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OLGU SUNUMU / *CASE REPORT*

Temporary Cranial Nerve Paralysis Due to Carotid Cavernous Fistula

Karotiko-kavernöz Fistüle Bağlı Geçici Kranyal Sinir Paralizileri

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ABSTRACT

Carotid cavernous fistulas are abnormal vascular shunts between the carotid artery system and the cavernous sinus, which may result in life threatening conditions. Carotid cavernous fistulas are usually classified as direct or indirect. The etiologies of direct carotid cavernous fistulas are frequently trauma, ruptured cavernous carotid artery aneurysm, arterial dissection or iatrogenic causes. Endovascular treatment techniques are the first choice for treatment. Embolization of a direct carotid cavernous fistula by transarterial balloon detachment is a well-known treatment procedure. Herein, we present a case of carotid cavernous fistula and discuss its etiology, diagnosis, potential complications and treatment choices in the light of the current literature.

Key words: *abducens nerve; carotid-cavernous sinus fistula; endovascular procedures; paralysis*

ÖZET

Karotiko-kavernöz fistüller, karotis arter sistemi ile kavernöz sinüs arasındaki hayatı tehdit edebilecek anormal vasküler şantlardır. Genellikle direkt ve indirekt olarak sınıflandırılırlar. Etyolojilerinde sıklıkla travma, rüptüre karotid arter anevrizması, karotid arter diseksiyonu yer almakta veya iyatrojenik olabilmektedir. Endovasküler teknikler ilk seçenek tedavi metodlarıdır. Transarteriyel ayrılabilir balon yerleştirilmesi iyi bilinen bir tedavi yöntemidir. Burada, bir karotikokavernöz fistül olgusu sunuyor ve güncel literatür eşliğinde karotiko kavernöz fistülün etiyoloji, tanı, potansiyel komplikasyonları ve tedavi seçeneklerini tartışıyoruz.

Anahtar kelimeler: *abdusens siniri; karotid kavernöz sinüs fistülü; endovasküler işlemler; paraliz*

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Introduction

Acquired arterio-venous fistulas (AVFs) are mostly caused by trauma, spontaneously or after surgery, and these lesions can involve all body areas. A carotid cavernous fistula (CCF) is an aberrant shunt between the cavernous sinus and the internal carotid artery or branches of the external carotid artery[1]. The CCF results in an increase in pressure inside the cavernous sinus. Signs and symptoms of a CCF include pulsatile orbital swelling, systolic murmur, palpable thrill, conjunctival injection, proptosis, decreased visual acuity, elevated intraocular pressure, and cranial nerve palsies[1–3]. Endovascular techniques are the most preferred treatment modalities to prevent the complications of CCF[4]. Herein, we describe a case of temporary abducens paralysis due to CCF, which was treated with endovascular balloon angioplasty.

Case Report

A 60 year old woman applied to our clinic with complaints of pain, epiphora, swelling, redness, diplopia and enlargement of the left eye which developed in one month. She had a history of blunt head trauma in a traffic accident 2 months ago.

A complete ophthalmic examination revealed the best corrected visual acuity as 20/20 in the right eye and 4/20 in the left eye. While the anterior segment and fundus examinations were unremarkable in the right eye, slit lamp examination revealed eyelid edema with conjunctival hyperemia, chemosis, and proptosis with a dilated fixed pupil in the left eye (Figure 1a). The direct and indirect pupillary reflexes to light were

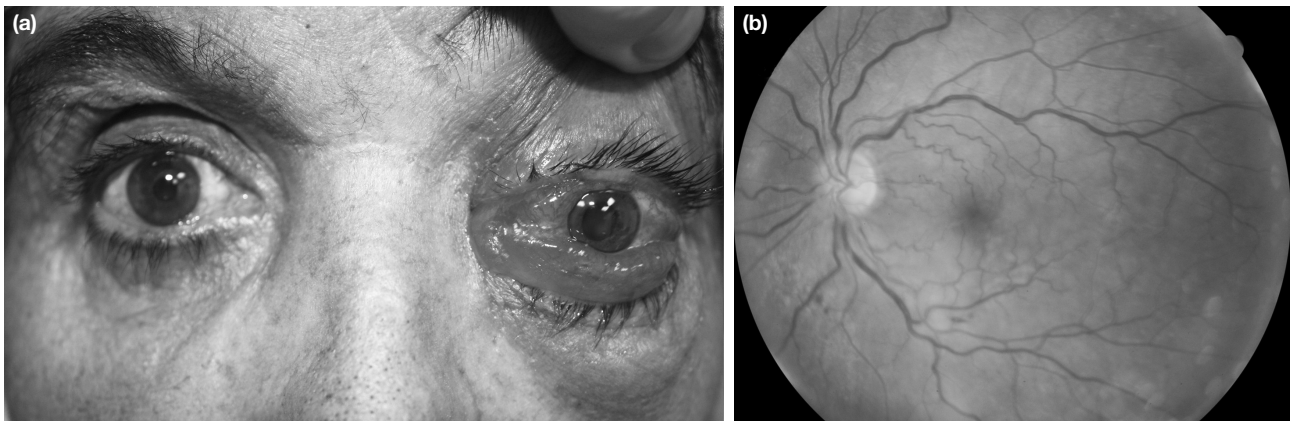

Figure 1. a, b. *Conjunctival chemosis, hyperemia, chemosis, proptosis and fixed dilated papilla on the left side* ***(a)****. Venous dilatation and increased tortuosity, arterial narrowing and flame shaped hemorrhages* ***(b)****.*

positive in the right eye while in the left eye pupilla was fix dilated and did not react with light and there was no indirect light reflex. Extra-ocular movements were limited in all directions in the left eye indicating the III, IV and VI nerve palsies while the right extra-ocular movements were normal. Fundus examination of the left eye showed obvious venous dilatation and increased tortuosity, arterial narrowing and flame shaped hemorrhages (Figure 1b).

Intraocular pressure was 16 mmHg in the right eye and 28 mmHg in the left eye. Gonioscopy revealed an open angle in both eyes. An ophthalmic bruit was heard in her left eye. Topical antiglaucomatous medication, including dorzolamide combined with timolol maleate was administered to the left eye.

After consultation of the patient to the neurosurgery department, neurology department and radiology department, magnetic resonance angiography imaging was performed and revealed a high flow carotid cavernous fistula. Cerebral digital subtraction angiography revealed a high flow carotid cavernous fistula with early venous filling in the arterial phase (Figure 2).

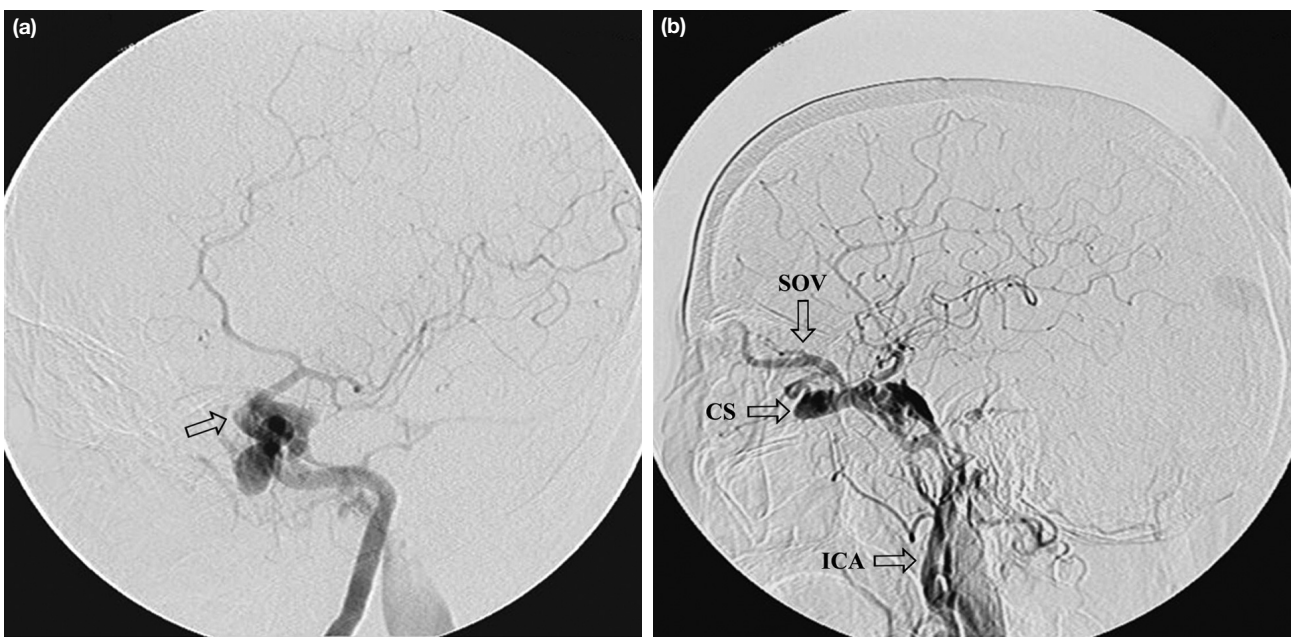

Figure 2. a, b. *Cerebral angiography showing a left-sided high flow carotid cavernous fistula that drained into the cavernous sinus (CS) (arrow) in the early arterial phase* ***(a)****. Internal carotid artery (arrow) and ICA* ***(b)****. Retrograde filling of the superior ophthalmic vein and cavernous sinus (arrows), CS and SOV. SOV, Superior Ophthalmic Vein; CS, Cavernous Sinus; ICA, Internal Carotid Artery.*

Endovascular balloon angioplasty was performed and two detachable balloons were placed in the fistula and the fistula was obliterated successfully (Figure 3).

Following the balloon angioplasty all signs and symptoms resolved in 10 days except the anisocoria, diplopia and restriction of abduction of the left eye (Figure 4). However, restriction of abduction of the left eye regressed over 4 months. At the 15th month follow-up visit, the only remaining pathologic finding was mild mydriasis in the left eye with totally normal pupil reactions (Figure 5).

Discussion

Carotid-cavernous fistulas are the most common type of arteriovenous malformation located above the neck in the body, and they are divided into two groups: direct/high flow and indirect/low flow. Usually direct CCFs' clinical findings are dramatically prominent, including pulsating exophthalmos, retro-orbital bruit, chemosis, decreased visual acuity, ophthalmoplegia, subarachnoid hemorrhage, and proptosis, while indirect CCFs have mild and fewer symptoms such as proptosis, chemosis, arterialized conjunctival veins, retro-orbital pain, elevated IOP and diminished visual acuity[1–3].

Carotid cavernous fistulas' classification systems, complications and treatment modalities have been well described in the literature[5–7]. Retrograde cortical venous drainage is defined as an important risk factor, which is a sign for elevated intracranial venous pressure

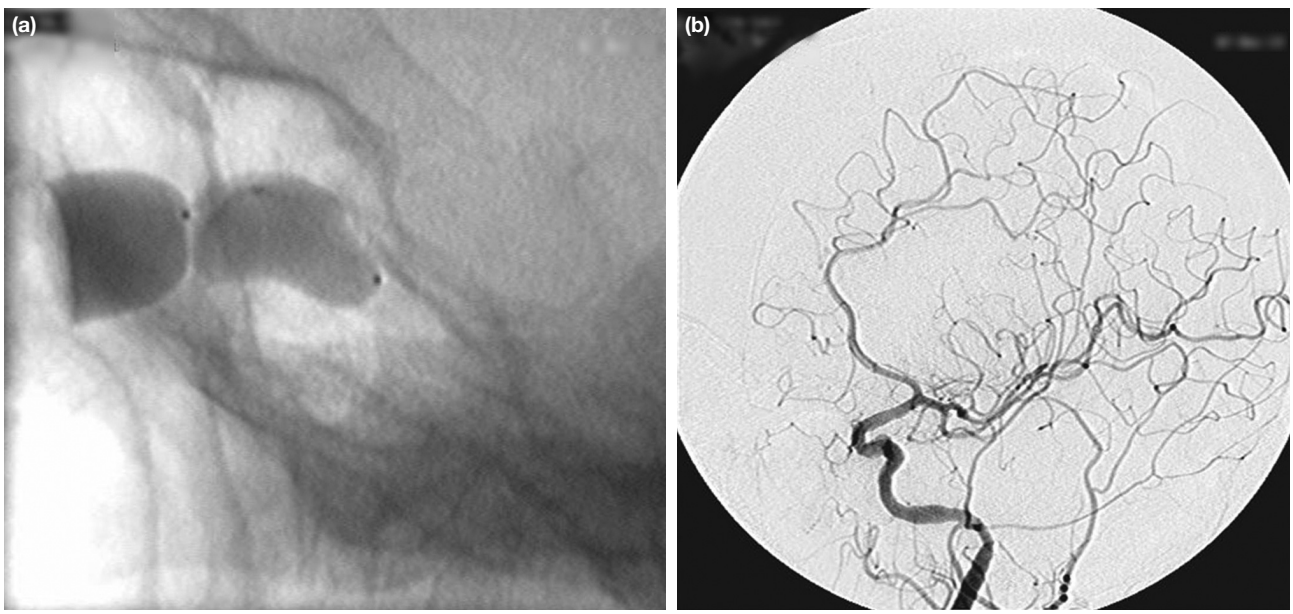

Figure 3. a, b. *Two detachable balloons were placed in the fistula* ***(a)****. The high flow carotid cavernous fistula was occluded with detachable balloons without residual arteriole venous shunting* ***(b)****.*

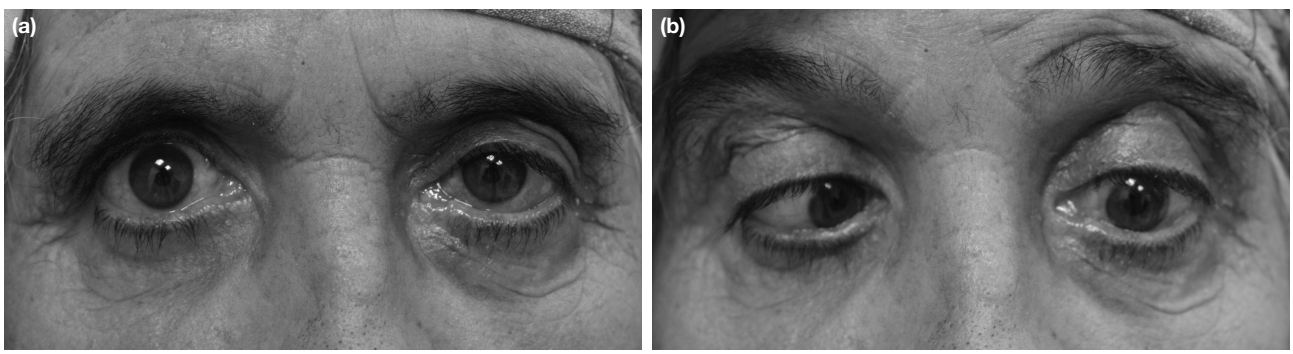

Figure 4. *All signs and symptoms resolved in 10 days, except anisocoria, diplopia and limitation of abduction of the left eye.*

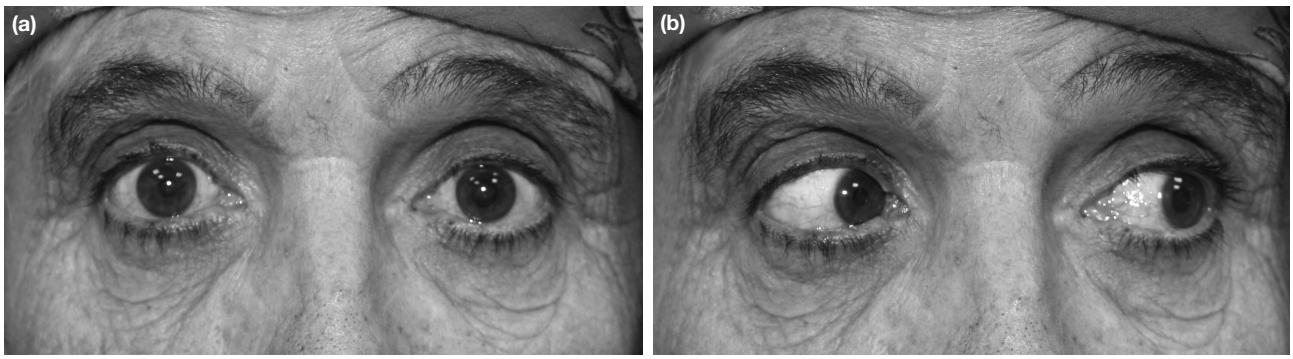

Figure 5. *Limitation of the abduction of the left eye regressed spontaneously in 4 months; only mid dilation of the left pupil persisted.*

predisposing to cerebral infarction and hemorrhage. Meyers et al. reported that if patients with cortical venous drainage are untreated, the risk of intracerebral hemorrhage ranges from 30% to 40% and may be fatal[8].

The main aim of CCF treatment is to obliterate the fistula while avoiding the complications with preserving the normal flow of blood through the internal carotid artery. Previously, surgical ligation of the fistula, conservative management and endovascular treatment techniques were reported in the literature[5]. Currently, trans-arterial or trans-venous embolization is the first treatment choice of most CCFs.

Reported complications of endovascular treatment include cerebral infarction, decreased visual acuity, diabetes insipidus, retroperitoneal hematoma, femoral vein thrombosis and ophthalmoplegia in 2–5% of patients[8,9]. Previously in the literature there have been some CCF case reports with cranial nerve palsies[10,11].

In their study including 48 patients with direct CCFs who were treated with transarterial balloon detachment technique, Tsai et al. reported five patients with oculomotor nerve palsy, four of them with abducens nerve palsy and one with simultaneous third and sixth cranial nerve palsy[11]. They also documented that Guglielmi detachable coils had a lower risk of procedure-related oculomotor nerve deficit in the treatment of direct CCFs[11].

Luo et al. reported that 16.7% of patients who underwent embolization of the CCF with n-butyl cyanoacrylate experienced temporary cranial nerve palsies that resolved completely in each case within 6 months[12]. Kim et al., in their studies on transvenous embolizations of cavernous dural arteriovenous fistulas, reported six cases (10.7%) who developed cranial nerve signs after transvenous embolization, including sixth nerve palsy[13].

Over-packing of the cavernous sinus is one possible reason for transient cranial nerve symptoms; other possible causes are progressive thrombosis of the cavernous sinus and direct injury of the nerve by coil or microwire/microcatheter[14,15]. The abducens nerve is particularly vulnerable to injury from vascular engorgement and trauma due to its proximity to the internal carotid artery and unsecured course through the cavernous sinus. In our case, it is unclear whether the cause of abducens paralysis was the caroticocavernous fistula or the balloon angioplasty treatment. Although the reason for the palsy is unclear, the abducens nerve paralysis was temporary, and the patient was treated successfully with endovascular balloon angioplasty.

Compressive lesions such as intracranial aneurysm, space occupying lesions and carotid cavernous fistulas may cause 3rd nevre palsy with pupil involvement, with a reduced pupillary light reflex and reduced accommodation, as a result of the superficial location of the autonomic nerve fibers. In our case, the ophthalomoplegia resolved much earlier than the anisocoira. Anisocoria persisted while pupil reactions were within normal limits at the 15th month follow-up visit.

Park et al., in their studies on clinical evaluation and natural history of acquired 3rd, 4th and 6th nerve palsies, reported that 67.6% of patients showed total recovery in all total nerve palsy cases[16]. In the same study they noted that only one out of 13 patients showed complete recovery from pupil involvement in 3rd nerve palsy which was due to Tolosa-Hunt syndrome[16]. Zhang et al., in their study, used some recovery criteria that accept either partial or completely normal pupillary reactions as complete recovery[17]. According to their criteria, our case showed complete recovery with mild mydriasis and completely normal pupillary reactions.

It is possible to diagnose CCF earlier in cases where the onset of ophthalmological signs and symptoms are earlier. It should be taken into consideration that, recovery of cranial nerve palsies may take longer duration, although most of the symptoms and signs disappear quickly after endovascular treatment. In conclusion, the relatively rare entity may be managed with a multi discipline approach and by using the modern diagnostic and therapeutic modalities.

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