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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

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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 interpeduncular 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.

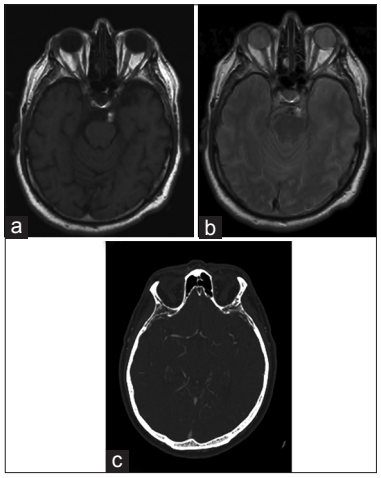

Figure 1: (a,b) T1 and T2 MRI sequences revealing a left-sided heterogeneously enhancing lesion adjacent to the cerebral peduncle. (c) CT angiogram revealed no evidence of aneurysm malformation

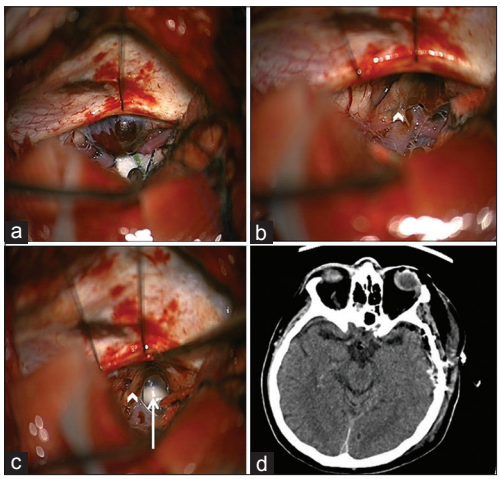

Figure 3: (a) Intraoperative image disclosing a raspberry-like lesion arising within the left oculomotor nerve in the interpeduncular cistern. (b,c) Postresection image revealing the decompressed third nerve with preservation of nerve continuity despite its deformity (arrow head) and the intact contralateral oculomotor nerve (long arrow). (d) Postoperative CT scan showing gross total resection

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].[3-5,7,9,12,14] Three patients presented with a pure oculomotor nerve deficit,[3,4] two cases had mixed

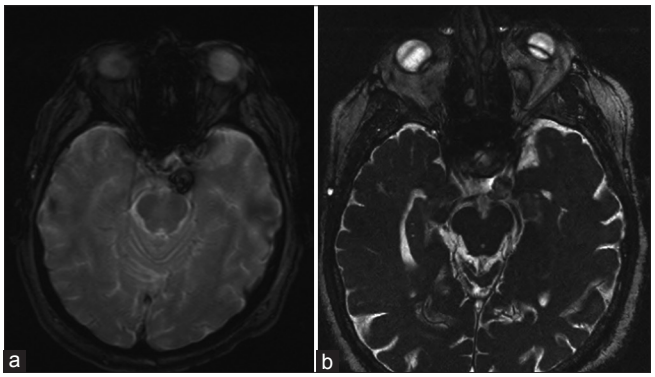

Figure 2: (a) Gradient echo MRI revealing round hypointense lesion with ferromagnetic susceptibility signals. (b) CISS MRI sequence revealing a left-sided heterogeneously enhancing lesion

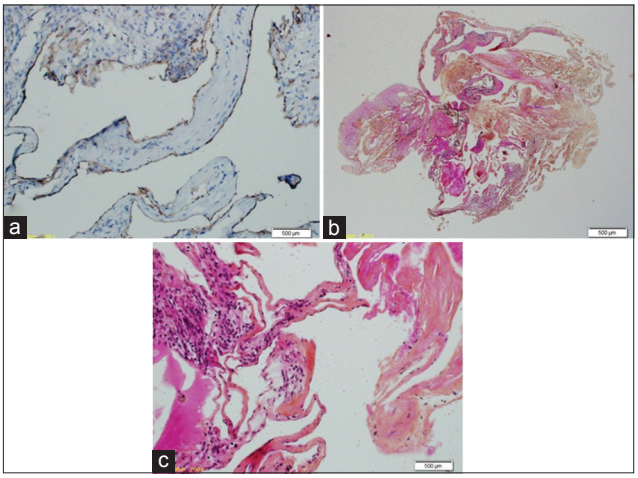

Figure 4: (a-c) H and E stain showing tightly packed vascular channels with no intervening nervous tissue containing thrombotic and blood products with varying thickness of hyalinized endothelial walls lacking smooth muscle or elastic tissue

Table 1: A summary of reported oculomotor cavernous angiomas

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
1983 Scott *et al.*	14/M	10-day history of progressive diplopia, R eye pain, headache and diminished visual acuity PE: Mydriasis and complete ptosis	CT scan: Homogenously enhancing lesion postero-lateral 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 quadrantanopsia	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 Subsebquent 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-transsylvain	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 heterogenous 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 mulberrylike lesion immediately posterior to the oclumorotor 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, P com: Posterior communicating

ophthalmic and oculomotor nerve symptomatology[5,9] and one case had an optic nerve deficit in addition to ophthalmic and oculomotor nerve involvement.[7] 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[14] and the other had pituitary dysfunction with ipsilateral optic nerve deficit.[12]

In the six patients with oculomotor nerve palsy, three cases presented acutely with complete oculomotor nerve palsy in less than 24 h,[3,9] two cases presented with a partial deficit that worsened to a complete palsy over 7-10 days[4,7] and one patient developed slow progressive partial oculomotor palsy over a 6-month period.[9] Cases with severe acute oculomotor nerve deficit at presentation had MRI findings suggesting intralesional hemorrhage such as hyper-T1 and hyper-T2 signals.[3,9] Radiological findings of acute hemorrhage were not found in patients with a more slowly progressive symptomatology.[4,5,7,12]

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.[3] Of the remaining four patients without recovery, three did not have surgical preservation of nerve integrity.[4,7,9] One case with partial oculomotor palsy developed immediate postoperative worsening after partial resection followed by subsequent improvement of nerve function.[5] One case without preoperative oculomotor nerve deficit developed persistent oculomotor paralysis after partial resection.[12]

DISCUSSION

Cerebral CAs are circumscribed, mulberrry-like lesions consisting of thin hyalinized capillary channels without intervening parenchyma.[8] 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.[6,12,14]

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[7] and/or the ophthalmic nerve inferolaterally at its entry in the lateral wall of the cavernous sinus.[5] Similarly, a large third cranial nerve CA in the parasellar region can induce pituitary dysfunction by mass effect.[12] Interestingly, cisternal trigeminal nerve CAs can also present with multiple cranial nerve deficits, including oculomotor nerve palsy.[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.[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 *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.[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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