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Case Report

Twig-like MCA: A rare cause of intracranial bleeding ☆

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ABSTRACT

Twig-like or unfused middle cerebral artery (MCA) is a rare congenital vascular anomaly defined by the absence of the M1 segment. It can be found incidentally or can be revealed by cerebral ischemia or hemorrhage. Although rare, neuroradiologists should be familiar with such findings in order to differentiate them from differential diagnoses such as Moyamoya disease and steno-occlusive disorders of the MCA.

We report a case of a twig-like MCA revealed by intracranial bleeding in an 84-year-old woman.

Introduction

Twig-like or unfused MCA is a rare congenital vascular variation of the MCA, with a prevalence of less than 1% in the literature. It is defined by the replacement of the M1 segment by a plexiform arterial network of small dysplastic vessels [1]. Its diagnosis can be incidental or it can be revealed by cerebral ischemia or hemorrhage [2]. Cerebral digital subtraction angiography (DSA) is the imaging modality of choice to make this diagnosis and to eliminate differential diagnoses such as Moyamoya disease and steno-occlusive disorders of the MCA [3].

We report a case of a twig-like MCA revealed by intracranial bleeding in an 84-year-old woman.

Case presentation

An 84-year-old woman with a medical history of hypertension, type 2 diabetes, dyslipidemia, gout, and a surgical evacuation of a chronic subdural hematoma 1 month before admission presented to the emergency department with sudden-onset headache and vomiting, followed by impaired consciousness with a Glasgow Coma Scale of 13 at admission.

A nonenhanced head CT scan (Fig. 1) revealed an anterior interhemispheric hematoma associated with subarachnoid and interventricular hemorrhage.

Within a few hours of admission, the patient's consciousness deteriorated, and her GCS decreased from 13 to 8. A second CT scan showed the development of hydrocephalus,

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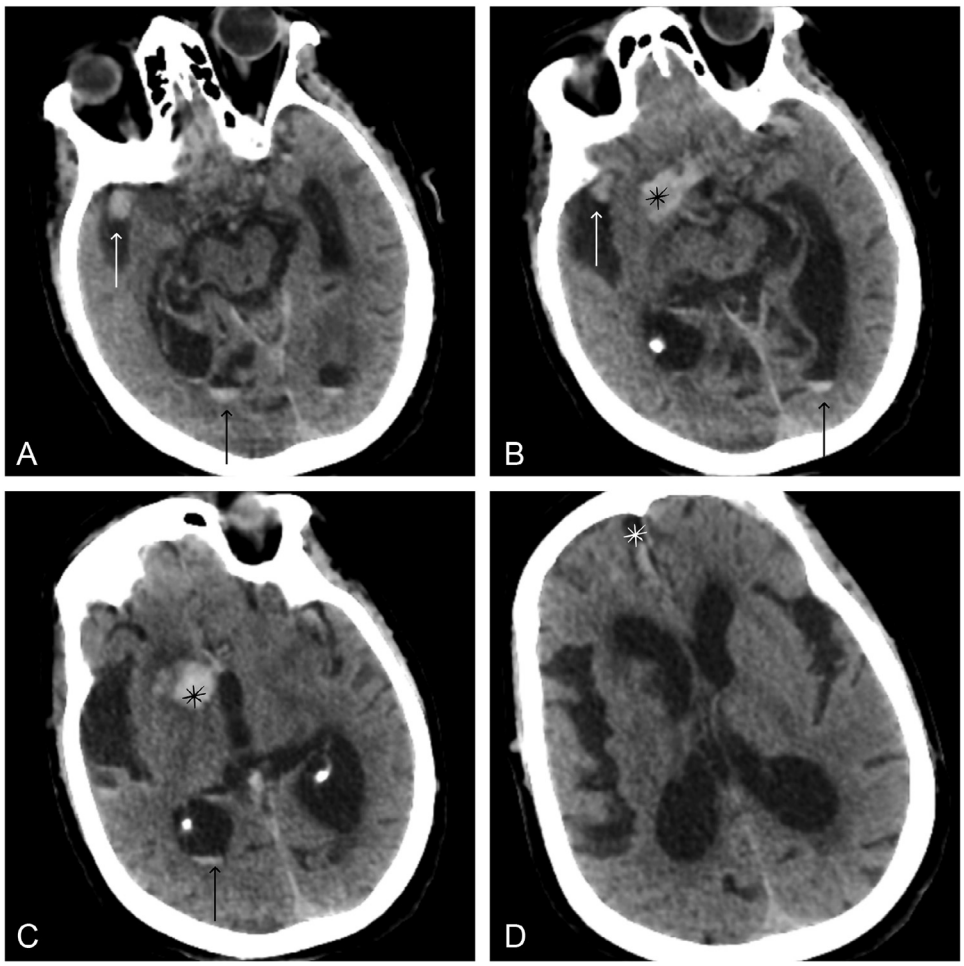

Fig. 1 – Axial images of a nonenhanced cerebral CT scan showing subarachnoid hemorrhage in the sylvian (white arrows) and interhemispheric fissures (white asterisk) with intracranial hematoma (black asterisk) and intraventricular hemorrhage in the occipital horns (black arrows).

prompting the performance of an external ventricular drain under general anesthesia.

Postoperatively, cerebral digital subtraction angiography (Fig. 2) with 3D rotational acquisition (Fig. 3) demonstrated the absence of the M1 segment, replaced by a plexiform arterial network between the internal carotid and the sylvian bifurcation. Further analysis of the 3D acquisition identified an aneurysm originating from the arterial twig. There was no evidence of intracranial arteriosclerotic degeneration, transdural anastomosis, or other vascular anomalies. The diagnosis of twig-like MCA with an associated aneurysm explaining the hemorrhage was established.

Discussion

Anatomic variations of the MCA are relatively rare, with duplication and fenestration being the most frequent, with estimated prevalences of 0.2%-2.9% and 1%, respectively [4]. Twig-like MCA is an uncommon vascular variation, predominantly reported in case reports [5], with a prevalence varying between 0.1% and 1% in the literature [6]. Twig-like MCA consists of a plexiform network of small vessels that replace the M1 segment, sparing the ICA terminus and the anterior cerebral artery [6]. Its pathogenesis is still not fully understood, although it might be caused by an evolutionary arrest of the developing MCA in its fetal form due to an unknown underlying mechanism or fetal vascular insult [5,7]. According to Akkan et al. [8], the extent of the twig-like appearance depends mainly on how early the interruption of MCA development occurred. This can lead to 3 different presentations: a plexiform network replacing the entire MCA, a plexiform network terminating at the bifurcation, or a plexiform network ending before the bifurcation begins. In our case, the plexiform network ended before the bifurcation.

Twig-like MCA may be observed on cross-sectional imaging [6], although it is often misdiagnosed on these modalities as arteriovenous malformation or pseudo-occlusion of the MCA [7]. Therefore, cerebral angiography with 3D acquisition should be recommended whenever unilateral steno-occlusive MCA is found on MRA or CTA [2]. In our patient, the diagnosis was confirmed by cerebral angiography.

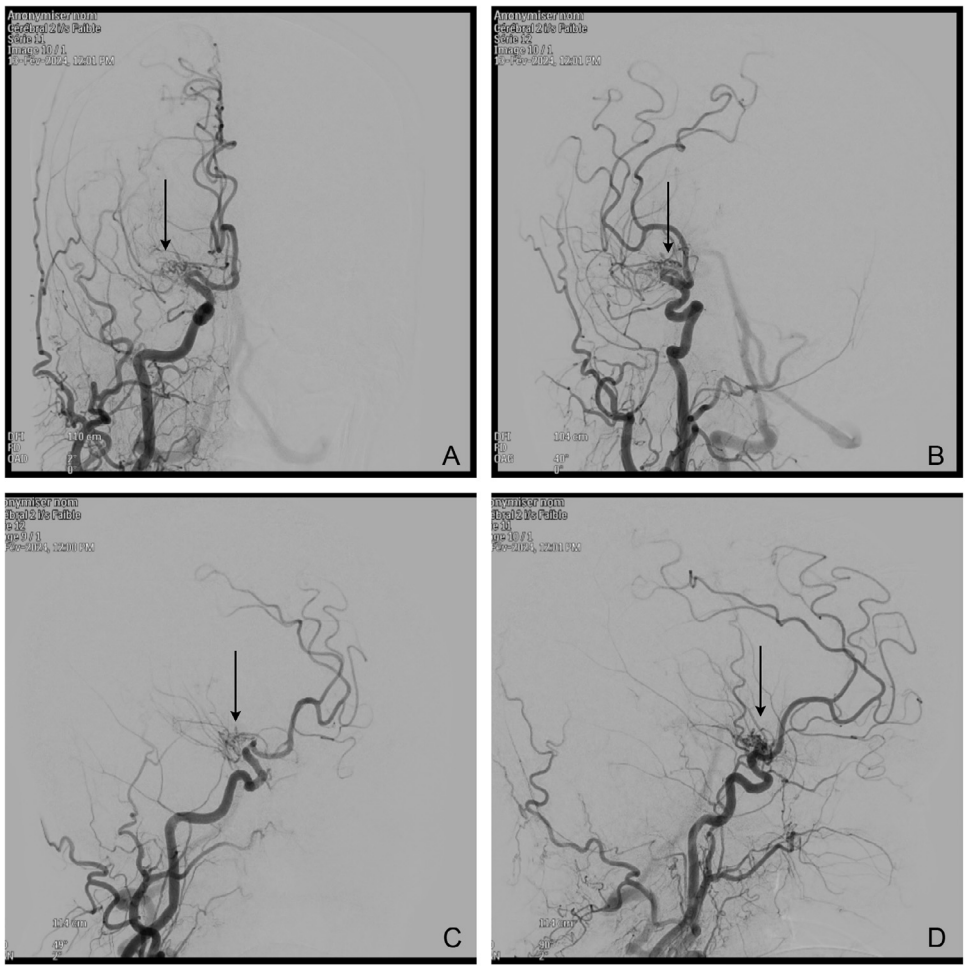

Fig. 2 – Cerebral angiographic images showing Twig like MCA : Posteroanterior (A), oblique (B) and lateral (C, D) projections of the right common carotid artery showing the absence of the M1 segment and its replacement with a plexiform network compatible with twig like MCA (arrows). Note the absence of transdural anastomosis nor intracranial atherosclerotic stenosis.

Unfused or twig-like MCA can be incidentally found on cerebral angiography performed for other reasons, but it is often diagnosed when symptomatic, as infarction in the MCA territory can be found in up to 33.3% of cases, according to the largest series in the literature [9]. These infarcts are often lacunar and focal. Twig-like MCA can also present with intracranial hemorrhage. Indeed, hemorrhagic events have been reported in 54% of patients according to the literature and previous reports [5]. Subarachnoid, intracranial, or intraventricular hemorrhage have all been reported [2]. The mechanism of hemorrhage may be due to rupture of collateral vessels, including the plexiform arterial network, or flow-related aneurysms [2]. Aneurysm formation is also a known complication of twig-like MCA since its first report in 2005 [3]. Aneurysms may be found at the level of the twigs or proximal to the MCA, most frequently at the anterior communicating artery, suggesting they form due to hemodynamic stress [2,3,9].

To the best of our knowledge, there are 43 reported cases in the international literature of aneurysms associated with twig-like MCA [7,10]. The main differential diagnoses of twig-like MCA are Moyamoya disease and steno-occlusive disorders of the MCA. Several imaging findings may help differentiate twig-like MCA from these conditions. Moyamoya disease is a nonatherosclerotic progressive, usually bilateral, steno-occlusive arteriopathy that affects the ICA terminus and the proximal segments of the MCA and ACA. Atherosclerotic steno-occlusive disease of the MCA is another differential diagnosis to consider, typically seen in older patients with multiple cardiovascular risk factors. It is usually multifocal, involving both extracranial and intracranial circulations, whereas twig-like MCA is usually unilateral, involving a single MCA, and is more often diagnosed at a younger age [6].

Due to its low prevalence and limited data in the literature, there are no consensus guidelines on the management of twig-like MCA. Conservative management with regular imaging follow-up is recommended when asymptomatic. ECA to MCA bypass and encephaloduroarterial synangiosis can be successful strategies to enhance vascular supply in cases of is-

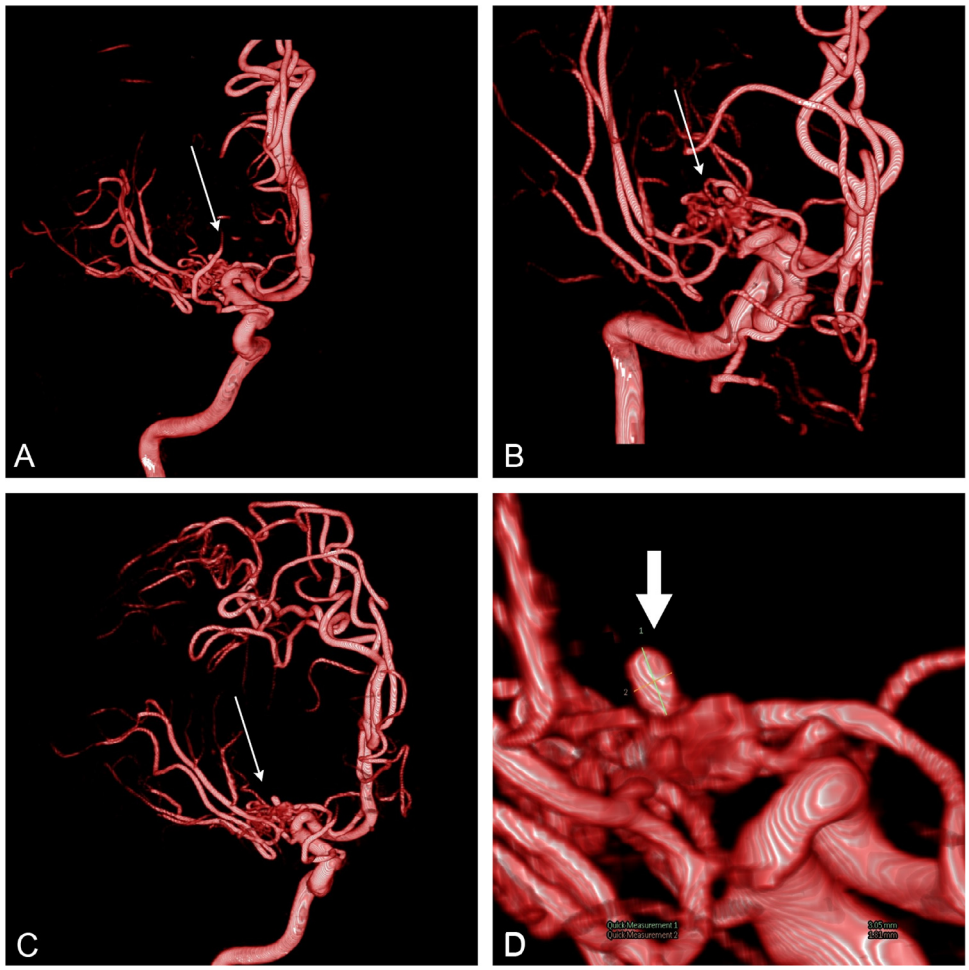

Fig. 3 – 3D acquisition images of the right ICA: 3d images (A, B And C) of the right ICA showing the absence of the M1 segment and its replacement with a plexiform network compatible with twig like MCA (thin arrows). Note the presence of an aneurysm in the plexiform network (thick arrow in d).

chemic events. Endovascular management is limited to treating associated aneurysms [6].

Patient consent

Written informed consent for publication has been obtained from the patient's legal representative.

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