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Intraventricular meningiomas: A case series and literature review.

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ABSTRACT… Objective: Intraventricular meningiomas (IVMs) are rare type of meningiomas. Majority of IVMs are located in lateral ventricles. **Study Design:** Case Series. **Setting:** Civil Hospital Karachi. **Period:** January 2013 to January 2018. **Material & Methods:** 15 patients were assessed with histologically verified IVMs, clinical features, radiological findings, surgical approaches, outcome and literature review. **Results:** The most common presentations included raised intracranial pressure (66.7%), visual deficits (40%), cognitive changes and dysphasia. All lesions arose in the lateral ventricles. Preoperative diagnosis was confirmed on MRI. Excision was performed using the posterior parietal and parieto-temporal approach for lateral ventricle tumors. Total excision was done in 13 out of 15 patients and two patients with residual tumor underwent stereotactic radiosurgery. Biopsy report showed WHO grade-I lesion in all cases. Postoperative complications included CSF leakage, transient hemiparesis and dysphasia. Glasgow Outcome Score of 5 was found in majority of cases (87%) on follow-up. **Conclusion:** These results depict that IVMs can be excised completely with minimum postoperative morbidity. However, resection requires planning to avoid eloquent cortical damage.

Key words: Intraventricular, Meningioma, Surgical Approaches, Total Excision, Outcome.

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INTRODUCTION

Meningiomas are the most common benign primary intracranial tumor, approximately 35.9% of all primary intracranial neoplasms.[1] It is predominant in females, with female to male ratio of 1.8:1. Intraventricular meningiomas (IVMs), the only intra-axial meningiomas, are rare type of meningiomas. It represents 1-2 % of all meningiomas.[2] Majority of IVMs are located in trigone of lateral ventricles, preferably on left side.[3]

Intraventricular meningioma (IVM) has three distinctive features that differentiates them from other meningiomas. Firstly, they are extremely rare compared to other locations. Secondly, they have no dural attachment. And lastly, they present a neurosurgical challenge because of their location, where complete tumor resection without complications such as visual field defects may be challenging due to the proximity to the optic radiations.[5]

Intraventricular meningiomas (IVM) are uncommon tumors and this is evidenced by the limited worldwide published data. According to a recent Indian study, the ventricular system provides enough room for tumors to enlarge, so smaller tumors do not present early. However, presentations of larger lesions vary from raised intracranial pressure, with or without localising signs, to visual and cognitive impairment.[3,4] This fact was also supported by another larger case series of 42 patients by Danica Grujicic et al. which showed that 83% of the patients presented with raised ICP. Other common presentations were visual deficits (78.6%) and altered cognition (50%). Considering the surgical challenge, another significant aspect of this study was the good outcome of patients associated with total

excision; 83% patients had 6-month Glasgow Outcome Scale of 5.[6]

Regarding management, the treatment of choice for intraventricular meningiomas remains surgery. Grujicic et al, Ma et al and Nanda et al agreed upon transcortical parieto-occipital and transtemporal routes as the most recommended options for sufficient safe resection.[6,8,9] To add this, Faquini et al concluded in his series that these approaches can also be employed for total resection with minimum postoperative morbidity, even in absence of high tech tools.[7]

The availability of limited data and scanty research papers depict clearly the rarity of pathology, specifically in our population. So in this case series, we share our experience of 15 patients with biopsy proven intraventricular meningiomas, the clinical features, radiological findings, management, outcome and literature review for better understanding of this pathology and further, this will help in better management of such leisons.

MATERIAL & METHODS

This case series was conducted from January 2013 to January 2018. Patient's consent for study was taken at time of admission. In this retrospective series, we assessed the case records of 15 patients diagnosed as intraventricular meningioma. These patients were admitted in Neurosurgery ward, Civil Hospital Karachi and underwent treatment during a period of 5 years. Out of 15 patients, 10 were male and 5 female. Mean age at presentation was 37 years. We evaluated clinical presentation, radiological features, surgical procedures plus their complications, histological diagnoses, and postoperative outcome. Preoperative diagnosis was confirmed on Gadolinium enhanced MRI images. Surgical excision was treatment of choice for all patients, by using parieto-occipital (trans-sulcal) and parieto-temporal approaches. Histopathological diagnoses were made according to WHO grading by histopathologist. Postoperative outcome was analyzed with the help of Glasgow Outcome Scale (GOS). Only those patients with biopsy proven IVMs were included in the study. Patients with co-morbidities were excluded.

An extensive review of literature from January 2003 to January 2018 was done. From the reviewed case series, information regarding patient age, gender, presentation, radiology, treatment, complications and outcome was extracted.

RESULTS

In this study, 15 patients were included with a mean age of 37 years at presentation (range 27-50 years). Male-to-female ratio was found to be 2:1.

The most common presenting symptoms were headache 15(100%), nausea/vomiting 10(67%), blurring of vision 10(67%), visual field deficit 6(40%), impaired cognition and speech difficulty. Duration of symptoms range from 2-8 months. On neurological examination, the most common signs were papilledema 10(67%) and contralateral homonymous hemianopia 6 (40%). Diagnoses were confirmed on contrast enhanced MRI brain. All IVMs originated from lateral ventricles. The MR images revealed the tumor to be hypo- to isointense on T1-weighted images, and dense enhancement with gadolinium. Enhancement was homogenous in 11(73.33%) cases and heterogenous in 4 cases. Maximal tumor size ranged from 3-7.0 cm, with a mean size of 4 cms. CT scan showed calcifications in 4 cases and hydrocephalus in 10 (66.67%) cases. All patients underwent surgical excision. Total excision was done in 13 (86.67%) out of 15 cases and two patients with residual tumor underwent stereotactic radiosurgery afterwards. Excision was performed using the trans-sulcal parieto-occipital and transtemporal approaches for lateral ventricle tumors. All of the tumors were in the atrium (trigone) of the lateral ventricle, firm to hard in consistency. Intracapsular debulking of tumor was done with loop diathermy in piecemeals before separating from surrounding structures. Vascular pedicle (posterior choroidal artery) was identified, coagulated and divided. Postoperative complications included CSF leakage in 2 cases and transient hemiparesis and dysphasia in 2 cases, which improved gradually

in 2-3 months duration. Surgical mortality was 0%. Histopathology report showed WHO grade I lesion in all cases. On 6 monthly follow-up, thirteen patients (87%) showed Glasgow Outcome Score (GOS) of 5, while the remaining two patients who underwent SRS for residual tumor had GOS 4. Improvement was noted in ICP, visual fields and dysphasia.

Authors	Cases N	Sex M:F	Age Range	Symptoms				Signs	
				H/A	N/V	SD	IC	PE	VFD
Nakamura et al. 2003	16	1:1	25-77	8	-	3	2	4	7
Bhatoe et al. 2006	12	3:1	30-50	12	-	-	-	8	-
Lyngdoh et al. 2007	9	1:1.3	12-60	7	7	3	4	8	4
Menon et al. 2009	15	1:2	14-75	10	-	-	-	10	6
Ødegaard et al. 2012	22	1:2.7	26-81	16	9	-	-	-	2
Ma et al. 2014	43	1:2.6	14-61	23	4	2	2	-	4
Faquini et al. 2015	4	1:3	17-45	4	-	-	-	3	1
Nanda et al. 2016	18	1:1.25	26-87	11	1	-	-	8	2
Grujicic et al. 2017	42	1:2.5	1-67	35	35	-	21	35	31
Present study	15	2:1	27-50	15	10	4	4	10	6
Total	196	1:2	1-87	141	66	12	33	86	63

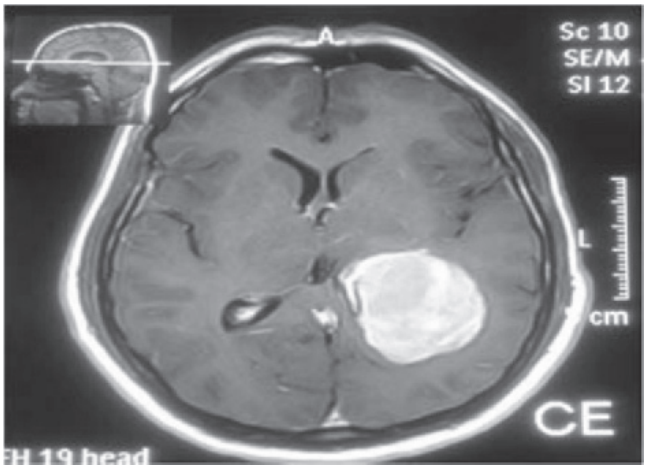
H/A= headache; N/V = nausea/vomiting; SD= speech difficulty; IC= impaired cognition; VFD = visual field defects; PE = papilledema

Table-I. Demographics and clinical presentations of different Case series from 2003-2018.

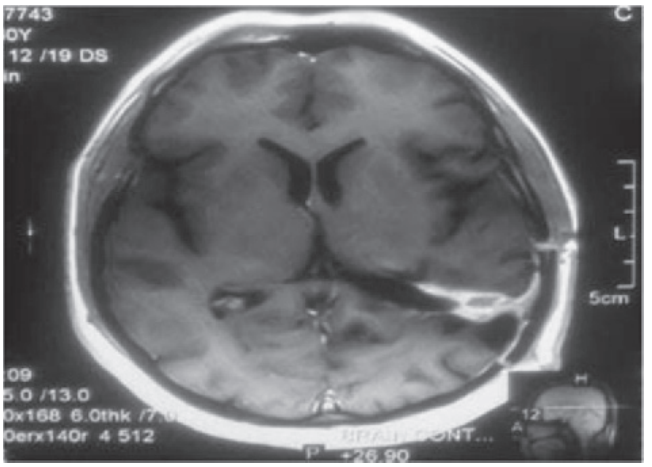
Authors	Cases N	Location	Side	Size cm	HCP	Surgical approach	Total removal	Recurrence	Mortality
Nakamura et al. 2003	16	L=13, III=1 IV=2	Lt=9 Rt=4	2-7	14	PO=10 TC=3 FT=1 SO=2	15	1	-
Bhatoe et al. 2006	12	L=9 III=1 IV=2	Lt=7 Rt=2	-	-	PO=9 FT=1 SO=2	12	-	1
Lyngdoh et al. 2007	9	L=7, IV=2	Lt=5 Rt=2	4.5-7.4	8	PO=2 TT=5 SO=2	8	-	0
Menon et al. 2009	15	L=15	Lt=8 Rt=7	-	-	PO=11 TT=4	13	2	-
Ødegaard et al. 2012	22	L=20, III=1 IV=1	Lt=8 Rt=12	1.9-7	11	PO=20 FT=1 SO=1	21	1	-
Ma et al. 2014	43	L=43	Lt=16 Rt=27	1.7-8.2	6	PO TT	43	0	-
Faquini et al. 2015	4	L=4	Lt=1 Rt=3	>3	3	PO=4	4	0	0
Nanda et al. 2016	18	L=15, IV=3	Lt=8 Rt=7	-	8	PO=12 TT=1 TC=1 SO=3	17	4	-
Grujicic et al. 2016	42	L=40, III=2	Lt=25 Rt=15	1-10	-	PO=20 TT=7 TP=12 TC=3	39	1	0
Present study	15	L=15	Lt=8 Rt=7	3-7	10	PO=8 TT=7	13	-	0
Total	196	L=181 III=5 IV=10	Lt=95 Rt=86	1-10	60	PO=96 TT=24 TP=12 TC=7 FT=3 SO=10	185	9	1

HCP= hydrocephalus; L= lateral ventricle PO= parieto-occipital; TT= trans-temporal; TC= transcallosal; FT= frontal transcortical; TP= temporo-parietal; SO= suboccipital

Table-II. Tumor features, surgical approach and outcome of different case series from 2003-18.



Preoperative image



Postop Image

Figures-1.

DISCUSSION

As IVM is one of the most infrequent type of meningioma, therefore, there is limited number of published clinical series in the literature, with most of them reporting relatively few patients indicating the rarity of pathology. Shaw, in 1854, first described IVM as a tumor arising at right trigone.[10] Further, in Harvey Cushing's case series of 313 meningiomas, only 1% was intraventricular.[11] In the following years, various case series were published, but included less number of patients. In 1965, Delandsheer thoroughly reviewed these cases and identified 175 cases of lateral ventricular meningiomas.[12] In 1986, Criscuolo and Symon further identified 400 cases in the world literature.[13] To add this, few recent case series have been published; including Nakamura et al, who revealed 132 additional cases of IVMs in 2003; Ma et al, who illustrated the surgical approaches of 43 meningiomas of trigone and Grujicic et al. who presented case series of 42 patients with IVMs in 2016, resulting in a total of 617 cases.[2,6,8] Amongst them, 497 occurred in the lateral ventricles (80.5 %), 85 in the third ventricle (13.7%), and 35 in the fourth ventricle (5.6%). Tables-II summarizes the main findings of published case series of patients with IVMs since 2003.

Regarding origin of IVMs, it has been elaborated in literature that arachnoid cell nests are present in the normal choroid plexus stroma, and examination of the choroid plexus usually unveils collections of these cells.[14] IVM in the trigone is presumed to arise from its choroid plexus (present in inferio-medial wall of the trigone). The initial growth of tumor is from the choroid plexus into the lumen of the ventricle. The vascular pedicle, comprising of anterior or posterior lateral choroidal arteries, enters tumor from an inferomedial direction. The venous drainage of the trigone is made by the medial and lateral atrial veins.[7] Optic radiation, originating from lateral geniculate nucleus, pass over the roof and the lateral wall of the temporal horn as well as on the inferolateral aspect of the atrium.

Regarding gender of patients, literature shows female predominance for IVMs ranging from 41 to 82%, with ratio of 2:1 for female-to-male.[15] However, in our series we identified a male predominance of 66.7 % with a ratio of male to female of 2:1. Further risk increases as age progresses.[16] In our series, the age ranged from 27-50 years and the mean age was 37 years.

According to literature review, IVMs enlarge gradually and acquire larger size before symptoms develop, unless the location of tumor within ventricular system causes obstruction of CSF circulation. According to Cushing and Eisenhardt (1938), there are five clinical presentations of IVMs in trigone: (1) raised ICP symptoms; (2) contralateral homonymous hemianopia; (3) contralateral sensorimotor deficit; (4) cerebellar affection (>50%), and (5) dysphasia and/or

paralexia in left-sided tumors.[11] Also, Winkler suggested that pathophysiology may include both causes, i.e., intracranial hypertension as well as mass effect on surrounding parenchyma.[17] In our study, the clinical presentations complied strongly with these theories. Regarding duration of symptoms, the reported range is from a few months up to 20 years.[11] However, in our series, duration of symptoms were comparatively smaller, ranging from 2 to 8 months.

In the literature review, different diagnostic modalities were illustrated, yet in recent times, CT and MRI are the most accurate diagnostic choice, with later being more useful in terms of detailed brain anatomy, lesion location and surgical planning. According to a review of 400 intraventricular meningiomas by Criscuolo and Symon, majority of the tumors arose within the lateral ventricles (80%), followed by third (15%) and fourth (5%) ventricle, respectively.[13] This stratification correlates well with the amount of choroidal plexus tissue within these ventricles. In our series, all of the IVMs arose from lateral ventricles. Further, in published cases, they are found to be more common on the left side, as shown in Table-II. Rather in our series, significant laterality was not identified (53.3% on left side).

IVMs have a classic radiologic appearance; well circumscribed but the typical dural attachment is absent.[18] On CT scan, they are usually hyperdense with homogenous contrast enhancement and may have calcified areas (47%) and hydrocephalus.[15,6] Further, localized hydrocephalus can be visualzed in ipsilateral trigone and temporal horn in IVMs of lateral ventricle origin.[18] In our case series, calcifications were not that common (27%) but hydrocephalus was significant (66.7%); more than Odegard et al and Nanda et al series (50, 44%) as in Table-II. MRI appearance of IVM was similar as reported in literature; usually iso- to hypointense on T1WI and iso- to hyperintense on T2WI along with dense enhancement on contrast.[19] Further, it is reviewed that precise knowledge of cortical anatomy is obligatory to undergo safe resection. Therefore in recent advances of neuroradiology, the use of DTI is quite noteworthy, for delineating the relationships between tracts and tumors, specifically for parenchymal lesions.[20] However this privilege of diffusion tensor imaging was lacking in our setup, but our complication rate was comparatively similar to different series utilizing such resources.

Total surgical resection of IVMs is the treatment of choice, yet challenging enough due to its deep seated intraventricular location, the proximity of motor, sensory, and language cortex, plus optic radiation and the vascular structures. According to Fusco and Spetzler, the choice of the surgical approach is defined by the best route to long axis of tumor, minimization of transcortical transgression, preoperative neurological deficit, and proximity to eloquent areas and tracts.[21] So in our series, tumor resection was done via transsulcal parieto-occipital (PO) approach in 8 patients and transtemporal (TT) approach in 7 patients. Also, these approaches were identified as the most common employed routes in literature of 196 IVM resections in last 15 years; PO 49% and TT 12%. Further, in our cases, total excision was accomplished in 13 (87%) comparable with Grujicic et al and Nanda et al (93%, 94%). Table-II.

Regarding the precision and safety of approach, the use of intraoperative ultrasound and neuronavigation improves accuracy in locating and excising the tumor completely, with limited intraoperative complications and post-operative neurological sequelae.[8] In our cases, trajectory was entirely based upon anatomical knowledge and neuroradiological assessment. Still extent of excision, complication rate and outcome was comparable with the neuronavigation guided resections.

According to Grujicic et al, mortality rates ranged from 0 to 42% in previous case series. Subsequently, in last 15 years, published data revealed a significant decline in operative mortality. In our series of 15 patients, surgical mortality was 0%. On 6 monthly follow-up, 87% showed Glasgow Outcome Score of 5. It was in accordance with Grujicic et al series (83.3%).[6]

CONCLUSION

IVMs commonly acquire larger dimensions

before being diagnosed. Total excision should be the main goal. The main challenge is to resect IVMs in the trigone, while avoiding damage to geniculucalcarine tracts. So, the surgical approach should be precisely planned, in accordance with tumor location. Parieto-occipital and trans-temporal approaches are the most commonly recommended surgical routes.

Although precision can be improved by mapping these tracts preoperatively by DTI based tractography and intraoperatively by neuronavigation and evoked potential monitoring, yet application of core anatomical and neuro-radiological knowledge can also lead to accomplishment of comparable results.

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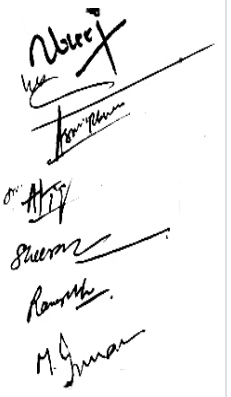
1	Urooj Fatimah Siddiqui	Study Design.	
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