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An Ischemic Stroke in a Sub-Saharan African Patient Revealing a Carotid Web: A Case Report and a Systematic Literature Review

Léhleng Agba[1,2*], Donissongui Soro[1], Awissoba Awidina-Ama[1], Liliane Ngoudjo[1], Hugo Yaïche[1], Olivier Heinzlef[1]

[1]Neurology Department, Centre Hospitalier Intercommunal de Poissy/Saint-Germain-En-Laye, Poissy, France
[2]Neurology Department, CHU-Kara, Université de Kara, Kara, Togo
Email: *thierrielle@gmail.com

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Abstract

One third of ischemic strokes have no known cause, and basic understanding of the mechanisms of these "cryptogenic" strokes is lacking. However, observational studies are increasingly bringing to light an etiology that has long remained hidden, the carotid web. The authors report a case of carotid web in a 32-year-old patient, gardener, from sub-Sharan Africa, with no medical history or known cerebrovascular risk factors. Indeed, due to a sudden weakness of his left limbs, the MRI was performed and revealed an ischemic stroke positive on diffusion-weighted within the territory of the right middle cerebral artery. FLAIR sequences were normal, while T^{2*} sequences revealed an M1 thrombus ipsilateral to the brain lesion. After thrombolysis and thrombectomy, the patient's weakness improved significantly. Digital subtraction angiography of supra-aortic trunks revealed the carotid web, which was incriminated as the etiology of this ischemic stroke in the absence of any abnormality on the other ancillary examinations that were performed. To prevent a recurrence, the patient was given Aspegic 250 mg/day and received physical therapy. The clinical course was improved and at 3 months, there was no new cerebrovascular event and his Rankin score was 1.

Keywords

Carotid Web, Ischemic Stroke, Sub-Saharan Africa

1. Introduction

One third of all ischemic strokes have no known cause [1] and there is a fundamental gap in knowledge about the mechanisms of these "cryptogenic" strokes.

However, it is essential to identify the underlying cause of cerebral ischemia for effective secondary prevention. Recent studies have shown that a carotid web (CW) is a novel risk factor for cryptogenic ischemic stroke [2] [3]. CW is also known as carotid bulb fibromuscular dysplasia (FMD). First described in 1968 as a discrete ridge [4], it is best defined radiographically as a distinctive, intraluminal filling defect commonly affecting the posterolateral wall of the carotid artery at the most proximal portion of the internal carotid artery (ICA) [2]. Momose and New first used the term "web" in 1973 to describe this entity on carotid angiography in 4 patients in a series of 7000 patients during 8 years at Massachusetts General Hospital [5]. CW is often misdiagnosed or missed because of the lack of relevant experience of radiologists or clinicians, especially in patients in a precarious situation. We report a young male patient with ipsilateral CW associated with acute right middle cerebral artery occlusion who was successfully treated by stent retriever embolectomy and we review the literature on this uncommon cause of ischemic stroke.

2. Case Report

A 32-year-old sub-Saharan African patient, right-handed, gardener, who did not have a previous medical history or known cerebrovascular risk factors, was admitted to the emergency department of the local hospital for a sudden left upper limb weakness 30 minutes after lunch. He was not on any medication. He was a nonsmoker and had no other specific medical history. Symptoms began on May 24, 2020 at 3:30 p.m. with a fall at home due to weakness in the left limbs. He was taken to the emergency room at 5:05 p.m. and the examination revealed a blood pressure of 127/83 mm Hg, a pulse of 84 per minute, a weight of 68 kg for a height of 198 cm, *i.e.*, a body mass index (BMI) of 17.3 $kg{\cdot}m^{-2}$. Consciousness was normal, there was no speech disorder. Left hemiplegia was noted, predominantly in the upper limb, and hypoesthesia was present in the limbs with deficits. The National Institutes of Health Stroke Scale (NIHSS) score was 13. Magnetic resonance imaging (MRI) performed at 5:50 pm showed a right middle cerebral artery infarction on diffusion-weighted images (**Figure 1(a)**), no abnormalities in Fluid-attenuated inversion recovery (FLAIR) sequence, and M1 thrombus on the T2*-weighted images (**Figure 1(b)**) with occlusion of right middle cerebral artery on angiography sequences (**Figure 1(c)**). Thrombolysis was initiated at 6:25 pm after no abnormalities or contraindications on a biological tests (**Table 1**) and electrocardiogram (ECG). The latter showed a regular sinus rhythm with a heart rate of 78 per minute. The patient was referred for thrombectomy in an interventional radiology unit. This treatment, which began at 07:20 pm, was performed by direct thromboaspiration and stentriever with introduction of Cangrelor at the end of the procedure allowing TICI 2b recanalization. Digital subtraction angiography (DSA) of supra-aortic trunks revealed a CW ipsilateral to the vascular lesion (**Figure 2**). A brain scan performed at 24 hours after thrombolysis showed a superficial right sylvian ischemia without hemorrhagic

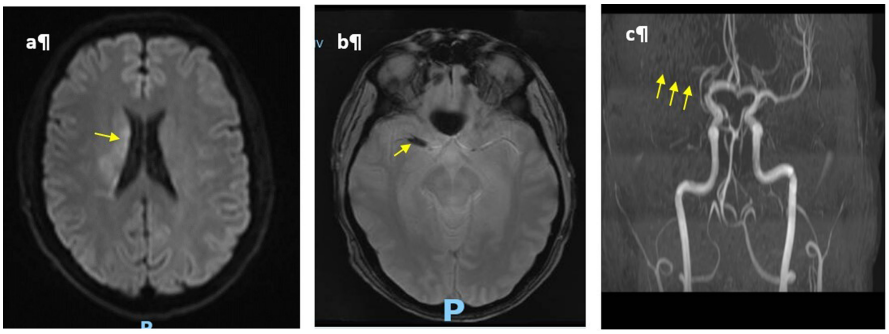

Figure 1. MRI performed 2 hr. 20 min after symptom onset shows acute ischemic lesions in the deep region of the right MCA on diffusion weighting (a); T2*-weighted sequences show M1 thrombus on the right, and angiographic sequences show occlusion of the right MCA (c).

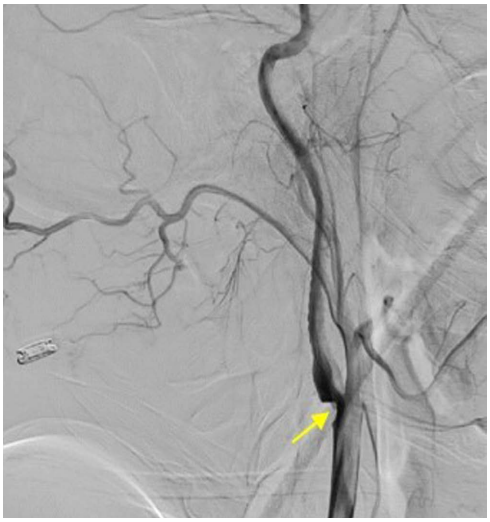

Figure 2. Digital subtraction angiography (DSA) of supra-aortic trunks showing a small triangular filling defect (yellow arrow) along the posterior wall of the right carotid bulb what is consistent with the web.

complications (**Figure 3**). The physical examination of the patient showed an improvement of the motor deficit with, at day 2, a NIHSS score of 5 and a Rankin score of 2. At day 4, the patient was transferred to a rehabilitation suite on Aspegic 250 mg daily. He was scheduled to have an endarterectomy of his CW. After three months of drug treatment, his condition progressed well without recurrence and with a Rankin score of 1.

3. Literature Review

3.1. Background and Epidemiology

CWs were first described 4 decades ago in a study of catheter angiograms at the Massachusetts General Hospital [5], and subsequent case reports have added about 50 cases to the literature [6]. CWs have had many different names in the

Table 1. Results of biological blood tests.

Parameters	Values	Reference values for Males
CHEMISTRY		
Urea (mmol/L)	2.4	0.9 - 6.2
Creatinine (µmol/L)	63	56 - 119
Uric Acid (µmol/L)	389	126 - 418
Glucose (mmol/L)	4.2	3.5 - 6.3
Sodium (mmol/L)	144	135 - 151
Potassium (mmol/L)	4.1	3.6 - 5.2
Chloride (mmol/L)	116	101 - 115
Phosphorus (mmol/L)	1.3	0.7 - 1.5
Alanine aminotransferase, ALT (U/L)	53	8 - 54
Aspartate aminotransferase, AST (U/L)	59	17 - 60
Alkaline phosphatase, ALP (U/L)	298	101 - 353
Gamma-glutamyl transferase, GGT (U/L)	67	9 - 71
Lactate dehydrogenase, LDH (U/L)	355	274 - 745
Protein, total (g/L)	70.5	46.7 - 86.4
Albumin (g/L)	49.0	32.7 - 49.8
Bilirubin, Total (µmol/L)	7.6	3.8 - 32.0
Bilirubin, Direct (µmol/L)	1.3	0.9 - 4.1
Cholesterol, Tot al (mmol/L)	3.87	1.8 - 5
Cholesterol, HDL (mmol/L)	0.78	>1
Cholesterol, LDL (mmol/L)	2.61	<4.10
Triglycerides (mmol/L)	2.35	<1.80
Troponine (ng/L)	0.05	0 - 0.04
D-dimer (ng/mL)	300	0 - 250
C-reactive protein (mg/L)	7	0 - 5
COMPLETE BLOOD COUNT		
Haemoglobin (g/L)	14.5	135 - 165
Haematocrit (%)	48	41 - 50
Red blood cells $\times 10^{12}$/L	5.23	4.37 - 5.63
White blood cells, Total $\times 10^{9}$/L	7.88	4.5 - 11
Granulocytes $\times 10^{9}$/L	6.35	1.7 - 7
Lymphocytes $\times 10^{9}$/L	3.74	1.5 - 4
Monocytes $\times 10^{9}$/L	0.81	0 - 1
Platelets	378	150 - 400

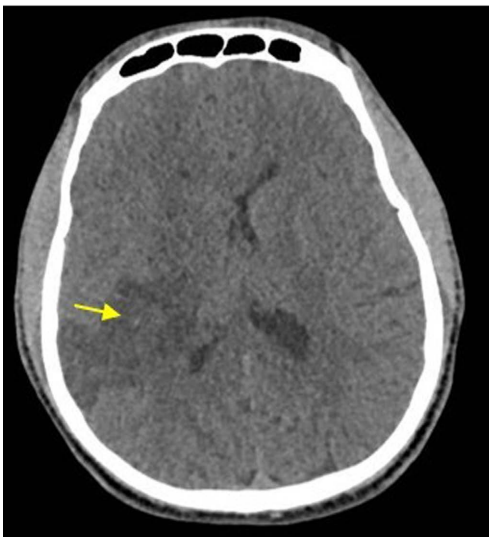

Figure 3. The non-contrast CT scan performed 24 hours after thrombolysis and thrombectomy showed a. Territorial infarction in the territory of the right middle cerebral artery without hemorrhagic complications.

literature, including carotid weblike formations or septums, carotid pseudovalvular folds, carotid shelves, carotid diaphragms, and thrombotic carotid megabulbs. "Carotid web" is the most widely used description [7]. Case-control studies have found that CWs are present in 9% to 37% of patients younger than 60 years with cryptogenic stroke, and that a CW increases the risk of ischemic stroke approximately 10- to 20-fold [6] [8] [9]. The mean age of patient with CW is reported to be relatively low, 52.5 years (ranged 16 - 78 years) by Zhan *et al.* in their systematic review study conducted in 2018 [10]. However, a more recent largest US epidemiologic study of this affection conducted in 2021 reported the following epidemiologic characteristics: a mean age of 63 years, ratio of 62.5% female and 50% of African American [11]. In the same year, Mac Grory *et al.* reported that, three of four patients with symptomatic CW were Black [12] is in accordance with our reported case. A handful of studies has also been carried out in sub-Saharan Africa, the most recent being that of Gaye *et al.* in Senegal. Published in March 2022, it was a descriptive study of 6 cases which revealed an average age of 41 ± 6 years and a sex ratio of 1 [13]. Therefore, it is clear that most of the reviews suggest that this is an affection found mainly in young adults, especially in the case of cryptogenic ischemic stroke. It has been theorized across multiple observational studies that CW is a cause of ischemic stroke and may be associated with a high risk of recurrent stroke. A 2018 systematic review demonstrated an ischemic stroke recurrence rate of 56% (with a median of 12 months to the recurrent stroke, range, 0 - 97 months) in patients with CW receiving medical management. The risk of stroke recurrence is increased 8-fold in patients with an ipsilateral CW as compared with patients without CW. Moreover, the recurrent ischemic strokes were in the same vascular territory as the CW,

and the risk of recurrent stroke appeared to persist after 2 years [14].

3.2. Histopathology and Pathophysiology

CW corresponds to a linear filling defect usually on the posterolateral side of the carotid bulb, just beyond the carotid bifurcation [3]. Despite sharing a location typically associated with atherosclerotic disease, histologically, CW has been shown to represent a nonatherosclerotic entity [10]. Surgical pathology of resected CWs demonstrates intimal fibrous proliferation without evidence of atheromatous deposits, consistent with a focal variant of intimal FMD. This atypical, intimal pathology is in contrast to the classic, medial FMD often seen as a string of beads on imaging [10]. CW has recently been recognized as a cause of stroke of undetermined cause, representing as many as 9.4% to 37% of cryptogenic strokes [3] [6]. The pathogenesis of ischemic stroke caused by a CW is not fully understood, but it could be attributed to the morphological characteristics and regional hemodynamic changes in a CW [2]. In fact, according to Choi PM, turbulence and stasis in the cul-de-sac upstream of the web may create a thrombogenic milieu, and embolic stroke may occur when the size of the thrombus is sufficiently large [2]. This hypothesis is supported by cerebral angiography demonstrating marked stasis of intravenous contrast distal to the CW [10] [15] and the appearance of thrombus superposed along the upper surface of the diaphragm on vascular imaging in some patients with CW [2] [3] [16].

3.3. Diagnosis

The diagnosis of the CW is made exclusively with the help of imaging of the supra-aortic vessels. Once highlighted, the web will be incriminated as responsible for the ischemia on condition that it is located on the homolateral side of the cerebral infarction and in the absence of any other identifiable cause. A recent study provided evidence on the correlation between a CW and ischemic stroke in patients in whom alternative causes of stroke are not identified [6]. A CW may be an underrated risk factor for stroke. Sajedi *et al.* found that patients with a CW accounted for 21.2% of cryptogenic stroke cases [4]. Doppler ultrasound is a widely used examination tool for the carotid artery without any radiation injury. The representative ultrasonographic manifestation of CWs is a membranous structure protruding from the carotid sinus or the initial segment of the ICA that fluctuates with blood flow. Additionally, hemodynamic changes near the CW can also be observed by Doppler ultrasound, which is superior to computed tomographic angiography (CTA) in showing morphological and hemodynamic evidence of thrombus formation. However, a CW is often misdiagnosed as an atherosclerotic plaque by the operator because of a lack of clinical experience and skills [3] [9]. CTA allows detailed anatomical imaging of the craniocervical vessels with multiplane reconstruction in a short period, and has a high sensitivity and specificity for diagnosing a CW. CTA also differentiates between a CW and atherosclerosis, and other lesions. However, CTA cannot provide informa-

tion about flow dynamics and the composition of the lesion. Additional disadvantages of exposure to radiation and iodinated contrast agent have also been found [2] [6] [7] [8] [17]. Magnetic Resonance Imaging (MRI) has the advantages of nonradiation and non-invasiveness. A sagittal image shows the shape and position of a CW, and an axial image shows the characteristics of the luminal structure. Only a few studies have shown the application of high-resolution MRI (HRMRI) in the diagnosis of a CW. A small-sample study by Zhu *et al.* suggested that carotid ultrasound combined with CTA and HRMRI is an effective and reliable method for diagnosing a CW [18]. Therefore, HRMRI could also be considered as a useful diagnostic method to detect CW [19]. Digital subtraction angiography (DSA) is an imaging tool with high temporal and spatial resolution, and it is the criterion standard for craniocervical angiography in carotid imaging. DSA is characterized by a remnant of contrast that can be observed over a CW [7]. However, a disadvantage of DSA is that it has a risk of losing the CW in the invasive operation by standard posteroanterior and lateral projections. A misdiagnosis usually occurs during emergency endovascular treatment [19].

3.4. Treatment and Prognosis

Due to the lack of high-quality clinical studies, there is currently no uniform standard for the treatment of carotid arteries. Therefore, the optimal treatment options for preventing relapse in patients with symptomatic CW are unclear. One of the major knowledge gaps in deciding the optimal treatment is the lack of studies on the risk of recurrent stroke in patients with a symptomatic CW. Most patients are treated with antiplatelet therapy, but some physicians advocate using anticoagulation therapy as a better choice because of focal blood stasis in the carotid artery caused by CW [7]. According to the literature review, more than half of symptomatic CW patients treated with this standard medical therapy had stroke recurrence with a median time to recurrent stroke of only 12 months [10]. Due to the histopathological feature of a CW, a carotid stent or carotid endarterectomy (CEA) is the optimal treatment [16]. Some studies have shown that CEA or carotid stenting is safe and effective for carotid arteries. A recent review of 70 patients showed that 60% of them had a symptomatic CW, and they underwent CEA or carotid artery stenting without any complications or stroke recurrence for a median follow-up of 14 months [10].

4. Conclusion

For a long time, ischemic stroke in young adults has always been associated with arterial dissection as the most frequent etiology. Therefore, if no dissection or atherosclerosis is found during explorations, rare causes such as CW are not thought of. Its presence should be kept in mind in order to systematically look for it in ischemic strokes in young adults, especially in ischemic recurrences in the same cerebral artery territory.

Ethics Statement

The authors declare that they received informed consent from the patient and approval from the Ethics committee (approval number 2022-CHI-PSGL-078) of Poissy-Saint-Germain-En-Laye Hospital before writing and submitting this work for publication.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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