Resection of an oculomotor nerve cavernous angioma

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Abstract

Background: Cavernous angiomas (CAs) of cranial nerves are rare, and their occurrence on the third cranial nerve is particularly rare. Surgical management of such CAs involving the third nerve is controversial. We describe a case of a symptomatic CA of the oculomotor nerve and review the literature in order to ascertain the relevance of surgical intervention.

Case Description: A 71-year-old male patient presented with a 2-month history of progressive oculomotor nerve paralysis. CA of the oculomotor nerve was suspected on magnetic resonance imaging (MRI). The patient underwent complete resection of the CA through a subtemporal approach, preserving the integrity of the nerve. Histopathological analysis confirmed the diagnosis of CA. Despite optimal resection, the patient did not improve postoperatively.

Conclusion: CAs of cranial nerves can cause rapid or progressive neurological deterioration. Whereas delayed treatment often leads to irreversible deficits, early nerve-sparing surgical excision of the CAs may potentially restore function.

Key Words: Cavernoma, cavernous angioma, oculomotor nerve, third nerve palsy

INTRODUCTION

Cavernous angiomas (CAs) are common cerebrovascular malformations often diagnosed in the fourth or fifth decade of life.1,11 These lesions occur rarely in cranial nerves, most commonly within the optic nerve.5,7 They are sometimes found in the oculomotor nerve, but this is exceptionally rare. Their surgical management remains controversial.

MATERIALS AND METHODS

We describe a case of a symptomatic CA of the oculomotor nerve presenting with acute nerve palsy that failed to improve following surgical resection. We review the literature on surgical cases of the oculomotor nerve CA and discuss the outcome in terms of recovery of nerve function.

CASE REPORT

A 71-year-old male without significant medical history presented with sudden binocular diplopia and left frontal headache. Physical examination 48 h after onset of symptoms revealed a partial third cranial nerve palsy on the left side. Initial cerebral magnetic resonance imaging (MRI) demonstrated an 8 mm oval lesion in the anterior left interpupillary cistern that was hyperintense on T1 and heterogeneous on T2-weighted images [Figure 1a and b]. On the computed tomography angiography (CTA), the lesion...
enhanced but did not correspond to an intracranial aneurysm [Figure 1c]. The patient was observed. Over the following week, the left third cranial nerve deficit worsened significantly. The ipsilateral pupil was fully dilated and unreactive. Complete ptosis and paralysis of the oculomotor nerve-related muscles were also observed. A second MRI showed ferromagnetic susceptibility signals on gradient echo images [Figure 2a]. Constructive interference steady state (CISS) T2-weighted sequences better delineated the lesion and its relation to the oculomotor nerve [Figure 2b]. The suspected diagnosis was CA of the third cranial nerve. Given the recent complete third nerve paralysis and the risk of recurrent hemorrhage, surgery was performed.

**Intervention**

Surgery was performed 2 months after onset of symptoms. Through a left subtemporal approach, exposure of the interpeduncular cistern revealed a raspberry-like lesion bulging from the third cranial nerve, anterolateral to the left cerebral peduncle [Figure 3a]. A well delineated plane of dissection allowed removal of the lesion while preserving the integrity of the nerve [Figure 3b and c].

**Postoperative course**

Postoperative computed tomography (CT) scan showed no complication [Figure 3d]. Histopathological analysis showed a lesion with high vascularity and hyalinized channels lined by a single layer of endothelial cells, findings that confirmed the diagnosis of a cavernous angioma [Figure 4]. Six months after surgery, the patient remained with a complete oculomotor palsy.

**Literature review**

We found eight surgical cases of oculomotor nerve CA [Table 1]. Three patients presented with a pure oculomotor nerve deficit, two cases had mixed...
### Table 1: A summary of reported oculomotor cavernous angiomas

| Year/ authors | Age/sex | Symptom progression and physical examination | Radiological findings | Acute hemorrhage | Surgical approach | Intra-operative appearance | Degree of resection | Preservation of nerve integrity (Y/N) | Outcome |
|---------------|---------|------------------------------------------|----------------------|----------------|----------------|--------------------------|-------------------|------------------------------------|---------|
| 1983 Scott et al. | 14/M | 10-day history of progressive diplopia, R eye pain, and diminished visual acuity PE: Mydriasis and complete ptosis | CT scan: Homogenously enhancing lesion posterolateral to the dorsum sellae Conventional angiography: Small vascular blush. No aneurysm | Y | R frontotemporal approach | Bulbous, tortuous reddish mass | Total | N | Severe hemiparesis 5 days post-operatively, with subsequent improvement. Persistent third nerve palsy at 3-month follow-up |
| 1986 T. Yamada et al. | 33/M | One-week history of progressive L blepharo-ptosis and double vision. PE: Mydriasis and limited EOMs | CT scan: Isodense mass in L prepontine area CT angiography: No aneurysm | Y | L frontotemporal approach Multicystic dark reddish global nodule | Total | N | Persistent L oculomotor nerve palsy |
| 1990 Matias-Guiu et al. | 36/F | One-year history of galactorrhea, amenorrhea and occasional headaches PE: L temporal quadrantopia | CT scan: Homogenously enhancing expansive tumor in the L CS Conventional angiography and MRI: Extra-axial tumor in the L temporal fossa | N | L temporal approach | Reddish tumor in the left cavernous sinus | Subtotal | Y | Residual oculomotor nerve paralysis at 6-month follow-up |
| 1993 Ogilvy et al. | 28/M | Sudden onset of R retro-orbital pain and partial ptosis which progressed to complete ptosis during the day PE: limitation in EOMs and complete ptosis | CT scan: Calcified hyperdense lesion with erosion of the posterior clinoid process MRI: Heterogeneous T1 and T2 hyperintensity with partial contrast enhancement | Y | R frontotemporal approach-transsylvian | Reddish-brown lesion | Total | N | Persistent L oculomotor nerve palsy |
| 2005 Park et al. | 33/M | Initial eye dryness and pain Progressive L pupil dilatation and ptosis with double vision for 6 months PE: limitation in EOMs, mydriasis and complete ptosis | MRI: Extra-axial T1 and T2 isointense mass abutting the left uncus and CS Homogeneous enhancement | N | L frontotemporal craniotomy | Tortuous red vessels proximal to the CS | Subtotal | Y | Worsened post-operative oculomotor nerve palsy Subsequent improvement at 2-month follow-up |

(Contd...)
Table 1: Contd....

| Year/ authors       | Age (years)/ sex | Symptom progression and physical examination                                      | Radiological findings                                                                 | Acute hemorrhage | Surgical approach | Intra-operative appearance | Degree of resection | Preservation of nerve integrity (Y/N) | Outcome                                            |
|---------------------|------------------|-----------------------------------------------------------------------------------|----------------------------------------------------------------------------------------|------------------|--------------------|--------------------------|---------------------|--------------------------------------|---------------------------------------------------|
| 2007 Itshayek et al. | 25/F             | 3-month history of pain in R occipital/hemifacial region                          | CT scan: Isodense mall in the R prepontine area. No enhancement. MRI: Heterogenous on T1 and T2 sequences. Subtle enhancement. Lesion located at the level of the oculomotor nerve adjacent to the CS | N                | R pterional craniotomy-transsylvian | Red, multiloculated lesion, raspberry-like lesion | No resection       | Y                                    | Asymptomatic at 18-month follow-up               |
| 2011 Wolfe et al.   | 69/M             | Suddent onset of complete L oculomotor nerve palsy 3 weeks prior to presentation | MRI: Hyperintense on T1 and heterogeneous on T2. No enhancement. MR angiography and conventional angiography: Suggestive of thrombosed Pcom aneurysm | Y                | L pterional craniotomy-transsylvian | Round, clot-containing mass arising from a mulberry-like lesion immediately posterior to the oculomotor nerve CS entry | Total              | Y                                    | Persistent oculomotor nerve palsy at 6-week follow-up |
| 2011 Wolfe et al.   | 26/M             | sudden-onset of complete R oculomotor nerve palsy                               | MRI: T1 hyperintensity in the R crural cistern                                          | Y                | R cranio-orbital transsylvian | NA                       | Total              | Y                                    | Resolution of oculomotor nerve palsy at 6-month follow-up |

Y: Yes, N: No, M: Male, F: Female, PE: Physical examination, CT: Computed tomography, R: Right, L: Left, EOM: Extra-ocular movements, CS: Cavernous sinus, MRI: Magnetic resonance imaging, Pcom: Posterior communicating

ophthalmic and oculomotor nerve symptomatology and one case had an optic nerve deficit in addition to ophthalmic and oculomotor nerve involvement. In two cases, despite the fact that the CA location was proven to be on the third cranial nerve intraoperatively, no oculomotor nerve deficit was found at presentation: One had only ipsilateral trigeminal symptoms and the other had pituitary dysfunction with ipsilateral optic nerve deficit.

In the six patients with oculomotor nerve palsy, three cases presented acutely with complete oculomotor nerve palsy in less than 24 h, two cases presented with a partial deficit that worsened to a complete palsy over 7-10 days and one patient developed slow progressive partial oculomotor palsy over a 6-month period. Cases with severe acute oculomotor nerve deficit at presentation had MRI findings suggesting intralesional hemorrhage such as hyper-T1 and hyper-T2 signals. Radiological findings of acute hemorrhage were not found in patients with a more slowly progressive symptomatology.

Seven of the eight cases of oculomotor nerve CA had partial or total resection of the lesion [Table 1]. Of the five cases with total resection, only one had recovery of nerve function with complete resolution of symptoms after 6 months. Of the remaining four patients without recovery, three did not have surgical preservation of nerve integrity. One case with partial oculomotor palsy developed immediate postoperative worsening after partial resection followed by subsequent improvement of nerve function. One case without preoperative oculomotor nerve deficit developed persistent oculomotor paralysis after partial resection.

**DISCUSSION**

Cerebral CAs are circumscribed, mulberry-like lesions consisting of thin hyalinized capillary channels without intervening parenchyma. Although most arise in the brain and sometimes in the spinal cord, such lesions may exceptionally involve cranial nerves. The optic nerve and the facial/vestibulocochlear complex are the ones most commonly affected.
Our review of this rare pathology showed that the clinical presentation of oculomotor nerve CA is variable. A third cranial nerve CA located in the oculomotor triangle can compress the optic nerve superomedially\cite{5} and/or the ophthalmic nerve inferolaterally at its entry in the lateral wall of the cavernous sinus.\cite{5} Similarly, a large third cranial nerve CA in the parasellar region can induce pituitary dysfunction by mass effect.\cite{12} Interestingly, cisternal trigeminal nerve CAs can also present with multiple cranial nerve deficits, including oculomotor nerve palsy.\cite{2} Although not found in our review, oculomotor nerve CA may potentially be responsible for subarachnoid hemorrhage, as has been reported in cases of optic nerve CAs and superficial brainstem CAs.\cite{10,13}

Including our case, five patients underwent CA resection with preservation of nerve integrity. Two of these had recovery of oculomotor nerve function. As seen in the case reported by Park \textit{et al.} in 2005, slowly progressive and incomplete oculomotor nerve deficit at presentation might allow a better chance of neurological recovery after surgery if the nerve is preserved. Although the completeness of oculomotor palsy may be a negative prognostic factor for nerve function recovery, one case reveals that early complete oculomotor palsy can be totally reversed with prompt total resection.\cite{3} Thus, surgical intervention within a short time of onset of symptoms may be associated with better postoperative improvement. Our case, which presented early with an incomplete oculomotor palsy, may have benefited from surgical resection, had it been performed earlier.

**CONCLUSION**

CAs of the third cranial nerve can cause rapid and progressive neurological deterioration. Whereas delayed treatment often leads to irreversible deficits, early surgical intervention with preservation of the nerve may potentially allow for improvement of nerve function.

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