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Case study

Surgical treatment of cavernous malformations involving medulla oblongata

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ABSTRACT

Surgical treatment of cavernous malformations (CMs) involving medulla oblongata is more difficult than the CMs in other sites because of the surrounding vital structures. However, the distinctive features and treatment strategies have not been well illustrated. Therefore, we enrolled a total of 19 patients underwent surgical treatment of CMs involving medulla oblongata in our hospital from August 2008 to August 2014. The clinical features, surgical management and clinical outcome of these patients were retrospectively analyzed, while our institutional surgical indications, approaches and microsurgical techniques were discussed. In our study, gross total resection was achieved in 17 patients and subtotal resection in 2. Two patients underwent emergency surgeries due to severe and progressive neurological deficits. The postoperative new-onset or worsened neurological deficits occurred in 6 patients. After a mean follow-up of 45.8 ± 22.2 months, the neurological status was improved in 10 patients and remained stable in 7. The mean modified Rankin Scale (mRS) was 2.58 ± 1.26 preoperatively, 3.11 ± 0.99 postoperatively and 1.84 ± 1.42 at the recent follow-up, respectively. During the follow-up period, no rehemorrhage and recurrence occurred, and the residual lesions remained stable. We recommended surgical resection of symptomatic CMs involving medulla oblongata via optimal approaches, feasible entry zones and meticulous microsurgical techniques in attempting to achieve safe resection and favorable outcome. The clinical features, surgical indications, timing and microsurgical techniques of this special entity should be distinctive from the brainstem cavernous malformations in other sites.

1. Introduction

Since brainstem cavernous malformations (CMs) are associated with higher hemorrhagic rates and poorer neurological outcome [1,2], microsurgical resection has been considered to be the optimal treatment [3,4]. In the last two decades, remarkable progress has been achieved in surgical treatment of brainstem CM and it is consensus that the primary goal of the surgery is to prevent further hemorrhage rather than neurological recovery [3–5]. Therefore, the surgical strategy of brainstem CMs has become how to balance the hemorrhagic morbidities and surgery-related morbidities. CMs involving medulla oblongata, as a more special entity of brainstem CMs, are of unique features comparing to the other sites. The CMs located in medulla oblongata and pontomedullary could be associated with potential life-threatening symptoms and the proximity to critical structures may pose a particular challenge to microsurgical resection. Therefore, we advocated that the surgery-related decision-making criteria, timing, surgical techniques and clinical outcome should be reconsidered.

Based on our experience in surgical treatment of 19 patients harboring CMs involving medulla oblongata, we aim to evaluate the clinical features and neurofunctional outcome of these patients and summarize our institutional experience on surgical indications, timing and microsurgical techniques of this disease.

2. Methods

2.1. Clinical chart

From August 2008 to August 2014, nineteen consecutive patients with CMs involving medulla oblongata underwent surgical treatment in our hospital. The clinical data including age, gender, symptoms and neurological signs at admission were retrospectively analyzed. The present study was approved by the West China Hospital Trials and Biomedical Ethics Committee. The mRS was used to evaluate the neurological status. Preoperatively,

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cranial Computer Tomography (CT), Magnetic Resonance Imaging (MRI) with contrast enhancement and Diffusion Tensor Tractography (DTT) were performed as routine.

2.2. Surgery

Our institutional surgical indication was CMs involving medulla oblongata which presented with symptomatic acute and subacute hemorrhage and were surgically accessible.

We preferred performing the surgery during the subacute stage due to the favorable dissection plane. However, for patients with severe and progressive neurological deficits such as weakness of gag reflex and respiratory dysfunction, emergency surgery should be performed.

Somatosensory evoked potentials, motor evoked potentials and brainstem auditory evoked potentials were monitored routinely. Recording needles were inserted into the orbicularis oris, orbicularis oculi muscles, the posterior pharyngeal wall and the tongue bilaterally. A stimulation probe was applied for cranial nerve nuclei and corticospinal tract mapping.

The selection of the surgical approach was tailored individually. The safe entry zone was determined by the integration of anatomical landmarks, pial presentation, preoperative DTT, intraoperative neuronavigation and nuclei mapping. Standard microsurgical exposure and resection was performed including low-power bipolar coagulation and sharp dissection. Generally, an *en bloc* resection and elaborate exploration of the wall of hematoma cavity was attempted to improve the possibility of complete resection. However, if the hemodynamic instability occurred during separation of the lesion, piecemeal resection should be adopted. The developmental venous anomalies (DVAs) and the hemosiderin-stained gliotic tissues were preserved.

2.3. Follow-up and clinical outcome

Postoperative complications and the new-onset or worsened neurological deficits were recorded, mRS was used to evaluate the neurological status postoperatively (3 days after the surgery). The follow-up was performed at 6 months after surgery and once a year thereafter. During the follow-up period, changes in neurological deficits (improved, stable or aggravated) and mRS were assessed in outpatient center. mRS scores of 1–2 was considered as favorable outcome. Experts of neuroimaging processed the cross analysis of the MRI to identify the recurrence and rehemorrhage.

3. Results

There were 7 males and 12 females with a mean age of 39.74 ± 10.68 years (range from 15 to 58 years). All the patients presented with symptoms of one or more hemorrhagic episodes (10 patients presented with single episode of hemorrhage, 6 patients with two episodes and 2 patients with three or more episodes. one patient underwent the surgery for CM located in medulla oblongata 10 years ago, and suffered from rehemorrhage this time). The calculative annual hemorrhage rate was 4.24% (assuming that all CMs generated since birth) and the annual rehemorrhage rate was 54.17%. The most common symptom was sudden onset of cranial nerve dysfunction ($n = 14$) including dysphagia ($n = 6$), dysarthria ($n = 4$), facial ($n = 3$) and abducent paralysis ($n = 3$), cardiac instability ($n = 2$) and neck pain ($n = 2$). Other symptoms and neurological signs included hemiparesthesia ($n = 11$), hemiparesis ($n = 9$), ataxia ($n = 5$), headache ($n = 2$) and respiratory dysfunction ($n = 3$) (Table 1). The mean mRS score was 2.58 ± 1.26 preoperatively. According to the locations of these lesions, all the cases were divided into 3 groups: upper dorsal medulla involvement ($n = 6$), lower dorsal medulla involvement ($n = 3$) and ventrolateral medulla involvement ($n = 10$).

Table 1
Symptoms and signs on admission.

Symptoms and signs	Lesion location, *n*		
	Upper dorsal medulla involvement	Lower dorsal medulla involvement	Ventrolateral medulla involvement
Cranial nerve deficits	4	4	10
Oculomotor paralysis	0	0	1
Abducent paralysis	1	0	2
Facial paralysis	2	0	1
Dysphagia	1	2	3
Dysarthria	1	1	2
Cardiac instability	2	0	0
Neck pain	0	1	1
Hemiparesthesia and body numbness	2	1	8
Hemiparesis	1	0	8
Ataxia	1	0	4
Headache	2	0	0
Respiratory dysfunction	2	0	1

3.1. Surgery

Seventeen patients underwent surgical treatment in subacute phase after hemorrhage (Fig. 1), and two patients were taken for the emergency surgery (one for acute severe dysphagia combined with coma for 30 min, and one for progressive respiratory dysfunction) (Fig. 2). The following approaches were adopted: suboccipital approach ($n = 9$, including telovelar approach in 6 cases) (Fig. 3), retrosigmoid approach ($n = 5$) and far lateral approach ($n = 5$). Gross total resections were achieved in 17 cases, while subtotal resections were performed in 2 cases. These two lesions were observed involving the obex, and when we attempted to shrink the lesion away from eloquent hemosiderin-stained gliotic tissue, drastic cardiac instability occurred. Intraoperatively, the DVAs were observed in 4 cases (21.05%), and all the typical caput medusae structures were preserved carefully. The surrounding hemosiderin-stained gliotic tissues were preserved in all cases.

3.2. Complications and follow up

Postoperative new-onset or worsened neurological deficits occurred in 6 patients including dysphagia ($n = 1$), dysarthria ($n = 1$), paresthesia ($n = 2$), ataxia ($n = 2$) and walking difficulty ($n = 1$). No surgical related hemiparesis occurred. The mean mRS 3 days after the surgery was 3.11 ± 0.99. One patient suffered from postoperative hydrocephalus, who was performed the Ventricle-Peritoneal shunt. Gastrostomy tube was placed in 6 patients who suffered from dysphagia preoperatively and two patients who presented with new onset of lower cranial nerves deficits. Tracheostomy was performed for the patients who suffered from respiratory dysfunction ($n = 1$) or severe aspiration pneumonia ($n = 2$). One patient required the ventilator due to preoperative respiratory failure despite an emergency surgery was performed, the patient died due to postoperative pneumonia eventually. Other complications included intracranial infection ($n = 1$) and aspiration pneumonia ($n = 2$).

All the patients underwent routine follow-up. After a mean follow-up of 45.8 ± 22.2 months (range from 18 to 78 months), the preoperative neurological status was improved in 10 patients and remained stable in 7. The postoperative new-onset or worsened symptoms were improved in 5 patients and remained stable in 1. At the recent follow-up, the mean mRS was 1.84 ± 1.42 and 15