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Foramen Magnum Meningiomas: Still a Big Challenge

Semih Can Çetintaş, Alperen Kaya, Bülent Kağan Tombak, Enes Karaca, Salih Alper Gürsoy, Rahşan Kemerdere, Taner Tanrıverdi

Department of Neurosurgery, İstanbul University-Cerrahpaşa, Cerrahpaşa Faculty of Medicine, İstanbul, Türkiye

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Abstract

Objective: Foramen magnum meningiomas (FMMs) are rare but challenging for neurosurgeons to remove because of their anatomic location where vital neuro-vascular structures exist. The difficulties encountered by a neurosurgeon during the surgery and important points during management of complications are underlined, which may help guide young neurosurgeons.

Methods: In this report, management of FMMs are provided in 7 patients who underwent surgical treatment between 2012 and 2022. In this small series, 4 women and 3 men were included. Mean age was 54.28 ± 13.61 years with a mean follow-up of 45.71 ± 40.18 months.

Results: Although gross total removal was achieved in all patients 3 showed complication (42.8%) during follow-up period. One showed recurrence.

Conclusions: Gross total removal in grade 1 FMM does not guarantee total cure, and strict follow-up is mandatory. We conclude that anterior or anterolaterally located FMMs can be safely removed by midline suboccipital subtonsillar approach. More importantly, every possible complication before surgery should be discussed in detail with the patients and/or their next of kin in order to avoid compensation lawsuits, which is a devastating problem among surgeons all over the world.

Keywords: Foramen magnum, meningioma, surgery

Introduction

Foramen magnum meningiomas (FMMs) are skull base meningiomas located in the craniocervical junction (CCJ). These tumors account for up to 3.2% of all meningiomas and are one of the most challenging pathologies for neurosurgeons.[1-3] Due to the vital neurovascular structures in the CCJ, such as the vertebral artery (VA), the cranial nerves IX-XII, the posterior inferior cerebellar artery, and the brain stem, FMMs generally cause multiple neurological deficits before and after surgery, which may seriously decrease the quality of life of the patients. It has been reported that the majority of FMMs arise from the anterior or anterolateral to the foramen magnum.[4-6] The relationship of the tumor with the vital neurovascular structures and the location itself make surgical removal very difficult, so almost every complication that may arise after surgery should be discussed with the patients and/or next of kin before surgery, given that at the present time physicians, especially surgeons, are faced with action for compensation cases.

Since FMMs are very rare, there are no larger clinical series in the current literature in which we encountered only 3 studies including more than 100 cases.[4-6] Our experience depends largely on small series published by tertiary centers.[7-12] With this small series, including only 7 patients, our main goal is to share our experience with young neurosurgeons and to underline some important points and to define how to manage these challenging tumors.

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Corresponding author: Semih Can Çetintaş, Department of Neurosurgery, Cerrahpaşa Medical Faculty, İstanbul University-Cerrahpaşa, İstanbul, Türkiye
e-mail: semihcan1992@gmail.com
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Methods

This study has been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments and approved by the Local Ethical Committee of İstanbul University-Cerrahpaşa, Cerrahpaşa Medical Faculty (Approval No: 755898, Date: August 18, 2023). A written informed consent form was obtained from all participants.

Patient Population

This small series included 7 patients who underwent microsurgery for FMMs in our neurosurgical department between November 2012 and January 2022, and all patients were operated on by the same surgeon with the same surgical approach. All patients had meningiomas arising from the anterolateral or lateral dura at the foramen magnum level, and the boundaries were described previously.[2,5]

Radiological Studies

The main radiological work-up included contrast-enhanced magnetic resonance imaging (MRI). Head MRI is the gold standard for these tumors in order to get an idea about tumor size, location, side of dural attachment, and relationship with the neurovascular structures. Encasement of the VA on preoperative MRI is one of the important findings before surgery, and the surgeon must have evaluated this situation with neuroradiologists. Furthermore, a head MRI may provide some findings related to the consistency of the tumor. In this small series, computed tomography was not obtained routinely. Only 3 patients needed cerebral angiography (CA) because of VA encasement by the tumor.

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Location Classification

As previously described,[2,13] tumor location was classified based on the axial MRI scans as (i) anterior, (ii) anterolateral, and (iii) posterior.

Surgical Approach

Surgical interventions were possible after we had informed consents signed by the patients. In this series, all patients were placed in a prone position with the head in a neutral position. A midline vertical skin incision was made from the inion up to the third vertebral lamina. The craniectomy was performed and directed toward the foramen magnum, which is then opened. Only the posterior third of the occipital condyle was removed in 3 patients, and the posterior arch of the C1 was removed in 5 cases. The dura was opened, and arachnoid membrane was incised. Arachnoid incision led to the cerebrospinal fluid release, which allowed for less cerebellar retraction. In 4 patients, real-time neuro-monitoring was used and instant changes were recorded. In our department, real-time monitoring was available since 2016. By using an ultrasonic aspirator (cavitronic ultrasonic surgical aspirator [CUSA]), the tumor was debulked first and sequentially dissected from the surrounding neurovascular structures. After tumor removal, meticulous hemostasis was carried out, and the dura was closed in a watertight fashion. The muscle and skin layers were closed in a standard fashion. All patients were admitted to our neuro-intensive care unit immediately after surgery.

Follow-up

In all patients, early MRI within 72 hours of the surgery was obtained. All patients were followed up both clinically and radiologically after a 6-month and 1 year interval. After 1 year, the follow-up intervals were prolonged depending on the clinical, radiological, and histopathologic findings. If the diagnosis was grade 1 or 3 meningiomas, patients were sent for radiation therapy and followed at 3- or 6-month intervals. As described previously,[5] extension of tumor removal was based on postoperative MRI findings and defined as "total" if no residual tumor or dural enhancement, "subtotal" if no residual tumor but dural enhancement, and "partial" if there is a presence of residual tumor.

Results

In this small series, 4 women and 3 men were included. The mean age was 54.28 ± 13.61 years (range 39-72 years), and mean follow-up was 45.71 ± 40.18 months (range 4-112 months) (Table 1).

Clinical Variables

Before surgery, the main presenting symptoms included cervicooccipital pain and paresthesia of the upper limbs. Dysphagia and unsteadiness were less common. In 2 patients, FMM diagnosis was incidental and had no symptoms. In the preoperative neurological examination upon admission, limb hypesthesia and monoparesis were noted in 3 patients. Cranial nerve deficits were encountered in 2 patients, including paresis of the vagus and glossopharyngeal nerves. Neurological examination in 2 patients was normal and FMM was diagnosed incidentally in these patients.

Surgical Features

In all patients, the prone position with the head in neutral was used instead of the park bench (lateral oblique), semi-sitting, and/or sitting positions, which may lead to severe air embolism during and air trap in the brain after surgery. Tumor removal was achieved through a sub-occipital, sub-tonsillar approach. It is clear that the extension of bone removal, including posterior arch of C1 lamina and occipital condyle, depends mainly on the tumor extension caudally, cranially, or laterally. In our small series, the occipital condyle was partially removed in 3 patients, and in 5 patients, the posterior arch of the C1 lamina had to be removed. In 6 patients, the VA was safely dissected from the tumor, but in 1 case (case 7), the VA was totally encased by the tumor, and preoperative embolization was performed. The patient tolerated the embolization well, and during surgery, the VA was sacrificed (Figure 1). In all patients, the extent of tumor resection was graded as Simpson grade II.

Radiological Outcome

Total tumor removal was achieved in all 7 patients. At the last follow-up, head MRI of the 6 patients showed no tumor. However, during the follow-up period, 1 patient showed recurrence 8 years after surgery. This was case 1) 39-year-old female, and head MRI showed anterolateral FMM on the right side. The first surgery was performed in 2012 and the tumor was totally removed. The histopathological diagnosis was grade 1 transitional meningioma. There was no neurological deficit after surgery. Because of the coronavirus disease 2019 pandemic, regular follow-up was not possible and in 2020, head MRI showed recurrence of the tumor, which encased the VA. The second surgery was performed in which partial removal was achieved. The second histopathological diagnosis was grade 2 atypical meningioma and the patient was sent for radiation therapy. At the last follow-up, the neurological examination was normal and the patient is still under our follow-up.

Table 1. Summary of the 7 Patients with Foramen Magnum Meningioma

No	Age	Gender	Side	Location	Removal	Grade	Recurrence	Follow-up (Months)
1	39	Female	Right	AL	Subtotal	1*	Yes	112
2	66	Female	Left	AL	Total	1	No	84
3	72	Female	Left	AL	Total	1	No	52
4	49	Male	Right	AL	Total	2	No	40
5	45	Male	Right	AL	Total	1	No	16
6	42	Male	Left	Lateral	Total	1	No	12
7	67	Female	Right	AL	Total	1	No	4

AL, anterolateral.
*The first histopathologic diagnosis was grade 1 meningioma, but after recurrence the second histopathologic diagnosis was grade 2 meningioma.

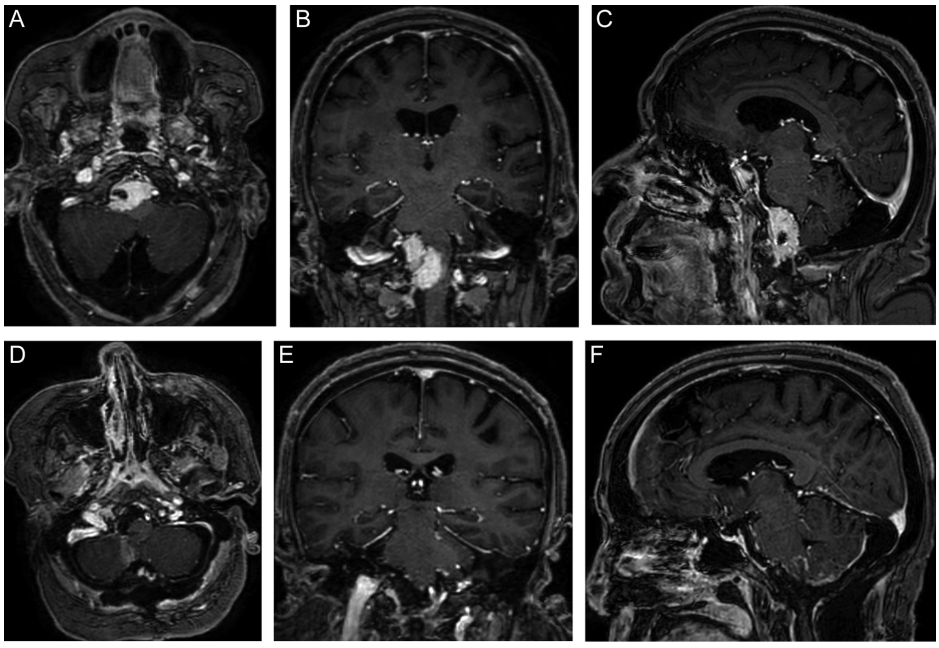

Figure 1. Contrasted preoperative (A through C) and early postoperative (E through F) head magnetic resonance imaging of a 67-year-old patient showing total encasement of the vertebral artery and total removal of the tumor. The vertebral artery was embolized and sacrificed during surgery. The histopathologic diagnosis was grade 1 meningioma, and the patient is still in our neuro-intensive care unit because of lower cranial nerve involvement.

Surgical Complications and Outcome at the Last Follow-up

At the last follow-up, 4 patients showed a normal neurological examination. Almost 10 years after surgery, 1 patient (case 2) developed hydrocephalus which was treated by a ventriculo-peritoneal shunt. After the procedure, the patient developed respiratory difficulty and was transferred to our neuro-intensive care unit. After recovery, the patient was transferred to our clinic from the neuro-intensive care unit. However, the patient refused to stay in the hospital and left voluntarily. One week later, we were informed by the patient's next of kin that the patient died. In 1 patient, the neurological examination showed that hypesthesia on the upper limbs was still present together with a decreased gag reflex. The last patient (case 7) developed severe hydrocephalus and a cerebrospinal fluid leak, which was treated by a ventriculo-peritoneal shunt. Postoperative head MRI showed ischemia on the right cerebellar peduncle, and the patient is suffering from difficulty in swallowing and respiration because of aspiration pneumonia. Thus, the patient needed a tracheostomy and percutaneous endoscopic gastrostomy. The patient is conscious without motor weakness and is still being followed up in our neuro-intensive care unit.

Discussion

Meningiomas in the foramen magnum are rare, but they still present challenges for neurosurgeons. The presence of vital neurovascular structures in this small anatomical area may inevitably cause devastating complications in some patients after surgery. It should be noted that neurological deficits may occur even before surgery. Thus, patients and/or their next of kin should be clearly informed in detail about the complications that may occur due to surgery. In today's modern era, we have advanced radiologic imaging modalities, improved anesthetic techniques, and modern surgical equipment such as neuro-navigation, CUSA, neuromonitoring, and improved surgical skills. However, despite these great advances, there are still controversies with respect to the surgical management of these tumors. Furthermore, no common consensus exists among neurosurgeons related to the type of surgical approach because it is clear that the neurosurgeons' experience and/or preferences are the main factors for determining the surgical approach.

The first controversy concerns the operative positioning of the patient. Some prefer sitting.[13] or semi-sitting.[8] position and underlined that this position decompresses the neuro-axis upon opening of the bone and dura. Others.[4,11] suggested a lateral oblique position for avoiding or decreasing the possible risk of air embolism. Depending on the present small series, we suggest that the prone position with the head in neutral is enough and safe for the removal of anterior and anterolateral FMMs. This position decreases the risk of air embolism and air trap in the brain, which may lead to death. We prefer the midline straight incision from the inion to the level of C-3 vertebra instead of an inverted hockey stick-like or paramedian straight incision. We have to underline that our aim is not to compare the type of surgical positioning and skin incision; rather, we want to underline that the experience of the operation team, including the neurosurgeons and anesthesiologists, determines the positioning and skin incision.

The second controversy is the removal of the bone. Occipital craniectomy was performed in all patients in this series, although some prefer to have occipital craniotomy. There has been no controversy related to occipital craniectomy and craniotomy among neurosurgeons. The main discussion is about the removal of the occipital condyle in FMMs. Drilling one-third to one-half of the occipital condyle has been suggested by some studies.[1,14] to have enough surgical corridor for the removal of the anteriorly located FMMs, and no cranio-cervical instability was observed. In this small series, we drilled the posterior third of the occipital condyle in the first 3 patients, but with increased experience, we did not need to drill the condyle in the last 4 patients, and we had enough surgical corridor to anterolateral and laterally located FMMs. As a consequence, we agree with Nanda et al[15] and Wu et al[4] who suggested that removal of the occipital condyle was unnecessary for

a safe and complete resection of anterior FMMs. Removal of the cervical lamina such as C1 depends on the caudal extension of the tumor and we had to perform removal of the posterior arch of C1 in 5 patients. We have to note that partial removal of the condyle depends on the surgeons' experience and there is no common consensus on the removal of the occipital condyle. Finally, our experience supported the suggestion from Dobrowolski et al[8] that midline suboccipital subtonsillar approach is enough and safe for the removal of anterior, anterolateral, and posterior FMMs.

The third issue that should be taken into consideration is the relationship between the FMM and VA. Neurosurgeons should have deep knowledge about this relationship before surgery because it determines the surgical strategy and has an important effect on the prognosis. It is well known that encasement of the VA by the FMM may lead to a worse prognosis compared to others.[4-6] Cerebral angiography is the sole method to show the relationship and should be performed before surgery if there is a close relationship between the FMM and VA on the preoperative MRI. In the present small series, 3 patients needed CA, and in 1 case, the VA was embolized for safe resection. The patient tolerated the embolization well, and the tumor was totally removed. In the other 2 patients, the embolization was not tolerated, but the tumors were removed totally because of their soft consistency. We suggest embolization in the case of total encasement of the VA if the patient is able to tolerate it.

The fourth issue is the management of surgical complications because postoperative management is as important as the surgery itself. The aim of surgery is to have total tumor removal with minimal, ideally without neurological deficits. It has been reported that recurrence is one of the important risk factors for a worse prognosis, and clearly, recurrence is lower in gross total resection.[10] It is more difficult to resect recurrent FMMs, which generally encase the VA and become stiffer, as seen in our case. Surgical morbidity and mortality have been reported at around 40% and 25%, respectively.[3-6] Furthermore, anterior location and VA involvement are the 2 factors that are strongly associated with the risk of surgical complications.[4-5] A higher mortality rate has been reported in patients who already have severe neurological dysfunction during the preoperative period.[3-6] Depending on the current literature and our own experience, we underline that lower cranial nerve (cranial nerves IX to XII) temporary and/or permanent dysfunction is the most common complication, followed by cerebrospinal fluid (CSF) fistula and hydrocephalus. In our patient group, we had 2 patients with severe hydrocephalus, which was treated with ventriculoperitoneal shunt insertion. At the long-term follow-up, only 1 patient showed a decreased gag reflex, which did not need special treatment. Watertight dural closure decreases the rate of CSF fistula and if the fistula still exists, lumbar catheter should be tried. If it fails, ventriculoperitoneal shunt should be considered. Severe lower cranial nerve dysfunction was seen in 1 case, and the patient has both tracheostomy and gastrostomy and is being followed up in our neuro-intensive care unit at the time of writing this paper.

Limitations

The authors of this paper are aware of the limitations of the study. We think that there are 2 main limitations. First, the sample size is very small because this series included a single surgeon's experience. It should be appreciated that FMMs are very rare, and the majority of series in the current literature have a limited number of patients, Only 3 papers included more than 100 patients, including 114 patients by Wu et al,[4] 107 patients by Bruneau and George.[5] and 185 patients by Li et al.[6] By reporting our small series, we just wanted to point out the main problems encountered in the management of FMMs and share our experience with young neurosurgeons. Second, the follow-up period is not long enough and ranged from 4 to 112 months in this series. For a meningioma, the follow-up period should be very long in order to provide more comprehensive results.

Conclusion

Although we have advanced radiological, neurophysiological, and surgical equipment, together with anatomical knowledge and surgical experience, the management of FMMs is still a big challenge. We conclude that anterior or anterolaterally located FMMs can be safely removed by a midline suboccipital subtonsillar approach. The condylar resection is not necessary in most cases, and postoperative management is as important as the surgical removal itself. Respiratory and swallowing difficulties caused by dysfunction of the lower cranial nerves need special attention and have a negative impact on the prognosis of the patients. Lastly, every possible complication before surgery should be discussed in detail with the patients and/or their next of kin in order to avoid compensation lawsuits, which is a very devastating problem among surgeons all over the world.

Ethics Committee Approval: Ethics committee approval was received for this study from the ethics committee of İstanbul University-Cerrahpaşa, Cerrahpaşa Faculty of Medicine (Approval No: 755898, Date: August 18, 2023).

Informed Consent: Written informed consent was obtained from the participants who participated in this study.

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