

## Case report

### Isolated abducens palsy of advanced age: a rare presentation of dural carotid cavernous fistula: a case report



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#### Abstract

Abducens palsy is the most common isolated cranial nerve palsy due to its long peripheral course. Multiple anatomical relationships, particularly within the cavernous sinus and orbita, make the nerve vulnerable. 67 year-old female patient was admitted with worsened headache and lateral gaze restriction of the left eye, which appeared recently. She had no prior history of trauma. Prominent appearance of the left cavernous sinus on cranial magnetic resonance imaging, raised the need of digital subtraction angiography which revealed the presence of bilateral type D dural arteriovenous fistula of cavernous sinuses. Cavernous sinus pathologies, which are usually known to manifest with multiple ocular motor palsies because of the close relationship between 3<sup>rd</sup>, 4<sup>th</sup> and 6<sup>th</sup> nerves inside, might rarely present with isolated abducens palsy. The clinician should pay particular attention to headache in such kind of patients and dural carotid-cavernous fistula should be taken into account, even in the absence of previous trauma history.

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## Introduction

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Abducens palsy is the most common isolated cranial nerve palsy due to its long peripheral course [1]. Multiple anatomical relationships particularly within the cavernous sinus and orbita, make the nerve vulnerable. Hypertension, diabetes, trauma, multiple sclerosis and neoplasms are among the most common accounted etiological factors [2]. Cavernous sinus pathologies, which are usually known to manifest with multiple ocular motor palsies because of the close relationship between 3<sup>rd</sup>, 4<sup>th</sup> and 6<sup>th</sup> nerves inside, might rarely present with isolated abducens palsy [1]. Similarly, carotid-cavernous fistulas (CCFs) are scarce causes of isolated manifestation.

## Patient and observation

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67-year-old woman with one year history of intermittent headache, which was prominent in the left periorbital region, was first evaluated in another hospital and found to have normal Brain CT and MRI scan. She was recommended analgesic and antidepressant treatment. 2 months before admission to our clinic, the headache worsened, became continuous and finally medial deviation of the left eye appeared. This time she was suffering from diplopia. Neurological examination was normal except left abducens palsy. The medical and family history was unremarkable. A comprehensive blood survey (including ESR, CRP, hemogram, serum biochemistry, TFT, vitamin B12, folate and ASO levels) was normal; vasculitic and tumour markers were negative. Transthoracic echocardiogram was normal. An orbita MRI and MR venogram were performed for exclusion of Tolosa Hunt Syndrome; brain MRI was repeated. Orbita MRI and MR venogram were normal. Asymmetric enlargement of the left cavernous sinus and contiguous tubular signal-void regions were detected on the brain MRI. There was asymmetric dural enhancement on the anterior part of the temporal pole, on the middle cranial fossa. Cranial arterial MR angiography (TOF) showed enlargement of cavernous sinuses, more prominent on the left side and homogenous enhancement after contrast administration (Figure 1). The left opthalmic artery and left superior opthalmic vein were increased in caliber when compared to the right side.

Since the findings were not enough to make definite diagnosis, DSA was performed, which revealed the presence of early venous drainage to the cavernous sinuses both from external carotid

arteries and cavernous segments of the internal carotid arteries. The venous drainage was predominantly supplied by superior opthalmic and angular veins. The lesion was angiographically classified as bilateral type D dural arteriovenous fistula of cavernous sinuses (Figure 2). Neurosurgery, Interventional Radiology and Neurology Departments agreed on endovascular embolization therapy. The patient was informed about the management but she disapproved the treatment and was discharged.

## Discussion

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Incidence of abducens palsy is 11/10000. Besides multiple systemic causes mentioned above, the definite etiology cannot be determined in %34 of the patients [2]. CCFs are rare causes of isolated manifestation, and refer to abnormal vascular shunts, allowing blood to flow either directly or indirectly from the carotid artery into the cavernous sinus. They have been classified according to the hemodynamic properties, etiology, or anatomy of the fistula [3]. The classification of Barrow and colleagues depends on the anatomy of the fistula, where direct CCFs connect carotid artery and cavernous sinus directly; and indirect ones allow blood flow from the branch vessels of the carotid artery [4]. According to this classification, our patient had bilateral type D CCF (shunt between cavernous sinus and meningeal branches of both internal and external carotid arteries). The majority of spontaneous CCFs are idiopathic, low-flow in nature and tend to appear in middle-aged women without any prior trauma history [4]. Common symptoms are exophthalmia, proptosis, chemosis, conjunctival injection, secondary glaucoma, headache, tinnitus and/or dylopia. The variability of symptoms and findings depend on the size of the fistula, localisation within the cavernous sinus, rate of flow and drainage pattern, particularly [3,5]. In addition to this, CCFs associated with retrograde cortical venous flow often manifest with neurological symptoms related to venous congestion or infarction [3].

Diagnosis is made by aid of clinical and angiographical findings. Orbital sonography, computed tomography and MRI are non-invasive diagnostic methods, however DSA is gold standard and superior to others for demonstrating the location and drainage pathway of the fistula [3,5]. Spontan occlusion can occur %36 in the course of dural CCFs, however treatment is required for many of the patients [4,5]. Surgical intervention is a choice for treatment,

nonetheless endovascular management including transarterial and transvenous embolization is the first line treatment for most of CCFs resulting in favourable outcomes [3,5].

## Conclusion

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The first target of evaluation in an individual with abducens palsy is correct anatomic localisation of the lesion. Our case with isolated abducens palsy is a rare presentation of CCF. Although it is reasonable to expect multiple ocular motor palsies in the course of cavernous sinus pathologies, isolated abducens palsy might scarcely occur alone. The clinician should pay particular attention to headache in such kind of patients [1] and dural CCF should be taken into account, even in the absence of previous trauma history.

## Competing interests

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The authors declare no competing interests.

## Authors' contributions

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YD, NB and Akyol GA wrote the first draft manuscript and conducted the literature review. OY, IE, OB and AK reviewed the draft manuscript and provided comments. YD finalised the manuscript which was subsequently approved by all authors. All authors have read and agreed to the final manuscript.

## Figures

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**Figure 1:** T1 (a) T2 (b) weighted axial and post contrast T1 (c) weighted coronal images show enlargement of both cavernous

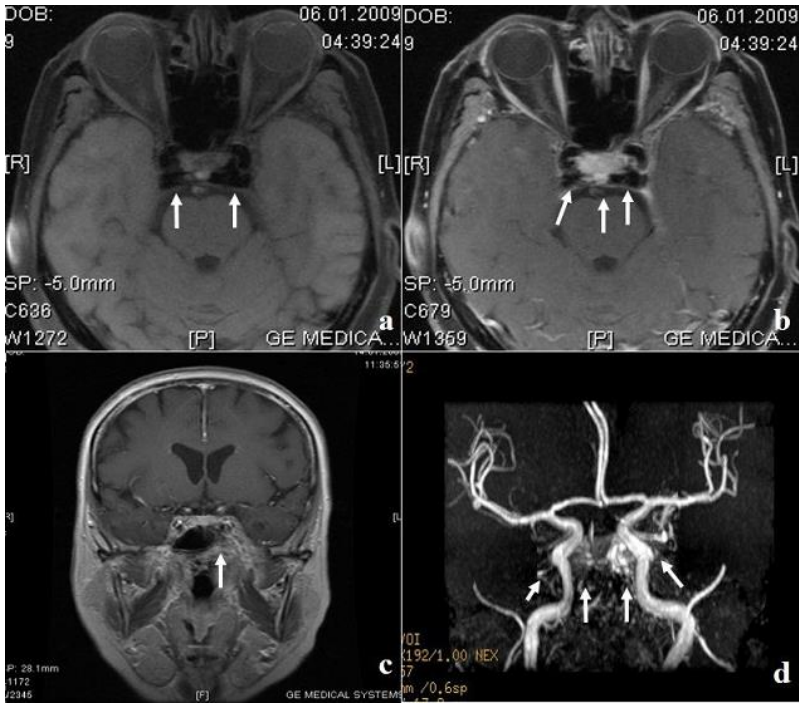
sinuses which is more prominent on the left side, which show flow voids at the posteromedial aspect and homogenous contrast enhancement; cranial arterial MR Angiography (TOF); (d) demonstrates flow-related enhancement in both cavernous sinuses, particularly on the left side (arrows)

**Figure 2:** Conventional angiography (DSA) shows early venous drainage from cavernous segments of the left and right internal carotid arteries into both cavernous sinuses (postero-anterior view of left internal carotid injection (a,b) and oblique view of right internal carotid artery injection (c,d) respectively)

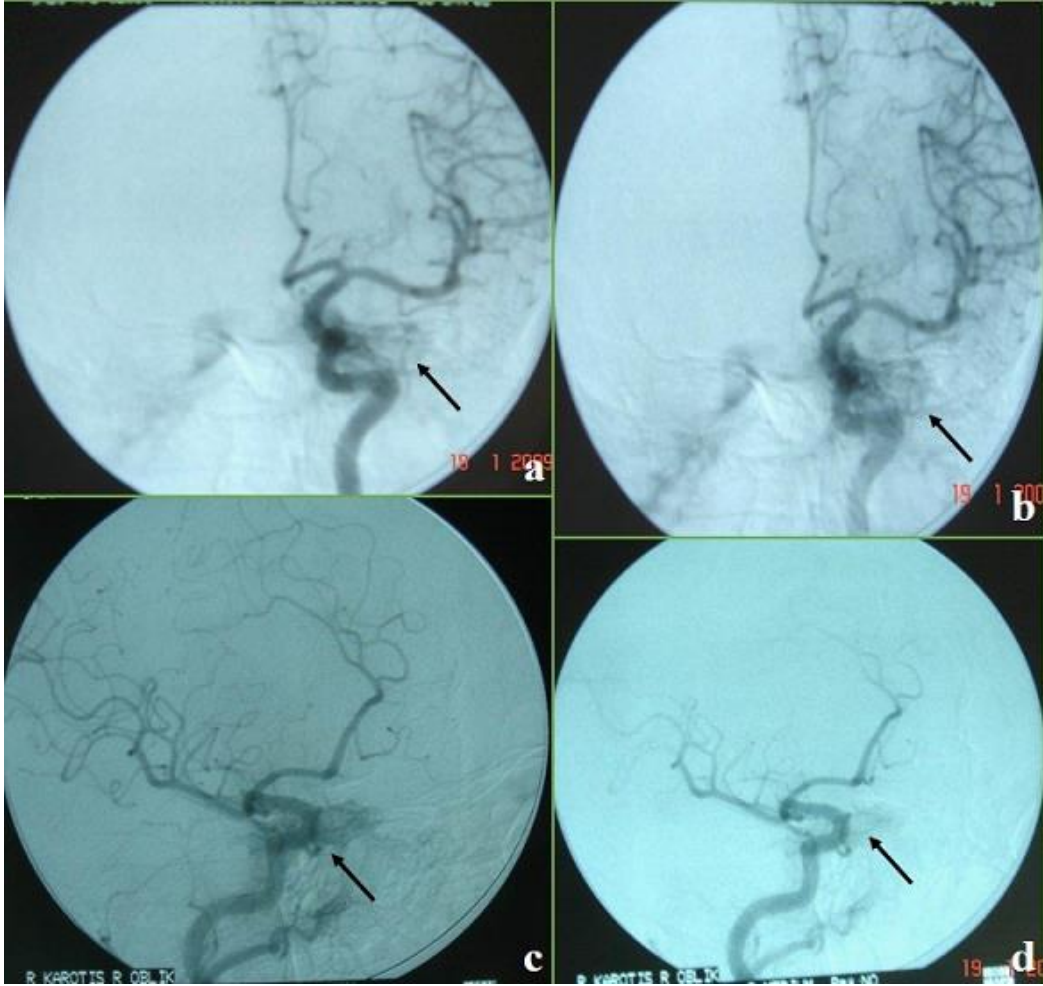
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**Figure 1:** T1 (a) T2 (b) weighted axial and post contrast T1 (c) weighted coronal images show enlargement of both cavernous sinuses which is more prominent on the left side, which show flow voids at the posteromedial aspect and homogenous contrast enhancement; cranial arterial MR Angiography (TOF); (d) demonstrates flow-related enhancement in both cavernous sinuses, particularly on the left side (arrows)



**Figure 2:** Conventional angiography (DSA) shows early venous drainage from cavernous segments of the left and right internal carotid arteries into both cavernous sinuses (postero-anterior view of left internal carotid injection (a,b) and oblique view of right internal carotid artery injection (c,d) respectively)