

Functional outcome after surgical treatment of cavernous malformation involving ocular motor cranial nerves: A systematic review



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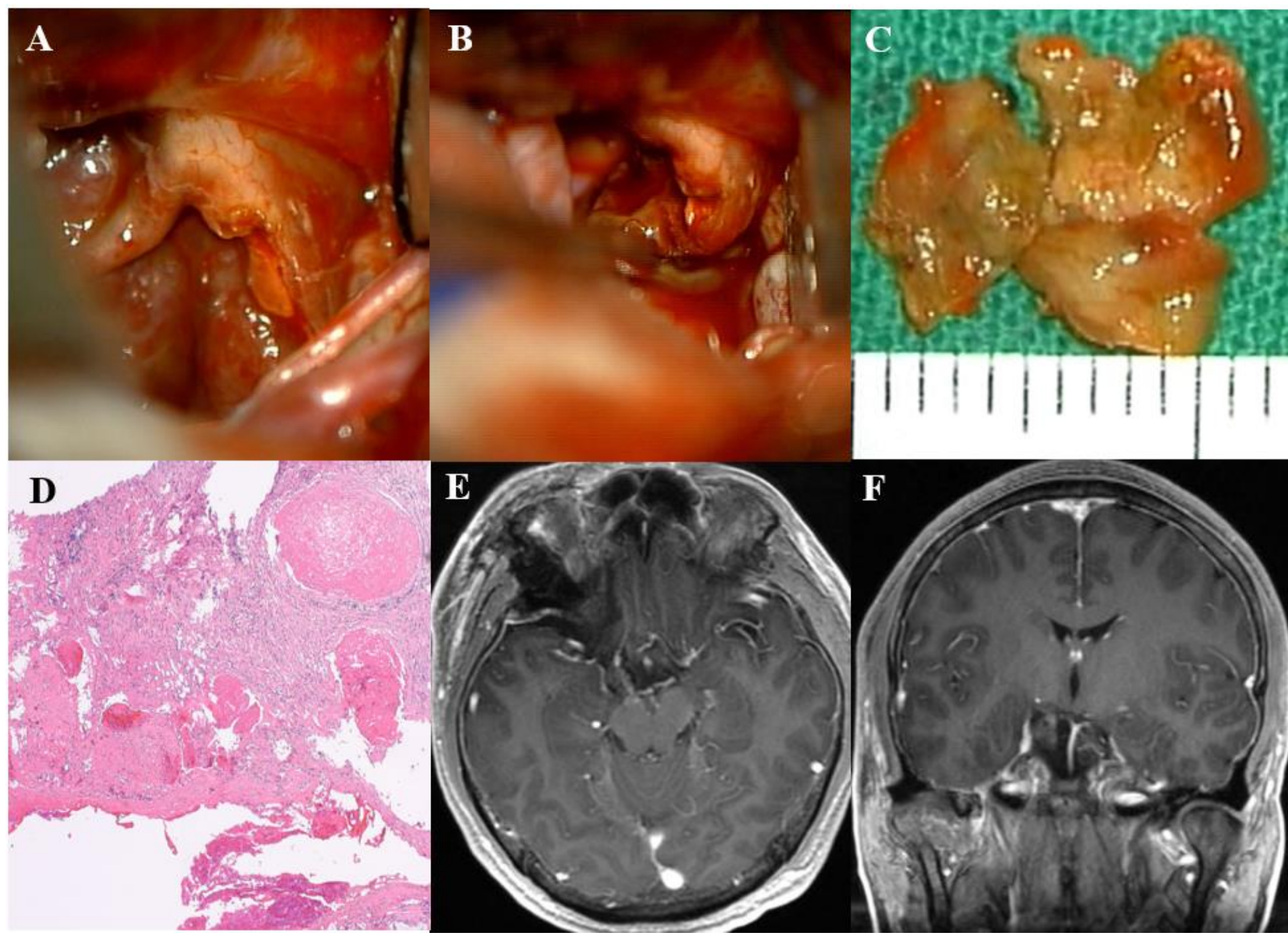
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Introduction

- Cavernous malformation (CM) is an angiographically occult vascular malformation with an incidence of 0.3% to 0.5% in the general population.
- Although rare, **CMs may arise from the cranial nerves (CNs)**, and most cases occur in the CN II and VII/VIII complex.
- Mortality related to CMs of the CNs rarely occur. Therefore, **functional preservation** gains importance for the management of these lesions.
- **CMs of the ocular motor CNs III, IV, and VI** are extremely rare and associated with diplopia and strabismus.

Methods and Materials

- Reviewed 'PubMed', 'SCOPUS', 'Web of Science', and 'Google Scholar' databases to identify relevant case reports and studies of CMs of the ocular motor CNs published (between January 1980 and December 2018)
- Based on the **Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA)** guidelines.
- The algorithm used the terms '**cavernous malformation**', '**cavernous angioma**', and '**cranial nerve**' as the search terms.
- The inclusion criteria were as follows: **(1) surgically confirmed CM of CN III, IV, and VI (by histology or intraoperative view)**
(2) radiological confirmation of disease.



Intraoperative view after removal of the mass. The residual lesion is present, and CN III continuity is preserved. Histopathologic examination of hematoxylin-eosin stained surgical specimen (x40) shows a typical cavernous malformation with endothelium-lined, sinusoidal cavities without other features of normal blood vessels, such as muscular or adventitial layer

Results

- Median age: 46 years (range, 3 mos-71 yrs), 27 pts (18 men, 9 women)
- CN III in 17 (63.0%), CN VI in 2 (7.4%), and CN IV in 8 (29.6%)
- After surgery, functional changes revealed **improvement in 9 (40.9%), no change in 10 (45.5%), and worsening function in 3 (13.6%)** patients.
- **Functional preservation was achieved in 12 (54.5%) of 22 patients.**
- **GTR or STR and nerve continuity preservation** showed a better result for postoperative **functional improvement ($p = 0.008$) and preservation ($p = 0.004$)** than other surgical treatment methods with a statistical significance ($p = 0.008$). GTR and nerve transection did not result in improvement.

Table 1 Literature review of cavernous malformation affecting the ocular motor cranial nerves

Author & year	CN	S/A	Side	Treatment	Nerve function		Functional change	Functional preservation
					Pre-Tx	Post-Tx		
Blizzard (2018) [1]	III	M/3mon	Lt	Conservative care	Incom	Improved Incom	Improved	Yes
Diaz (2010) [2]	III	M/46	Lt	GTR & n. continuity preservation	Incom	No deficit	Improved	Yes
Inui (2016) [3]	III	M/51	Rt	GTR & n. continuity preservation	Com	Incom	Improved	Yes
Itshayek (2007) [4]	III	F/25	Rt	Exploration (no resection)	No deficit	No deficit	Unchanged	Yes
Lombardi (1994) [5]	III	F/46	Lt	GTR & n. continuity preservation	Com	Incom	Improved	Yes
Matias-Guiu (1990) [6]	III	F/36	Lt	STR & n. continuity preservation	No deficit	Incom	Worsened	Yes
Muzumdar (2001) [7]	III	M/45	Lt	GTR & n. continuity preservation	Incom	Improved Incom	Improved	Yes
Obaid (2014) [8]	III	M/71	Lt	GTR & n. continuity preservation	Com	Com	Unchanged	No
Ogilvy (1993) [9]	III	M/25	Rt	GTR & n. transection	Com	Com	Unchanged	No
Park (2005) [10]	III	M/33	Lt	STR & n. continuity preservation	Incom	Improved Incom	Improved	Yes
Patro (2008) [11]	III	M/54	Rt	Conservative care	Com	Incom	Improved	Yes
Rotondo (2014) [12]	III	F/34	Rt	GTR & n. continuity preservation	Incom	Improved Incom	Improved	Yes
Scott (1983) [13]	III	M/14	Rt	GTR & n. transection	Com	Com	Unchanged	No
Wolfe (2011) [14]	III	M/26	Rt	STR & n. continuity preservation	Com	No deficit	Improved	Yes
Wolfe (2011) [14]	III	F/69	Lt	GTR & n. continuity preservation	Com	Com	Unchanged	No
Yamada (1986) [15]	III	M/33	Lt	GTR & n. transection	Com	Com	Unchanged	No
Present patient (2019)	III	M/43	Rt	STR & n. continuity preservation	Com	Incom	Improved	Yes
Diaz (2010) [16]	VI	M/56	ND	ND	ND	ND	Improved	Yes
Moon (2011) [17]	VI	F/54	Rt	GTR & n. transection	Incom	Com	Worsened	No
Bassetti (1994) [18]	IV	F/50	Lt	GTR & end to end anastomosis (and/or n. graft)	Com	Com	Unchanged	No
Grafteo (2017) [19]	IV	M/57	Lt	GTR & n. transection	Com	Com	Unchanged	No
Kraschl (2014) [20]	IV	M/70	Rt	GTR & end to end anastomosis (and/or n. graft)	Com	Incom	Improved	Yes
Lombardi (1994) [21]	IV	F/50	Rt	GTR & end to end anastomosis (and/or n. graft)	Com	Com	Unchanged	No
Manjila (2011) [22]	IV	F/31	Lt	GTR & n. transection	No deficit	Com	Worsened	No
Sindou (1992) [23]	IV	M/65	Rt	GTR & end to end anastomosis (and/or n. graft)	No deficit	No deficit	Unchanged	Yes
Sürücü (2007) [24]	IV	M/53	Lt	GTR & end to end anastomosis (and/or n. graft)	Com	No deficit	Improved	Yes
Yaghi (2011) [25]	IV	M/70	Lt	ND	ND	ND	ND	ND

CN: cranial nerve, GTR: gross total resection, STR: subtotal resection, Incom.: incomplete nerve deficit, Com.: complete nerve palsy, ND: not described, n.: nerve, S/A: sex/age in years (unless specified), M: male, F: female, Rt: right, Lt: left, Tx: treatment.

Table 2 Functional outcomes after surgical treatment for cavernous malformation of the ocular motor cranial nerves (n = 22)

Nerve function	GTR/STR & nerve continuity preservation (n = 11)	GTR & anastomosis (n = 5)	GTR & nerve transection (n = 6)	p-value*
Improved: Unchanged: Worsened	8: 2: 1	1: 4: 0	0: 4: 2	0.008
Functional preservation (Yes: No)	9: 2	3: 2	0: 6	0.004

* Fisher's exact test was used for this analysis

Conclusions

Based on the systematic review, we recommend surgical treatment for patients with progressive deficits due to CMs involving the ocular motor CNs. A prompt surgery must aim to achieve nerve continuity preservation for functional recovery and preservation of ocular motor CNs.

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