Case Report

Eagle syndrome as a rare cause of recurrent transient ischemic stroke in Senegal (West Africa)

Birame Loum1, Cheikh Ahmedou Lame1*, Cheikhna B. Ndiaye1, Kamadore Toure2, Mouhamadou Mansour Ndiaye3

1Department of ENT, Hospital principal de Dakar, Dakar, Senegal
2Department of Public Health, 3Department of Neurology, Cheikh Anta DIOP de Dakar, Dakar, Senegal

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*Correspondence:
Dr. Cheikh Ahmedou Lame,
E-mail: cheikhlame@gmail.com

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ABSTRACT

Eagle syndrome is a rare condition, often characterized by nonspecific symptoms. It is due to an abnormally long or compressive styloid process on surrounding structures. Exceptionally, it can cause neuro-vascular manifestations. We report an observation of Eagle syndrome discovered incidentally in presence of recurrent transient ischemic stroke. A 74-year-old man with no cardiovascular risk factors, was admitted to our department following 4 episodes of transient ischemic stroke with right hemiplegia and aphasia, always rapidly resolving. Head and neck CT scan showed 2 long styloid processes with a marked impingement of the left one against the ipsilateral internal carotid artery. Intraoral styloidectomy was performed. The patient recovered fully and remained free of symptoms without neurological impairment, at 6 months. Eagle syndrome is a rare condition which may lead, exceptionally, to repetitive transient ischemic stroke. Surgical styloidectomy must be considered to reduce the risk of new vascular events and prevent serious complications such as dissection of the internal carotid artery.

Keywords: Eagle syndrome, Internal carotid artery, Ischemic stroke, Styloidectomy

INTRODUCTION

Eagle syndrome corresponds to symptomatic elongation of the styloid process (superior to 3 cm) and/or calcification of the stylohyoid ligament.1,2,3 It manifests itself in a variety of non-specific symptoms related to the proximity of neurovascular structures (carotid arteries, internal jugular vein and cranial nerves).4 Stroke occurrence is exceptional in this case. We report an unusual case of recurrent transient ischemic stroke that reveals an Eagle syndrome.

CASE REPORT

A 74-year-old man, with no pathological history, was admitted in December 2020 to a clinic in Dakar (Senegal) for the occurrence of recurrent transient ischemic stroke. He had presented 4 episodes of neurological deficit with right hemiparesis and aphasia, occurring after head rotation to the left and down side. These signs were quickly resolved within 24 hours. The patient did not use alcohol or tobacco. No cardiovascular risk factors (high blood pressure, diabetes, dyslipidemia, heart disease) were found in him.

Biological tests including blood count, hepatic, renal workup, and lipid assessment (cholesterol and triglycerides) were normal.

No abnormality was found on electrocardiographic and transthoracic echocardiography monitoring. Supra-aortic trunks doppler ultrasound did not reveal atherosclerotic plaque.
Brain CT was normal. However, CT angiography of the supra-aortic arteries showed long styloid processes measuring 37 mm on the left and 32 mm on the right side; with impingement of the left one against the internal carotid artery (Figures 1 and 2).

Diagnosis of transient ischemic stroke secondary due to internal carotid artery compression by a long styloid process was retained and clopidogrel 75 mg per day prescribed.

Intraoral left styloidectomy was performed (Figures 3, 4 and 5) and the patient was discharged 4 days after. The check carried out a week later showed no anomalies.

Postoperative follow-up was simple, without neurological impairment, at 6 months.

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**Figure 1:** CT angiography of the neck with 3D reconstructions showing long styloid process (white arrows).

**Figure 2:** Head and neck CT angiography showing the impingement of the long styloid process against the left internal carotid artery (black arrow).

**Figure 3:** Operative view of the stylohyoid ligament after left tonsillectomy.

**Figure 4:** Operative view of the long styloid process after stylohyoid ligament section.

**Figure 5:** Resection of styloid process.
DISCUSSION

Eagle syndrome was first described in 1937 by Watt Eagle.\textsuperscript{1,2,5,6} It is characterized by abnormal ossification of the styloid process, the stylohyoid ligament and rarely the lesser cornu of the hyoid bone that causes an increase in the thickness and length of the styloid process.\textsuperscript{1,3} The elongated styloid process may cause compression of adjacent structures with various symptoms.\textsuperscript{1,7,8} The incidence of Eagle syndrome varies between 4 and 28%.\textsuperscript{5,7,9} In 93% of cases, the elongated styloid process is bilateral but rarely symptomatic.\textsuperscript{2,14,6}

Clinical presentation is subdivided into two major syndromic entities: The classic syndrome can occur unilaterally or bilaterally, and the symptoms may include neck pain, ear pain, dysphagia and dysphonia, tinnitus, odynophagia, sensation of a foreign body in the throat, and dizziness. All of these symptoms which can occur together or separately, at varying frequencies, are attributed to cranial nerves irritation V, VII, IX, or X; all very close to the styloid process.\textsuperscript{1,5,6,7} Stylocarotid syndrome in which the elongated styloid process is in contact with the internal carotid artery causing compression or carotid artery dissection with secondary occurrence of ischemic strokes.\textsuperscript{2,5,8}

Eagle syndrome diagnosis is made by imaging.\textsuperscript{6} Head and neck CT scan is the gold standard by visualizing the entire stylohyoid chain and measuring the length of the styloid process Eagle syndrome is confirmed by a styloid process superior to 3 cm.\textsuperscript{2,6,9,10} This elongated styloid process can exceptionally cause mechanical conflict with the internal carotid artery, thus defining the stylocarotid syndrome.\textsuperscript{6,7,10}

This conflict can lead either to direct arterial compression (corresponding to a hemodynamic mechanism), or to carotid dissection or thrombosis. It can also lead to the formation and/or rupture of an atherosclerotic plaque.\textsuperscript{10} The consequence is the occurrence of stroke which can be transient and repetitive as was the case with our patient.\textsuperscript{1,4,6,10}

In symptomatic cases, surgical procedure is the most chosen treatment.\textsuperscript{1,5,6} It prevents vascular events repetition and complications such as dissection of the internal carotid artery.\textsuperscript{4} This surgery involves resection of the styloid process by external cervical or intraoral approach.\textsuperscript{1,5,6} Intraoral approach was performed in our patient with uneventful outcome. After 6 months of follow-up, no recurrence of stroke was observed in our patient.

CONCLUSION

Repeated transient ischemic stroke is a rare manifestation of Eagle syndrome. Surgical resection of the long styloid process should be considered to prevent recurrence of stroke and complications, such as internal carotid artery dissection.

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