approximately 50% stenosis of the common carotid bulbs and mild stenosis of the origin of external carotid arteries. The patient did not undergo any form of surgical revascularisation procedures and died of severe aspiration pneumonia approximately 2 months after the second stroke. BICAO portends a poor outcome and carries a risk of recurrent ischaemic events. The best management strategy for this vascular occlusion remains unclear.

BACKGROUND

Occlusion of both internal carotid arteries is an extremely rare entity and carries a grave prognosis. The pertinent medical literature on the best medical treatment and long-term outcome of patients with such occlusions is limited by the small numbers of patients studied and the short duration of follow-up in the majority of these cases.^{1–8}

CASE PRESENTATION

A 52-year-old right-handed man was brought to our emergency department because of sudden onset of right-sided hemiparesis and speech disturbance. The patient developed ischaemic stroke 1 year earlier that had resulted in dense spastic left hemiparesis and hemianaesthesia. He was a smoker, hypertensive and hyperlipidaemic. His medications were aspirin, ramipril, amlodipine and atorvastatin. The patient lived in a rural area near the Iraq–Iran border. The patient's records mention that he was hospitalised for 2 weeks after developing his first stroke; when he was discharged, he demonstrated left-sided grade 2 pyramidal weakness, hemianaesthesia and hemi-neglect. He had no dysphasia or dysphagia. During the course of the following months, his left-sided weakness was more or less the same; he was bed-ridden and was entirely dependent on his wife, who also performed physiotherapy for him. The blood pressure on admission was 180/105 mm Hg with a regular pulse rate of 87 bpm. He demonstrated global aphasia, and new right-sided weakness of grade 2. The patient had

Figure 1 Non-contrast brain CT scan of the patient at the level of the basal ganglia and thalami, which was performed at the time of admission. There is a new infarction at the left middle cerebral artery territory. An old and large infarction can be seen at the area of the right middle cerebral artery.

dysphagia and a nasogastric tube was placed. An urgent non-contrast brain CT scan of the patient revealed a new infarction at the region of the left middle cerebral artery (figure 1).

INVESTIGATIONS

Routine blood tests were within their normal reference range, apart from serum total cholesterol of 380 mg/dL and serum triglyceride of 294 mg/dL. A 12-lead ECG was unremarkable. Transthoracic echocardiography revealed mild diastolic dysfunction with an ejection fraction of 61%. Carotid Doppler-duplex ultrasound examination revealed bilateral high-grade common carotid artery stenosis with complete occlusion of both internal carotid arteries' origin. A review of the patient's past carotid Doppler, which was performed 11 months prior, revealed the same findings. The patient sustained a blast injury during the Iraq–Iran war; there were three shells in his body. This had precluded the use of MR studies.

TREATMENT

Clopidogrel replaced aspirin as the antiplatelet agent. We increased the daily doses of his previous medications.

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OUTCOME AND FOLLOW-UP

The patient was hospitalised for 10 days. On the date of discharge, he demonstrated global aphasia and spastic quadriplegia. He was dependent on a nasogastric tube for feeding and medication ingestion. An indwelling Foley's urinary catheter was used. The patient's family decided to treat him in Iran and cerebral angiography was carried out there, 1 month after discharge (figure 2 and video 1). We have no transcranial Doppler or single-photon emission CT/positron emission tomography scan facilities to assess cerebral blood flow at our hospital in Iraq. The decision was to keep him on the same medical treatment. The patient developed severe aspiration pneumonia 3 weeks later and died.

DISCUSSION

Bilateral internal carotid artery occlusion (BICAO) is an extremely rare entity and carries a grave prognosis.¹ According to Mead *et al*² only eight (0.4%) of 2228 transient ischaemic attacks/patients with completed stroke had BICAO. Persoon *et al*³ found that the annual stroke rates in patients with BICAO range between 0% and 13%. The collateral circulation in BICAO comes from the vertebrobasilar system with cross-filling of the middle cerebral artery through the circle of Willis, an external carotid/ophthalmic anastomosis, or a combination of the two.⁴

The pertinent literature on the best management and long-term outcome of patients with BICAO is limited by the small numbers of patients studied and the short duration of follow-up in the majority of these studies.^{5–8}

Wade *et al*⁸ analysed 74 patients with atherosclerotic occlusion of both internal carotid arteries. They found that 18 (53%) patients developed further strokes with an ischaemic event rate of 15% per patient per year. AbuRahma and Copeland¹ analysed and followed up 21 patients with BICAO for 1–11 years (an average of 6 years). Thirteen patients underwent some sort of vascular surgical intervention. The overall mortality rate was 52%; in the surgical group it was 38%; and 75% in the medical group. In the medical group, 75% of patients developed

Video 1 Conventional cervical/cerebral angiography of the patient. Note that both common carotid arteries are stenosed by approximately 50%, both origins of the external carotid arteries are mildly stenosed and both internal carotid arteries are completely blocked. The vertebrobasilar system is intact and provides cross-filling of the middle cerebral artery through the circle of Willis. At the end of the video, the contrast dye passes through the left vertebral artery (blue arrow), basilar artery and posterior cerebral arteries (red arrows). After 2 s, the arterial territories of both middle cerebral arteries (inside the red circles) and anterior cerebral arteries (inside the yellow circle) become visible; this means that these areas have been supplied through the circle of Willis, from the posterior circulation. When the venous phase appears after 3 s, the dye seems to pass very smoothly and rapidly into the sagittal and transverse/sigmoid venous sinuses (orange arrows) and thereafter into the internal jugular veins (green arrow). Accordingly, the anterior circulation in this patient is dependent on the intact posterior circulation in order to bypass the occluded internal carotid arteries (VA, vertebral artery; PCA, posterior cerebral artery).

Figure 2 Conventional cervical/cerebral angiography of the patient, which was carried out approximately 6 weeks after the second ischaemic stroke. Both internal carotid arteries are completely occluded (red arrows) and there is no distal flow of the dye.

multiple ischaemic strokes while 15% of patients in the surgical group developed only one ischaemic stroke.

Catala *et al* reported on 19 patients with BICAO. Nine had brain CT scan evidence of completed stroke; all of these infarcts were located at the middle cerebral territory or at the junctions between two major cerebral arteries. In addition, they concluded that patients suffering athermanous BICAO may survive with minimal neurological deficits.⁹

However, Patel *et al* reviewed 30 812 carotid duplex examinations that were carried out over a period of 12 years and found 45 patients with BICAO. They concluded that once patients with BICAO develop ischaemic stroke (or myocardial infarction), their survival figures decline considerably.¹⁰

Accordingly, there is a wide variation in severity of the clinical presentation of carotid artery stenosis/occlusion, ranging from entirely asymptomatic cases to those resulting in fatal ischaemic stroke; cerebral collateral circulation has been recognised as an important aspect of cerebral circulation affecting the risk of ischaemic stroke itself, as well as other manifestations of stroke presentation, such as stroke patterns in patients with carotid artery disease.¹¹

Henderson *et al*¹² concluded that patients with collaterals supplying the hemisphere distal to a severe internal stenosis have a lower risk of stroke and TIA, both disabling and non-disabling, than patients without collaterals. Compared with flow

territory maps in control subjects, the flow territory maps in patients with bilateral ICA occlusion demonstrate that the vertebrobasilar arteries contribute significantly more to MCA and ACA flow on both sides. This finding is consistent with the results of previous studies of patients with symptomatic bilateral ICA occlusion. It was found that, when both ICAs were occluded, flow through the basilar artery was increased 2.5-fold compared with that in control subjects, indicating that the basilar artery is the main supplying artery.^{13 14} Accordingly, Romero *et al*¹¹ concluded that the presence of good collateral circulation is a favourable prognostic factor and its absence is considered an unfavourable prognostic indicator in acute and chronic settings for patients with symptomatic carotid artery disease.

The two principal mechanisms for ischaemic stroke in patients with carotid artery occlusion/severe stenosis are hypoperfusion and embolism; in this setting, decreased cerebral perfusion and insufficient collateral circulation may exacerbate the impact of embolism, as microemboli are more likely to cause symptomatic brain ischaemia due to impaired washout in hypoperfused areas.^{11 15 16} According to Thanvi and Robinson,¹⁷ embolism is probably the most common mechanism, accounting for nearly two-thirds of strokes in internal carotid artery occlusion; the emboli originate from the distal tail of the occluded artery. The subject of future risk of completed stroke or transient ischaemic attacks in patients with internal carotid artery occlusion is complex; the highest risk is seen in patients with bilateral occlusion.¹⁷

Friedman *et al*⁶ performed external carotid artery revascularisation on 10 patients with BICAO; they had a mean follow-up of 44.7 months. Only one patient developed a transient ischaemic attack. The authors concluded that external carotid artery revascularisation may be an effective and durable treatment for patients with BICAO and that medical treatment alone or extracranial-intracranial bypass do not appear to offer these patients any protection from symptoms of cerebrovascular insufficiency.

Our patient developed an ischaemic stroke at the right middle cerebral artery 1 year prior. He was diagnosed with BICAO at that time by Doppler-duplex analysis. The patient was given medical treatment only. The second ischaemic stroke hit the

patient at the contralateral arterial area after approximately 1 year. Both internal carotids were occluded and spontaneous recanalisation was not found.

Although conventional cerebral angiography revealed the presence of good collateralisation and cross-filling of anterior circulation through the vertebrobasilar system, the patient developed a new contralateral disabling ischaemic stroke. The patient later died of severe aspiration pneumonia.

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Contributors OSMA managed the patient, took and edited the pictures and video, and drafted the manuscript.

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Learning points

- Bilateral internal carotid occlusion (BICAO) is a rare disease and carries a poor outcome.
- The collateral circulation in BICAO comes from the vertebrobasilar system with cross-filling of the middle cerebral artery through the circle of Willis (as in our patient), an external carotid/ophthalmic anastomosis, or a combination of the two.
- External carotid artery revascularisation may be an effective and durable treatment for patients with BICAO.

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