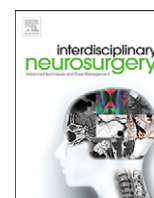


Contents lists available at [ScienceDirect](http://ScienceDirect.com)

Interdisciplinary Neurosurgery: Advanced Techniques and Case Management

journal homepage: www.inat-journal.com

Case Reports & Case Series (CRP)

Awake craniotomy for cortical language mapping and resection of an arteriovenous malformation adjacent to eloquent areas under general anesthesia – A hybrid approach



Pree Nimmannitya^{a,b}, Yuzo Terakawa^{a,*}, Taichiro Kawakami^a, Naohiro Tsuyuguchi^a, Hidetoshi Sato^a, Toshiyuki Kawashima^a, Kenji Ohata^a

^a Department of Neurosurgery, Osaka City University Graduate School of Medicine, Osaka, Japan

^b Division of Neurosurgery, Department of Surgery, Faculty of Medicine, Thammasat University, Pathumthani, Thailand

ARTICLE INFO

Article history:

Received 14 June 2015

Revised 25 August 2015

Accepted 25 October 2015

Keywords:

Awake craniotomy

Brain mapping

Arteriovenous malformation

Surgical treatment

ABSTRACT

Surgery of arteriovenous malformation (AVM) is sometimes challenging and carries a high risk of morbidity, especially when the AVM is located in an eloquent area of the brain. Unlike gliomas, awake craniotomy has not been widely used for resection of AVM. The authors present a case of an AVM in the left frontal lobe which was successfully removed with the aid of awake craniotomy with cortical language mapping. In conclusion, awake craniotomy for functional cortical mapping is beneficial for AVM resection, especially when the lesion is located in or adjacent to eloquent areas of the brain. A hybrid approach with functional mapping in the awake condition and AVM resection under general anesthesia may be useful in selected cases. Furthermore, en bloc resection with the nidus embedded in the brain parenchyma may be a useful means of removal to reduce operation time and intraoperative blood loss if there is no apparent functional cortex surrounding the AVM, as in the present case.

© 2015 The Authors. Published by Elsevier B.V. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

1. Introduction

Awake craniotomy is now widely available and has recently been used for various types of functional brain mapping in both language and non-language areas. Awake craniotomy with functional brain mapping is especially useful if the lesion is located in the brain parenchyma adjacent to eloquent areas, including motor, sensory, and language areas of the brain. Accordingly, it is often utilized for removing intrinsic brain tumors, such as gliomas, to achieve maximum resection of the lesion with preservation of brain function.

Surgery of arteriovenous malformation (AVM) is sometimes challenging and carries a high risk of morbidity, especially when the AVM is located in an eloquent area of the brain [1]. Unlike glioma, AVMs do not contain brain tissue in the nidus, and the border between the AVM and surrounding normal brain tissue is often distinct. Therefore, awake craniotomy has not been widely used for resection of AVM [2]. Here, we report a case of AVM in the left frontal lobe that was successfully removed using awake craniotomy with language mapping, and discuss the utility of functional brain mapping for resection of AVM in eloquent areas.

2. Case report

A 38-year-old right-handed woman presented with the first episode of generalized convulsions at the age of 25 years. The patient received medical checkup and was diagnosed as having an AVM of Spetzler-Martin grade 3 (S2-E1-V0). The patient had already undergone four times endovascular treatment for nidus embolization using *N*-butyl cyanoacrylate and gamma knife radiosurgery by the age of 27 years. The nidus was slightly reduced in size but complete obliteration of the nidus had not been achieved. Nonetheless, the patient had been followed up by regular magnetic resonance imaging (MRI) examinations without additional treatment and had been well for 13 years. Follow-up MRI obtained at the age of 38 years incidentally detected asymptomatic minor intracerebral hemorrhage from the AVM nidus mainly located in the left superior and middle frontal gyri with a maximum diameter of 6 cm (Fig. 1). On angiography, the nidus was fed by cortical branches of the anterior cerebral artery, i.e., frontopolar, anterior internal frontal, and middle internal frontal arteries as well as cortical branches of the middle cerebral artery, i.e., orbitofrontal and prefrontal arteries. The main drains were the ascending cortical veins draining into the superior sagittal sinus (Fig. 1). Although the patient remained neurologically intact, aggressive resection of the nidus was planned to prevent subsequent hemorrhage.

As the nidus was large and located in the dominant frontal lobe, we planned awake craniotomy for language mapping. We used techniques in functional cortical mapping that have been reported elsewhere [3].

* Corresponding author at: Department of Neurosurgery, Osaka City University, Graduate School of Medicine, 1-4-3 Asahi-machi, Abeno-ku, 545-8585 Osaka, Japan. Tel.: +81 6 6645 3846; fax: +81 6 6647 8586.

E-mail address: terakawa@med.osaka-cu.ac.jp (Y. Terakawa).

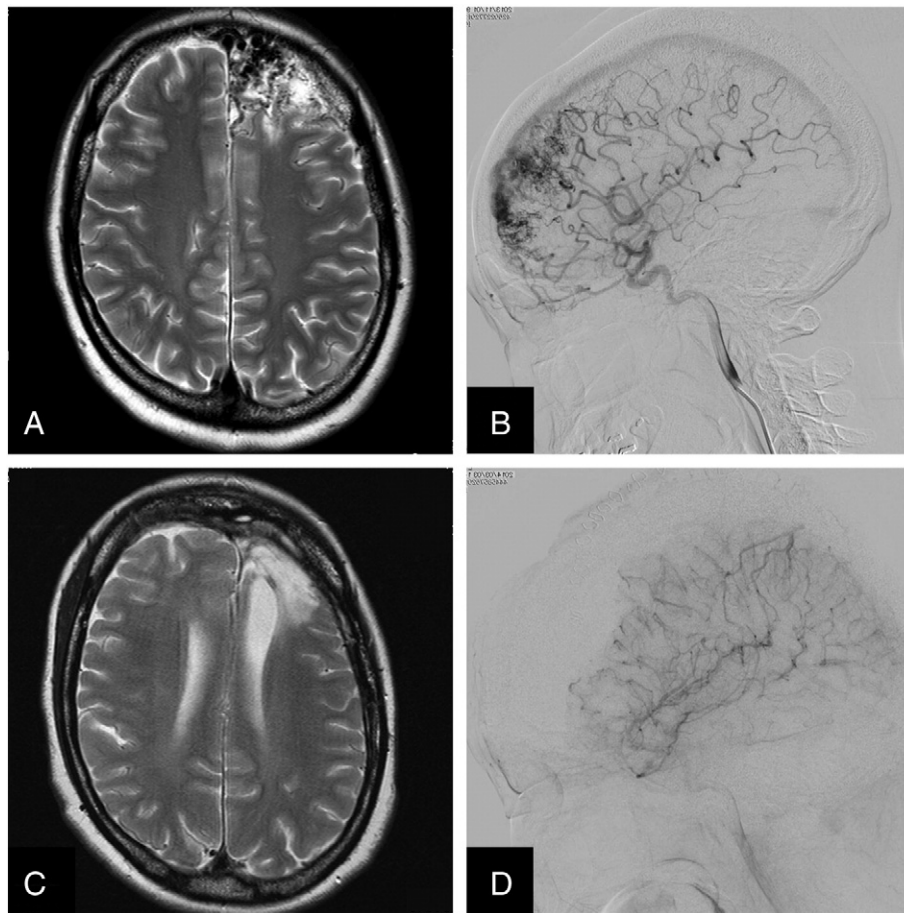


Fig. 1. Comparative pictures of preoperative and postoperative imaging study. (A) Preoperative axial T2 weighted magnetic resonance imaging (MRI) study of the brain showing a 6-cm arteriovenous malformation (AVM) located in the left frontal lobe. (B) Preoperative digital subtraction 4-vessel angiogram (DSA) showing a large AVM nidus, which is fed by cortical branches of the anterior cerebral artery and the middle cerebral artery. The main drains are the ascending cortical veins draining into the superior sagittal sinus. Postoperative axial T2 weighted MRI (C) and postoperative DSA (D) showing total resection of the AVM.

We used asleep–awake–asleep protocol with a laryngeal mask during both asleep phases. We performed the motor and visual naming tasks during the awake phase. After identifying the motor and language areas by cortical mapping, general anesthesia was induced and resection of the nidus was performed. Intraoperative images before and after resection of the AVM is shown in Fig. 2. As the nidus was large and there was a certain distance between the nidus and areas positive for language function, we decided to perform left frontal lobectomy with the nidus embedded inside the brain parenchyma to reduce the operative time and decrease intraoperative blood loss. Total removal of the nidus was carried out successfully (Figs. 1 and 2). The total awakening time from extubation to re-intubation of a laryngeal mask was 1 h and 2 min and the resection time was 3 h 52 min. The estimated intraoperative blood loss was only 140 ml.

Postoperatively, the patient complained of slight difficulty of speech, but this symptom resolved completely within a few days. Otherwise, the postoperative course was uneventful and neuropsychological tests showed no deterioration of cognitive or language functions (Fig. 3).

3. Discussion

Even in the microsurgical era, surgical morbidity is still unsatisfactory especially in cases of AVM located in eloquent areas of the brain. A previous study in 150 patients with surgically treated AVMs demonstrated significantly higher rates of surgical morbidity in patients with eloquently located AVMs (20.2%) compared to patients with non-eloquently located AVMs (8.2%) [1]. Of note, the authors of this previous report found that the risks of postoperative complications, such as

infection, hydrocephalus, or deep venous thrombosis, were also higher in patients with eloquently located AVMs, besides the inherent risk of manipulating eloquent areas of the brain. It was speculated that this finding was probably due to higher incidence of neurological worsening after surgery that could potentially lead to prolonged hospitalization. Therefore, safe resection of AVM with minimal additional neurological deficits appears to be indispensable to reduce surgical morbidity in patients with eloquently located AVMs.

Recently, awake craniotomy with functional brain mapping has widely been used for resection of intrinsic brain tumors, such as gliomas in or adjacent to eloquent areas; however, the use of awake craniotomy in AVM surgery is, to our knowledge, still limited to small series. Burchiel et al. previously reported eight cases of AVM in which functional cortical mapping was used to remove AVM. A total resection was achieved without causing additional neurological deficits in seven of eight cases [4]. In another previous study of 12 AVMs in which motor and language mapping were performed, five cases underwent language mapping in the awake condition as in the present case. In all five of these cases, the functional cortex was identified, resulting in its preservation during AVM resection [2].

There are several advantages to utilizing awake craniotomy with functional cortical mapping in AVM surgery. First, previous reports suggest that motor or language function may shift, because of brain plasticity, from its anatomical location to an adjacent gyrus or even to the contralateral hemisphere in the presence of long-standing lesions, including AVMs [5,6]. Accordingly, precise localization of language function seems to be feasible only with the patient in the awake condition. Second, surgeons can determine the extent of the functionally safe

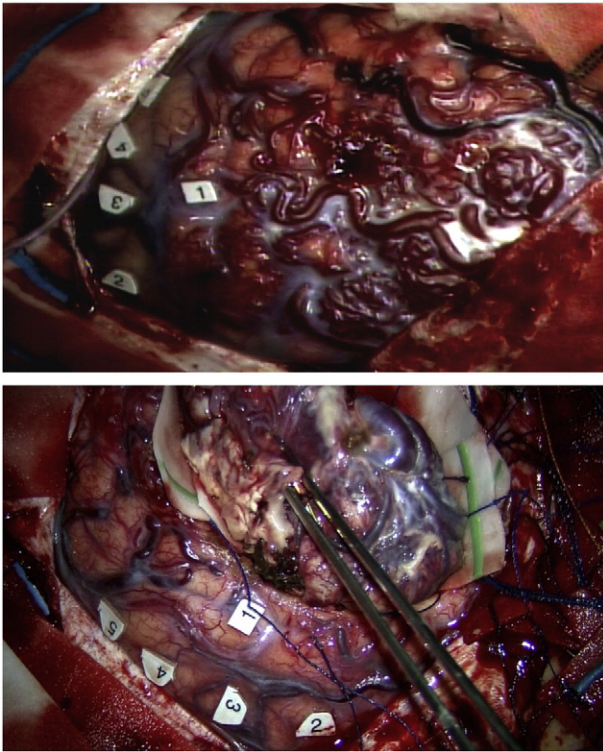


Fig. 2. Intraoperative images before (upper) and after (lower) resections of an arteriovenous malformation (AVM). The language area labeled as “1” was located adjacent to the AVM. The primary motor cortex was labeled as “2 to 5”. Complete AVM resection was performed while preserving these functioning cortices.

resection zone on the basis of precise brain functional mapping. Finally, in selected cases, surgeons may be able to choose en bloc resection with nidus incorporated in the non-functioning brain cortex, as performed in the present case, instead of unfolding the nidus complex. On the other hand, several issues remain to be resolved in awake craniotomy for AVM resection. First, preparations must be made for unexpected hemorrhage or possible hemodynamic changes during AVM resection, as these conditions may lead to significant worsening of the patient's condition. Second, the patient's fatigue should not be underestimated during AVM resection. Therefore, resection of the AVM itself may need to be performed under general anesthesia. In such cases, precise subcortical mapping becomes impossible, and consequently, the possibility of injuring subcortical fibers remains during AVM resection. Nevertheless, we consider that en bloc resection based on brain mapping is useful to reduce operation time and intraoperative blood loss, and therefore it should be considered as one of the resection techniques in AVM surgery.

4. Conclusion

We described a case of AVM that was successfully removed without additional neurological deficits with the aid of awake craniotomy with functional cortical mapping. Awake craniotomy for functional cortical

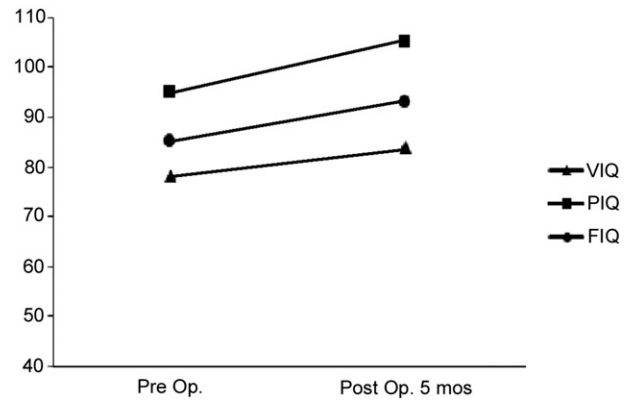


Fig. 3. A bar graph showing preoperative and postoperative Wechsler Adult Intelligence Scale scores. *VIQ = visual intelligence quotient; PIQ = performance intelligence quotient; FIQ = full-scale intelligence quotient.

mapping is also beneficial for AVM resection, especially when the lesion is located in or adjacent to eloquent areas of the brain. Therefore, awake craniotomy should be considered as one option for the surgical strategy. A hybrid approach with functional mapping in the awake condition and AVM resection under general anesthesia may be useful in selected cases. Furthermore, en bloc resection with the nidus embedded in the brain parenchyma may be a useful means of removal to reduce operation time and intraoperative blood loss if there is no apparent functional cortex surrounding the AVM, as in the present case.

Acknowledgments

All authors have no conflicts of interest to declare.

Pree Nimmanniyta is supported by a fellowship from Takeda Science Foundation.

We are grateful to Ms. Arisa Miyamoto at the Department of Neurosurgery, Osaka City University Graduate School of Medicine, Osaka, Japan, for her support in neuropsychological assessment.

References

- [1] C. Schaller, J. Schramm, D. Haun, Significance of factors contributing to surgical complications and to late outcome after elective surgery of cerebral arteriovenous malformations, *J. Neurol. Neurosurg. Psychiatry* 65 (1998) 547–554.
- [2] A. Gabarrós, W.L. Young, M.W. McDermott, M.T. Lawton, Language and motor mapping during resection of brain arteriovenous malformations: indications, feasibility, and utility, *Neurosurgery* 68 (2011) 744–752.
- [3] H. Duffau, L. Capelle, J. Sichez, T. Faillot, L. Abdennour, J.D. Law Koune, S. Dadoun, A. Bitar, F. Arthuis, R. Van Effenterre, D. Fohanno, Intra-operative direct electrical stimulations of the central nervous system: the Salpêtrière experience with 60 patients, *Acta Neurochir. (Wien)* 141 (1999) 1157–1167.
- [4] K.J. Burchiel, H. Clarke, G.A. Ojemann, R.G. Dacey, H.R. Winn, Use of stimulation mapping and corticography in the excision of arteriovenous malformations in sensorimotor and language-related neocortex, *Neurosurgery* 24 (1989) 322–327.
- [5] R.M. Lazar, R.S. Marshall, J. Pile-Spellman, L. Hacin-Bey, W.L. Young, J.P. Mohr, B.M. Stein, Anterior translocation of language in patients with left cerebral arteriovenous malformation, *Neurology* 49 (1997) 802–808.
- [6] R.M. Lazar, R.S. Marshall, J. Pile-Spellman, H.C. Duong, J.P. Mohr, W.L. Young, R.L. Solomon, G.M. Perera, R.L. DeLaPaz, Interhemispheric transfer of language in patients with left frontal cerebral arteriovenous malformation, *Neuropsychologia* 38 (2000) 1325–1332.