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Case Reports & Case Series

Microsurgical excision of giant dominant lobe insular cavernoma presenting acutely: Sometimes you win, sometimes you learn

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ABSTRACT

Introduction: Eloquent location and difficult access make insular surgery challenging. Thus operative adjuncts like neuronavigation, intraoperative monitoring and awake surgery are now considered the standard of care for these lesions. However, these may not be possible in all cases and resource constraints may place limits to what could be achieved. There still remains a possibility of achieving reasonable results with good surgical technique and meticulous surgery.

Case report: We report 2 cases of giant (> 6 cm) left insular cavernoma, excised using pterional-transylvian-transinsular approach, in a resource constrained and emergency setting. Both the cases had a sudden presentation with one case presenting with sudden increase in hemiparesis while the other presented with headache resulting from bleed in the background of a history seizures for 6 months. While clearing the clot in the first case, the hemosiderin rim was removed while it was deliberately avoided in the second patient. While the first case developed neurological worsening after surgery, eventually improving at follow-up, the postoperative course was uneventful in the second patient. Postoperative scan revealed a complete cavernoma excision in both the patients.

Conclusion: Operating outside ideal conditions is always challenging, but not impossible. Spontaneous hemorrhage in and around these lesions may also be a blessing in disguise during surgery. While we do not advocate this as the ideal methodology, emergent conditions and lack of gadgets often necessitate evasive action and push ourselves. A combination of sound anatomical knowledge and experience in operating in this area can allow their safe excision with considerable safety. Considering the global neurosurgical burden and paucity of skilled manpower and resources, an undertaking of such a nature might be helpful to other readers in similar conditions.

1. Introduction

Insula, the fifth cerebral lobe, is not just a hidden structure anatomically, its arcane nature percolates into the diverse array of functions it is involved with. The indispensability of the adjoining neurovascular structures adds further to the surgical challenge. These include the middle cerebral artery (MCA) with its important branches and perforators on the lateral as well as the medial surface of the insula, the close vicinity of the internal capsule and centrum semiovale carrying important descending motor fibres and important language related association fibres adjoining the perinsular sulci on the dominant side [1]. Attempts of resection of pathologies inside the insula has an inherent risk of damage to these adjoining structures, particularly in sizeable and infiltrative lesions [2]. That is the reason why maximal safe resection of insular gliomas is generally not possible without awake surgery and intraoperative neuromonitoring [2]. In developing countries, the treating neurosurgeon faces certain a unique situations when faced with the microsurgical excision of these lesions. The vital gadgets like neuronavigation, intraoperative ultrasound, intraoperative neuromonitoring system etc. are not readily available in many centers, there is a lck of trained personnel to operate this gadgets, the patients often come from lower socio-economic background and fail to comply during awake craniotomies and most importantly, the tertiary care neurosurgery centres are over burdened with a plethora of neurosurgical cases.

Abbreviations: MRI, magnetic resonance imaging; CT, computed tomography; CA, cavernous angioma; IC, insular cavernoma; MRC, medical research council; MCA, middle cerebral artery

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Table 1
Literature review of 25 reported cases of surgically managed insular cavernomas.

S.No.	Author	Cases	Side	Age (years)	Presentation	Approach	Additional tool	Outcome	Size (mm)
1	Bertalanffy et al. (1991)	6	L	43 M	GnSz	Trans-sylvian	None	Intermittent FSz	20
			R	42F	CPSz			Frequency dec	15
			R	55F	HHeminop			Hemiper,HemiHy	20
			R	49 M	L heminopsia			hemianopsia worsened; transient left hemiparesis	30
			R	63F	Vertigo			transient left hemi- hypaesthesia	10
			L	55 M	L hemiperesis			Thalamic infarct; motor deficit	15
2	Casazza et al. (1996)	3	NM	NM	NM	NM	NM	Removal of the hemosiderin ring did not correlate with better outcome	NM
3	Tirakotai et al. (2003)	8	3 L:5R	33–58 (mean = 45.3) 3 M:5F	Headache(3); hemihypesthesia (2); hemiparesis (1); GnSz (1); CPSz (1); memory disturbance (1)			No deficit and all Sz control	20 (1); 15 (3); 12 (1); 10 (1); 4 (1)
4	Duffau et al. (2005)	1	L	33 M	Sz (History of trauma)	Fronto parietal flap Trans-sylvian	NN with intra operative mapping	Sz free (no AEDs)	7
5	Esposito et al. (2006)	1	L	34F	Recurrent headache	Tans sulcus between the triangular and opercular parts	NN with MRI 3D corticotopography	No deficit	11
6	Roberto et al. (2010)	4	R	48F	GnSz	Trans-sylvian	Intra operative USG (4) and NN (3) awake craniotomy with cortical mapping (1)	Sz free (no AEDs) [2]	NM
			L	26F	Hemiparesis;aphasia; nausea and vomiting			And decreased Sz frequency [2]	
			R	40F	dysesthesia; right facial palsy; nausea and vomiting				
			L	41 M	CPSz; Hemiparesis;aphasia;				
7	Matsuda et al. (2012)	2	L	36F	Sz	Total excision with partial excision of gliosis in one and total in another	Awake craniotomy and subcortical mapping		NM
				33 M				Sz free (no AEDs)	

Thus, more often than not, surgery for lesions in areas like insula are carried out without the luxury of the gadgets that have become “mandatory” across the developed countries.

Cavernous angiomas represent a relatively uncommon cerebrovascular anomaly [3]. The insula is a rare site for these lesions with limited reports available in the literature (Table 1). Giant cavernomas are defined as those that measure > 6 cm. The size of intracranial lesion is always an important surgical consideration and giant cavernoma in the dominant insular lobe is, needless to say, represents a formidable situation. We hereby present two cases of giant IC managed microsurgically without intraoperative monitoring under general anesthesia. The idea is to highlight two specific things: 1. It is possible to excise these lesions in resource constrained set-ups and 2. Awareness of the critical points during surgery can avoid intraoperative complications.

2. Methods

We report two cases of large to giant insular cavernoma (> 5 cm) among 28 patients of supratentorial CA [M:F = 18:8] operated at our center between January 2012 and December 2018. Both lesions were on the dominant side and histopathologically confirmed as cavernous hemangiomas. The extent of excision was confirmed by postoperative magnetic resonance imaging (MRI) after 3 months of surgery. Patient consent for publication and institute ethics committee approval were taken for this study.

2.1. Case descriptions

2.1.1. Case 1

A 47-years-old lady presented with progressive right hemiparesis for 4 months with sudden severe headache and acute worsening of hemiparesis 2 days prior to the admission. There was no prior history of seizures. On examination, she had right hemiparesis (power of 3/5, Medical Research Council grade) with hyperreflexia. Cranial MRI showed a well-defined lesion (maximum dimension 6.5 cm along vertical plane) occupying the entire left insular lobe. It was hypointense on T1 and hyperintense on T2 WI, showing a uniform post-contrast enhancement. A prominent hemosiderin rim was seen all around the lesion, intermingling with the internal capsule medially (Fig. 1 A–C). We operated on her using a left pterional-transylvian-transinsular approach. Working between the branches of the MCA, insular corticectomy was performed and the cavernoma was excised completely. There was intraoperative evidence of hemorrhage in certain areas of the cavernoma. While removing the clot the hemosiderin rim was also evacuated. In the immediate post-operative period, the patient had aphasia and right hemiplegia. Histopathology suggested the diagnosis of cavernous hemangioma. In the course of follow up (28 months), her aphasia and hemiplegia gradually improved (MRC grade 4/5). A follow up MRI suggested complete excision of the cavernoma along with a well-defined cavity (Fig. 1D-F).

2.1.2. Case 2

An 18-year-old boy presented with recurrent episodes of generalized tonic clonic seizures for 6 months and sudden severe headache and vomiting 15 days prior to admission. On examination, there was no neurological deficit. Cranial computed tomogram (CT) suggested a subacute hematoma in the left insula-basal ganglia area with compression of the ventricles (Fig. 2a). On MRI, the lesion was multicystic, 6.2 cm in transverse dimension, heterogenous in intensity on T1 and hyperintense on T2 WI (*Zabramski Type I*), the latter showing fluid levels inside the lesion (Fig. 2b–d). It was capped laterally by an elongated, T1 iso, T2 hyperintense zone, suggesting acute hemorrhage. Compared to the previous case, the posterior part of the insula was free and the medial compression was mainly on the caudate nucleus, anterior limb and genu of the internal capsule. The posterior limb of the internal capsule was not so severely compressed/affected by the lesion or the hematoma. Superiorly, the lesion was compressing the coronal radiata. This patient was also operated using transylvian trans-insular corridor. Immediately after cortisectomy the bulging and yellowish discolored insula, hematoma surrounding the cavernoma extruded under pressure. Working along the hematoma, we encountered the dusky red mulberry like cavernoma. It was dissected from all around and removed completely. Learning from the previous case, the gliotic and hemosiderin laden rim abutting the internal capsule was intentionally left behind medially. The patient recovered without any deficits postoperatively. Immediate postoperative CT as well as follow-up MRI showed complete excision of the cavernoma and an intact internal capsule (Fig. 2e–h). The hemosiderin rim, left intentionally during surgery, could be seen clearly in the postoperative MRI (Fig. 2f). Histopathological examination of the resected specimen was consistent with the diagnosis of cavernous hemangioma. At 12 months' follow-up, the patient was seizure free and functionally intact.

Both the cases continue to be in regular follow-up. Considering the emergent nature of the presentation and deteriorated condition, outcomes have been satisfactory.

3. Discussion

Intracranial cavernous malformations are relatively uncommon congenital vascular anomalies. Pathologically, these represent a bunch of sinusoidal vessels without any draining vein or feeding artery. These do not have neural tissue incorporated in to the mass. Most often, these affect the superficial parts of the cerebral hemisphere and rarely affect deeper locations like the insula, basal ganglia or the brainstem [4]. Propensity for bleed, expansion by microhemorrhages, mass effect on the surrounding structures and perilesionaln epileptogenesis are the major considerations for treating these lesions [5]. Venous angiomas are often associated with these lesions and their role in the occurrence as well as recurrence in CM has been described previously [6]. These lesions are angiographically occult and need MRI to decipher their details. Their appearance on MRI depends on the presence and stage of the bleed as noted by Zambraski [7].

Insula, a part of the mesocortex, forms an anatomical and functional interface between the neocortex and the allocortex. It remains covered by the overlying fronto-temporo-parietal opercula. Intimate relationship with the MCA, its branches and important perforators, presence of internal capsule and corona radiata superomedially and important association fibres related to language function along the limen insula and peri-insular sulcus on the dominant side, combine to make surgery in this area a daunting task [1].

Insular cavernomas are rarely reported. Literature review revealed 25 odd cases reported so far [8]. Fewer number of large to giant ICs causing mass effect or hemorrhage have been reported. It is well documented that deep seated CMs have a higher propensity for bleed. Moreover, in certain locations, the presence of compact white matter bundles like the internal capsule (insular-basal ganglionic CM) or long tracts (brainstem) often predispose to neurologic deficits, either acutely from even the slightest of hemorrhages in and around CMs or as seen in our patients, or by chronic compression and microbleeds (as evidenced by hemosiderin rim affecting the internal capsule). Seizures are the most common mode of presentation of intracranial CMs. Insula has a great potential to become epileptogenic in diseased states like low grade glioma [9]. Thus, like our second patient, IC producing seizures is not uncommon.

Despite disagreements on the best course of management of supratentorial cavernomas, giant IC causing symptoms remains an undebatable indication for surgery. Literature on IC surgery have reported complications in the range of 1–14% [10,16]. Most complications are attributable to vascular injuries or mechanical injuries to the adjoining white matter tracts. Therefore, a number of intraoperative adjuncts have come into play in insular surgery of late. These include intraoperative monitoring like sensory/motor evoked potentials for

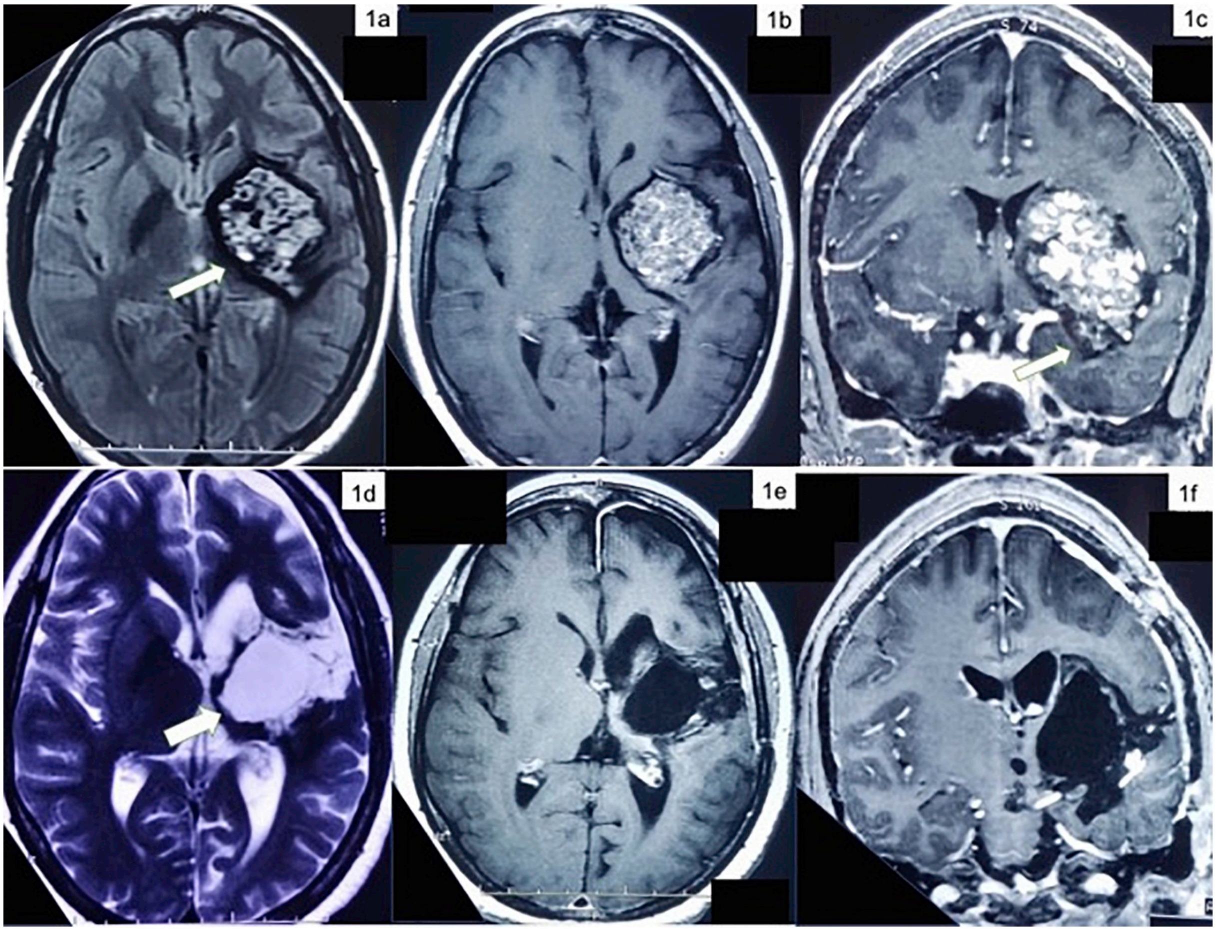

Fig. 1. On T2W FLAIR (a) and T1WI contrast axial image (b), the lesion was occupying the entire insula with a well-defined hemosiderin rim all around and involving the internal capsule (shown with arrow, 1a). Coronal section showed uniform contrast enhancement and involvement of the entire insula by the cavernoma of approximate size 6.5 cm (c). Postoperative MRI shows surgical cavity and complete cavernoma excision (d,e,f). Only a small rim of tissue (marked with arrow) is present medial to the surgical cavity suggesting a possible damage to the internal capsule (d).

subcortical tracts, direct cortical stimulation for cortical mapping as well as subcortical mapping [11]. Table 1 shows the major surgical series' on IC and their results.

In developing countries, the necessary gadgets for operating on these patients is not always available. Then there are issues related to trained man power, doctor-patient ratio, poor socio-economic patient background often precluding ideal intraoperative patient assessment and awake craniotomy. Thus, many a times, the neurosurgeon faces the challenge of operating on such daunting lesions in sub-optimal situations. In this regard, adherence to meticulous neurosurgical techniques and exercising caution at critical areas during the surgery can improve the patient outcome. We intend to highlight these operative nuances and our own experience with such lesions.

Operative morbidities in insular surgery in general are largely due to the suboptimal surgical exposure including the MCA branches, opercular retraction and the lack of timely identification of "at risk" structures like the lenticulostriate perforators and the internal capsule. A standard pterional craniotomy is sufficient to expose the sylvian fissure and the adjoining opercula. Neuronavigation is useful but not mandatory for this purpose. Moreover, release of CSF after sylvian fissure opening makes its use after duratomy a lot less reliable, although Dorward et al. have argued against this point [12]. Proper patient positioning is necessary to allow a wide splitting of the sylvian fissure, allowing frontal and temporal lobes to separate away. Depending on the part of the insula to be targeted, the degree of head rotation will vary, a step where neuronavigation is not mandatory. It is important to visualize the MCA early in the surgery. Corridors developed between the MCA branches after coagulation of the short insular perforators provide direct access inside the insula. At this stage, some surgeons use intraoperative ultrasound to decide the surgical trajectory. However, we have noticed a bulge and discoloration of the insula in both our cases. A small corticectomy generally brings into view the lesion or the hematoma. Thereafter, the lesion itself guides further dissection. Visualization of the lenticulostriate perforators via color doppler, as suggested by Steno et al. is very useful, especially in the deeper aspect of the lesion [13]. Intermittent visualization of the M1 segment in the sylvian fissure and microscopic visualization of deeper portion of the insula near the apex generally gives an opportunity to identify these perforators without any adjunct. This may not be as easy in cavernomas as it is in low grade gliomas. Avoidance of opercular retraction is very important, particularly on the dominant side. Drainage of hematoma early on can reduce any brain bulge and reduce the detrimental effects of retraction, if it is needed. Dynamic retraction provided by the suction cannula and intermittent use of brain spatula generally suffice. Hematoma surrounding the cavernoma helped us in creating a dissection plane all around the cavernoma in our second patient whereas a continuous bipolar shrinkage of the cavernoma and a circumferential dissection of the lesion was performed in the first patient. We need to remain within the anatomical borders of insula, demarcated by the peri-insular sulci, all around except medially. On the medial side, similar to the excision of low grade glioma, the limit is basal ganglia and its typical nutmeg appearance on the upper posterior part and the lenticulostriate perforators inferomedially [1]. It is to be remembered that the posterior lobe of the insula overlies the posterior limb of internal capsule [8].

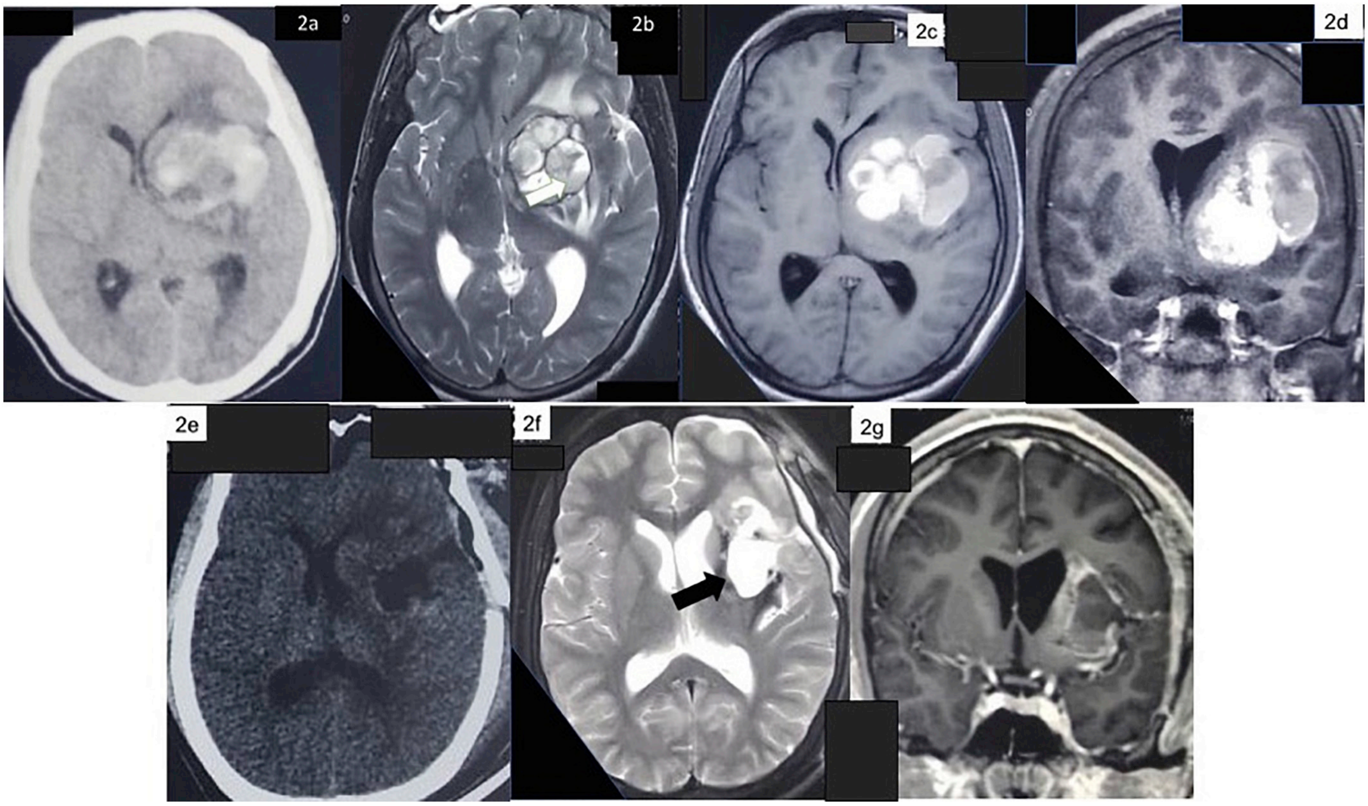

Fig. 2. Computed tomogram (a) shows a heterogeneously hyperdense lesion in the left insula-basal ganglia area with compression of the ventricles. It appears as multicystic mulberry like lesion, being hyperintense on T1WI and mixed signal intensity on T2WI, with fluid levels, affecting mainly the anterior part of the insula of approximate maximum size 6 cm (b,c,d). Post-operative computed tomogram axial brain (e) shows well defined surgical cavity and excision of the cavernoma (e). Follow-up MRI showed complete cavernoma excision, remaining hemosiderin rim along the preserved internal capsule (shown with arrow) and intact postero-inferior insula (f,g).

Moreover, it is covered by superior temporal gyrus and hence this portion of insula is not only difficult to access but also has the inherent dangers of neurological deficits after surgery.

We report here two patients of giant insular cavernoma, both presenting in emergent settings: one with mass effect and the other one with hemorrhage. The take home message here is that while both patients underwent the same procedure, they had different outcomes. In the second case, the lesion did not involve the posterior insular lobe and the posterior limb of the internal capsule was away from the lesion. We also believe that our decision against the excision of hemosiderin rim medially might have contributed considering the risks to the internal capsule and the lenticulostriate perforators. Postoperative scan clearly showed the left behind hemosiderin rim. On the other hand, the first patient had no bleed around the lesion to work around. It was occupying the entire insula and the hemosiderin rim was abutting the internal capsule itself. The fact that the patient had neurologic deficits preoperatively indicated either a compression or atleast a partial destruction of the internal capsule by the lesion, something that could be best deciphered by a preoperative tractography. This leads us to ponder if we could have avoided excision of the hemosiderin rim. Pathologically, the rim comprises gliotic tissue, stained chronically with blood breakdown products. It is not known to cause recurrence of cavernoma, unless there are venous channels as a part of the associated venous angioma. It can, however, cause seizures and hence the principles of epilepsy surgery dictate its removal [14]. However, its excision should be avoided in eloquent areas, particularly in the absence of intraoperative patient monitoring [15]. Moreover, it is the cortex, not the white matter that is involved with the production of seizures and hence the hemosiderin rim excision towards the white matter side of these cavernomas may not be attempted during surgery.

This hypothesis has also been supported while removing similar dominant lobe cavernomas and per-tumoral gliosis and hemosiderin rims have been shown to harbour function tissue [16]. While there is an argument to be made for awake surgery [17], it need to be understood that awake surgery in cases of intracranial-bleed and brain bulge is neither advisable nor was considered to be an option by our anesthesiologist. Thus despite being handicapped at many places we were able to “do more with less” and wish to highlight the same for our readers in similar conditions and encourage a safe approach to these benign but potentially dangerous pathologies.

4. Conclusion

Excision of giant insular cavernoma is possible in resource limited settings and in emergent presentations. Hematoma around cavernoma provides a surgical plane for excision. Excision of hemosiderin rim abutting the internal capsule should be avoided. Posterior insular involvement and hemosiderin rim involving the internal capsule are markers of possible intraoperative injury to the internal capsule. It is possible to resect these lesions in the absence of modern gadgets using a combination of sound anatomical knowledge, experience of surgery in this area and adherence to meticulous microsurgical techniques.

Declaration of interests

The authors also report no conflicts of interest.

Appendix A

Table A1
Contribution details.

	Contributor 1–3	Contributor 4–5	Contributor 6–7
Concepts	Yes	Yes	
Design	Yes	Yes	
Definition of intellectual content	Yes	Yes	Yes
Literature search	Yes	Yes	Yes
Clinical studies	Yes	Yes	Yes
Experimental studies			
Data acquisition	Yes	Yes	
Data analysis	Yes	Yes	Yes
Statistical analysis			
Manuscript preparation	Yes	Yes	Yes
Manuscript editing	Yes	Yes	Yes
Manuscript review	Yes	Yes	Yes
Guarantor		Yes	

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