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Case Report

Recurrent posterior fossa intracranial capillary hemangioma in a pregnant woman: A case report and review of literature

Mohammad Ali Abouei Mehrizi [a], Humain Baharvahdat [a,b], Sajjad Saghebdoust [a,b,*]

[a] Department of Neurosurgery, Faculty of Medicine, Mashhad University of Medical Sciences, Mashhad, Iran
[b] Section of Neurovascular Intervention, Department of Neurosurgery, Ghaem Hospital, Mashhad University of Medical Sciences, Mashhad, Iran

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ABSTRACT

Introduction: Intracranial capillary hemangioma (ICH) is a rare tumor with a slightly higher incidence in women. Surgical management of ICH during pregnancy requires a joint decision, for it has been a contentious issue owing to the inadequate number of cases.

Presentation of case: A 23-year-old female with an ICH in the posterior fossa underwent subtotal tumor resection (STR). One year later, she was referred to the neurosurgery department in the 8th week of pregnancy due to a progressive headache, vomit, and a bulging occipital mass. Subsequent to an abortion, preoperative angiography and Bleomycin injection were performed, and the tumor was totally resected thereafter. Over a one-year follow-up, her symptoms faded, and she experienced no recurrence.

Discussion: ICH, a highly vascular entity, poses a substantial intraoperative bleeding risk. During the first surgery, intraoperative bleeding deterred the surgeon from a gross total resection (GTR), leading to a recurrence due to the growth of residual tissue during pregnancy. Having aborted the fetus, the patient underwent angioembolization to lessen the risk of intraoperative bleeding before reoperation, rendering it more possible for the surgeon to achieve GTR. Accordingly, quality of resection and a multidisciplinary approach is necessary to ensure optimal treatment.

Conclusion: There is a growing consensus that not only is STR a contributory factor in ICH recurrence, but female hormones and increased cardiac output in pregnancy might play a pivotal role in the progression of the tumor. Thus, paying further attention to pregnant or peripartum patients with suspected ICH should assume greater significance.

1. Introduction and importance

Hemangioma is a benign tumor characterized by rapid vascular growth, followed by a course of involution. The condition was first proposed by Virchow in 1867 [1]. This type of tumor is usually found as a congenital mass during the neonatal period. It can involve different parts of the body's skin, including the head and neck (60%), trunk (25%), and extremities (15%), as well as internal organs. In some cases, the internal organ involvement is present without evident cutaneous lesion. In this regard, some patients may be asymptomatic until adulthood [2].

Rarely hemangiomas can involve the central nervous system (CNS). CNS involvement includes meninges, cauda equina, nerve roots, and spinal cord, but nerve roots and cauda equina are the most commonly affected parts [3]. A relative predominance of the female gender and hormonal changes can affect tumor growth, and pregnancy can be a trigger for tumor enlargement. Moreover, an increase in cardiac output and considerable fluid retention during pregnancy is deemed to accelerate tumor progression [4]. It is reported that hemangiomas are usually located in cranial convexity, middle fossa, and posterior fossa in 26%, 28%, and 25% of the cases, respectively [5]. To the best of our knowledge, we report the first case of a recurrent extradural hemangioma in the posterior fossa one year after ICH removal surgery due to an unplanned pregnancy. This case report has been reported in line with the SCARE Criteria [6].

2. Case presentation

A 23-year-old female presented with symptoms of severe progressive headache, blurred vision, and gait disturbances. The patient did not

* Corresponding author. Department of Neurosurgery, Faculty of Medicine, Mashhad University of Medical Sciences, Mashhad, Iran.
E-mail address: sajjadsaghebdoust921@gmail.com (S. Saghebdoust).

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report diplopia, facial weakness, otalgia, otorrhea, or any mastication disorder. The psychosocial, family, and drug history also had no notable findings. Moreover, physical examination showed no focal neurological deficit (FND). Corneal reflex, ocular motility, and pupillary function were normal, and no other abnormalities were noted. The patient underwent computed tomography (CT) scan with contrast injection revealing a large enhanced tumor in the left hemisphere of the cerebellum (Fig. 1-A), and preoperative magnetic resonance imaging (MRI) revealed the presence of a heterogeneous extra-axial enhancing mass (Fig. 1-B). With suspicion of an enlarged ICH, the patient underwent open surgery by the first author. The tumor vascularity caused intraoperative bleeding deterred the neurosurgeon from gross total resection (GTR) of the tumor, and the tumor residue was seen in the postoperative MRI. The microscopic evaluation of the extracted extradural mass demonstrated a benign vascular neoplasm consisting of dilated cystic vessels with thin endothelium, confirming the diagnosis of hemangioma. After the first surgery, the patient was told to comply with a follow-up program due to subtotal resection (STR) of the tumor, and she was asked to visit our clinic anytime she experienced any new symptoms indicative of tumor recurrence.

The patient had an unintentional pregnancy one year after the surgical treatment. During the 8th week of pregnancy, she was referred due to our neurosurgical department with severe progressive headache, vomit, and a bulging mass in the left occipital bone. On account of her progressive symptoms indicative of increased intracranial pressure and with the diagnosis of ICH recurrence, a collective decision was made by a group of neurosurgeons and gynecologists to proceed with abortion in order to perform surgical intervention afterward. Subsequently, minimally invasive angiography and embolization using Bleomycin were conducted by our neurointerventionist so as to devascularize the tumor (Fig. 2), and open surgery was performed thereafter. The patient was told to adhere to a scheduled follow-up program over a period of 12 months. Accordingly, she was visited at 6 and 12 months postoperatively, experiencing no FND. Moreover, her postoperative MRI showed nothing indicative of tumor recurrence.

3. Clinical discussion

Hemangiomas are considered tumors of infancy; however, they can present in around 10% of the cases during adulthood. The most common affected sites are skin and oral mucosa, but uncommonly other internal parts can be involved, too. The involvement of the central nervous system is very rare, and a recent literature review by Massman et al. [7] showed that less than 50 cases had been proposed to occur in CNS.

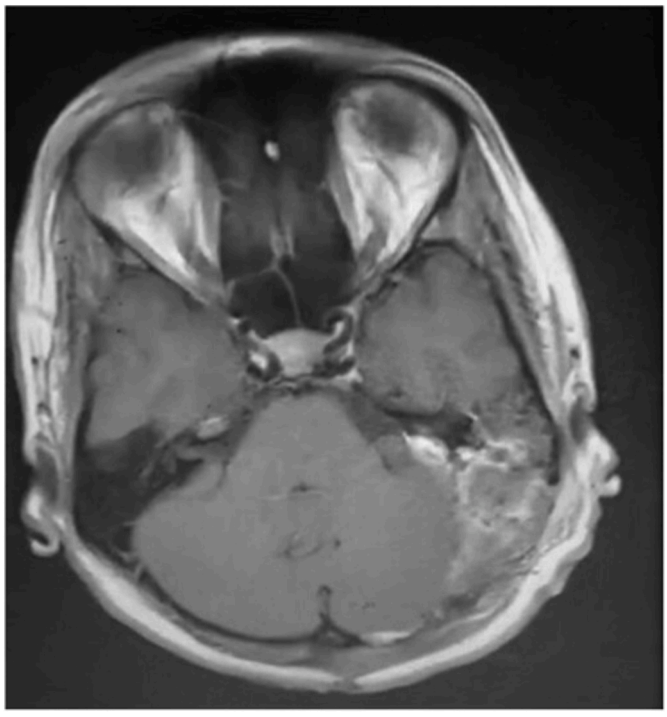

Fig. 2. Post-gadolinium axial T1-weighted MRI showed successful tumor devascularization after angioembolization with Bleomycin prior to the second craniotomy.

Moreover, of 25 women reported to have harbored ICH, 5 cases were pregnant. We provide a comprehensive review of the pertinent literature regarding the individuals with an ICH during the peripartum period (Table 1).

To date, only seven women, including the present case, were diagnosed with ICH during the perinatal period, and four were pregnant at the time of ICH diagnosis [4,7]. Having reviewed the literature, we report our patient as the only one who experienced an ICH recurrence during pregnancy. It is reported that the production of angiogenic and unknown growth factors during pregnancy might have a pivotal role in the rapid growth of this entity. Furthermore, it is proposed that higher blood flow in pregnant women can also be another pathophysiology for tumor progression. Still, due to a dearth of adequate cases, the exact underlying mechanism of the disease flare during pregnancy is not

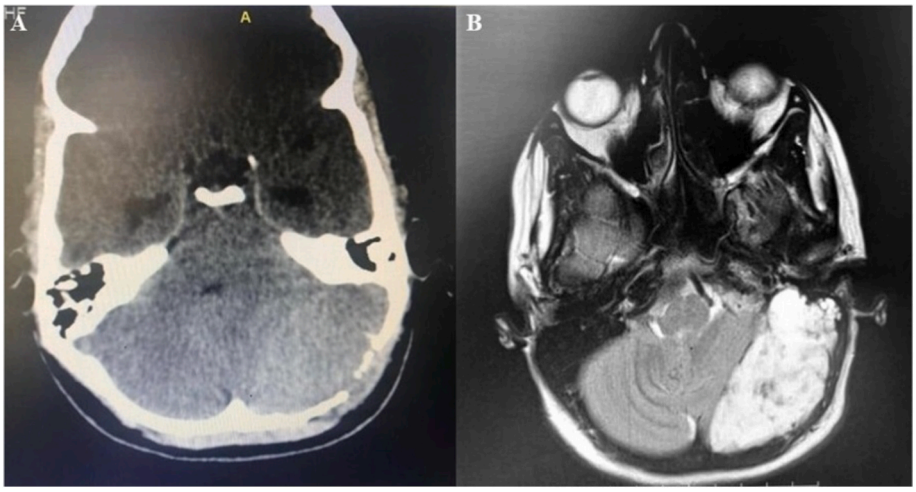

Fig. 1. Preoperative computed tomography showed an iso to a hyperdense mass lesion in the left cerebellar hemisphere with mass effect and adjacent bone destruction (A). Axial T2-weighted MRI revealed a T2-hyperintense extra-axial mass with extensive vasogenic edema (B).

Table 1
Summary of cases with Intracranial capillary hemangioma diagnosed during the peripartum period ICH: Intracranial capillary hemangioma, GTR: Gross total resection, STR: Subtotal resection, CN: Cranial nerve, eTSS: endoscopic transsphenoidal surgery, NA: Not available.

Author/ Year	Age (year)	History of ICH prior to pregnancy	Time of diagnosis	Delivery	Location	Dura attachment	Symptoms	Treatment	Follow-up
Present case/ 2022	23	Yes	8 weeks of gestation	Abortion	Posterior fossa	Yes	Headache, vomit	Angioembolizaion with Bleomycin and craniotomy (GTR)	No recurrence (12 months)
T. Ishikawa et al.[6]/ 2022	28	No	30 weeks of gestation	Spontaneous	Dorsum sellae	Yes	CN 6 palsy	eTSS (GTR)	No recurrence (18 months)
Massman et al.[5]/ 2021	23	No	5 weeks after delivery	NA	Cavernous sinous	Yes	None	eTSS (GTR)	No recurrence (12 months)
Mirza et al.[12]/ 2012	28	No	Few months after delivery	NA	Transverse sinus	Yes	Migraine-like headache	Craniotomy (GTR), perioperative Dexamethasone infusion	No recurrence (12 months)
	41	No	3.5 months after delivery	NA	Convexity (Occipital dura)	Yes	Headache during pregnancy and vague visual disturbance after delivery	Craniotomy (GTR), perioperative Dexamethasone infusion	No recurrence (13 weeks)
Smith et al.[13]/ 2007	26	No	36 weeks of pregnancy	Cesarean	Middle cranial fossa	Yes	Headache, vomit, photophobia, confusion and CN 6 palsy	Craniotomy (GTR), preoperative Dexamethasone infusion	Symptom resolution (4 months)
Simon et al.[7]/ 2004	31	No	38 weeks of gestation	Cesarean	Cerebellar tentorium	Yes	Headache, vomit	3 Craniotomies (2 STR, 1 GTR)	No recurrence after the third craniotomy (41 months)

thoroughly known [7,8].

The most common symptom of ICH is headache [7], as per the case of our study, accompanying nausea and/or vomiting, and probably FND. However, these symptoms are non-specific, and imaging and histology are indispensable for the diagnosis. ICHs appear as well-circumscribed, largely homogeneous, avidly enhancing lesions that are hypointense to isointense on T1-weighted MRI and hyperintense on T2-weighted imaging. The mass presents itself in the CT scan as a cystic lesion with slightly high attenuation encircled by a vast area of low attenuation, and contrast imaging can add to the accuracy of diagnosis. By all accounts, the final diagnosis is established through microscopic evaluation, depicting a mesh of vascular spaces made of thin epithelium. The presence of CD31 and CD34 markers in immunohistochemistry is contributory as well [9].

The cornerstone of the treatment is an entire resection of the tumor, whereas biopsy and piece resection is accompanied by a high risk of bleeding and is contraindicated. In this regard, it is sensible to infer that our case experienced recurrence owing to the STR of the tumor, which would have been prevented if total resection had been achieved in the first surgery. In order to reduce the risk of bleeding, the surgeon may decide to use preoperative embolization. However, the vascular nature of the tumor should be defined before any preoperative intervention [10]. It is believed that corticosteroid therapy can be used as an adjunct treatment due to its anti-angiogenic nature, and it can also be helpful in the regression of tumor residuals. It is proposed that recurrence seldom happens [11], whilst our case showed that it could have a recurrence, especially during hormonal changes of pregnancy. Furthermore, there is a consensus that medical treatment not accompanying partial resection can also be a risk factor for tumor recurrence [12].

4. Conclusion

Intracranial capillary hemangioma is a scarce tumor presenting non-specific symptoms like headache and neurological deficits. Due to a paucity of information, no consensus of opinion has been reached about the nature of the disease. Nonetheless, the standard surgical and medical treatments have demonstrated a favorable prognosis in affected individuals. Based on pertinent literature [10] and our experience, not only can ICHs recur in case of STR, but they also tend to grow dramatically due to hormonal changes during pregnancy. Although making an inference based on a single case is challenging and requires further investigation, it would be wise to pay more attention to pregnant or peripartum patients harboring a suspected ICH.

Ethical approval

Ethical approval clearance was not required in the treatment of the patient in this report.

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Author contribution

Mohammad Ali Abouei Mehrizi contributed in the data collection, interpretation, and leadership responsibility for the research activity planning and execution, including mentorship external to the core team.

Humain Baharvahdat contributed in data collection and interpretation.

Sajjad Saghebdoust contributes in the study concept or design, writing the paper and interpretation.

Registration of research studies

1. Name of the registry: None.
2. Unique Identifying number or registration ID: None.
3. Hyperlink to your specific registration (must be publicly accessible and will be checked): None.

Guarantor

The guarantor of this study is Sajjad Saghebdoust MD.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Provenance and peer review

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Declaration of competing interest

The authors declare that there is no conflict of interest regarding publication of this paper.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2022.104913.

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