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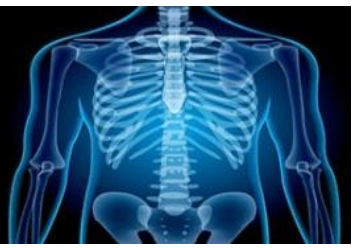

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An intriguing unique presentation of middle cerebral artery dolichoectasia with its fusiform aneurysm: A case report

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Abstract
Cerebral dolichoectasia is an uncommon angiopathic disorder, which signifies dilatation, elongation, and tortuosity of intracranial arteries, commonly involving the vertebrobasilar arterial system with rare involvement of anterior and middle cerebral arteries. As a consequence of the weakening of vessel walls, altered cerebral hemodynamics, and compression of nearby neural structures, this condition can cause serious clinical symptoms and complications. The majority are isolated cases and are related to various risk factors such as increasing age, metabolic disorders, chronic hypertension, and hereditary diseases affecting the vascular interstitial framework. Computed tomography cerebral angiography is sensitive in the diagnosis and cerebral digital subtraction angiography is considered standard for diagnosing dolichoectasia.
We report a rare case of a 55-year-old male patient who was admitted with a history of trauma, with loss of consciousness on presentation. CT brain angiography was performed which was suggestive of dolichoectasia with fusiform aneurysm of the middle cerebral artery, which was confirmed on digital subtraction angiography.

Keywords: Middle cerebral artery, dolichoectasia, fusiform aneurysm, atherosclerosis, dilatation

Introduction
Cerebral Dolichoectasia is an uncommon cerebral arterial vessel disorder, characterized by dilatation, elongation, and tortuosity of arteries. In the intracranial arterial system, this disorder is more centrally oriented, with common involvement of arteries of the vertebrobasilar system, followed by the carotid arteries, with rare involvement of the peripherally located anterior and middle cerebral arteries [1]. Fusiform and dolichoectatic aneurysms are often asymptomatic and incidentally detected on CT for other reasons, however, they may present with ischemic strokes, intracranial hemorrhage, or symptoms related to compression of brain structures. Though usually asymptomatic, this disorder can demonstrate varied features ranging from symptoms due to compression of adjacent brain structures to vascular phenomena including transient ischemic attacks, stroke, or bleeding [2]. The prevalence of intracranial dolichoectasia in the general population is 0.06 to 5.8% [1].
We report a rare case of dolichoectasia with a long-segment fusiform aneurysm of the middle cerebral artery with concealed rupture and thrombosis based on prominent radiological findings of Computed tomography angiography (CTA) and digital subtraction angiography (DSA).

Case
A 55-year-old man was brought to the emergency and accident unit with an alleged history of a road traffic accident, while under the influence of alcohol. After the incident, he experienced few vomiting episodes, followed by loss of consciousness. There was no documentation indicating whether the patient experienced an episode of seizure or not. The patient was vitally stable, maintaining saturation with a Glasgow Coma Scale (GCS) score of 15 on admission. The patient was a known case of Diabetes mellitus. No other relevant medical or family history was available. The patient had no past surgical history. On external examination, he had few contused lacerated wounds over the lower lip, with a loss of part of the lower lip and the root of the nose. He also had abrasions below the left eye and lateral to the right lateral canthus.

The patient was subjected to an emergency computed tomography, non-contrast, and contrast with angiography of the head with injection of the contrast bolus, with the range from the arch of aorta to the cranial vault, with a layer thickness of 1 mm.

A well-defined heterogeneously hyperdense peripherally enhancing area around the dilated right MCA in the right frontotemporal and ganglio-capsular region was visible on CT brain imaging non-contrast phase (Figure 1) that represents a subacute bleed following a concealed rupture. The angiographic phase revealed a fusiform dilatation and elongation of the MCA with tortuosity along its entire course (Figures 2 and 3). An additional vessel is seen coursing inferior to the hyperdense area, likely designating the dilatation of a branch of the dolichoectatic MCA (Figure 4). The carotid arteries, vertebral arteries, and the rest of the intracranial arteries were normal. Areas of subarachnoid bleed or ischemic regions were not visualized on CT.

The above-mentioned findings were confirmed by cerebral digital subtraction angiography (DSA), which revealed a dysplastic right MCA with a giant fusiform aneurysm with partial thrombosis (Figure 5). There was no contrast leak from the aneurysm into the adjacent peri-aneurysmal heterogeneously hyperdense area as observed on CTA.

Intracerebral hemorrhage was treated with IV mannitol, and prophylactic antibiotics were started due to suspicion of aspiration. The patient was intubated due to persistent hypoxemia with an oxygen saturation below 90% on high-flow O_2. After being intubated, the patient was placed on 100% FiO2 and given a Positive End-expiratory Pressure (PEEP) initial dose of 6 cm H_2O, which was subsequently increased to 10 cm H_2O. However, the patient succumbed to Acute respiratory distress syndrome (ARDS) precipitated by aspiration pneumonitis.

Figure

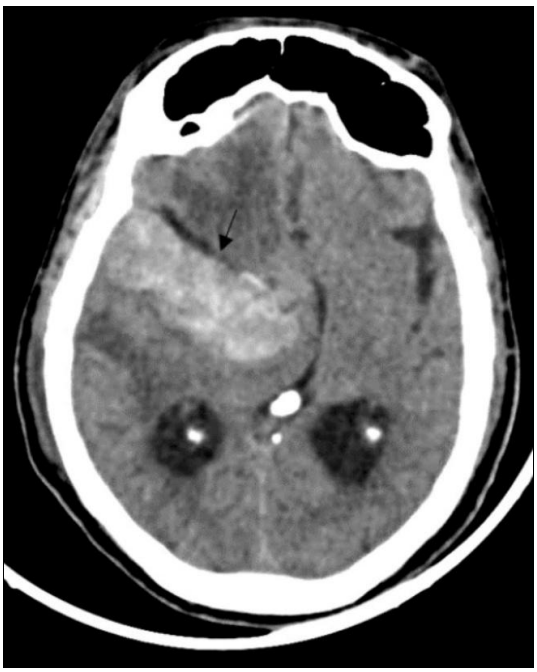

Fig 1: Axial non-contrast computed tomography of the brain shows a well-defined hyperdense area (marked by a black arrow) representing bleed in the right temporal and ganglio-capsular region with effacement of the right lateral ventricle and midline shift to the left

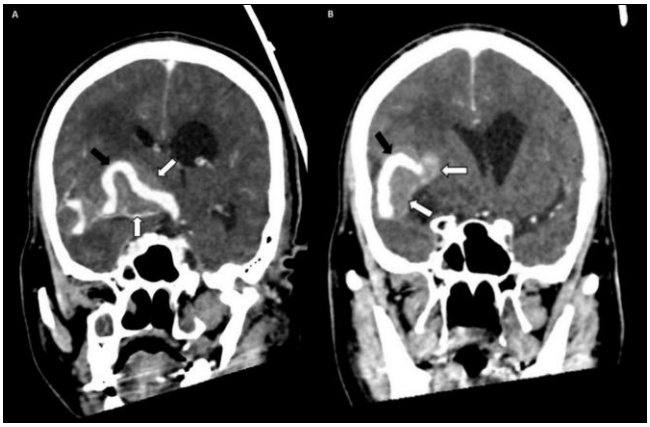

Fig 2: Images A and B are coronal MPR reconstructed sections of CTA showing dilatation and tortuosity of the right Middle cerebral artery (as marked by black block arrow) along its entire course, with surrounding hyperdense peripherally enhancing area suggestive of bleed (as marked by white block arrow).

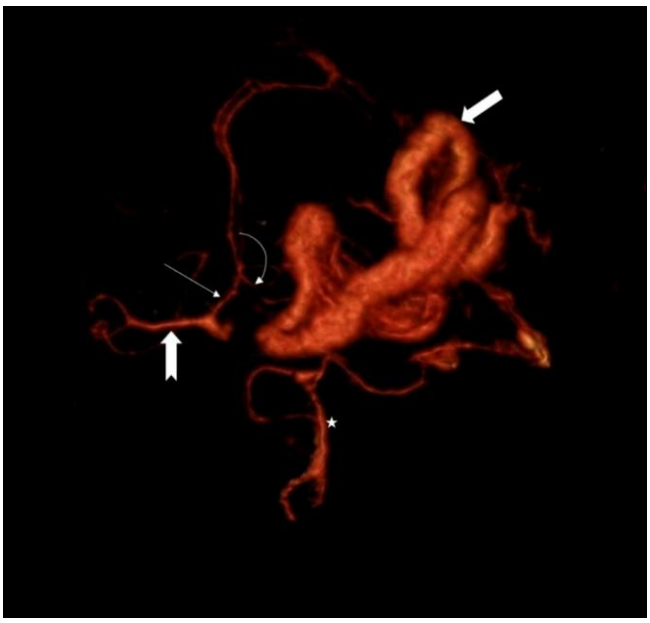

Fig 3: 3D CT volume rendered image with bone subtraction of the Circle of Willis showing a dilated tortuous right middle cerebral artery (marked by white block arrow). Other annotated structures include the left middle cerebral artery (marked by a white notched arrow), right anterior cerebral artery (marked by a white curved arrow), left anterior cerebral artery (marked by a white line arrow), and basilar artery (marked by a white star).

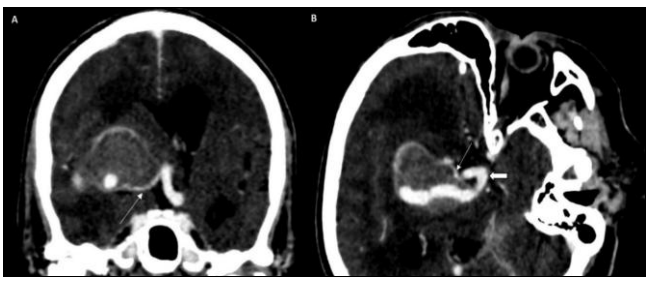

Fig 4: Coronal MPR reconstructed image (Image A) of the CT brain of angiographic phase showing another vessel (marked by white line arrow) inferior to the hyperdense area, likely a dilated branch of dolichoectatic MCA. An axial curved planar reformation (CPR) CT angiography phase (Image B) for better visualization of dilated MCA (marked by white block arrow) and dilated branch of MCA (marked by white line arrow) arising from dolichoectatic MCA.

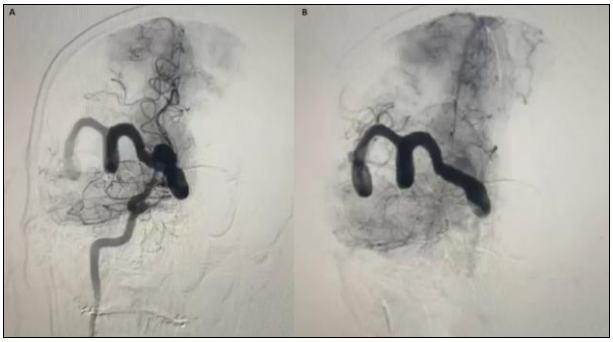

Fig 5: Coronal sections of cerebral digital subtraction angiography (Image A and B) show dilated and dysplastic right MCA with its fusiform aneurysm

Discussion

The rare angiopathic condition known as Cerebral dolichoectasia is defined by dilatation, tortuosity, and elongation of intracranial arteries [1]. The term "Intracranial arterial dolichoectasia" originates from the Greek language, where the word 'dolichoectasia' signifies both dilatation and elongation of the intracranial arteries [3]. Various terms, including mega-artery, cirsoid aneurysm, fusiform aneurysm, serpentiform aneurysm, mega dolicho-artery, and dilatative arteriopathy have been used to represent this elongation and dilatation of cerebral arteries [4]. This condition is associated with risk factors such as advanced age group, metabolic disorders, chronic hypertension, and hereditary diseases affecting the vascular interstitial structure formation [2]. It has a male preponderance [5]. A range of various hereditary disorders, such as Marfan's syndrome, Ehlers-Danlos syndrome type IV, neurofibromatosis type 1, tuberous sclerosis, pseudoxanthoma elasticum, Pompe's disease, Fabry's disease, autosomal dominant polycystic kidney disease, and acquired immunodeficiency syndrome, have been linked to this condition [1]. A fusiform aneurysm is a type of non-saccular arterial dilatation characterized by circumferential ballooning of the vessel, which overlaps with the definition of dolichoectasia. However, along with arterial dilatation, the latter also incorporates elongation and tortuosity (dolichosis) of the artery [4].

Atherosclerosis is the original hypothesis for the pathophysiology of intracranial dolichoectasia, however, many investigators revealed no clinical significance of atherosclerosis, rather it can be superimposed on the ectatic arterial wall [6]. Out of the proposed pathophysiological processes, the widely accepted one suggests abnormal vessel remodeling and anomalous deposition of connective tissue within the vessel wall, attributed to the disparity between matrix metalloproteinases and antiprotease activity in the connective tissue [6]. In this instance, atherosclerosis abetted by underlying diabetes mellitus is presumed to be the cause of MCA dolichoectasia.

The prevalence of intracranial dolichoectasia in the general population is 0.06 to 5.8%, commonly involving the vertebrobasilar arterial system or distal internal carotid arteries, rarely affecting the middle cerebral artery and anterior cerebral artery, with only a few documented case reports of MCA dolichoectasia [1]. Because of this, to determine dolichosis in the anterior and middle cerebral artery, Gutierrez et. al. have suggested a visual evaluation method that relies on the assessment of compression of surrounding structures and relative tortuosity of the vessel as compared to the contralateral artery [4]. In 2007, Passero and Rossi suggested threshold values for the diameter of vessels other than the basilar artery to demonstrate arterial ectasia. The proposed cutoff values include ≥ 7 mm for the internal carotid artery, ≥ 4 mm for the middle cerebral artery, and ≥ 4 mm for the vertebral artery [2]. The MCA in our patient was dilated with a diameter measuring 9 mm along its entire length.

Though asymptomatic, patients may present with a variety of symptoms due to compression of adjacent brain structures, such as headache, pulsatile tinnitus, hemiplegia, vertigo, visual symptoms, with signs and symptoms related to cranial nerve palsies, like facial weakness, etc. [5]. Hemodynamic complications include TIA or stroke and intracranial hemorrhage. Hydrocephalus is a rare complication of this disorder.

For the diagnosis, CT cerebral angiography is sensitive as it provides a high definition of vessel morphology. Other option includes MR imaging and time-of-flight (TOF) MR angiography. Though invasive, digital subtraction angiography (DSA) is considered standard for diagnosis as it provides real-time information about luminal flow and assists in further potential therapeutic intervention [6].

In the GENIC study involving 510 patients, the authors demonstrated that "small vessel disease", characterized by features such as multi-lacunar infarcts, is notably more prevalent in patients with intracranial dolichoectasia [7]. In 2010, an association between dolichoectasia of intracranial arteries, coronary ectasia, and abdominal aortic aneurysm was proposed by Pico *et al*., for which a prophylactic cardiological examination and abdominal sonography are recommended in patients with intracranial dolichoectasia [7]. Currently, there is no available specific treatment for the prevention of arterial dilatation and tortuosity [8]. Numerous surgical procedures have been advocated in the treatment of intracranial dolichoectasia, including clip reconstruction, wrapping, distal bypass, aneurysmorrhaphy with thrombectomy as well as resection, and end-to-end anastomosis [1, 8]. However, the dolichoectatic MCA often presents significant challenges for revascularization, and any surgical intervention could potentially cause cerebrovascular accidents, and thus is frequently avoided [1].

Conclusion

Intracranial dolichoectasia is not an uncommon angiopathy, but dolichoectasia of the middle cerebral artery is rare, which shows a male preponderance and is seen to occur at any age. Though usually asymptomatic, the clinical features might resemble symptoms of cerebrovascular accidents or those because of brain lesions. Therefore, a high degree of clinical suspicion, detailed history, and radiological imaging are a must for prompt diagnosis. Many surgical procedures have been reported for the treatment; however, MCA involvement poses a surgical difficulty due to the risk of brain ischemia. Follow-up imaging is highly recommended to track the progress of the disease.

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Conflict of interest
The authors have no conflict of interest.

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