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Stereotactic Surgery for Cavernous Malformation in Eloquent Area: Alternative in Selected Cases

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Abstract

Introduction: Cavernous malformations (CM) are infrequent vascular lesions, generally presenting with epileptic seizures and bleeding. Symptomatic or those with recurrent bleeding should be considered for surgical removal. CM of eloquent areas usually require awake surgery; when this it is not possible, other precision instruments such as stereotactic tools can be used to plan and perform safe surgeries with good functional results.

Methods: A 36-year-old male patient presented with seizures, including an episode of status epilepticus, and recurrent bleeding. MRI revealed a left frontal opercular lesion with typical characteristics of CM, close to the primary language area. The patient did not accept awake surgery, so a stereotactic system was used to define the exact point of approach.

Results: Complete macroscopic resection of the lesion was achieved without surgical complications and without functional deficits.

Conclusion: Precision neurosurgical instruments such as the stereotactic framework and the neuronavigator, can contribute to plan surgeries when other modern technology (tomography or intraoperative resonance) is not available or when the patient does not collaborate, as in this case.

Keywords: Cavernous malformations • Stereotactic surgery • Eloquent area

Introduction

Cerebral cavernous malformations (CM) are vascular abnormalities that are composed of abnormal hyalinized capillary groups surrounded by hemosiderin deposits and a gliotic margin [1,2]. CM is the most common vascular abnormality and account for 10%-25% of all vascular malformations; 70%-80% of them are supratentorial. Magnetic resonance imaging (MRI) in echogradient T2 sequence shows the typical characteristics of the lesion [3-5]. The annual risk of bleeding is 0.7-1.1% per lesion in patients without a history of bleeding, but increases to 4.5% in patients with previous bleeding [6-9]. Female sex is related to a worse prognosis [10,11].

The first description of stereotactic-guided CM resection was published by Davis and Kelly in 1990. Stereotactic procedures, whose main utility is the biopsy of brain lesions, are also indicated in the planning of surgical approaches for small brain lesions, with a precision of 1 mm [12-14]. Brain lesions located near or within eloquent areas represent a challenge for neurosurgeons because of the risks of permanent postoperative neurological deficit, with significant deterioration in the quality of life of the patient [15-17]. We present the case of a patient with a cavernoma close to the language areas, who did not accept awake surgery and whose lesion was resected using a minimal stereotactic-guided approach.

Case Presentation

A 36-year-old male patient was admitted to the emergency of our institute with a 2-year history of tonic-clonic epileptic seizures with transient postictal motor aphasia, fully controlled with levetiracetam (500 mg/12 h), and an episode of status epilepticus secondary to bleeding from the CM. Neurological and neuropsychological examination found moderate neurocognitive deterioration (alterations in attention, concentration, reading comprehension and auditory verbal and working memory) without motor or sensory deficits and preserved language.

Echogradient T2 MRI images demonstrated a left paraopecular lesion with typical characteristics of CM (lobulated lesion surrounded by a hypointense halo) (Figure 1). Functional MRI demonstrated the expression language areas (Broca) towards the anterior border of the lesion and the comprehension language area (Wernicke) towards the posterior part (Figure 2). A medial displacement of the arcuate tract was seen in the tractography of frontotemporal association pathways (Figure 3).

The patient did not accept awake surgery, stating that he was very anxious and would not tolerate this type of procedure. We reviewed the published literature in stereotactic-assisted eloquent area surgeries and decided to perform a stereotaxia-guided invasive approach.

Surgical techniques

Surgical technique is illustrated in Figure 4. We placed initially the Micromar Stereotaxy frame. The points for the pins were infiltrated with 2% lidocaine without epinephrine, and the level of the frame was previously defined by analyzing the MRI. With the frame in place, a contrast enhanced brain CT was performed using a stereotaxia protocol; subsequently the coordinates of the approach path (skin, skull, cerebral cortex) were calculated, integrating the MRI references, just behind the left frontal pars opercularis. Once the surgical field was prepared, the projection of the lesion towards the skin was marked, guided by the stereotactic coordinates. A 5 cm italic "S" incision was made in the left frontotemporal region and then a triangular craniotomy of approximately 3 cm per side was also made with stereotactic guidance, as well as finally the cortical entry point. A 5 mm corticotomy was performed, direct to the lesion as marked by the coordinates. The advantage of CM is that it is not infiltrative; it maintains a plane of perilesional gliotic tissue, which greatly facilitated resection. Usually the gliotic tissue impregnated with hemosiderin should be respected, but we decided not to do it especially towards the medial part so as not to damage white matter, due to the proximity of the arcuate fasciculus. Finally, a complete resection was achieved without procedural complications.

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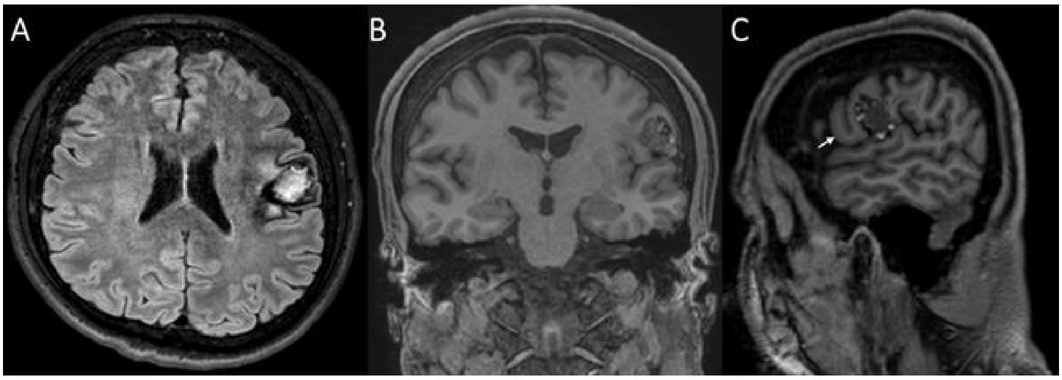

Figure 1. Preoperative MRI. A) FLAIR sequence showing a left paraopercular lesion with perilesional hypointensity B) and C) T1 without contrast with a lesion close to the left frontal pars opercularis (white arrow).

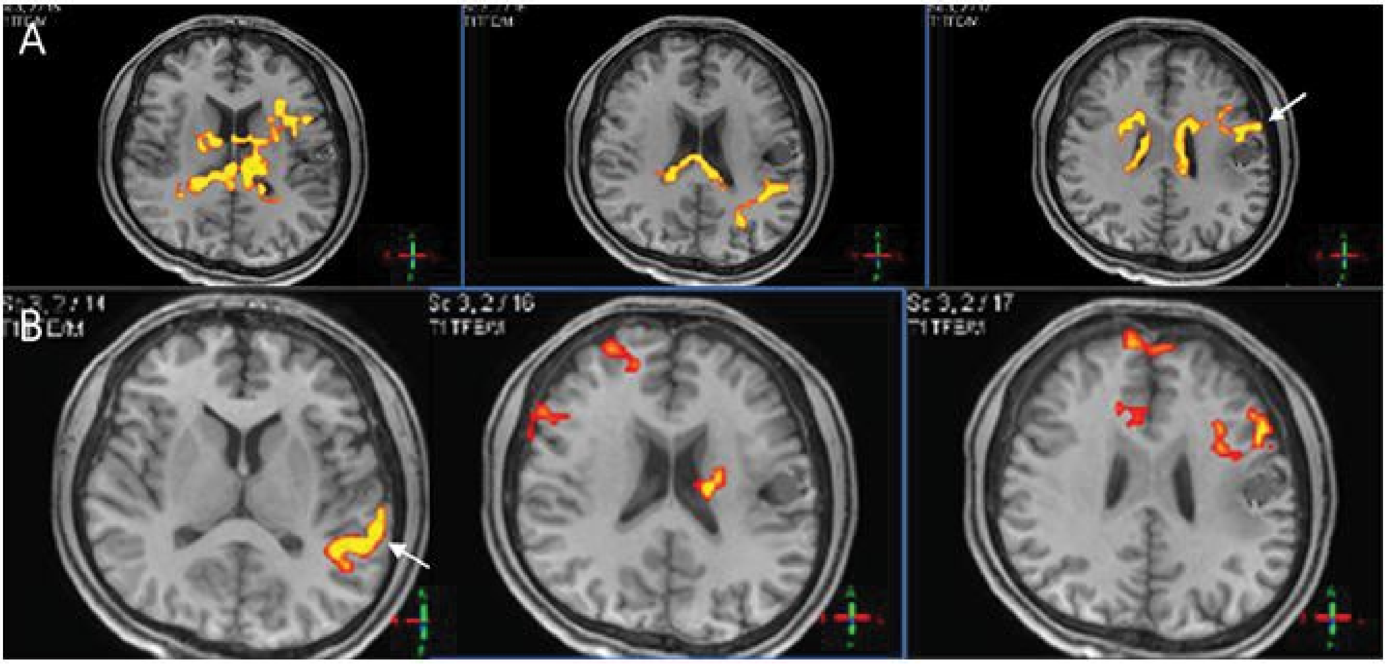

Figure 2. Functional mri: A) broca's area (white arrow) and B) wernicke's area (white arrow).

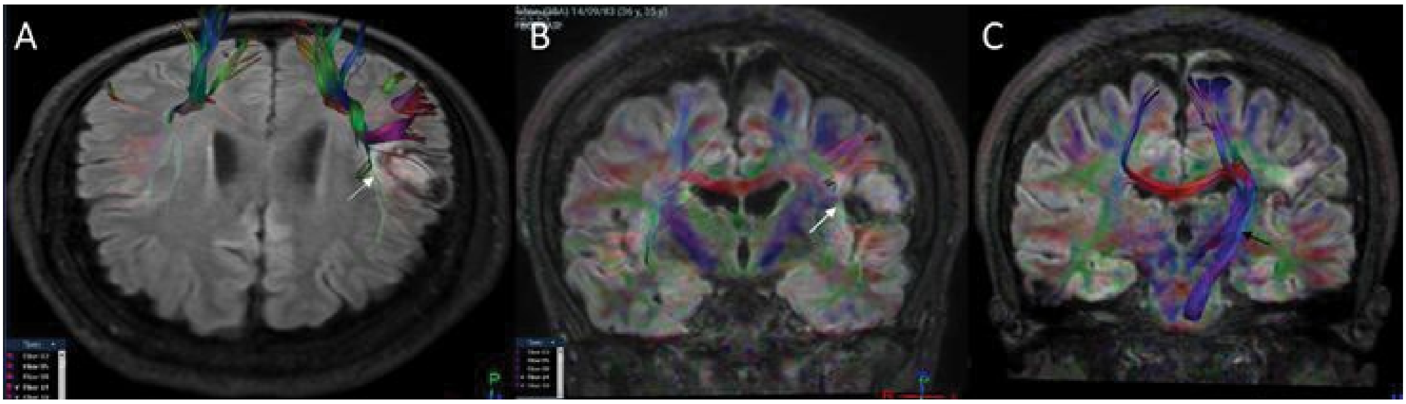

Figure 3. Tractography: A and B) arcuate fasciculus (white arrow) green color displaced medially peripheral to the lesion and C) corticospinal tract in blue color away from the lesion (black arrow).

Outcome After surgery, the patient was awakened, and there were no signs of postoperative neurological injury. He was discharged on the fourth postoperative day with full control of his epileptic seizures and preserved language (expression, comprehension). Postoperative tomography showed total resection of the lesion, with a small left frontal pneumocephalus. The pathology report confirmed the diagnosis of cavernous malformation.

Results and Discussion

CM is congenital vascular malformations, composed of wide and adjacent sinusoidal vascular spaces, without interposition of brain parenchyma. In Gadolinium MRI CM are seen as well-defined rounded or ovoid lesions, with minimal mass effect, with little or no enhancement after contrast administration

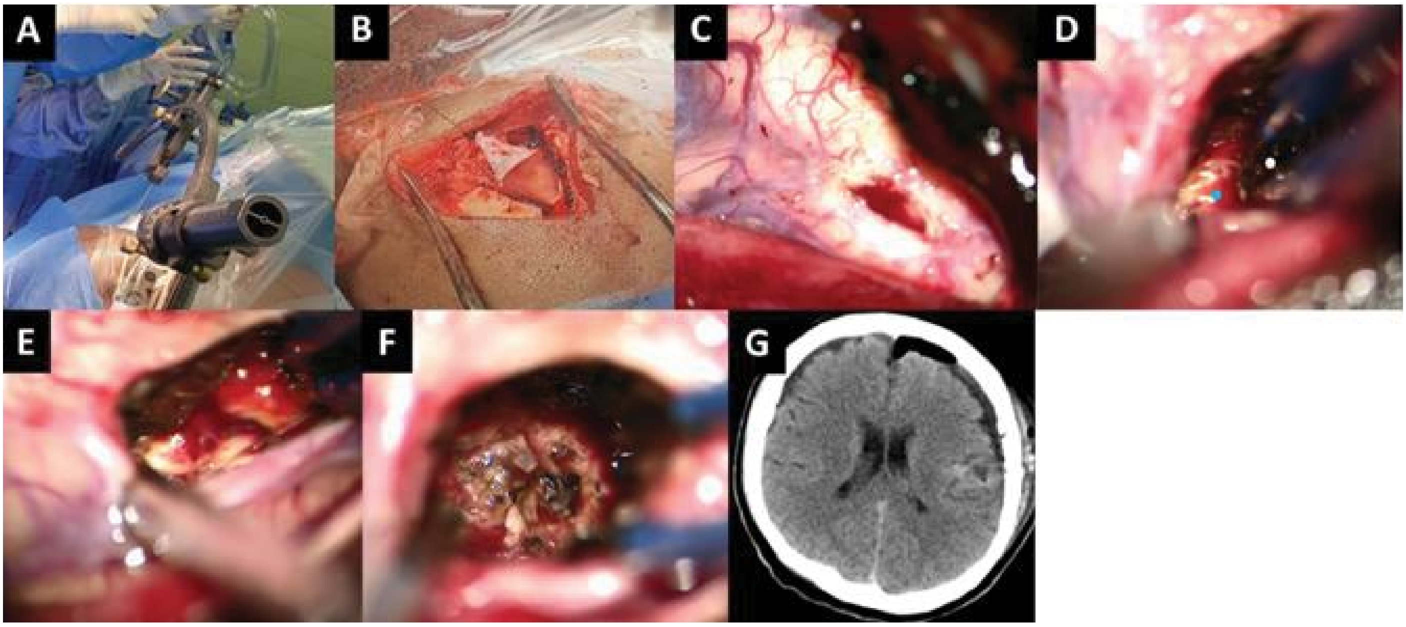

Figure 4. Intraoperative images: A) stereotactic system with the coordinates of the approach point, B) minimal centered triangular craniotomy, C) minimal stereotactic-guided corticotomy, D and E) dissection of cm of brain tissue (light blue point: white matter; white point: cm), F) final view without macroscopic evidence of cm and G) immediate postsurgical CT shows complete cm removal. Small quantity of pneumocephalus is illustrated in the left frontal subdural region.

[18]. There are familiar forms of CM and so far three associated genes have been identified (CCM1, CCM2, CCM3) [19]. Table 1 shows the main series of cases of CM surgery assisted with stereotactic techniques.

Since the first stereotactic-guided CM surgery was performed, many reports have been documented. Stereotactic-guided microsurgery offered significant advantages in treating deep brain lesions, or lesions that cannot be reliably located due to their small size or lack of obvious landmarks. Ebeling U, et al. [20] reported 16 stereotactic-guided microsurgical procedures; small superficial lesions in 6 patients and deep subcortical lesions in 10 patients. No patient developed a new postoperative neurological deficit. Three additional series of stereotaxia in CM were published in 1994. Giulioni M, et al. reported their experience with 18 pediatric patients (age range: 10 months-17 years) operated on for symptomatic CM [21]. The stereotactic guidewire was used in four patients whose lesions were deep subcortical in non-eloquent areas. They achieved complete resection in all cases, without mortality. Lerch KD, et al. reported 46 patients with small cerebral vascular malformations (less than 3 cm) operated with stereotactic guidance. Thirty eight patients had deep subcortical lesions, 3 had mesial temporal lesions, and 5 in the brainstem. They also obtained complete microsurgical resection of the lesions in all cases, with a surgical morbidity of 6.5% and no mortality. Operated on 7 patients with CM, six of which were in eloquent brain areas, with not postoperative neurological deficits [22,23].

Two more studies were presented in 1995 that confirmed the benefits of the stereotactic technique [24]. Presented 10 patients who presented with epilepsy (8 men, 2 women, mean age 32 years). The mean postoperative hospital stay was 5.1 days without surgical complications. In a mean follow-up period of 22 months after resection, all patients experienced better seizure control. Boecher-Schwarz HG, et al. [25] Stated that stereotactic-guided neurosurgery offers the advantage of planning and select the least traumatic approach, due to the exact location of the lesion. They reported 12 patients (age range 16-54 years) with supratentorial CM (size range 0.5-1.8 cm) treated along two years, with stereotactic-guided microsurgery. The skin incision and craniotomy (mean diameter 2.8 cm) were planned stereotactically, with a corticotomy of less than 1 cm. No relevant neurological morbidity related to surgery occurred in any patient half a year after surgery.

In another series of nine CM patients treated by stereotactic-guided surgery presented in Brazil in 2000 by radical resection was considered as the treatment of choice for CM [26]. Postoperative morbidity occurred in two cases: one patient had a seizure on the third postoperative day and the other presented dysphasia and hemiparesis on the second postoperative day, both with full recovery. They achieved total resection of the lesion in all cases without neurological deficit [27]. In 2003 in Germany presented their experience of stereotactic-assisted surgeries in 19 of 39 patients. In 10 of the patients, the CMs were located in an eloquent area. They achieved complete resection in all without postsurgical neurological morbidity [28]. Reported ten patients with small and deep intracranial CM resected with stereotactic assistance. They performed a minicraniotomy (<3 cm in diameter). They achieved complete resection in all ten patients; they did not present complications such as bleeding or infections. The use of a stereotactic system offers significant advantages in the management of small CM lesions, located in eloquent areas. It allows an exact preoperative location of the lesion with smaller skin incisions and craniotomies, less brain manipulation and consequently less morbidity and complications.

Our patient did not accept awake surgery, so we decided to use a stereotactic guide, which allowed us to calculate the approach point with great precision. Once the approach was completed, the resection of the MC was greatly facilitated by taking advantage of the non-functional gliotic plane that surrounds it, that can be circumferentially dissected and removed without damaging the surrounding parenchyma. This characteristic differentiates it from other lesions such as gliomas that do not have defined margins, and where awakening the patient is essential [19,29]. The stereotactic guide for CM surgery is safe and effective, and in our case it did not present functional complications.

Most CMs cause seizures, focal neurological disorders, and headache. Surgical resection of symptomatic CM located in non-eloquent areas is indicated [3,30]. Surgery for CM in eloquent areas provides a clear benefit compared to natural history; it should be performed in all CM in symptomatic eloquent areas. However, deciding to resect a CM becomes more complicated when it is located in an eloquent area and is hardly symptomatic or asymptomatic. The use of stereotaxy, neuronavigator and intraoperative functional MRI images significantly reduces the risk of complications and establishes microsurgical resection as a favorable treatment method for most cases [31-33].

Table 1. Summary of stereotactic surgery for cavernous malformation of eloquent area, cases reported in the literature.

Authors and Year	n	Clinical presentation	Location	Degree of resection	Complications	Outcomes	Follow-up time
Ebeling U, et al. 1993	10	Seizures, focal signs, asymptomatic	Temporal, frontal, parietal, primary motor	Total: 9 (reoperated for residual: 1)	No	No	No data
Giulioni M, et al. 1994	17	Seizures, headache	Temporal, frontal, parietal, occipital, parieto/occipital, cerebellum	Total: 17	No	No	1-17 y
Lerch KD, et al. 1994	46	Bleeding, seizures, focal signs, headache, asymptomatic	Frontal, primary motor, parietal, occipital, paratrigonal, temporal, brainstem, cerebellum	Total: 46	No	Hemiparesis: 3	6 m-5 y
Kunz U, et al. 1994	7	Seizures, headache, asymptomatic	Occipital, primary motor, Broca´s, frontal	Total: 7	No	No	No data
Casey AT, et al. 1995	10	Seizures	Temporal, frontal, parietal, occipital	Total: 10	No	No	5-48 m
Boecher-Schwarz G, et al. 1996	12	Seizures, asymptomatic, bleeding, focal signs	Frontal, temporal	Total: 12	No	Left hypoesthesia: 1 mild aphasia: 1 mild left hand paresis: 1	7-78 w
Meness MS, et al. 2000	9	Seizures, focal signs, headache	Temporal, frontal, parietal, parieto-occipital	Total: 9	No	No	No data
Grunert P, et al. 2003	19	Bleeding, seizures, focal signs, headache, asymptomatic	Temporal, frontal, parietal, occipital, Insula	Total: 18 (reoperated for residual: 1)	No	Aphasia and hemiparesis: 1 hemianopia: 1	No data
Slotty PJ, et al. 2013	10	Bleeding, seizures	Temporal, frontal, parietal	Total: 10	No	Hemiparesis: 1	3 m

Conclusion

While lesions in the language area require awake surgery, in cooperative patients, and assisted by intraoperatory neuroimaging (CT or MRI). Whenever this is not possible, other precision instruments such as a stereotactic frame, or a neuronavigator, may allow us to locate lesions in eloquent areas with sufficient precision, with the aim of minimizing the risks of lesions in these areas. CMs are well-defined lesions, which generally allow complete resection, without postsurgical neurological deficit, even if they are located close to eloquent areas. Stereotaxia continues to be a useful tool that allows high precision surgery and approaches to be planned with minimal morbidity.

Conflict of interest

None.

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