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Progression of cerebellar chronic encapsulated expanding hematoma during late pregnancy after gamma knife radiosurgery for arteriovenous malformation

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Abstract

Background: The etiology and appropriate management strategy of chronic encapsulated expanding hematoma during pregnancy after gamma knife radiosurgery for arteriovenous malformation (AVM) remain unclear.

Case Description: A 34-year-old female developed chronic encapsulated expanding hematoma during late pregnancy, after angiographic disappearance of cerebellar AVM following two courses of gamma knife radiosurgery. The present case implicates pregnancy as a potential promoter of growth and enlargement of chronic encapsulated expanding hematoma, which may become life-threatening and require surgical intervention.

Conclusion: Immediate surgical management after delivery may be associated with a favorable outcome, so close follow-up management and patient education are very important in women planning pregnancy.

Key Words: Arteriovenous malformation, gamma knife, pregnancy, radiosurgery

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INTRODUCTION

Gamma knife radiosurgery is an effective treatment for cerebral arteriovenous malformation (AVM) resulting in angiographic disappearance in more than 80-90% of cases. Actual rates of obliteration of cerebellar AVMs, with median target volume of 3.85 cm^3 and median marginal dose of 21 Gy, were 53% at 3 years and 76% at 5 and 10 years.[1] However, relatively rare complications such as cyst formation and chronic encapsulated expanding hematoma may develop more than 5 years after gamma knife radiosurgery even if angiographic disappearance has been achieved.[4,10,15,16,19] Furthermore, little is known about the mechanisms and appropriate management of chronic encapsulated expanding hematoma during pregnancy. We present a case of cerebellar chronic encapsulated expanding hematoma encountered during late pregnancy, 4 years after angiographic disappearance of AVM nidus following two courses of gamma knife radiosurgery, with a cumulative dose of 44 Gy to the margin at the 50-60% isodose line, carried out at an interval of 4 years.

CASE REPORT

A 20-year-old female presented with sudden onset of severe headache associated with nausea followed by disturbance of consciousness, and was admitted to another hospital. The diagnosis of cerebellar and subarachnoid hemorrhage from AVM supplied by the

posterior inferior cerebellar artery (PICA) was based on the findings of computed tomography (CT) [Figure 1a] and cerebral angiography. Emergency evacuation of the hematoma was performed via a midline suboccipital approach. Four months after surgery, vertebral angiography demonstrated left cerebellar hemispheric AVM supplied by the PICA [Figure 1b]. Fourteen months after the initial hemorrhage, gamma knife radiosurgery was performed to treat the AVM nidus with a volume of 0.487 cm^3 at another institution using a Leksell Gamma Knife model B unit (Elekta AB). The procedure was planned using GammaPlan software based on stereotactic digital subtraction angiography and magnetic resonance (MR) imaging. A prescribed dose of 20 Gy was delivered to the lesion margin at the 50% isodose line. Three years after the first radiosurgery, vertebral angiography showed a small residual nidus in the left cerebellar hemisphere [Figure 1c]. The patient underwent repeat radiosurgery at the previous institution using a Leksell Gamma Knife model C unit (Elekta AB) 4 years after initial radiosurgery. The target volume of the nidus was 1.5 cm^3, a larger volume than that at the initial radiosurgery, and was intended to improve the treatment efficacy. The procedure was planned using GammaPlan software and a prescribed dose of 24 Gy was delivered to the lesion margin at the 60% isodose line. Vertebral angiography obtained at 5 years after the second radiosurgery revealed complete disappearance of the AVM [Figure 1d]. However, T2-weighted MR imaging and postcontrast T1-weighted MR imaging obtained at 7 years after the second radiosurgery revealed an enhanced lesion adjacent to the cyst formation in the left cerebellar hemisphere [Figure 2a, b]. The patient was lost to follow up during the 18 months after the last examination. The patient subsequently presented with headache and nausea, which had persisted over 3 weeks, at age 34 years in the 32nd week of pregnancy, and was referred to our institution 9 years after the second radiosurgery.

The patient had headache and nausea, but no other neurological deficits were identified except for House–Brackmann grade 3 facial palsy persisting since her childhood. Other medical history was unremarkable. On admission, CT demonstrated an irregularly shaped, heterogeneous high density hematoma with perifocal edema in the vermis extending to the left cerebellar hemisphere [Figure 2c]. Her infant was delivered by cesarean section immediately after admission and osmotic therapy was started. Despite conservative management, disturbance of consciousness developed and deteriorated due to the extensive perifocal edema and hydrocephalus. Three-dimensional CT angiography revealed no vascular abnormality around the lesion.

Midline suboccipital craniotomy was performed and cerebrospinal fluid was released from the ventricular drainage. A very firm, reddish angiomatous nodular granuloma with adjacent cyst was visualized in the cerebellar hemisphere. Indocyanine green videoangiography confirmed the absence of abnormal vasculature around the lesion. The lesion contained angiomatous capsule and firm organized hematoma. Gross total resection was achieved without injury to the surrounding structures. No AVM nidus was observed during surgery. Her symptom was completely resolved

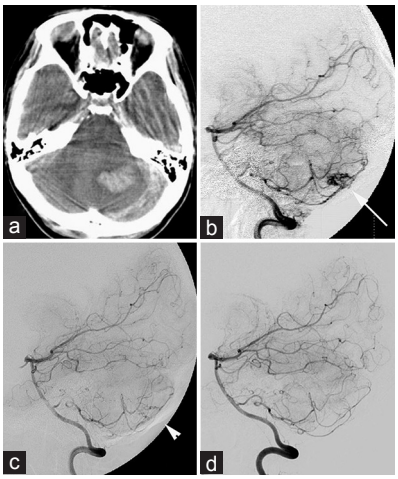

Figure 1: (a) CT scan at initial onset demonstrating left cerebellar hemorrhage with subarachnoid hemorrhage. (b) Left vertebral angiogram before first gamma knife radiosurgery showing a left cerebellar hemispheric AVM supplied by posterior inferior cerebellar artery (*arrow*). (c) Left vertebral angiogram at 3 years after first radiosurgery revealing residual nidus in the left cerebellar hemisphere (*arrowhead*). (d) Left vertebral angiogram obtained at 5 years after the second radiosurgery revealing no residual AVM nidus

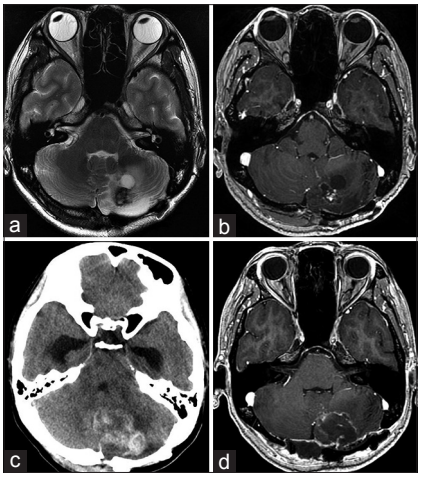

Figure 2: Axial T2-weighted MR image (a) and postcontrast T1-weighted MR image (b) demonstrating appearance of the enhanced lesion adjacent to the cyst formation in the left cerebellar hemisphere at 7 years after second radiosurgery. (c) CT scan showing an irregularly shaped, mixed density lesion with extensive edema in the left cerebellar hemisphere. (d) Postoperative gadolinium-enhanced T1-weighted MR image demonstrating total removal of the lesion

immediately after surgery and the postoperative course was uneventful. Postoperative MR imaging revealed total resection of the hematoma with the adjacent cyst [Figure 2d]. Postoperative angiography confirmed disappearance of the AVM. Her baby's growth and development was also normal.

Histological examination of the lesion obtained during surgery demonstrated encapsulated hematoma consisting of a dense collagenous outer layer and a granulated, newly vascularized, angiomatous inner layer with extensive multinodular hemorrhage at various stages of organization [Figure 3a]. Hemosiderin deposits and coagulation necrosis were also observed [Figure 3b]. The microvasculature in the inner layer demonstrated inflammatory infiltration in the vascular walls and thickening of the vessel walls with hyaline degeneration, which are characteristic findings of vasculitis [Figure 3c]. Immunohistochemical examination demonstrated strong staining for CD34 in the microvasculature [Figure 3d].

DISCUSSION

Chronic encapsulated expanding hematoma after gamma knife radiosurgery

Chronic encapsulated expanding hematoma is a rare but very important late onset complication after gamma knife radiosurgery for AVMs, and may develop even if angiographic disappearance has been achieved. Surgical treatment may be required due to progression in some cases.[10,15,19] Chronic encapsulated expanding hematoma is often accompanied by cyst formation, which tends to occur in patients followed up for longer than 5 years after gamma knife radiosurgery.[16] Larger nidus volume and higher radiation dose may be risk factors for delayed cyst formation,[4] but cyst formation may still occur despite a relatively small nidus and low prescribed margin dose.[19] Total obliteration can be achieved after repeat stereotactic radiosurgery (SRS) for incomplete obliteration after initial SRS.[8,9,13,23] Delayed cyst formation occurred in 4.6% of cases at a median of 108 months after repeat SRS. In the present case, chronic encapsulated expanding hematoma occurred 9 years after the second radiosurgery for the relatively small residual nidus. The cumulative radiation dose was 44 Gy to the lesion margin, which was presumably high enough to induce the hematoma.

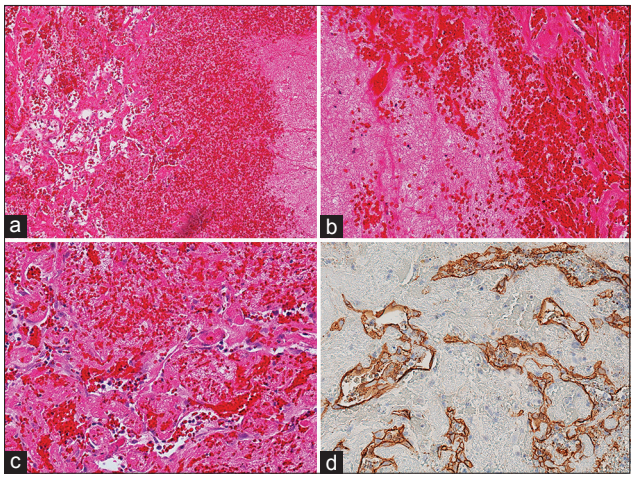

Figure 3: (a) Photomicrographs of the chronic encapsulated expanding hematoma demonstrating angiomatous abnormal vessels, multifocal hemorrhage, and coagulation necrosis. HE, original magnification ×100. (b) Areas of coagulation necrosis and extensive hemorrhage. HE, original magnification ×200. (c) Angiomatous region showing thickening of the vessel walls with hyaline degeneration and inflammatory infiltration. HE, original magnification ×200. (d) Immunohistochemical examination demonstrating strong staining for CD34 in the microvasculature. Original magnification ×200

Chronic encapsulated expanding hematoma during pregnancy

The present case of cerebellar chronic encapsulated expanding hematoma occurred during pregnancy, 9 years after the second radiosurgery. Such occurrence of chronic encapsulated expanding hematoma during pregnancy has not been reported previously, and the etiology and appropriate management strategies remain unclear. Several studies have demonstrated rapid enlargement of intracranial meningiomas during pregnancy.[11,14,22] The rate of presentation increased in the second and third trimesters. Several mechanisms, such as increased blood volume, vascular engorgement, increase in tumor-associated vascularity, increase in intracellular fluid, and increased edema, may explain both the rapid increase in tumor size during pregnancy as well as the frequent partial regression postpartum.[11,12,14,22] Recent studies showed that pregnancy and the puerperium are associated with increased risks of hemorrhage and aggressive behavior in cavernous malformations and other vascular lesions.[3,17] In the present case, the chronic encapsulated expanding hematoma became symptomatic in the third trimester, suggesting relatively rapid progression during pregnancy because this period was only 18 months after the last follow-up examination.

Recent experimental studies have revealed that representative histological changes in smaller arterioles or the microvasculature after irradiation are likely to be caused by microvasculitis, which consists of hyaline degeneration, fibrinoid necrosis, lymphocytic infiltration, and adventitial fibrosis.[2,6,7,18,21] Histological examination of the present case revealed extensive multifocal hemorrhage with multi-stage organization from abnormal angiomatous vessels with hyaline degeneration adjacent to coagulation necrosis. These findings are compatible with those of the experimental studies. On the basis of these findings, we suggest that repeated hemorrhage from the abnormal fragile

vasculature with subsequent multi-stage organization in the lesion occurred during pregnancy due to increased blood volume and vascular engorgement, and this may have caused the rapid enlargement of the hematoma and increased perifocal edema resulting in the progressive deterioration of neurological symptoms during late pregnancy.

The optimum timing for neurosurgical intervention in pregnant patients remains to be elucidated. The indications for surgery and delivery must be determined in relation to the severity of the neurological symptoms in the mother, the aggressiveness of the lesion, and the gestational period.[12] The general recommendation is that neurosurgical intervention should be avoided in the late second and third trimester, because of the high risk of intracranial hemorrhage associated with increased maternal intravascular volume. However, cesarean delivery under general anesthesia with subsequent neurological decompression should be considered for patients with risk of cerebellar herniation.[5,20] Chronic encapsulated expanding hematoma in the cerebellum may cause severe clinical problems and is potentially life-threatening because of the proximity to the brainstem and fourth ventricle. The urgency of such condition increases the likelihood of surgical intervention during pregnancy. Most obstetricians and pediatricians would consider that the delivery should be delayed to 32 weeks of gestation to ensure fetal maturity and survival. In the present case, the patient only complained of headache and was relatively stable on admission, so that cesarean section could be performed under general anesthesia immediately after admission, because the gestational age was 32 weeks and the condition of her infant was stable. After delivery, her neurological status rapidly deteriorated due to increased perifocal edema and development of hydrocephalus, so that midline suboccipital craniotomy was performed. Her neurological deficits were immediately resolved after surgery.

The present case implicates pregnancy as a potential promoter of growth and enlargement of chronic encapsulated expanding hematoma, which may become life-threatening and require surgical intervention. Accurate diagnosis and immediate surgical management after delivery are likely to result in favorable outcome. We suggest that cesarean section followed by craniotomy is indicated for patients with chronic encapsulated expanding hematoma who are neurologically unstable with conservative therapy in late pregnancy.

CONCLUSION

The present case shows that chronic encapsulated expanding hematoma after gamma knife radiosurgery may develop and increase the risk of hemorrhage, with more aggressive behavior during late pregnancy. Craniotomy and total removal of the lesion after delivery by cesarean section under general anesthesia resulted in good outcome. However, the patient should be warned of the risk of this life-threatening complication prior to attempts at becoming pregnant. Therefore, follow-up examinations should be regularly scheduled for young women of child bearing age after gamma knife radiosurgery for AVMs, despite the confirmation of angiographic disappearance of AVM nidus, because of the difficulty in predicting rapid progression of the chronic encapsulated expanding hematoma during pregnancy.

REFERENCES

1. Bowden G, Kano H, Tonetti D, Niranjan A, Flickinger J, Lunsford LD. Stereotactic radiosurgery for arteriovenous malformations of the cerebellum. J Neurosurg 2014;120:583-90.
2. Chuang VP. Radiation-induced arteritis. Seminars in roentgenology 1994;29:64-9.
3. Dias MS, Sekhar LN. Intracranial hemorrhage from aneurysms and arteriovenous malformations during pregnancy and the puerperium. Neurosurgery 1990;27:855-65.
4. Izawa M, Hayashi M, Chernov M, Nakaya K, Ochiai T, Murata N, *et al*. Long-term complications after gamma knife surgery for arteriovenous malformations. J Neurosurg 2005;102 Suppl: 34-7.
5. Jayasekera BA, Bacon AD, Whitfield PC. Management of glioblastoma multiforme in pregnancy. J Neurosurg 2012;116:1187-94.
6. Kamiryo T, Kassell NF, Thai QA, Lopes MB, Lee KS, Steiner L. Histological changes in the normal rat brain after gamma irradiation.Acta Neurochir (Wien) 1996;138:451-9.
7. Kamiryo T, Lopes MB, Berr SS, Lee KS, Kassell NF, Steiner L. Occlusion of the anterior cerebral artery after Gamma Knife irradiation in a rat. Acta Neurochir (Wien) 1996;138:983-90.
8. Kano H, Kondziolka D, Flickinger JC, Yang HC, Flannery TJ, Awan NR, *et al*. Stereotactic radiosurgery for arteriovenous malformations, Part 3: Outcome predictors and risks after repeat radiosurgery. J Neurosurg 2012;116:21-32.
9. Karlsson B, Kihlstrom L, Lindquist C, Steiner L. Gamma knife surgery for previously irradiated arteriovenous malformations. Neurosurgery 1998;42:1-5.
10. Kurita H, Sasaki T, Kawamoto S, Taniguchi M, Kitanaka C, Nakaguchi H, *et al*. Chronic encapsulated expanding hematoma in association with gamma knife stereotactic radiosurgery for a cerebral arteriovenous malformation. Case report. J Neurosurg 1996;84:874-8.
11. Lusis EA, Scheithauer BW, Yachnis AT, Fischer BR, Chicoine MR, Paulus W, *et al*. Meningiomas in pregnancy: A clinicopathologic study of 17 cases. Neurosurgery 2012;71:951-61.
12. Lynch JC, Gouvea F, Emmerich JC, Kokinovrachos G, Pereira C, Welling L, *et al*. Management strategy for brain tumour diagnosed during pregnancy. Br J Neurosurg 2011;25:225-30.
13. Maesawa S, Flickinger JC, Kondziolka D, Lunsford LD. Repeated radiosurgery for incompletely obliterated arteriovenous malformations. J Neurosurg 2000;92:961-70.
14. Moscovici S, Fraifeld S, Cohen JE, Dotan S, Elchalal U, Shoshan Y, *et al*. Parasellar meningiomas in pregnancy: Surgical results and visual outcomes. World Neurosurg 2013 Jul 10. pii: S1878-8750 (13) 00765-1. DOI: 10.1016/j.wneu. 2013.06.019.
15. Nakamizo A, Suzuki SO, Saito N, Shono T, Matsumoto K, Onaka S, *et al*. Clinicopathological study on chronic encapsulated expanding hematoma associated with incompletely obliterated AVM after stereotactic radiosurgery. Acta Neurochir (Wien) 2011;153:883-93.

16. Pan HC, Sheehan J, Stroila M, Steiner M, Steiner L. Late cyst formation following gamma knife surgery of arteriovenous malformations. J Neurosurg 2005;102 Suppl: 124-7.
17. Safavi-Abbasi S, Feiz-Erfan I, Spetzler RF, Kim L, Dogan S, Porter RW, *et al*. Hemorrhage of cavernous malformations during pregnancy and in the peripartum period: Causal or coincidence? Case report and review of the literature. Neurosurg Focus 2006;21:e12.
18. Schultheiss TE, Kun LE, Ang KK, Stephens LC. Radiation response of the central nervous system. Int J Radiat Oncol Biol Phys 1995;31:1093-112.
19. Shuto T, Ohtake M, Matsunaga S. Proposed mechanism for cyst formation and enlargement following Gamma Knife Surgery for arteriovenous malformations. J Neurosurg 2012;117 Suppl: 135-43.
20. Tewari KS, Cappuccini F, Asrat T, Flamm BL, Carpenter SE, Disaia PJ, *et al*. Obstetric emergencies precipitated by malignant brain tumors. Am J Obstet Gynecol 2000;182:1215-21.
21. Watanabe T, Saito N, Hirato J, Shimaguchi H, Fujimaki H, Sasaki T. Facial neuropathy due to axonal degeneration and microvasculitis following gamma knife surgery for vestibular schwannoma: A histological analysis. Case report. J Neurosurg 2003;99:916-20.
22. Weyand RD, MacCarty CS, Wilson RB. The effect of pregnancy on intracranial meningiomas occurring about the optic chiasm. Surg Clin North Am 1951;31:1225-33.
23. Yen CP, Jain S, Haq IU, Jagannathan J, Schlesinger D, Sheehan J, *et al*. Repeat gamma knife surgery for incompletely obliterated cerebral arteriovenous. malformations. Neurosurgery 2010;67(1):55-64.