



Case Report

Dural arteriovenous fistula mimicking temporal arteritis

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ABSTRACT

Introduction: Dural arteriovenous fistula (dAVF) is a very rare disease characterized by an abnormal vascular communication between arteries and veins in dural mater. It frequently presents with intracranial haemorrhage. Common presenting symptoms are headache and seizures.

Case Report: Here we report a case of dAVF in which the patient's symptoms mimic a temporal arteritis in a 23-year-old woman. She presented with painful mass at forehead for 9 months with frontotemporal headache. Magnetic resonance imaging demonstrated dural arteriovenous fistula.

Conclusion: Since both diseases have different prognosis but similar presentation, it is important to ensure that there is no dural arteriovenous fistula in patient with suspected temporal arteritis.

1. Introduction

Dural arteriovenous fistulas (dAVFs) are rare vascular condition that can occur anywhere in intracranial dural mater. It involves communication between dural arteries and veins or venous sinus. There are various areas in dura that may harbour dAVFs leading to a wide spectrum of intracranial and extracranial presenting symptoms. Some dAVFs may lead to fatal intracranial haemorrhage. In view of the high rate of intracranial haemorrhage as clinical presentation, it is important to recognize the early symptoms and signs of dAVF; to minimize the complication. However, the disease can also remain to be completely silent.

2. Case report

23 years old single lady presented with painful serpentine-like mass at right forehead since past 9 months. The mass gradually showed increment in size. The mass has never bled or ulcerated before. She never had any pre-existing skin lesion or infection at the face or head region.

She had daily right sided fronto-temporal headache. There was no seizure, blurry vision, vomiting, epistaxis, nasal blockage, reduced hearing, tinnitus or facial weakness. She had no previous head trauma

and was pre-morbidly well. She had no prior usage of hormonal contraceptive.

Clinical examination showed prominent vessels at right forehead extending to right temple which covered a 4 cm x 5 cm right supraorbital area. The vessels were soft, pulsating and compressible. It showed an immediate vessel refilling. It was tender on gentle palpation (Fig. 1). A bony indentation measuring 0.5 cm x 0.5 cm was felt at the right forehead.

There was no facial palsy. Her vision was equally 20/20. Ear, nose and throat examination were unremarkable.

A Computed Tomography (CT) scan showed bony defect at right frontal bone corresponding to the depressed area during clinical examination (Fig. 2).

Cerebral Magnetic Resonance Imaging (MRI) confirmed the diagnosis of right supraorbital dural arteriovenous fistula (dAVF). On MR Arteriogram (MRA), there was irregular flow within the lesion; supplied by right superficial temporal artery and right supraorbital artery. Intraorbital part of the supraorbital artery was convoluted. This artery arose from right meningo-ophthalmic artery which subsequently arose from right middle meningeal artery (MMA). The right MMA were enlarged compared to the left side. All internal carotid and cerebral arteries were normal. (Fig. 3).

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Fig. 1. Prominent and dilated right superficial temporal and supraorbital arteries were shown. There was no overlying skin change.

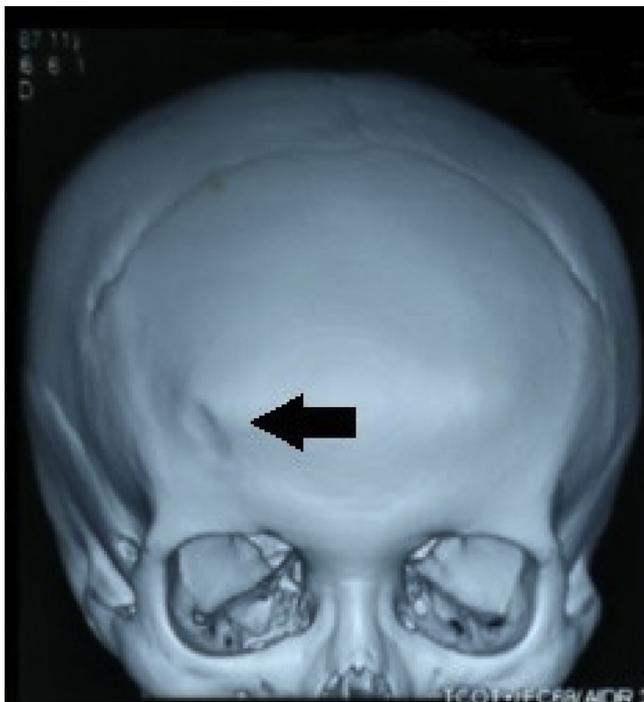


Fig. 2. CT reconstruction demonstrating a bony defect at supraorbital region (arrow pointed at the bony defect).

On MR Venogram (MRV), it predominantly drained via subcutaneous veins and to a lesser extent into right superficial temporal vein. No definite drainage into intracranial vein or orbit. Superior, inferior sagittal, straight, transverse and sigmoid sinuses were patent.

Embolization done to her right dAVF. Post embolization, she had transient right temporal blindness. No infarct seen on CT scan.

She is planned for another embolization in 1-year-time.

3. Discussion

The anatomic description of dural arteriovenous fistula (dAVF) was first described by Rizolli in 1881. The first angiographic description of dAVF was by Sachs in 1931 which he demonstrated direct connection of meningeal arteries and the venous system in the dura. The average age of occurrence at 57 years [1].

dAVF commonly presents with intracranial haemorrhage. Other presentations are headache, seizures or ocular symptoms [1]. Factors that are commonly related to dAVF with intracranial haemorrhage are leptomeningeal drainage, location outside a major venous sinus, variceal dilatation and sinus stenosis [2].

The gold standard for diagnosing dAVF is by catheter angiography [2]. Agid R et al (2009) reported that all anterior cranial fossa dAVF were supplied by ethmoidal branches of ophthalmic arteries and 62% of them has contribution from external carotid arteries which mainly comes from branches of middle meningeal arteries [1].

Treatment modalities are open surgical disconnection, interventional neurology with embolization and radiosurgery. The choice of intervention may depend on several factors such as age, size and location of dAVF, presenting complaints, neurological deficit as well as surgical risks.

Giant cell arteritis (GCA) is a disease affecting commonly seen in people over 50 years of age. It is the most common systemic vasculitis in above 50 year old with incidence of 3.5 per 100,000 per year [3]. The diagnostic criteria for GCA by American College of Rheumatology is presence of 3 out of 5 criterias which are age more than 50 years, new headache, superficial temporal artery tenderness or decreased pulsation, elevated ESR more than 50 mm per hour and abnormal temporal artery biopsy showing necrotizing arteritis, characterized by a predominance of mononuclear cell infiltrates or a granulomatous process with multinucleated giant cells. The condition is the commonest form of vasculitis caused by inflammation to medium and large vessel, mainly affecting the head and neck. It is also known as temporal arteritis as the disease usually affects the temple. The presenting complaints include headache, scalp tenderness and visual loss. It should be treated early in order to minimize complications such as blindness, stroke and aneurysm. GCA is mainly treated with corticosteroids (40–60 mg of Prednisone, typically given for up to two years. Immunosuppressive drugs such as Methotrexate and Azathioprine are use in chronic cases or in those affected by steroid side effects [4].

Another differential diagnosis that should be taken into consideration is cerebral venous sinus thrombosis (CVST) affecting commonly female with mean age of 35 years. Headache is the first and commonest symptom of CVST; followed by seizures, paresis, papilloedema and change in mental status. Usage of oral contraceptive increased the risk to develop CVST. CVST is treated by anticoagulant and endovascular treatment with thrombolytic agent or via mechanical thrombectomy. [5]

In this case, her atypical presentation of dAVF in which she presented with painful superficial temporal and supraorbital arteries rather than the commoner intracranial haemorrhage may create a confusion in diagnosis. Her clinical presentation and clinical findings are similar to

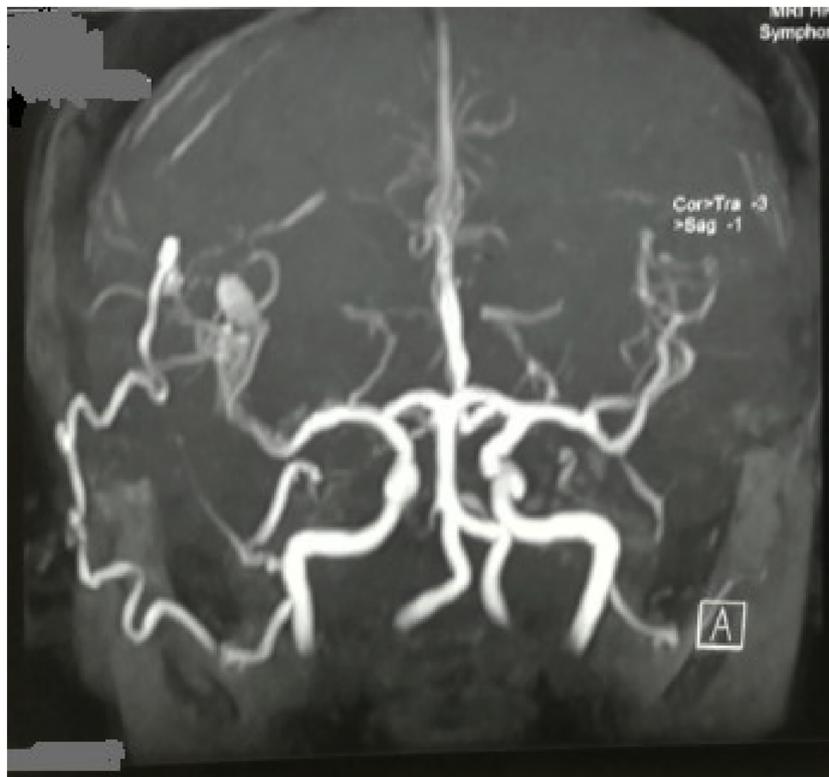


Fig. 3. MR Arteriogram showed dilated and tortuous right superficial temporal and supraorbital arteries.

GCA, but the finding of depressed frontal bony skull make the diagnosis unlikely. Therefore, we highlighted the importance in recognizing subtle clinical findings that may assist in diagnosing dAVF; a rare disease with fatal complications.

4. Conclusion

Early identification of dAVF is important to prevent further complication and minimize mortality as dAVF is itself a treatable condition. Recognizing subtle signs may lead to diagnosis and its appropriate management. This eventually leads to better outcome.

Conflict of interest

None.

Acknowledgement

Nil.

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