Indonesian Journal of Neurosurgery

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Suboccipital midline approach with C1 laminectomy and C2 partial laminectomy for anterolateral foramen magnum meningioma: A case report

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ABSTRACT

Introduction: Foramen magnum meningiomas (FMMs) arise from meningotheial cells of arachnoid layer in the craniospinal junction. FMMs are rare and comprise only 1.8 to 3.2% of all meningiomas. Patients with FMMs generally have vague symptoms and are often misdiagnosed. Surgical management of FMMs poses a challenge because foramen magnum is a highly complex territory of the skull base and contains many important and vital structures. Up until this date, the surgical approaches for FMMs that arise anterolaterally remain varied. We aimed to present a case of anterolateral FMM that was successfully managed through posterior suboccipital midline approach.

Case presentation: A 49-year-old female presented with chief complaint of tetra paresis for the past 4 months. Initially, she was experiencing tingling and numbness on her right neck radiating to her right hand. Her symptoms were worsened, radiating to all of her four extremities along with progressive motor weakness that made her unable to walk. Whole spine MRI was performed and anterior meningioma extending to the right side at the level of foramen magnum was found. The posterior suboccipital midline approach with C1 laminectomy and C2 partial superior laminectomy was performed. The patient made a good recovery immediately after the surgery and there was no recurrence of her symptoms during 4 months follow-up.

Conclusion: Surgical approaches for FMM remain varied according to several studies. However, the surgical strategy should be patient-tailored to achieve the maximal resection and prevent morbidity. As for our case, posterior suboccipital midline approach is safe and feasible surgical procedure to treat anterolateral FMM.

Keywords: foramen magnum, meningioma, suboccipital midline approach

Cite This Article: Christi, A.Y., Hakim, I.A., Pangaribuan, V.S.G., Wicaksono, P., Purnawan, A., Baskoro, W. 2021. Suboccipital midline approach with C1 laminectomy and C2 partial laminectomy for anterolateral foramen magnum meningioma: A case report. *Indonesian Journal of Neurosurgery* 4(2): 59-63. DOI: 10.15562/ijn.v4i1.143

INTRODUCTION

Meningiomas are the most common primary intracranial neoplasms, arise from meningotheial cells of arachnoid layer.¹ The cell of origin of meningiomas is believed to be the cells that make up the arachnoid villi, known as arachnoid cap cells.^{2,3} Arachnoid villi are most numerous in the area of the superior sagittal sinus, followed by the cavernous sinus, tuberculum sellae, lamina cribrosa, foramen magnum, and torcular herophili.² Meningiomas are slow-growing benign tumor, more likely to occur in females than males with a ratio of 1.8:1.¹ The incidence peaks at 45 years of age, median is 65 years old and the risk increases with

age.¹ Meningioma is representing 36.4% of Central Nervous System (CNS) tumors and account for 14.3 to 19% of all primary intracranial neoplasms.^{2,4} However, only 1.8 to 3.2% meningiomas arise at the foramen magnum.⁴

Foramen magnum meningioma (FMM) may originate at any location within the foramen perimeter.² FMMs include all meningiomas that arise within the following anatomical borders: anteriorly from the lower third of the clivus to upper margin of C2 body, laterally from jugular tubercle to the upper margin of C2 laminae, and posteriorly from the anterior edge of the squamous occipital bone to the C2 spinous process.^{5,6} FMM can be divided into two main types: craniospinal

and spinocranial. Craniospinal FMM originates intracranially and extends downward, while spinocranial FMM originates in upper spinal canal and extending intracranially.²

Patients with FMMs generally have vague symptoms and are often misdiagnosed or undetected for periods of time since the tumors progress relatively slow.^{2,7} The mean length of symptoms prior to diagnosis of FMM is 30.8 months, even in the era of Magnetic Resonance Imaging (MRI).⁴ Clinical triad that identifies FMM are unilateral cervical pain; development of unilateral arm sensory and motor deficits, which progress to the ipsilateral leg, then contralateral leg, and finally contralateral upper extremity (known as

classic foramen magnum syndrome); and cold, clumsy hands with intrinsic hand atrophy.^{2,4}

The primary strategy for meningioma removal is gross total removal, which typically entails the removal of the dura, bone, and vascular sinuses that have been infiltrated by the tumor.³ Surgical management of FMM poses formidable challenge due to the anatomical relationship with the critical vascular structures, brain stem, lower cranial nerves (CNs), upper cervical nerves and craniovertebral junction.^{4,8} As response to the difficulties resulting from the location of FMM and these complex anatomies, several surgical approaches have been described in the literatures; including the anterior transoral, transcervical retropharyngeal, antero-lateral, lateral, posterior sub-occipital midline, and posterolateral approaches.^{4,5,7,9}

In this article, the authors presented the surgical management of anterolateral FMM using posterior suboccipital midline approach with C1 laminectomy and C2 partial superior laminectomy.

CASE PRESENTATION

A 49-year-old female presented to the Neurosurgery Department of Soeradji Tirtonegoro General Hospital with chief complaint of tetra paresis for the past 4 months. Initially, she was experiencing tingling sensation and numbness on her right neck radiating to her right finger tips since 6 months before. She was also experiencing headache and neck pain-stiffness at that time. Her symptoms were worsening with tingling sensation and numbness radiating on all of her four extremities; along with progressive motor weakness that made her unable to walk. Patient also reported urine retention and made her once sent to Emergency Department for urinary catheter insertion. She denied any neck injury, tremor or gait disturbance before. Patient was a farmer and denied any repetitive neck motions while conducting her daily activities. There was no history of fever, sudden weight loss, infectious diseases nor neoplasm. Patient had history of using 3-month injectable hormonal contraception for more than 5 years.

The neurological examination

revealed normal cranial nerve function, absence of meningeal signs, diminished muscle strength in all four extremities with grades 3/5 for all muscle groups on right extremities and grades 4/5 on left extremities, hypotonic muscle tone, normal physiological reflexes, absence of pathological reflexes, and positive Spurling test. Patient still had urinary retention and treated with urinary catheter.

Laboratory tests were within normal range. Whole spine MRI was performed and revealed an anterior intradural extramedullary solid mass in the level of foramen magnum resulting in compression of the spinal cord to the posterolateral side (**Figure 1**). The mass was oval shaped with regular border and extending laterally to the right side with a

craniocaudal diameter of ± 28.9 mm and cross section width of ± 12.3 mm. After intravenous contrast injection, it showed strong homogenous enhancement with dural tail down to the C3 level suggestive a meningioma of the foramen magnum (**Figure 2**).

After getting informed consent, patient underwent surgery. The operation was conducted for 6 hours and 30 minutes. The procedures that we performed were suboccipital decompression, total laminectomy of C1, partial superior laminectomy of C2, and excision of the tumor. Intradural extramedullary tumor was found and removed by piece meal fashion. On gross analysis, the specimen was aggregate of brownish-white soft tissue fragments with spongy consistency.

Figure 1. MRI without contrast administration. A. Sagittal T1W MRI showed isointense intradural mass (white arrow) on the level of foramen magnum. B. Axial T1W MRI showed intradural mass of foramen magnum extends laterally to the right side, resulting in compression of the spinal cord to the posterolateral side.

Figure 2. MRI with contrast administration. A. Sagittal T1W image showed strong homogenous enhancement mass with dural tail down to the C3 level. B. Axial section showed the mass as an oval shaped with regular border, suggestive of a meningioma of the foramen magnum.

Histopathology examination revealed tumor growth arranged in whorled pattern. The cells were monophormic with round nuclei and nuclear pseudo-inclusion; confirming the diagnosis of meningothelial meningioma (WHO grade I).

Patient was monitored in intensive care unit for two days after the surgery. In the inpatient unit, we performed the follow-up neurological examination. The motor examination revealed an improving muscle strength on all four extremities. Patient had normal physiological reflexes, negative pathological reflexes, intact cranial nerve IX, X, XI, XII, and absence of meningeal sign. Headache, neck pain, neck stiffness, numbness and tingling sensation on her extremities had been relieved. However, patient was still using Foley catheter for urination.

A long-term follow up was done during 4 months post-surgery with excellent motor movement and disappearance of the tingling sensation. Her urinary retention has also resolved. There was no complication following the surgery and the patient was able to do her daily activities.

This case was classified as anterior FMM which extends laterally on the right side. Posterior suboccipital midline approach was chosen to minimize excision on muscles, preserve the bone, and avoid manipulation of the vertebral artery. Spinal cord retraction can be minimized with paramedian excision of duramater. The detailed surgical procedures were as follows: 1) We placed the patient in the prone position with the head in the neutral position (**Figure 3**); 2) Vertical midline skin incision was made frominion to the C2, exposing the suboccipital, lamina of C1, and lamina of C2; 3) Multiple burr hole on occipital were performed to decompress the bone; 4) Total laminectomy of C1 and partial superior laminectomy of C2 was made while still preserving the posterior muscles and spinous process; 5) Incision of duramater was made in the right paramedian level, 5 – 10 mm from the midline (**Fig. 4A, Fig. 4B**); 6) Debulking of the tumor and releasing the base of the right side (**Fig. 4C, Fig. 4D**); 7) The tumor was able to be excised, achieving Simpson grade 2 (**Fig.**

4E); 8) A watertight dural closure was done, muscular and aponeurosis layers are tightly closed (**Fig. 4F**).

DISCUSSION

Foramen magnum meningiomas (FMMs) arise from meningothelial cells of arachnoid mater (not dura) in the craniospinal junction.^{1,3} FMMs are relative rare and comprise only 1.8 to 3.2% of all meningiomas.⁴ Yet, meningiomas itself comprise 70% of benign tumors that arise at the foramen magnum (FM).⁹ The border of this lesion has been defined by Bruneau *et al.*^{5,6} which anteriorly it is bordered by the lower third of the clivus to the upper margin of C2, laterally from the

Figure 3. Patient was in prone position and incision was marked.

Figure 4. Intraoperative images of tumor resection. **A.** Paramedian duramater excision exposing spinal cord (black arrow head) and meningioma (asterix) extending to right lateral. **B.** Releasing the duramater attachment of the meningioma. **C.** Part of the meningioma (asterix) was exposed. **D.** Debulking of the meningioma around right C2 nerve root (black arrow). **E.** Removal of the meningioma (asterix) was done with piece-meal fashion around right C1 nerve root (black arrow). **F.** Closure of duramater using watertight dural closure.

jugular tubercle to the upper margin of C2 lamina and posteriorly from the anterior edge of squamous occipital bone to the spinous process of C2.

In this case report, the patient was a female and the diagnosis of FMM was made at 49 years of age. As mentioned in literature, females are more likely to suffer from meningioma than males with a ratio of 1.8:1 and the incidence peaks at 45 years of age.¹ The patient also had history of using 3-month injectable hormonal contraception for more than 5 years. This finding is compelling, as study by Wahyuhadi *et al.*¹⁰ reported association between meningioma and exogenous hormone exposure. Patients with history of hormonal contraception usage had 12.31 times higher risk for meningioma, with the 3-month injectable contraception has the highest risk rate compared to 1-month injectable and oral contraception.¹⁰

FMM management needs a comprehensive anatomical knowledge, due to many neural and vascular structures in FM.^{4,11,12} The neural structures involved are lower cranial nerves (CN IX-XII), upper cervical nerves (C1 and C2), cerebellar tonsils, inferior vermis, fourth ventricle, medulla, and spinal cord.^{4,7} Major arterial structures within FM are vertebral arteries (VA), posterior inferior cerebellar arteries (PICAs), anterior and posterior spinal arteries, and the meningeal branches of vertebral, external, and internal carotid arteries.^{11,12}

Many classifications have been proposed to help surgeon choose the most appropriate approach.^{2,4,6,9} Based on its vertical extensions, FMM can be divided into two main types: craniospinal (originates intracranially and extends downward) and spinocranial (originates in upper spinal canal and extending intracranially).² According to the compartment of development, FMM can be classified into intradural (which is the most common location), combination of intra and extradural, and rarely, strictly extradural.^{1,6} According to their anteroposterior and lateromedial orientation of dural insertion, FMM can be subdivided into posterior, lateral, or anterior.^{4,6} The lesion is considered posterior if its insertion is posterior to the

dentate ligament; lateral, if its insertion is between the midline and the dentate ligament; or anterior, if its insertion is on both sides of anterior midline.⁶ Most FMMs arise anterolaterally (68 – 98%), followed by posterolateral, purely posterior, and the least one is anterior only.⁴ FMM can also be classified according to the size relative to the foramen magnum. The lesion is considered small if less than one third of the transverse dimension of foramen magnum; medium, if one third to one half of foramen magnum size; and large, if the lesion is more than one half of foramen magnum transverse dimension.⁴ Relation of FMM to the VA can also classify FMM into 3: type-A is tumors arise below VA and grow upwards; type-B is tumors arise above VA and grow downwards; and type-C is VA coursing across the lesion with or without encasement.⁹

In our case, the lesion was anterior intradural extramedullary FMM that extends laterally to the right side. For managing this anterolateral FMM, we chose the suboccipital midline approach with C1 laminectomy and C2 superior partial laminectomy. The surgical procedure was chosen with consideration of familiarity and preservation of anatomical structures for craniocervical stability, as the surgeon planned to not using implant.

Suboccipital craniotomy, or craniectomy, with or without cervical laminectomy represents the classic posterior approach to the foramen magnum meningiomas and is familiar to most neurosurgeons.⁴ This approach has become the standard management for all posterior or postero-lateral located lesions.⁷ Resection of the posterior arch of atlas provides a more decent exposure of the dura over the craniocervical junction and is used by many of surgeons during a suboccipital craniotomy.⁹ A limited posterior laminectomy is also a routine for FMMs extending to the cervical spine that needs opening of the cervical canal.⁹

On the other hand, anterior located lesions have been considered to require more complex approaches such as the anterior transoral, the transcervical retropharyngeal, the posterolateral approach (far lateral approach), or the anterolateral approach (extreme-lateral

approach).⁴⁻⁷ The far lateral approach is preferred for intradural FMM located in the lateral or anterior, anterior FMM that extends beyond the midline, and extradural FMM that developed on the posterior part of lateral foramen magnum wall.^{5,8,13} Whereas, the extreme lateral approach has only been recommended for FMM with extradural extension through the bony structures.⁵

Even though there are alternative approaches for anterior FMM described recently in the literatures, the conventional suboccipital midline approach can still be considered. The consideration of conducting this approach is due to the fact that FMMs are rarely strictly anterior and majority of the tumors push the spinal cord to posterolateral.^{4,14} Hence, FMMs are seen immediately in the surgeon's point of view with this approach.¹⁵ Some studies have reported several advantages of performing suboccipital midline approach for anterior or anterolateral FMM.¹⁴⁻¹⁷ First, it is more familiar to most neurosurgeons and less complex than the far lateral or extreme lateral approach.^{4,17} Second, this posterior approach can provide wide enough working space after the tumor has started to be debulked, moreover in patients with large foramen magnum lesions.¹⁵⁻¹⁷ Thus, drilling the condyles nor lateral mass is not needed.^{14,17} Third, craniocervical stability is preserved through limited bone resection.¹⁵ Fourth, postoperative recovery is fast and clinical presentation is improving with low case of cerebrospinal fluid leak.^{15,17} Fifth, the overall rate of morbidity and mortality were minimal.^{15,17} In addition, the technique that needs to be noted while conducting the suboccipital midline approach is the paramedian incision to the duramater, because it allows the surgeon to progressively pull the tumor from anterior to lateral while minimizing spinal cord retraction.^{14,15}

In our case, the surgery went uneventful with no complication observed post-surgery. The complaints were diminished immediately after the surgery, with remarkable motor function during 4 months follow-up.

CONCLUSION

FMM remains a challenge for neurosurgeons due to many neural

and vascular structures in the location. Extensive skull base approach may help in the resection of FMM, but it is not essential for all patient with FMM. Ultimately, the management that were chosen should be tailored for each individual to achieve the maximal resection and preventing surgical morbidity. As for our case, suboccipital midline approach with C1 laminectomy and C2 partial laminectomy is a safe and feasible surgical procedure to treat anterolateral FMM.

CONFLICT OF INTEREST

Authors declare no conflict of interest regarding the publication of this paper.

FUNDING

Authors have received no funding or any financial inducement.

AUTHOR CONTRIBUTION

Ayu Yoniko Christi: assisted in the operation, edited the manuscript, and updated the references; Irfaanstio: drafted the manuscript, collected the data, and followed up the patient; Vega Pangaribuan: drafted the manuscript, edited the manuscript, and examined the patient; Pandu Wicaksono: assisted in the operation, reviewed the manuscript and updated the references; Andrianto: lead surgeon during the operation, and reviewed the manuscript; Wisnu Baskoro: admitted the patient into hospital, reviewed the manuscript and assisted in the operation.

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