

## Spontaneous recanalisation of a chronic internal carotid artery occlusion<sup>☆</sup>



### Recanalización espontánea de una oclusión crónica de arteria carótida interna

Dear Editor,

Internal carotid artery (ICA) occlusion is an important cause of stroke: stroke most frequently occurs at the time of occlusion or during the following year, due to low perfusion and cerebral ischaemia or artery-to-artery embolism. In symptomatic stenosis with more than 70% of arterial lumen, selected symptomatic patients with stenosis of more than 50%, or asymptomatic patients with a more than 70% stenosis, endarterectomy or carotid stent placement has been shown to be beneficial for secondary prevention of ischaemic strokes. However, there is no recommended surgical or interventional procedure for carotid occlusion and current recommendations focus on monitoring the contralateral ICA, controlling cardiovascular risk factors, avoiding distal hypoperfusion, and awaiting the development of natural compensatory mechanisms. The possibility of spontaneous recanalisation of the occluded artery is frequently overlooked.

We report the case of a patient with ICA occlusion which was confirmed by arteriography and followed up with ultrasound. At 18 months, spontaneous recanalisation was observed, with a critical stenosis of that artery, which poses the following questions: How frequent is spontaneous recanalisation of an occluded ICA? When does it happen? How should we react? For how long should we follow up patients with occluded ICAs?

### Clinical case

We present the case of a male smoker, aged 52, with type 2 diabetes mellitus and no history of arterial hypertension. He was admitted due to left hemiparesis with faciobrachial predominance and sudden-onset ipsilateral hypoaesthesia, with no language impairment, visual field deficit, or any other associated neurological symptoms. A brain MRI showed an infarct in the right middle cerebral artery (MCA) territory. An echo-Doppler study of the supra-aortic trunks (SAT) revealed an occlusion of the right proximal ICA, a stenosis of less than 50% of the left ICA, and multiple, well-defined atherosclerotic plaques in both carotid sinuses and in the right common carotid artery. These findings were confirmed by angiography of the SATs (Fig. 1A), which revealed no radiological findings typical of fibromuscular dysplasia, vasculitis, or arterial dissection. The patient's clinical symptoms sta-

bilised and he was discharged after 8 days of hospitalisation, receiving 300 mg of acetylsalicylic acid and 80 mg atorvastatin daily.

A follow-up echo-Doppler study of the SATs performed 6 months later showed the same results as those obtained during hospitalisation.

Eighteen months after his discharge, the patient visited our department due to a one-week history of dizziness without vertigo and no other associated clinical symptoms. Physical examination revealed no new findings besides the sequelae of the previous cerebral ischaemic event. Vascular risk factors were adequately controlled, although LDL cholesterol levels had decreased only to 90 mg/dL. An echo-Doppler study of the SATs revealed a filiform flow in the right ICA (which was occluded in the previous studies). This was confirmed by a new angiography of the SATs, which revealed a critical stenosis secondary to an atherosclerotic plaque at this level (Fig. 1B). The right hemisphere was adequately supplied by the left ICA. Lastly, considering the compensatory flow through the collateral arteries of the circle of Willis, the absence of significant interhemispheric asymmetries in the evaluation of haemodynamic reserve, the lack of microembolism detected by the transcranial Doppler ultrasound, and the fact that the patient had remained asymptomatic for the 18 months he was receiving the prescribed treatment, it was decided, in consensus with the interventional neuroradiology and vascular surgery departments, to continue the conservative treatment and periodic follow-up with ultrasound studies.

### Discussion

Spontaneous recanalisation of an occluded ICA was believed to be an infrequent phenomenon; however, the number of published cases and some case series<sup>1-4</sup> make us believe that this phenomenon is probably more frequent than thought. It most frequently occurs shortly after the occlusion<sup>5</sup>; however, published rates of late recanalisation range from 2.3% to 11%.<sup>1,6,7</sup>

The mechanism by which an occluded ICA is recanalised is subject to debate. Acute occlusions may be recanalised by endogenous lysis, decreased endothelial oedema located at the level of the occlusion, or intraplaque haemorrhage.<sup>8</sup> Several hypotheses have been proposed for the recanalisation of chronic occlusions, including the activation of endothelial thrombolytic mechanisms; the histological characteristics of the plaque are also believed to influence the probability of recanalisation.<sup>4</sup>

Carotid occlusions have long been described as a stable condition not requiring surgery, unlike severe, symptomatic carotid stenosis (from 70% to 99% of arterial lumen), which may be considered to involve high embolic risk and, therefore, benefit from endarterectomy or angioplasty. After spontaneous recanalisation, an occluded ICA develops severe stenosis; the therapeutic approach to be taken with these patients may change as a result of this, especially in those who have experienced an improvement in the sequelae caused by the previous stroke, and with the brain parenchyma supplied by the occluded ICA being partially preserved.

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**Figure 1** (A) Angiography of the SATs showing the occlusion of the right ICA. (B) Angiography of the SATs showing recanalization of the right ICA.

Intra-arterial angiography is the standard test of reference to observe an occlusion or differentiate it from an ICA with severe stenosis<sup>9</sup>; however, this is an invasive procedure and may be associated with a certain risk of complication. The availability of the echo-Doppler study of the SATs, and its good sensitivity and specificity to detect ICA pseudo-occlusions (94% and 100%, respectively),<sup>9,10</sup> support the use of this technique for proper follow-up of these patients. In doubtful cases, an echo-Doppler study of the SATs together with a CT angiography or contrast-enhanced MRI angiography may avert the need to perform an angiography.<sup>11</sup> The duration and frequency with which follow-up echo-Doppler studies of the SATs should be performed in patients with carotid occlusions are not clear.

This case is an example of the possibility of late spontaneous recanalisation of an occluded ICA. This event may be more frequent than expected. Therefore, we deem it interesting to consider following up patients with an occluded ICA through echo-Doppler studies of the SATs with the aim of promptly detecting candidates to invasive treatment due to recanalisation.

## Conflicts of interest

The authors have no conflicts of interest to declare.

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## Tolosa-Hunt syndrome with spontaneous remission and recurrence<sup>☆</sup>



### Síndrome de Tolosa-Hunt con remisión espontánea y recurrencia

Dear Editor,

Tolosa-Hunt syndrome (THS) is an idiopathic condition included in the differential diagnosis of painful ophthalmoplegia. The condition is caused by granulomatous infiltration of the lateral wall of the cavernous sinus and characterised by one or more episodes of unilateral orbital pain associated with paralysis of one or more oculomotor nerves. Magnetic resonance imaging (MRI) reveals granulomatous infiltration of the cavernous sinus. The condition is treated with corticosteroids, leading to the complete resolution of symptoms within 72 hours. Up to half of patients experience recurrences even several months or years after the first episode; recurrences are usually ipsilateral. However, spontaneous remission may also occur. We present a case of spontaneous remission and subsequent recurrence of THS.

Our patient was a 31-year-old Peruvian woman who had been living in Spain for the past 5 years. She had a history of idiopathic right-sided peripheral facial paralysis in 2009, which resolved completely, and a 2-year history of episodic migraine without aura. The patient was receiving oral contraceptives. She visited the emergency department in October 2011 due to a one-week history of binocular diplopia and headache. Diplopia became worse with right horizontal gaze and did not change with distance. Headache was right hemicranial and pulsatile; it was associated with photophobia and had similar features to those of her usual episodes of migraine, increasing progressively until reaching a score of 8/10 on the visual analogue scale. The patient also reported paraesthesia in the area around the mouth and the right side of the jaw. No proptosis or any other eye abnormalities were reported.

In August of the same year, the patient had experienced a similar episode, which lasted a week and resolved spontaneously, leaving no sequelae. The results from the physical examination and the patient's vital signs were normal. A neuro-ophthalmological examination revealed anisocoria (right pupil: 4 mm; left pupil: 3 mm); changes in illumination

did not affect pupil size. Both direct and consensual light reflexes were normal. We also observed ptosis in the right eye and dysconjugate gaze in the primary gaze position; esotropia in the right eye and hypertropia in the left (positive cover-uncover test, negative alternate cover test). Examination of ocular movements revealed limited abduction and supradextroversion of the right eye; diplopia was most marked in these gaze positions. In the following days, the right eye also displayed limited adduction, infradextroversion, and infralevoversion. The rest of the neurological examination (including examination of the eye fundus, visual acuity, and the remaining cranial nerves) found no abnormalities. In summary, the results of the neuro-ophthalmological evaluation were compatible with paralysis of the right third and sixth cranial nerves.

In view of her symptoms, the patient was diagnosed with recurrent painful ophthalmoplegia. A complete blood count and biochemical study (including determination of the erythrocyte sedimentation rate [ESR] and an angiotensin-converting enzyme [ACE] test) only revealed hypertriglyceridaemia. The results from serology tests for Borrelia and syphilis and an immunological study (antinuclear, anti-neutrophil cytoplasmic, and anti-thyroid autoantibodies) were negative. A CSF analysis (biochemical analysis, culture, and Venereal Disease Research Laboratory test) disclosed no abnormalities. Post-contrast MRI sequences displayed asymmetrical uptake in the orbital apex and the lateral wall of the right cavernous sinus (Fig. 1). No asymmetries were found in the calibre or signal intensity of the optic nerve.

The patient was diagnosed with THS based on her symptoms and neuroimaging results, and after other aetiologies were ruled out. We started treatment with intravenous methylprednisolone dosed at 1 g every 24 hours for 3 days, followed by decreasing doses of oral prednisone. Within 24 hours from treatment onset, headache and diplopia improved progressively, eventually resolving completely.

THS is a rare, idiopathic, inflammatory process of the cavernous sinus. The syndrome affects both sexes equally and is more frequent in middle-aged individuals. Some authors, however, have reported cases of THS in children.<sup>1</sup> THS is characterised by unilateral orbital pain that may radiate to the retro-orbital, frontal, temporal, and even occipital regions. Pain is associated with diplopia due to involvement of one or more oculomotor nerves, and may also affect the ipsilateral first or second branches of the trigeminal nerve.<sup>2,3</sup> Cranial nerve involvement may occur simultaneously or up to 2 weeks after headache. Involvement of other cranial nerves, such as cranial nerve II, the third branch of cranial nerve V, cranial nerve VII, or the ipsilateral sympathetic pupillary pathway, has also been described.<sup>4</sup> Bilateral cranial nerve involvement is very infrequent. In 2004, the International Headache Society

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