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Case Report
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Symptomatic Bilateral Cavernous Carotid Artery Aneurysms Presenting as an Acute Painful Complete Ophthalmoplegia: A Case Report and a Review of the Literature

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Abstract

Background: Cavernous Carotid Aneurysm (CCA) is a rare entity, but it is predominately found in the elderly. Clinical data regarding the diagnosis and the management of bilateral cavernous carotid aneurysms in old patients are scarce. The aim of our study is to present a case of acute painful ophthalmoplegia in an old patient due to cavernous carotid aneurysm with an extensive literature review.

Case Report: A 92-year-old female patient presented with a complete ptosis of the right eye. Neurological examination showed right severe ptosis associated with complete ophthalmoplegia. Brain MRI revealed large bilateral CCAs causing compression of cranial nerves III, IV and VI in the right cavernous sinus. Medical treatment consisting of gabapentin and antalgic therapies had been initiated. At the three-month follow-up, the patient was clinically stable.

Conclusion: This case illustrates a rare presentation of CCA manifested by complete ophthalmoplegia in a very old female patient. This diagnosis should be considered as a possible etiology of cavernous sinus syndrome.

Keywords: Bilateral cavernous carotid aneurysms; Cavernous sinus syndrome; Painful ophthalmoplegia; Management strategies

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Background

Cavernous Carotid Aneurysms (CCAs) account for 15% of all Internal Carotid Artery (ICA) aneurysms [1]. Among them, 5% are bilateral [2]. CCAs are predominately found in females and they tend to manifest in older ages. This condition is rare with few reported cases in the literature. Herein, we present an unusual case of acute painful bilateral cavernous sinus syndrome due to CCA in a very old woman.

Case Presentation

A 92-year-old female patient, with a history of moderately controlled hypertension, presented to the emergency department with a severe sudden onset headache prominent on the right periorbital region with a complete ptosis of the right eye starting two days earlier. No history of febrile illness or trauma was found. The patient reported no fluctuations in her symptoms and being in excellent health. Neurological examination revealed right severe ptosis associated with complete right ophthalmoplegia (Figure 1: Panels A to D). Pupils were round, equal and reactive to light. Vision acuity was preserved. Intraocular pressure measurement was normal. Anterior segment and dilated fundus examinations of both eyes were unremarkable. The remainder of the neurological and somatic examinations was normal.

Contrast enhanced CT brain demonstrated a filling defect in the right cavernous sinus. Brain MRI revealed large bilateral CCA measuring approximately 17 mm × 28 mm on the right and 9 mm × 10 mm on the left, causing compression of the cranial nerves III, IV, and VI in the right cavernous (Figure 2). Blood tests, including complete blood count, erythrocyte sedimentation rate, C-reactive protein, as well as liver, renal, and thyroid function tests, were normal. Diagnosis of symptomatic bilateral CCA was retained.

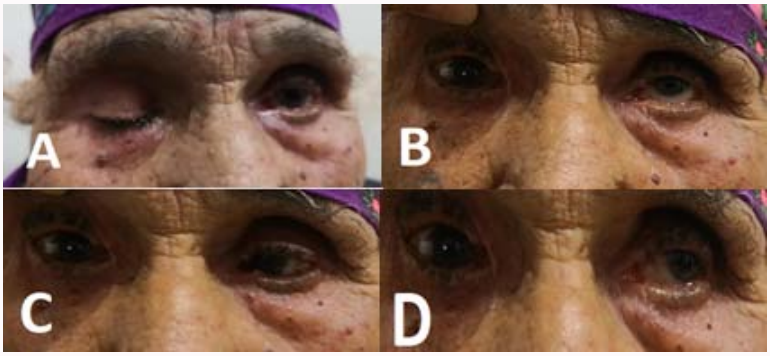

Figure 1: (A) severe right ptosis, (B) vertical gaze palsy of the right eye, (C, D) convergence and divergence dysfunction of the right eye.

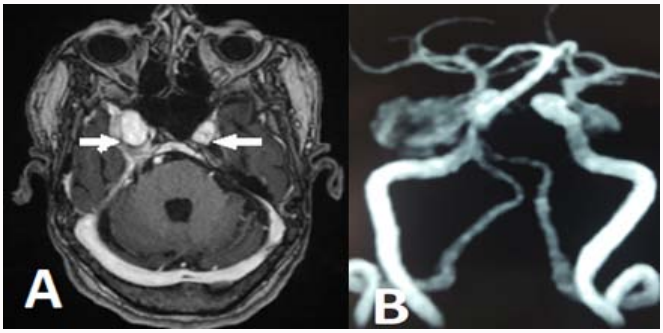

Figure 2: Brain MRI showing bilateral aneurysms (A) T1-weighted axial Brain Magnetic Resonance (MR) scan showing bilateral CCAs with compression of the cavernous sinus; (B)Magnetic resonance angiography sequence showing large bilateral cavernous carotid aneurysm (Arrows) measuring approximately 17 mm × 28 mm on the right side and 9 mm × 10 mm on the left side.

Table 1: Demographic and clinical data of old patients reported with bilateral CCAs.

Age (years)	Gender	Presentation	Neurological examination	Brain MRI	Treatment	Outcome
74 [9]	female	Diplopia	Hypertropia	Bilateral CCAs	NA	NA
76 [10]	male	Vertical diplopia with headache	Left mild mydriasis And ptosis	Giant bilateral CCAs	No treatment	Spontaneous subsidence of symptoms
85 [5]	female	Acute diplopia	Left IV and VI palsy	Bilateral CCAs	No treatment	NA
73 [6]	female	Diplopia	Bilateral paresis of lateral rectus muscles	Bilateral CCAs	Orthopedic correction of diplopia	NA
76 [4]	female	Fluctuating diplopia	Right lateral rectus palsy Left ptosis	Large bilateral CCAs	endovascular embolization of the left carotid aneurysm with flow diversion	Improvement of ptosis

CCAs: Cavernous Carotid Aneurysms; NA: Not Available

A conservative medical therapy was started based on antalgic drugs and gabapentin (300 mg x 3/day). Given the patient's advanced age and the high procedural risk, the surgical intervention procedure and interventional endovascular options were not considered.

At discharge, the patient's headache resolved, but complete ophthalmoplegia remained. At three-month follow-up, the patient was clinically stable.

Discussion

Bilateral CCAs are a rare entity accounting for 11% of all ICA aneurysms [2]. They are predominately found in elderly subjects and few cases have been reported in the literature. In order to assess the frequency, clinical presentation and management of bilateral unruptured CCAs in old patients, an extensive review of the literature was conducted (Table 1).

In this report, we describe a rare case of a unilateral severe ptosis associated with complete external ophthalmoplegia in a 92-year-old female patient. The symptoms resulted from bilateral CCA. A larger size was observed on the right side, probably explaining the unilateral clinical presentation due to the mass effect. The late clinical manifestation of CCA might be explained by the age-related structural weakness of the ICA wall, inducing aneurysm formation.

CCAs can remain asymptomatic or become symptomatic due to compression, rupture, or extension beyond the cavernous sinus [2,3]. As they are located in a venous pouch, the symptoms can mimic cavernous sinus thrombosis signs, including diplopia due to the involvement of the III, IV, and VI cranial nerves, the reduction in visual acuity caused by compressive optic neuropathy, and corneal hyperesthesia and trigeminal dysesthesia due to the injury of the first and the second branch of the trigeminal nerve [4,5].

It is worth noting that complete external ophthalmoplegia with pupil sparing from aneurysmal compression, as observed in our patient, is extremely rare. It can be explained by the involvement of only the superior division of the third cranial nerve, not including

pupillomotor fibers. To the best of our knowledge, only two cases have previously been reported with pupil-sparing third nerve palsy [3,6].

In the literature, all patients with bilateral CCAs experienced unilateral symptoms. In the present case, aneurysm on the left side was asymptomatic because of its small size. The natural evolution of CCA may lead to rupture resulting in a carotid-cavernous fistula, thrombus formation, subarachnoid hemorrhage, or distal embolization [2,3]. Fortunately, none of these complications occurred in our patient.

In the elderly population, it is important to establish the differential diagnosis between CCA and other serious conditions, including cavernous sinus malignancies, trigeminal neuralgia, pituitary tumors, and ocular myasthenia gravis, particularly when the parasympathetic third nerve is spared as in the present case [3,4].

Brain MRI remains the gold standard for the diagnosis of CCAs and for differentiating it from other etiologies. It should be performed in cases having multiple cranial nerve palsies because CCAs are a potentially treatable cause of extra ocular muscle palsy [3,4].

Management of these lesions is a daunting task for the physician. As these aneurysms have a low risk of morbidity and mortality, decisions regarding treatment should take into consideration the etiology, clinical presentation, size of the aneurysm, adequacy of cross circulation, and patients' preference [6,7].

Various treatment options are currently available, including conservative medical management, unilateral or bilateral carotid ligation with or without bypass, bilateral surgical clipping, bilateral endovascular occlusion, or endovascular flow diversion [7,8]. Due to their rarity, no definite guidelines exist and therapies should therefore be individualized. In the present case, conservative therapy was the treatment of choice due the patient's old age and the high risk of mortality [6-8]. It is worth noting that the use of gabapentin was reported to be effective in resolving headaches in a previous case report [5].

Conclusion

This case illustrates a rare presentation of CCA manifested by complete ophthalmoplegia in a very old female patient. This diagnosis should be considered as a possible etiology of cavernous sinus syndrome. A close follow-up is recommended to detect eventual complications, especially in old patients.

References

1. Vanikieti K, Poonyathalang A, Jindahra P, Cheecharoen P, Chokthaweesak W. Occipital lobe infarction: A rare presentation of bilateral giant cavernous carotid aneurysms: A case report. BMC Ophthalmol. 2018;18(1):25.
2. Yen PS, Teo BT, Chen SC, Chiu TL. Endovascular treatment for bilateral mycotic intracavernous carotid aneurysms. Case report and review of the literature. J Neurosurg. 2007;107(4):868-72.
3. Rosi Junior J, Welling LC, Yeng LT, Caldas JG, Schafranski M, Teixeira MJ, et al. Cavernous carotid artery aneurysms: epidemiology, natural history, diagnostic and treatment. An experience of a single institution. Clin Neurol Neurosurg. 2014;125:32-5.
4. Gagliardi D, Faravelli I, Villa L, Pero G, Cinnante C, Brusa R, et al. Bilateral cavernous carotid aneurysms: atypical presentation of a rare cause of mass effect. A case report and a review of the literature. Front Neurol. 2018;9:619.
5. Bodla AA, Ablett M, Inglis A. Bilateral intracavernous carotid artery aneurysms presenting as progressive cranial nerve palsies. Clin Exp Optom. 2007;90(3):207-8.
6. Caroli M, Bertani G, Fetoni V, Sasanelli F, Gaini SM, Samis Zella MA. Bilateral intracavernous carotid artery aneurysms presenting as abducens nerve palsy: Case report. J Neurosurg Sci. 2009;53(4):169-70.
7. Lustbader JM, Miller NR. Painless, pupil-sparing but otherwise complete oculomotor nerve paresis caused by basilar artery aneurysm. Case report. Arch Ophthalmol. 1988;106(5):583-4.
8. Ambekar S, Madhugiri V, Sharma M, Cuellar H, Nanda A. Evolution of management strategies for cavernous carotid aneurysms: A review. World Neurosurg. 2014;82(6):1077-85.
9. Mindel JS, Charney JZ. Bilateral intracavernous carotid aneurysms presenting as pseudo-ocular myasthenia gravis. Trans Am Ophthalmol Soc. 1989;87:445-57.
10. Díaz MB, Mercado FC, Lemme Plaghos LA. "Mirror-image" bilateral giants: Intracavernous carotid artery aneurysms. Interv Neuroradiol. 2006;12(3):251-6.