

# Asymptomatic Bilateral Internal Carotid Artery Occlusion Associated with Stenosis of the Right External Carotid Artery: Case Report

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## ABSTRACT

**Background:** Bilateral Internal Carotid Artery Occlusion (BICAO) is an extremely rare pathological entity which have grave prognosis. It is responsible for 0.4% of completed strokes. Asymptomatic BICAO remains exceptional. **Case report:** We present a case of a 72-year-old man with asymptomatic bilateral internal carotid artery occlusion associated with stenosis of the right external carotid artery. **Discussion:** Therapeutic abstention was indicated given the absence of symptoms and the advanced age of our patient. Due to the limited cases reported in the literature whose therapeutic outcomes are controversial, the proper management strategy of BICAO remains unclear. **Conclusion:** We concluded that the increase in the number of reported cases, preferably series with large sample, as well as a long-term follow up for these patients will allow for a better evaluation of each therapeutic decision select.

**Key words:** Asymptomatic; Bilateral occlusion; Internal carotid artery; Therapeutic abstention.

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## INTRODUCTION

Bilateral Internal Carotid Artery Occlusion (BICAO) is an extremely rare pathological entity, therefore, studies of the natural history of this disease are lacking in the literature.<sup>1</sup> It is responsible for 0.4% of completed strokes.<sup>2</sup> Bilateral asymptomatic occlusion remains exceptional. BICAO has grave prognosis and should be considered a marker for severe systemic disease.<sup>1</sup> The management strategy for these patients is poorly understood given the limited number of cases reported in the literature.

## CASE REPORT

We report the case of a 72-year-old man, with smoking, diabetes mellitus and high blood pressure as cardiovascular risk factors. He is admitted for management of crescendo angina. At admission, the cardiovascular examination revealed a bilateral carotid murmur. His neurological examination was with no abnormality. Electrocardiogram recorded a regular sinus rhythm without repolarization abnormalities. Transthoracic echocardiography revealed a normal left ventricular contractile function and cardiac enzymes were negatives.

Coronary angiography objectified *tritruncular* lesions (Figure 1,2) and the patient underwent angioplasty of the left *anterior* descending artery with placement of two active stents.

The supra-aortic trunks echo-Doppler, performed as part of evaluation of the carotid murmur, objectified a bilateral internal carotid artery occlusion associated with stenosis of the right external carotid artery in its origin. This result was confirmed by the angiography of supra aortic trunks (Figure 3,4). Although our patient is asymptomatic neurologically, we completed the assessment with a brain Magnetic Resonance Imaging (MRI). It was normal without signs of ischemic stroke (Figure 5). In the multidisciplinary meeting, therapeutic abstention was indicated for carotid lesions given the absence of symptoms and the advanced age of our patient. He was discharged with statins and antiplatelets aggregation.

## DISCUSSION

Most occlusions of the internal carotid artery occur in patients with a history of cardiovascular disease such as atherosclerosis, coronary heart disease and stroke.<sup>3</sup> 99.6% of occlusions affect only one side of the inter-

nal carotid artery.<sup>4</sup> Bilateral internal carotid artery occlusion is an extremely rare disease with a gloomy prognosis.<sup>2</sup>

It is reported in very few cases. We think that the rarity of the cases reported in the literature is the main cause of the misunderstanding of this pathology. So, we decided to share this observation because as the number of reported cases increases, we will have a database to explore in order to describe and understand its clinical, therapeutic and evolutionary aspect.

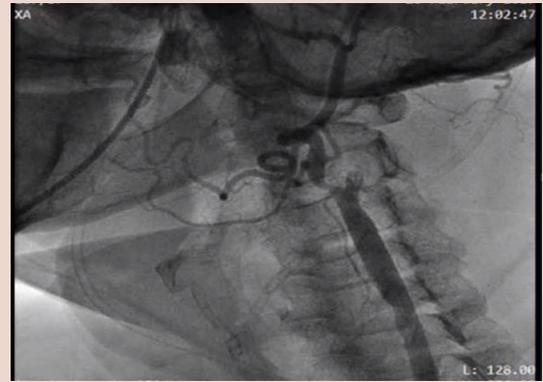
Patel *et al.* reported a retrospective review of 30,812 carotid duplex examinations performed over 12 years. The prevalence of BICAO in this series was 0.15% (Only 45 patients had BICAO).<sup>5</sup> Despite the fact that the mechanism of arterial stenosis/occlusion remains unclear, atherosclerosis is considered the most significant. Smoking, Hypertension, hyperlipidemia and diabetes mellitus are the principal risk factors of atherogenesis.<sup>6</sup> In the AbuRahma study, the majority of patients had more than one risk factor for atherosclerosis: smoking in 100%, hyperlipidemia in 67%, hypertension in 81% and diabetes mellitus in 33%.<sup>1</sup> In our case, patient had smoking, diabetes mellitus and high blood pressure as cardiovascular risk factors, which reinforces the hypothesis that atherosclerosis, is the most significant mechanism.

The clinical spectrum of BICAO varies from a completely asymptomatic occlusion to a fatal ischemic stroke.<sup>7</sup> According to the literature review, the majority of BICAO cases progress to fatal ischemic stroke. In reporting our case, we want to emphasize the interest of detecting this pathology in asymptomatic patients with a history of cardiovascular disease in order to treat it and thus prevent progression to fatal ischemic stroke.

AbuRahma *et al.* included twenty-one patients with BICAO; they were all symptomatic and their clinical presentations included hemispheric transient ischemic attacks, amaurosis fugax, strokes and non-hemispheric transient ischemic attack.<sup>1</sup> In our case, the patient was asymptomatic suggesting the presence of a satisfactory collateral circulation which was supplied by the anastomose of the distal branches of the external carotid artery with distal branches of the internal carotid artery and/or by the vertebrobasilar system into the Willis circle via the posterior communicating arteries.<sup>3,7</sup> The prevalence of asymptomatic BICAO is unknown. We reviewed another 2 asymptomatic BICAO cases. Chen *et al.* reported a case of a 46-year-old female BICAO patient only presenting mild clinical



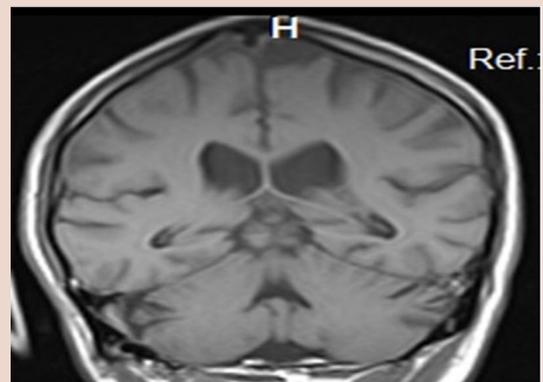
**Figure 1:** Coronarography showing tight stenosis of left anterior descending artery at its proximal and middle segment.



**Figure 4:** Angiographic image showing complete occlusion of the left internal carotid artery from its origin.



**Figure 2:** Coronarography showing a long occlusion of the right coronary artery from the proximal part of its first segment and extending to the second segment.



**Figure 5:** Normal brain MRI: it's showing no signs of cerebral infarction.



**Figure 3:** Angiographic image showing complete occlusion of the right internal carotid artery from its origin associated with external carotid stenosis.

cal symptoms, such as dizziness and nausea.<sup>3</sup> In the other case, Fatic *et al.* published the case of an asymptomatic bilateral internal carotid artery occlusion, bilateral external carotid artery stenosis and bilateral kinking of the vertebral artery.<sup>7</sup> In our patient, the evaluation of a carotid murmur allowed us to make the diagnosis of BICAO in a patient with ob-

vious atherosclerotic terrain given the presence of several cardiovascular risk factors and the notion of newly diagnosed ischemic heart disease.

The best management strategy for this disease remains unclear. Currently, there is little evidence to support management strategies and no official recommendations for asymptomatic bilateral carotid artery occlusion.<sup>7</sup> Patel's team described the natural history of BICAO in a cohort of 45 patients and evaluated the use of antiplatelets or anticoagulants in those. He concluded that survival favored patients on single antiplatelet (Odds Ratio [OR], 0.67;  $P \leq 0.008$ ) or with combination of anticoagulation (OR, 0.67;  $P \leq 0.018$ ).<sup>5</sup> On the other side, Osama Amin reported a case of a 52-year-old man who developed ischaemic infarction at the region of the right middle cerebral artery related to bilateral occlusion of the internal carotid artery on Doppler-duplex examination. He was treated medically by antiplatelet agents only and he presented; after one year; a new infarction at the region of the left middle cerebral artery. He died after 2 months. According to this observation, the prognosis of BICAO under medical treatment alone is poor and there is a risk of recurrent ischaemic events.<sup>2</sup> Revascularization approach; by vascular bypass, Carotid Endarterectomy (CEA) or Carotid Artery Stenting (CAS), is another choice supported by neurosurgeons. Friedman *et al.* reported that only 10% of BICAO patients who undergo external carotid artery revascularization present a transient ischemic stroke during the follow-up period without any effect of vascular bypass or medical treatment alone on symptom improvement.<sup>8</sup> Contrariwise, no significant difference between medical therapy and revascularization according to

the results of the meta-analysis conducted by Mylonas SN *et al.*<sup>9</sup> Persoon *et al.* revealed that the prognosis of BICAO patients treated by medical therapy is better than the surgical group.<sup>10</sup> This same result was demonstrated by a randomized study within a 2-year follow-up period.<sup>11</sup> Contrary to this result, AbuRahma *et al.* in their series of twenty-one patients with BICAO, demonstrated the superiority of surgical management compared to medical treatment. For a follow-up period of 6 years, the mortality rate was 38% in the surgical group and 75% in the medical group. The causes of death were myocardial infarctions and strokes.<sup>1</sup> Conservative therapy was selected by Chen *et al.* for her patient considering her mild clinical symptoms and satisfactory compensation.<sup>3</sup> Similarly, the asymptomatic BICAO patient reported by Fatic *et al.* has been successfully managed conservatively for over 5 years.<sup>7</sup>

When we analyzed the data from the literature series on the therapeutic strategy, we noticed a big contradiction. As a result, the therapeutic decision for our patient has become difficult. We chose conservative medical treatment taking into account the number of studies that reported the benefit of medical treatment versus revascularization over an acceptable duration of follow-up and given the absence of symptoms and the advanced age of the patient.

The duration of the follow-up of our patient, to this day, is 18 months. Comparing to the series, this duration remains short and insufficient to evaluate the effectiveness of our therapeutic behavior.

According to a literature review, BICAO have grave prognoses and associated with poor long-term survival. AbuRahma *et al.* reported an overall mortality rate at 52% for a follow-up period of 6 years.<sup>1</sup> In the series of Patel *et al.* which they described the natural history of 45 patients over a 31-month follow-up period at mean, mortality at 1, 3 and 5 years was 24%, 36% and 47% respectively.<sup>5</sup>

## CONCLUSION

BICAO is a rare vascular disease. Given its grave prognosis determined by the risk of occurrence of fatal ischaemic stroke, we highlight the interest of its screening in patients with obvious atherosclerotic terrain especially in the presence of a history of cardiovascular disease. We concluded that the increase in the number of reported cases, preferably series with large

sample, as well as a long-term follow up for these patients will allow for a better evaluation of each therapeutic decision select.

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Not applicable.

## CONFLICT OF INTEREST

The authors declare no conflict of interest.

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