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Case Report

Development of *De Novo* Cavernous Hemangioma after Radiosurgery for Cavernous Hemangioma

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We report a rare case of cavernous hemangioma (CH) which developed in adjacent location to a preexisting CH after gamma knife radiosurgery (GKRS). A 36-year-old woman underwent GKRS for a CH in the left lentiform nucleus. Three-and-half years after radiosurgery, MRI revealed a new CH in the left caudate nucleus. Surgical excision of the new lesion was performed. The pathological examination confirmed the diagnosis of CH. In radiosurgery for CH, it should be noted that a new CH may develop, which is likely to result from the interaction between radiation and predisposing factors of the patient.

KEY WORDS : Cavernous hemangioma · Cavernous angioma · Gamma knife · Radiosurgery · Radiation-induced tumor.

INTRODUCTION

Radiosurgery is one of the therapeutic options for cavernous hemangioma (CH). It was reported that radiosurgery confers a reduction in the risk of hemorrhage for high-risk CH[1]. Meanwhile, it is known that CH may be induced by radiation therapy or radiosurgery[3,7,12]. We report a case of CH which developed in adjacent location to a preexisting CH after gamma knife radiosurgery (GKRS).

CASE REPORT

A 36-year-old woman visited our hospital complaining of mild dysarthria and swaying tendency to the right side. A CH in the left lentiform nucleus and corona radiata was diagnosed with the typical appearance of repeated bleeding and associated venous anomaly (Fig. 1A, B). She under-went GKRS for the target volume of 1.1 cm^3 with a margi-nal dose of 13 Gy (50% of maximal dose). Neurological state improved gradually and remarkable shrinkage of the lesion was demonstrated on the follow-up MRI taken 1 year after treatment (Fig. 1C). Three-and-half year after GKRS she returned to the emergency room complaining of headache, nausea, and vomiting. While preexisting one remained stable with further decrease in size on the second follow-up MRI (Fig. 1D), a newly-developed lesion was identified in the left caudate nucleus (Fig. 1E). The new lesion also showed findings of CH with repeated hemorrhages. She underwent craniotomy immediately. The new lesion was completely excised via transcallosal transventricular approach (Fig. 1F). Pathologic examination revealed hemorrhagic tissue with collageneous stroma that contained a large number of hemosiderin-laden macrophages and inflammatory cells and cavernous thin walled vascular channels (Fig. 1G). The vascular channels were lined by a single layer of plump endothelial cells (Fig. 1H). This histopathologic finding was consistent with CH. Postoperatively, she recovered without neurological deficits.

DISCUSSION

CH, also known as cavernous malformation, is a relatively common vascular lesion, however, its etiology and natural course are not clear yet[6]. Traditionally, it has been considered a malformative lesion originating from embryonic development. But, de novo formation of CH has been well described after radiation therapy[2,4,5,8-10]. As for radiosurgery, 4 cases have been reported and the primary pathologies treated with radiosurgery included CH, metastatic brain tumor, vestibular

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schwannoma, and arteriovenous malformation[3,7,11,12]. Our case is the second case in which a new CH developed after radiosurgery for CH. Histopathology of this lesion is almost similar to that of conventional CH. However, excessive numbers of eosinophils within the lesion and plump endothelial lining cells of the cavernous vascular channels are uncommon findings in conventional CH.

Recently, three genes (CCM1, CCM2, and CCM3) associated with pathogenesis of CH have been identified in animal models and it was suggested that, in some cases, CH formation involves a genetic 2-hit mechanism and environmental second hits can produce lesions when there is a mutation to a single allele of the gene[6]. If preexisting CH reflects genetic predisposition of the patient and radiosurgery acts as an environmental second hit, radiosurgery for CH may control the original lesion and ironically induce an identical new lesion as in our case. It needs to be assessed by long-term longitudinal follow-up of a large number of patients to confirm whether the risk of a new lesion formation is higher in radiosurgery for CH than other pathologies.

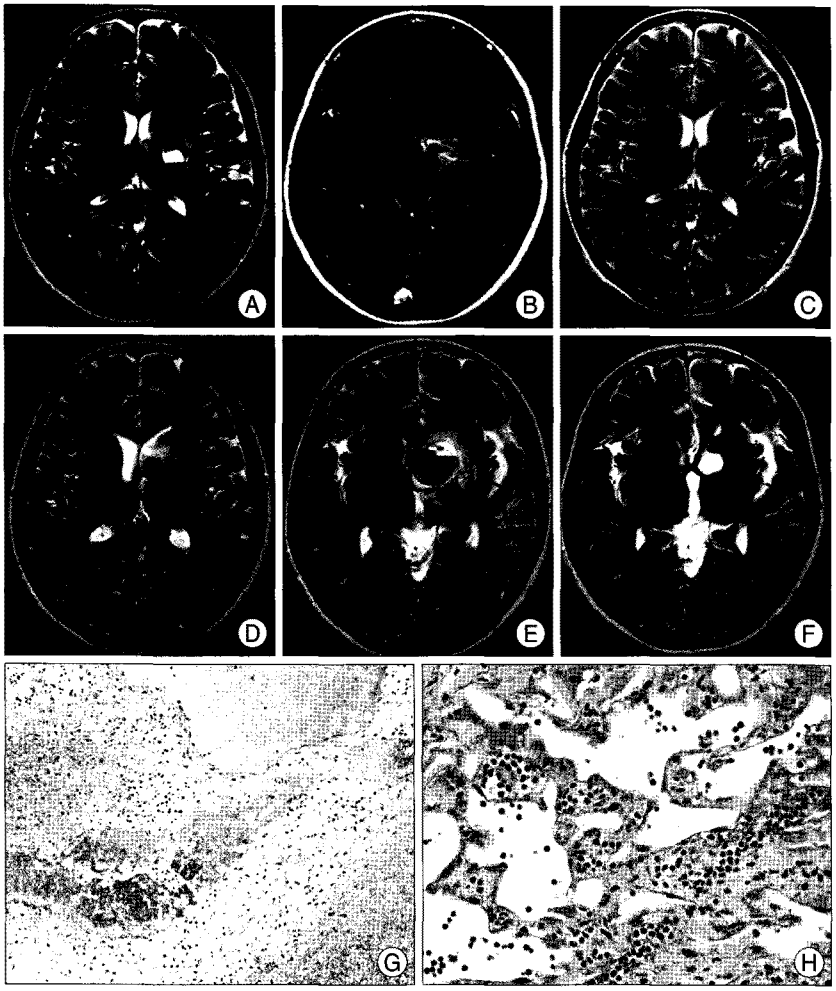

Fig. 1. Radiosurgery-induced cavernous hemangioma. A : The MRI shows a cavernous hemangioma (CH) with repeated bleeding in the left lentiform nucleus and corona radiata. B : An associated venous anomaly is observed beneath the CH. C : The MRI obtained 1 year after gamma knife radiosurgery demonstrates remarkable shrinkage of the CH. D : On the next MRI taken 3.5 years after radiosurgery, the preexisting CH remains stable with further decrease in size. However, newly-developed edema is shown in the caudate nucleus. E : Below the edema, a new CH is seen near the venous anomaly. F : Postoperative MRI reveals complete excision of the new CH. G : Photomicrograph of the new CH shows blood filled, cavernous vascular channels with hemorrhage (HE, × 100). H : The vascular channels are lined by a single layer of plump endothelial cells and intervening fibrous stroma contains excessive numbers of eosinophil (HE, × 200).

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