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Isolated painful abducens nerve palsy as a presentation of indirect carotid-cavernous fistula

*[1,2]Amirah Mohammad Razali *MD DrOph*, [2]Hsien Rong Tee *MD*, [3,4]Muhammad Zakwan Yahya *MD DrRad*, [5]Anna Misyail Abdul Rashid *MD MMed*, [6]Wan Hazabbah Wan Hitam *MD MMed*

[1]Department of Ophthalmology, Faculty of Medicine and Health Sciences, Universiti Putra Malaysia; [2]Department of Ophthalmology, Hospital Sultan Abdul Aziz Shah, Universiti Putra Malaysia; [3]Putra Clinical Centre of Neurovascular & Stroke (Putra-CNS), Hospital Sultan Abdul Aziz Shah, Universiti Putra Malaysia; [4]Department of Radiology, Kulliyyah of Medicine, International Islamic University of Malaysia; [5]Department of Neurology, Faculty of Medicine and Health Sciences, Universiti Putra Malaysia; [6]Department of Ophthalmology and Visual Science, School of Medical Sciences, Universiti Sains Malaysia, Health Campus, Kubang Kerian, Kelantan

Abstract

Indirect carotid-cavernous fistula (CCF) is a low-flow, vascular condition that may present with subtle and easily overlooked clinical features, sometimes resembling more common diagnosis such as microvascular neuropathy. We describe a 40-year-old man with poorly controlled diabetes and an active smoker, who presented with progressive left retrobulbar pain and binocular horizontal diplopia. Examination revealed an isolated left eye abduction limitation with the presence of mild corkscrew conjunctival vessels on the affected eye. Magnetic resonance imaging and angiography demonstrated bilateral distal internal carotid artery stenosis without an aneurysm. Digital subtraction angiography confirmed a left indirect CCF. The patient experienced early symptomatic improvement after angiography and was managed conservatively with near-complete resolution. This case highlights the importance of recognising subtle clinical signs and considering vascular causes in patients presenting with atypical cranial nerve palsies.

Keywords: Indirect carotid-cavernous fistula, painful ophthalmoplegia, abducens nerve palsy

INTRODUCTION

Carotid-cavernous fistula (CCF) refers to abnormal communication between the carotid arterial system and the cavernous sinus, leading to altered venous drainage and varying degrees of orbital congestion. Based on the Barrow classification, CCF can be divided into direct (Type A) and indirect (Type B-D) fistulas. Indirect CCFs are low-flow dural shunts supplied by meningeal branches of the internal or external carotid arteries and typically present more gradually.[1]

In contrast to direct CCFs, which are commonly associated with trauma and more striking ocular signs, indirect CCF may manifest with more subtle and less specific signs. Patients may have mild conjunctival injection, mild proptosis, intermittent diplopia or isolated cranial nerve palsies. The abducens nerve is particularly susceptible due to its location within the cavernous sinus.[2] Diagnosis may be challenging in the absence of classical features such as pulsatile proptosis, ocular bruit and chemosis. Patients with vascular risk factors such as diabetes, hypertension and smoking may be predisposed to spontaneous dural arteriovenous fistula formation.[3]

In everyday clinical practice, isolated abducens nerve palsy in patients with vascular risk factors is frequently attributed to a microvascular cause. However, this assumption may occasionally lead to missed or delayed diagnosis of alternative pathologies.

We describe a case of indirect CCF presenting primarily with isolated painful abducens nerve palsy with minimal orbital signs, illustrating

Address correspondence to: Amirah Mohammad Razali, Department of Ophthalmology, Faculty of Medicine and Health Sciences, Universiti Putra Malaysia. Email: amirahmrazali@upm.edu.my

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the importance of clinical assessment and maintaining a broad differential diagnosis.

CASE REPORT

A 40-year-old man with poorly controlled diabetes mellitus and a history of active smoking presented with a four-week history of progressively worsening left retrobulbar pain associated with binocular horizontal diplopia. He denied any history of trauma, headache or other neurological symptoms. On examination, visual acuity was 6/6 bilaterally with no relative afferent pupillary defect. Ocular motility showed limited abduction in the left eye with preservation of other extraocular movements. There was no proptosis. Anterior segment examination of the left eye revealed corkscrew conjunctival vessels (Figure 1), while the right eye appeared normal. Intraocular pressure was normal bilaterally. Fundus examination demonstrated mild bilateral non-proliferative diabetic retinopathy with no dilated or tortuous veins (Figure 2). HESS chart assessment showed approximately 15-degree underaction of the left lateral rectus muscle, in keeping with a left abducens nerve palsy (Figure 3). Blood investigations showed poor glycemic control (HbA1C 11.5%) together with elevated hemoglobin (18g/dL) and hematocrit (55.2%). Lipid levels were also raised (cholesterol 7.2 mmol/L). Other investigations, including inflammatory markers and infective screening, were unremarkable. Magnetic resonance imaging (MRI) and magnetic resonance angiography (MRA) of the brain showed bilateral internal carotid artery stenosis at the ophthalmic segment without evidence of aneurysm, vascular malformation or mass lesion. The size of bilateral ophthalmic veins was also within the normal limit and not dilated (Figure 4). Given the clinical suspicion of a vascular abnormality, digital subtraction angiography (DSA) was performed. This confirmed a left indirect carotid-cavernous fistula (Barrow type B), supplied by the meningeal branches of the internal carotid artery with drainage into the bilateral inferior petrosal sinuses (Figure 5).

Interestingly, the patient reported noticeable improvement in retrobulbar pain shortly after the angiographic procedure. In view of the mild symptoms and early improvement, a conservative approach was adopted. On follow-up, the pain resolved completely, with only minimal residual diplopia on left gaze.

DISCUSSION

Indirect CCFs are often difficult to recognise, particularly when classical features are absent. Compared to direct, high-flow fistulas, they often follow a more gradual course and may present with only subtle clinical signs.[2] As a result, the diagnosis may be overlooked, especially in patients with co-existing vascular risk factors. The

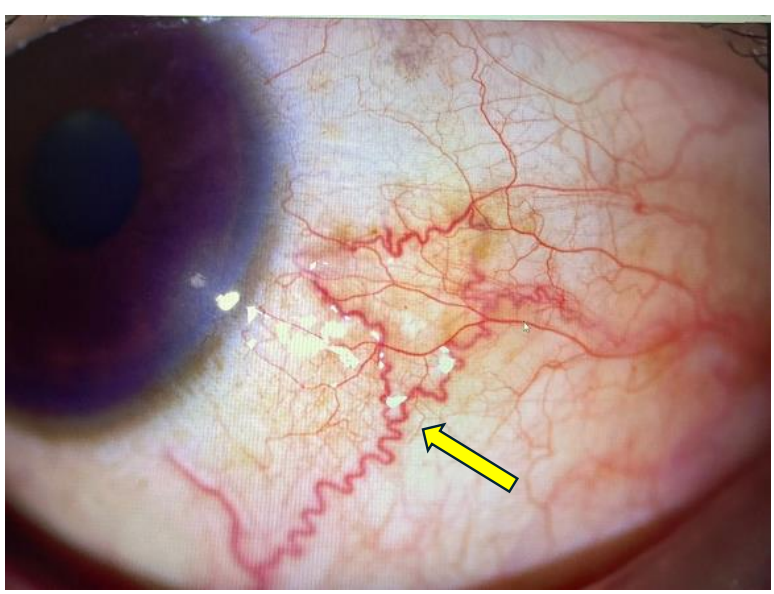

Figure 1. Anterior segment photo showing corkscrew conjunctival vessels (yellow arrow)

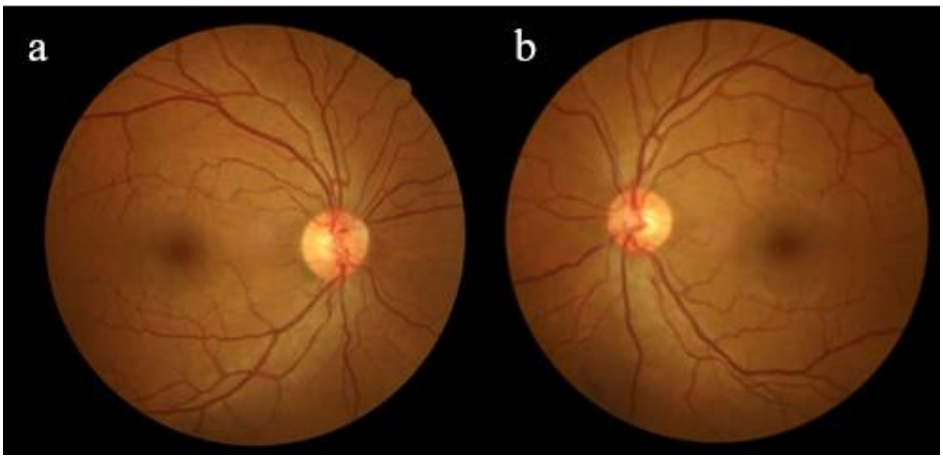

Figure 2. Fundus photographs of the right eye (a) and left eye (b) showing normal optic discs with no evidence of retinal venous dilation or tortuosity to suggest venous congestion.

abducens nerve is particularly vulnerable within the cavernous sinus due to its central location and proximity to the internal carotid artery. Increased venous pressure within the cavernous sinus can impair nerve function, resulting in isolated lateral rectus weakness. In patients with diabetes, such presentations may be misattributed to microvascular cranial neuropathy, which may delay further investigation.

In this case, the presence of retrobulbar pain and corkscrew conjunctival vessels provided important clinical clues. Episcleral venous congestion, even when subtle, should prompt consideration of an underlying vascular abnormality. Our patient had several vascular risk factors, including diabetes, smoking, hyperlipidemia and elevated hematocrit. It is possible that these factors contributed to altered hemodynamics and the development of the fistula. Increased blood viscosity associated with elevated hematocrit may further impair venous drainage and exacerbate cavernous sinus congestion.

Neuroimaging is essential for diagnosis. MRI and MRA may demonstrate signs such as cavernous sinus enlargement or abnormal venous structures, but DSA remains the gold standard

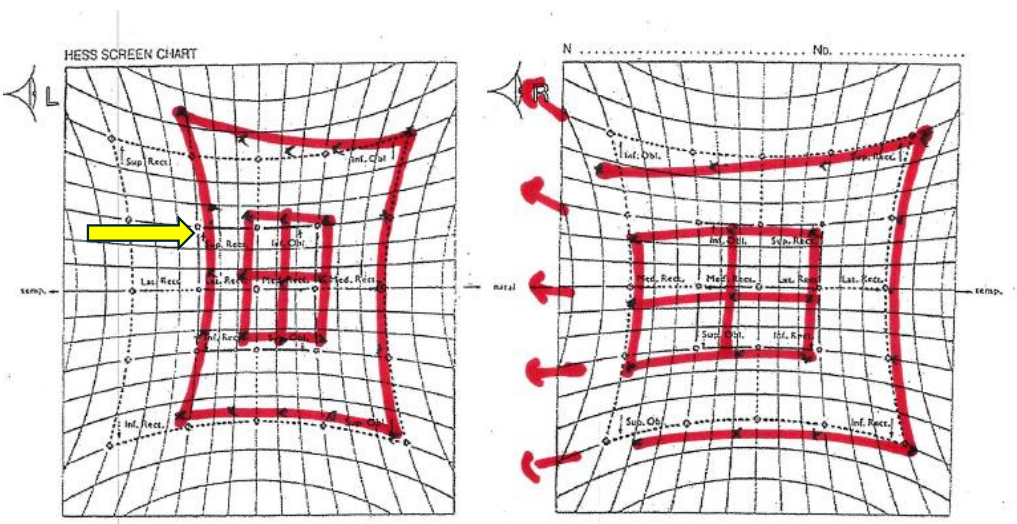

Figure 3. HESS chart shows underaction of the left lateral rectus muscle (yellow arrow) with overaction of the right medial rectus muscle.

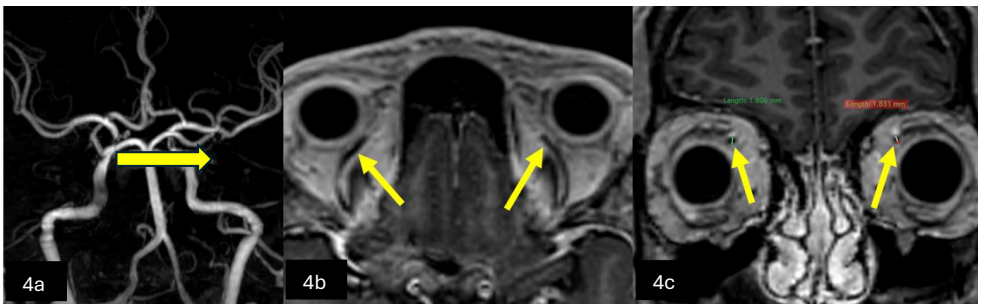

Figure 4a. 3D reconstruction of MRA shows stenosis at the ophthalmic segment of bilateral ICA. Figure 4b & 4c. T1WI post gadolinium image (axial and coronal) of bilateral orbits shows normal size and appearance of the bilateral superior ophthalmic veins (yellow arrows), measuring approximately 1.8mm each.

for definitive diagnosis and classification.[3,4] An interesting feature in this case was early symptomatic improvement following diagnostic angiography. Spontaneous resolution of the CCF following DSA has been described, especially in low-flow shunts seen in indirect-type. Hemodynamic alterations and the effects of contrast media during angiography may promote thrombosis of the fistulous connection, possibly through mechanisms such as transient vasospasm, local vascular irritation or minor endothelial injury.[5]

Management should be individualised based on symptom severity and visual risks. Conservative treatment is appropriate in patients with mild symptoms and stable vision, as spontaneous resolution is possible. Endovascular treatment is generally reserved for patients with progressive symptoms or vision-threatening complications and is highly effective.[6]

Overall, this case serves as a reminder that not all isolated abducens nerve palsies in patients with vascular risk factors are microvascular in origin. Careful clinical evaluation, including

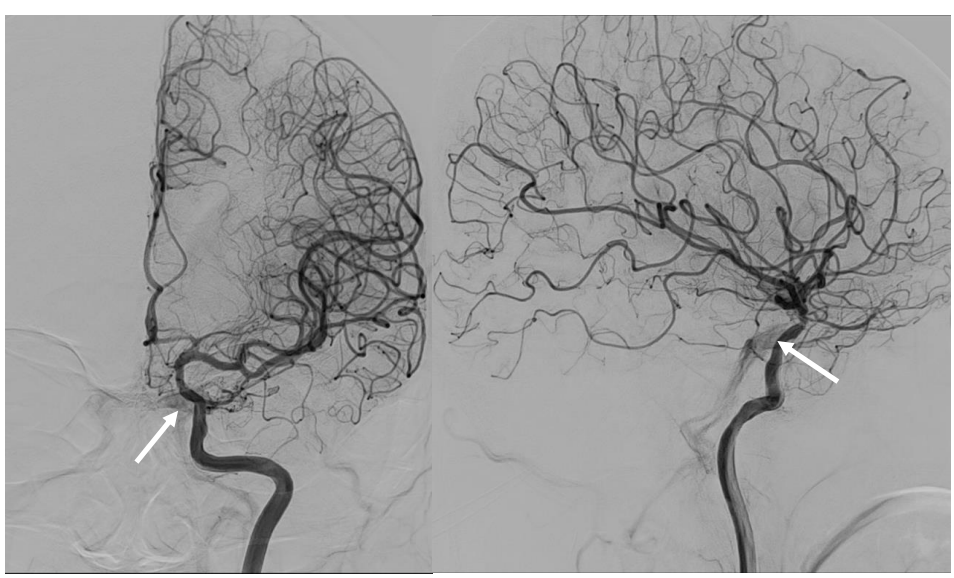

Figure 5. Left internal carotid artery (ICA) angiogram (AP and lateral views) show fistulous communication at the cavernous segment of left ICA and opacification of the cavernous sinus (white arrows) with drainage into the bilateral inferior petrosal sinus. No arterial supply from external carotid artery (ECA).

subtle ocular signs, remains essential.

In conclusion, indirect carotid-cavernous fistula may present in a subtle manner, including as an isolated painful abducens nerve palsy. Recognising their atypical features and considering further vascular imaging can help avoid misdiagnosis and ensure appropriate management.

DISCLOSURE

Ethics: Written informed consent for publication of clinical data and images was obtained from the patient.

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Conflict of Interest: None

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