



Case Report

Foramen magnum meningioma presented as cervical myelopathy in a pregnant COVID-19 patient: A case report

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ABSTRACT

Introduction: Meningioma is the second most common primary brain tumor. There are approximately 5.6 cases of meningioma per 100,000 pregnant women. Foramen magnum meningioma is rare, and the diagnosis, treatment, and prognosis are complex in pregnant women.

Case presentation: Herein, we report a case of foramen magnum meningioma in a pregnant woman at 32 weeks of gestation, who presented with chronic neck pain and cervical myelopathy. She tested positive for COVID-19 infection. Magnetic resonance imaging findings were compatible with foramen magnum meningioma, and the pathologic analysis revealed a WHO grade-I meningioma. The patient underwent cesarean section followed by tumor excision due to fetal distress and rapid deterioration.

Clinical discussion: Management of meningioma during pregnancy requires a multidisciplinary approach. No guidelines for surgical intervention, timing of pregnancy termination, or mode of delivery for pregnant patients with foramen magnum meningioma have been established. While it is best to prolong the pregnancy for as long as possible, a cesarean delivery is preferred to avoid increased intracranial pressure. Operative management of meningioma is warranted if the tumor is growing or symptomatic. This patient died due to the added complication of COVID-19. Although the prognosis of foramen magnum meningioma is usually favorable, COVID-19 comorbidity can increase illness severity.

Conclusion: Maternal and fetal health status must be evaluated to decide whether surgical excision and pregnancy termination are needed. In this case, COVID-19 infection and meningioma disease course required further investigation.

1. Introduction

Meningioma is the second most common primary brain tumor. This tumor, which is benign and slow-growing, affects females more frequently than males [1–3]. There are approximately 5.6 cases of meningioma per 100,000 pregnant women [4]. Foramen magnum meningioma is rare and accounts for 1.8%–3.2% of all meningiomas [3–5]. The clinical diagnosis is complex because of the anatomical location and because the initial symptoms are diagnostically nonspecific. The prognosis of foramen magnum meningioma is usually favorable with mortality rates range from 0% to 16.6% [3], but in this case, COVID-19 comorbidity can increase illness severity.

2. Case presentation

A 39-year-old female at 32 weeks of gestation presented to the emergency room with shortness of breath and progressive limb weakness. The patient had a 2-year history of worsening neck pain. She initially experienced weakness in her right arm 2 months before admission, followed by her left arm and both legs. There was no complaints of anosmia, seizure, blurred vision, slurred speech, dysphagia, hoarseness, or skewed face. She also experienced numbness from the neck down, but no urination or defecation disturbances were noted. She had a fever since 3 days before admission. The patient had no medical or family history of cancer, but she did have a history of contraceptive injection for 15 years. She had consumed ibuprofen and paracetamol for neck pain for 2 years.

Abbreviations: COVID-19, Coronavirus disease 19; IL-6, Interleukin 6; TNF- α , Tumor Necrosis Factor Alpha; VEGF, Vascular endothelial growth factor; MRI, Magnetic Resonance Imaging; PCR, Polymerase Chain Reaction.

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On physical examination, we found intact mental status, no meningeal signs, and cranial nerve palsy. Motor and sensory examination revealed tetraplegia and hypoesthesia below the C3–C4 spinal cord segments. Proprioception was abnormal in all extremities. Tendon reflexes were increased in both arms. Babinski and Chaddock reflexes were positive on both sides. PCR testing of a nasopharyngeal swab was positive for COVID-19. The chest radiography revealed paracardial infiltrates. Her blood gas analysis indicated respiratory failure. Abnormal laboratory results included increased D-dimer (2000 mg/dL) and hypokalemia. Our patient was admitted to the intensive care unit. She was intubated and treated with high-dose n-acetylcysteine, remdesivir, zinc, vitamin D, and vitamin C. Due to fetal distress, she underwent an emergency cesarean section, performed by our obstetrician under general anesthesia. The surgery was successful, but the baby did not survive.

Cervical MRI revealed a mass with homogenous contrast enhancement measuring 15 mm × 17 mm × 23 mm at the foramen magnum at the level of the second cervical vertebra that caused spinal cord edema (see Fig. 1). She was initially administered steroids; however, due to rapid deterioration, our experienced consultant neurosurgeon performed the tumor excision by craniotomy with a far lateral approach. Histopathological examination of the resected tissue revealed transitional and angiomatous type WHO grade 1 meningioma. A whorl pattern, consisting of proliferating meningothelial cells with oval to spindle nuclei, psammoma bodies, and the proliferation of blood vessels with hyalinization were observed. The patient's awareness increased briefly after surgery, but her condition continued to deteriorate. The patient was under close monitoring due to her worsening clinical condition. In the intensive care unit, her blood pressure decreased to 60/30 mmHg. She was administered norepinephrine, as the cause of shock was suspected to be sepsis. The patient eventually died due to septic shock and respiratory failure.

3. Discussion

Foramen magnum meningioma is a rare type of meningioma. The challenges in managing this patient were primarily due to the rarity of the tumor, its unusual nonspecific initial symptoms, and the accompanying pregnancy [6,7]. Prompt diagnosis is essential because this tumor is located close to areas that control vital functions. However, foramen magnum meningioma is often misdiagnosed. Bahrami et al. reported 2 cases of foramen magnum meningioma mistaken for carpal tunnel syndrome due to the similarity of symptoms [8]. Some reported cases of foramen magnum meningioma presented as occipital neuralgia, drop attacks, brainstem compression, and cervical myelopathy [5,9–11]. Delayed diagnosis may hinder treatment and increase morbidity and mortality [3].

Pregnancy in this patient may have contributed to tumor growth; in pregnancy, follicle-stimulating hormone and luteal hormone levels are low due to negative biofeedback. These hormones have been shown to inhibit tumor cell proliferation *in vitro* [4]. After the tenth week of pregnancy, the placenta produces estrogen and progesterone, and most meningiomas express progesterone receptors. Some studies reported that tumor growth is faster in the luteal phase of the menstrual cycle and in the second and third trimesters of pregnancy, when the level of progesterone is high [4,12]. Hemodynamic changes, including swollen cells, increased vascularity, intra or extracellular also occur during pregnancy, which could affect tumor size [4].

Management of meningioma during pregnancy requires a multidisciplinary approach. No guidelines for surgical intervention, timing of pregnancy termination, or mode of delivery have been established for pregnant patients with foramen magnum meningioma. Ideally, the pregnancy should be prolonged as long as possible [12]. Several complications and side effects of cesarean delivery, including neurological effects, have been reported [13,14]. However, cesarean delivery is

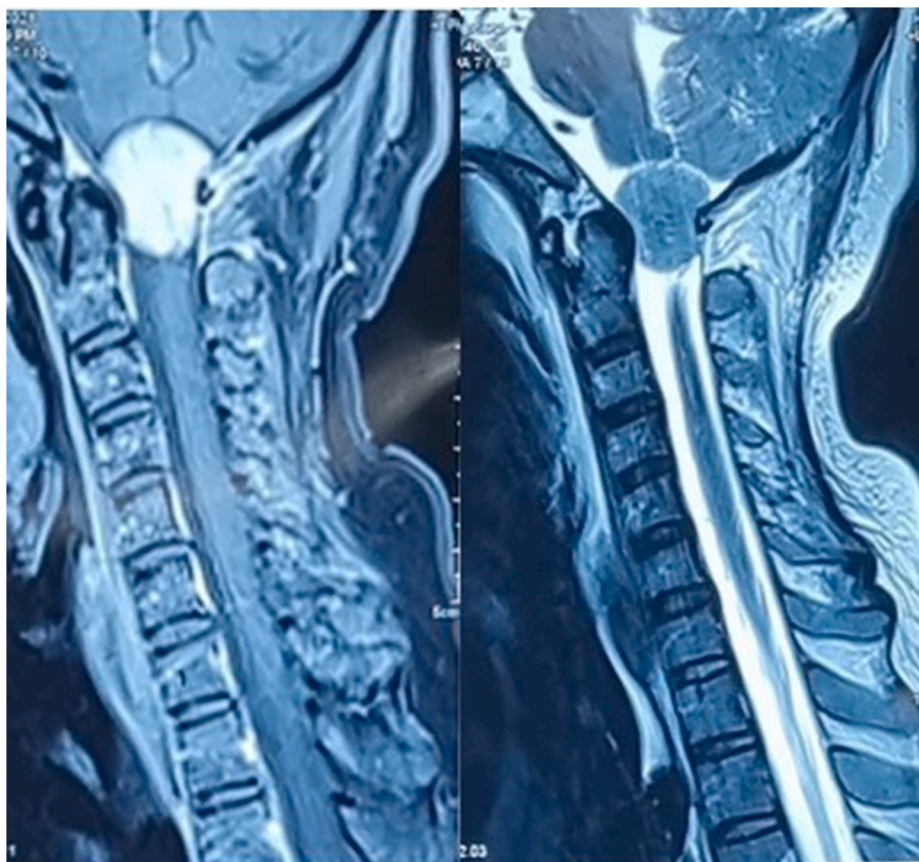


Fig. 1. Brain MRI, Sagittal view revealed a mass in the foramen magnum.

preferred to avoid increased intracranial pressure [12–15]. Operative management of meningioma is recommended if the tumor is growing or symptomatic [16]. Otherwise, it is recommended to follow up with routine MRI [15,16]. Tumor excision following pregnancy termination may be considered if the patient's life is threatened or a serious neurological deficit is present, such as decreased consciousness, midline shift, acute loss of vision, or hydrocephalus [15]. Our patient rapidly deteriorated, and fetal distress necessitated an urgent cesarean section followed by craniotomy. After tumor excision, the patient briefly regained consciousness but then she experienced septic shock, respiratory failure and died.

COVID-19 causes a hyperinflammatory state that may lead to tumor progression [17,18]. Interestingly, cytokine expression is increased in both meningioma and during COVID-19 infection [17,19]. In patients with SARS-CoV-2 who have the more severe form of COVID-19, neutrophils, TNF- α , and IL-6 were found to be elevated, while the monocyte, basophil, and eosinophil counts were decreased. Plasma IL-6 signaling may result in more inflammation and tissue damage [20]. COVID-19 infection is mediated by immune and nonimmune cells, activates the IL-6 signaling pathway and triggers the IL-6 inflammatory enhancer, a positive feedback loop that leads to the production of more IL-6, VEGF, and other chemoattractants. Due to its important role in immune-mediated inflammation, IL-6 supports tumorigenesis by directly stimulating tumor cells and an indirect effect on the tumor microenvironment. IL-6 stimulates growth, metastasis, and evasion of the immune system in several tumor types. Direct effects of IL-6 on cancer cells also include promotion of stem cell properties, induction of mesenchymal features, and therapeutic resistance. IL-6 orchestrates the interactions between cancer cells and the tumor microenvironment by preparing the foundation for cancer establishment in target organs and has also been shown to stimulate immune evasion by promoting stabilization of programmed death ligand [20,21].

Further investigation is needed to determine a molecular link between COVID-19 and the disease course of meningioma. Typically, the prognosis of foramen magnum meningioma is favorable, but our patient experienced the added complication of COVID-19 infection.

4. Conclusion

Complete history and thorough physical examination, followed by imaging, are critical for the diagnosis of foramen magnum meningioma. Due to its anatomical location, rapid diagnosis and determination of treatment modality are crucial. Maternal and fetal health status must be evaluated to decide whether surgical excision and pregnancy termination are needed. The way in which COVID-19 infection affects the disease course of meningioma requires further investigation.

The work has been reported in line with the SCARE 2020 criteria [22].

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Author contribution

All authors contributed to this work, commented on the manuscript at all stages, and finally the final version was approved for publication.

OJW : Study conception and design, data collection and draft manuscript preparation; DA: study design and manuscript editing.

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Guarantor

The guarantor was also the corresponding author.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Declaration of competing interest

None.

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