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CASE REPORT

Cyst Associated with Meningioma Treated Using Gamma Knife Radiosurgery

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Gamma-knife radiosurgery (GKS) has become an important treatment modality for meningioma. Recently, however, it has become known that GKS may occasionally be associated with cyst formation following treatment. The present report describes 2 cases of cyst formation associated with meningioma after treatment. From our experience with these 2 cases, we suggest raising awareness of the possible development of cyst after GKS and remind clinicians that a sophisticated decision-making process should be implemented when planning GKS for meningioma.

KEY WORDS: Radiosurgery · Meningioma · Complication · Cyst · Adverse effect.

INTRODUCTION

Meningioma is a common type of benign intracranial tumor ; however, it is not always easily removed using surgical methods.[1)2)] Gamma knife radiosurgery (GKS) has become an important treatment modality for meningioma, especially in cases in which surgical treatment is not easy such as skull base lesions.[3)4)] The increased use of GKS for such benign lesions has led to the recognition of late radiosurgical complications, which has attracted attention. Delayed cyst formation represents one such complication.[5)6)] Cyst formation following treatment for vestibular schwannoma and arteriovenous malformation has also been reported.[6)] Recently, several studies have reported that GKS may occasionally be associated with cyst formation following treatment for meningioma.[7-9)] According to these reports, the incidence of delayed cyst formation after GKS is estimated to be 0.7−1.7% in patients with meningioma, which appears to be lower than that in individuals with arteriovenous malformations. Nevertheless, reports describing this morbidity are scarce ; consequently, the true incidence remains unclear, and the pathogenesis of this morbidity is yet to be determined. The present report describes the clinical courses of 2 patients with meningioma who developed delayed cysts after GKS.

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CASE REPORT

Case 1

A 62-year-old man presented to our hospital with dizziness and underwent brain magnetic resonance image (MRI), which revealed a meningioma located in the right parasagittal area. The lesion extended beyond the falx to the opposite side, and also exhibited peritumoral edema (Fig. 1A and B). The patient underwent GKS (Perfexion, Elekta) using a marginal dose of 13Gy with 50% isodose line for the lesion (15cm^3) on January 2009. GKS was completed without any problems. At one year after GKS, follow-up MRI revealed central necrosis within the lesion and worsening peritumoral edema (Fig. 1C and D) ; however, the patient exhibited no clinical symptoms. MRI performed 2 years after GKS was similar to that of the first year, and the size of the tumor did not appear to be increasing. Four years after GKS, MRI revealed that the size of the tumor was decreased ; however, a new cyst developed in the anterior aspect of the tumor (Fig. 1E and F). The patient, however, exhibited no specific symptoms. Six years after GKS, the patient complained of mild, lower left extremity weakness and MRI, which was performed at that time, revealed enlargement of the cyst and worsening peritumoral edema (Fig. 1G and H). The patient underwent conservative management including mannitol and steroid, which resolved the symptoms.

Case 2

A 67-year-old woman presented to our hospital with meningioma located in the right convexity (Fig. 2A and B).

She had a surgical history for right trigone meningioma 10 years previously. The tumor subtype was pathologically confirmed to be meningothelial meningioma. The patient underwent GKS (Model 4C, Elekta) using marginal dose (13Gy) with 50% isodose line for newly lesion ($1cm^3$) at the right convexity on March 2007. GKS was completed successfully. One year after GKS, MRI revealed peritumoral edema, which was not apparent before GKS (Fig. 2C and D). The patient complained of headache and nausea ; follow-up MRI performed 2 years postoperatively revealed persistent edema and a small peritumoral cyst (Fig. 2E and F). The patient underwent conservative management. MRI performed 3 years after surgery revealed that the previous peritumoral cyst was larger and a new small lesion was observed around

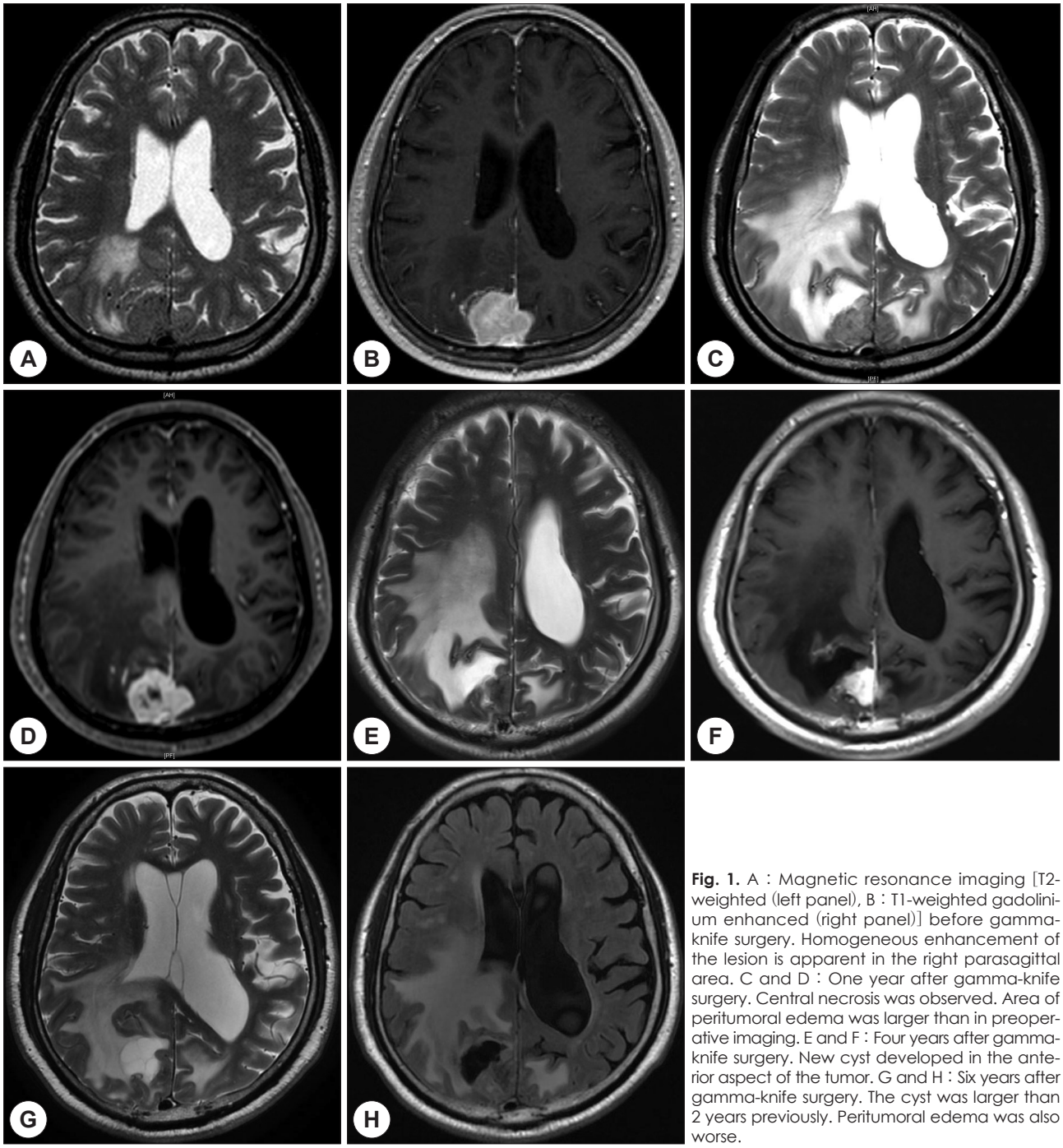

Fig. 1. A : Magnetic resonance imaging [T2-weighted (left panel), B : T1-weighted gadolinium enhanced (right panel)] before gamma-knife surgery. Homogeneous enhancement of the lesion is apparent in the right parasagittal area. C and D : One year after gamma-knife surgery. Central necrosis was observed. Area of peritumoral edema was larger than in preoperative imaging. E and F : Four years after gamma-knife surgery. New cyst developed in the anterior aspect of the tumor. G and H : Six years after gamma-knife surgery. The cyst was larger than 2 years previously. Peritumoral edema was also worse.

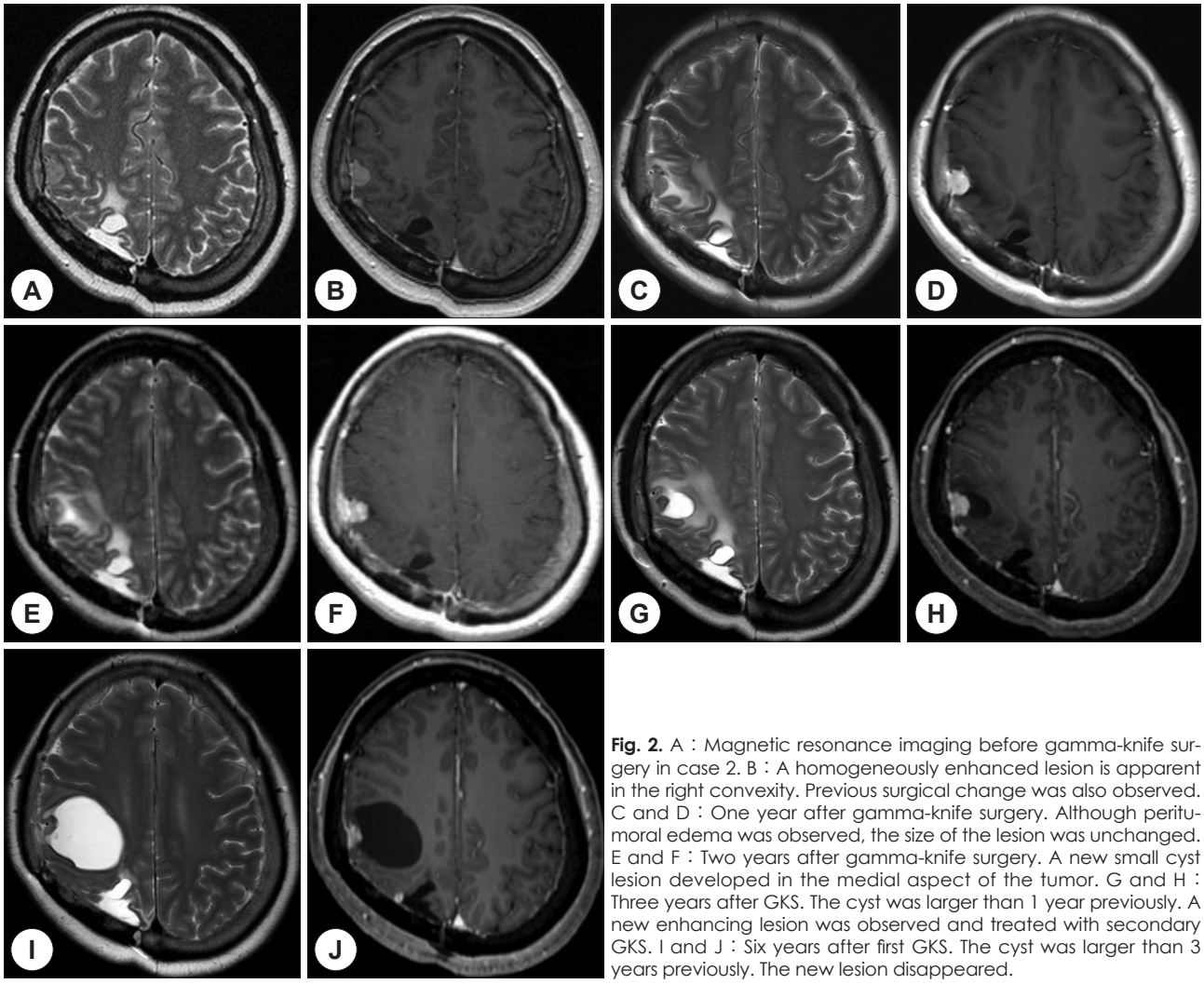

Fig. 2. A : Magnetic resonance imaging before gamma-knife surgery in case 2. B : A homogeneously enhanced lesion is apparent in the right convexity. Previous surgical change was also observed. C and D : One year after gamma-knife surgery. Although peritumoral edema was observed, the size of the lesion was unchanged. E and F : Two years after gamma-knife surgery. A new small cyst lesion developed in the medial aspect of the tumor. G and H : Three years after GKS. The cyst was larger than 1 year previously. A new enhancing lesion was observed and treated with secondary GKS. I and J : Six years after first GKS. The cyst was larger than 3 years previously. The new lesion disappeared.

the lesion (Fig. 2G and H). GKS was performed for the new lesion. Six years after the first GKS, follow-up MRI revealed that the size of the cyst increased further (Fig. 2I and J). The patient did not complain of any specific symptoms ; nevertheless, she remains under close observation.

DISCUSSION

The mechanism of cyst formation is not fully understood. Enlargement of a previous hematoma cavity and radiation-induced necrosis accompanied by breakdown of the blood-brain barrier have been proposed to explain cyst formation in cases involving arteriovenous malformation.[10] As for meningioma, Shuto, et al.[8] reported cyst formation or enlargement following GKS in five of 160 patients. Their histological and neuroradiological investigations identified various risk factors, including radiation-induced ischemic necrosis, increased vascular permeability, and intratumoral hemorrhage, and postulated that cysts may develop in the presence of such factors. In addition, they also reported that meningioma with intra- or peritumoral cystic components before GKS represented a high risk for marked cyst enlargement after GKS. Moreover, Igaki, et al.[5] reported 2 cases of delayed cyst formation after GKS in 3 meningioma lesions. They observed small, spotty cysts at the time of GKS in all 3 tumors that they encountered, with developing delayed cysts. In their patients experiencing delayed cyst formation as well as those in the study by Shuto, et al.[8] the incidence of having a cystic component within the pre-GKS tumor was exceptionally high. The majority of meningiomas, however, do not have intratumoral or peritumoral cysts. They suggested that there may be a risk for delayed cyst formation, and this should be followed-up carefully after stereotactic radiosurgery, especially in patients with a cystic component

within the meningioma, even if it is small and spotty. Regardless, the relationship between cyst development and maximal treatment dose in arteriovenous malformation was identified. Izawa, et al.[11] suggested that a high prescription dose induced alterations in brain tissue, which promoted delated cyst formation in arteriovenous malformation. In our study, the patients exhibited no cystic lesion before GKS. The treatment dose at the tumor margin in our cases (13Gy) was selected to ensure favorable tumor control ; however, a higher dose could potentially result in stronger radiation-induced histopathological changes. Considering the results from studies investigating arteriovenous malformation, a higher dose may have promoted delayed cyst formation in our patients.

There is no definite answer to the need for surgical or interventional treatment of postoperative cysts. Ferrante, et al.[12] reported histological results after surgical treatment in 9 patients with cystic meningioma. They recommended treatment of the cyst if it is enlarged within the tumor because the cystic wall of intratumoral cysts contain neoplastic cells. However, this differs in the case of peritumoral cysts. Zhao, et al.[13] reported a case involving a 17-year-old boy with an unusual large cystic meningioma (Nauta type II) in the right hemisphere. At surgery, it was found that the cyst contained a large amount of xanthochromic fluid and some semitransparent serumlike sediment. The intracystic nodule was confirmed to be a necrotic entity without tissue or cellular structure. Histological examination of the wall of peritumoral cysts is likely to be free from tumor cells. In addition, Weber, et al.[14] reported intraoperative findings and management of peritumoral cyst walls and cyst fluid in cystic meningiomas. They found that peritumoral cyst formation had meningioma cells in the cyst wall, and that cytological examination of the cystic fluid exhibited the presence of meningiothelial cells. They suggested that cystic meningiomas have the potential to spread through cystic fluid to the cyst wall in peritumoral cyst configurations. Moreover, Zhi, et al.[15] reported that in chordoid or papillary meningiomas, which assume aggressive metastatic properties, the cystic wall has frequently been shown to be composed of tumor cells, and recommended appropriate treatment that includes the cyst wall. In contrast, the cyst in our cases developed after GKS ; thus, it may be different from those described in previous reports. In addition, in a study by Shuto, et al.[8] the histologic examination of the cysts which was developed after GKS did not reveal tumor cells. Thus, it is difficult to ascertain a clear answer for treatment ; nevertheless, it is important to treat patient symptoms. The treatment may be surgical removal. Our study demonstrated improvement through conservative treatment.

CONCLUSION

GKS has recently been shown to be a safe and effective treatment option for intracranial meningioma. The increased use of GKS for such benign lesions has led to the recognition of late radio-surgical complications, which has attracted attention, with cyst formation representing one such complication. The development of new cyst or enlargement of preexisting cyst after GKS, present new clinical challenges. Thus, a more sophisticated decision-making process for treatment of cystic meningiomas may be required.

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