Angiomatous lesion and delayed cyst formation after gamma knife surgery for intracranial meningioma: case report and review of literatures

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Abstract: Gamma Knife has become a major therapeutic method for intracranial meningiomas, vascular malformations and schwannomas with exact effect. In recent years an increasing number of delayed complications after Gamma Knife surgery have been reported, such as secondary tumors, cystic changes or cyst formation. But angiomatous lesion and delayed cyst formation after Gamma Knife for intracranial lesion has rarely been reported. Here we report the first case of angiomatous lesion and delayed cyst formation following Gamma Knife for intracranial meningioma and discuss its pathogenesis.

Keywords: Gamma Knife, angiomatous lesion, delayed cyst, intracranial meningioma

Introduction

As the development of stereotactic radiosurgery, GKS has become a major therapeutic method for intracranial lesions with exact effect [1-4]. Although the morbidity and mortality of GKS for intracranial lesions is low, more and more delayed cyst formation or neoplasms (such as glioma and meningioma) associated with GKS have been reported in recent years [5-8]. However, delayed cyst formation after GKS for intracranial meningioma is rarely reported. Especially, angiomatous lesion and delayed cyst formation after GKS for intracranial meningioma has not been reported. The present case is the first documented case of angiomatous lesion and delayed cyst formation following GKS for intracranial meningioma.

Case report

History

A 68-year-old woman was admitted for headache and dizziness for 3 months. 12 years before admission, a high density lesion was occasionally found in the vicinity of her right cerebral falx and superior sagittal sinus with computerized tomography (CT) (Figure 1A), which was considered as meningioma through magnetic resonance imaging (MRI). At the same time, there were no other intracranial lesions. Worried about the complications of craniotomy, she underwent GKS with marginal dose of 12Gy and 50% isodose line. After radiotherapy, she did not follow-up.

Examination

We found no positive signs and performed MRI examination for her after admission. MRI showed that the original intracranial lesion did not enlarge with significant enhancement (Figure 1B-D). Furthermore there was a cyst about 5 cm×4 cm×4 cm without enhancement of the cystic wall and an irregular enhanced nodular lesion adjacent to the original intracranial lesion (Figure 1B-D).

Treatment

Craniotomy was performed for her to remove the original lesion, the cyst lesion and the nodular lesion. In operation, we found the original lesion had a smooth surface and based on cerebral falx and superior sagittal sinus. The
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Cyst contained transparent xanthochromic liquid without clear boundary between cystic wall and surrounding brain tissues. There was a 0.5 cm×1.0 cm×1.5 cm irregular reddish nodule with soft texture adhesion in the original lesion (Figure 1E). These lesions were completely removed. Laboratory examination indicated that the protein concentration in cystic fluid was 3.5 g/dl. Pathological examination showed the original lesion was meningioma of WHO class I with obvious collagenous fiber formation and calcification (Figure 1F). Furthermore, the reddish nodule was verified to be angiomatous lesion (Figure 1G). After operation, her symptoms were greatly improved and postoperative CT scanning was negative (Figure 1H).

Discussion

Meningiomas are the most common disease of intracranial lesions, for which the priority treatment is complete resection. However, meningiomas are often located in the skull base, parasagittal and functional areas with closely adhesion to important blood vessels and nerves, as well as close ties with the superior sagittal sinus. As a result, complete resection is very difficult for high surgical risks and severe complications. With the development of stereotactic radiosurgery, GKS is commonly used to treat some small intracranial meningioma or postoperative residual meningioma, which has higher tumor control rate with less complication and lower mortality [2, 3, 9]. Kondziolka reported 290 cases of meningioma treated by GKS, including 132 cases which used GKS as the first treatment, and the complete control rate for tumor was 91% [9]. Pollock reported 330 cases of intracranial meningiomas after GKS, including 192 cases which chose GKS as priority, and the tumor control rate was 94% [2].

Although the effect of GKS for intracranial lesions is good, an increasing number of delayed complications have been reported in recent years, such as secondary tumors, cystic changes or cyst formation [5, 6, 8, 10-12]. Nevertheless, late cyst formation after GKS for intracranial meningioma is rarely reported in literatures. Shuto et al reported five cases of

Figure 1. A: CT from 12 years before admission showing a high density lesion about 2 cm×1.5 cm in the vicinity of her right cerebral falx and superior sagittal sinus. B: T2-weighted axial MR image revealing that the primary lesion was hypointense without size change (white arrow) and there was a hypointensive nodule adjacent to it (black arrow) and a 5×4×4 cm cyst in her right occipital lobe with its fluid signal like cerebrospinal fluid. C: Contrast-enhanced T1-weighted axial MR image. D: Contrast-enhanced T1-weighted sagittal MR image. C and D: Demonstrating heterogeneous enhancement for primary lesion (white arrow), significant enhancement for the nodule (black arrow) and no enhancement for the cystic wall. E: Primary lesion and the irregular reddish nodule after operation. F: Photomicrograph of primary lesion indicating meningioma cells with obvious collagenous fiber formation and calcification. H&E, original magnification ×200. G: Photomicrograph of the reddish nodule rerealing the blood vessels with different diameter and arranged disorder. H&E, original magnification ×100. H: Postoprative CT showing no intracranial hematoma.
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delayed cyst formation after GKS for meningioma, and divided these cysts into three types [13]. The first type refers to the newly developed cysts located within tumors after GKS, which didn’t exist before GKS. The second type refers to the intratumoral cyst that existed before GKS, which became larger after GKS. The third type refers to peritumoral cyst that existed prior to GKS and enlarged following radiosurgery. In these 5 cases, there were 3 cases of first type, 1 case of second type and 1 case of third type. Igaki et al reported two cases of delayed cyst formation after GKS for meningioma, which were the second type [14]. Our present case is different from which Shuto and Igaki reported, which is a newly occurred peritumoral cyst following GKS for meningioma and whose pathogenesis is different from them. In terms of formation mechanism, these cysts which Shuto and Igaki reported came from the original cysts enlargement or cystic change of meningioma following GKS. Therefore, we believe that delayed cyst formation after GKS should be divided into four types. The first three types are as Shuto said. The fourth type, as the case we report, is new peritumoral cyst after GKS which didn’t exist before GKS.

At present, the pathogenesis for cyst formation after GKS for intracranial lesions is not documented. As for the mechanism for cyst formation after GKS for intracranial arteriovenous malformation, there are two main theories. One theory is that cyst formation may be associated with delayed hemorrhage after GKS [12]. Shuto suggested GKS could induce inflammatory in the parenchyma around the irradiated lesions, which could cause the formation of capillary vessels. These newly capillary vessels were so fragile that they induced repeated hemorrhage to delayed cyst formation [12]. The other theory is that cyst formation after GKS may have ties to blood-brain barrier destruction [10, 11]. Pollok thought GKS could increase permeability of the blood-brain barrier around the irradiated lesions, which result in the biologically active substances and fluid to extravasate from these vessels. Then the fluid accumulated to induce cyst formation [11]. In the case we report, cyst formation after GKS might have close link to angiomatous lesion.

Adult angiomatous lesion after GKS for intracranial lesions has been reported in recent years, but most of them originated from GKS for intracranial arteriovenous malformations [7, 15, 16]. Angiomatous lesion formation after GKS for intracranial meningioma hasn’t yet been reported. The current case is the first case of angiomatous lesion occurring after GKS for intracranial meningioma. For angiomatous lesion following GKS, the pathogenesis is unknown. It often occurs in patients with low-dose radiation therapy and is believed that it may be associated with neovascularization caused by local hypoxia resulting from necrosis of the vessel wall and vascular lumen narrowing following GKS [7, 15]. For the case we report, the lesion was located in the posterior part of the superior sagittal sinus, which rich cerebral superficial veins flow into. So we speculate that when GKS treated the meningioma, its adjacent drainage venous were under radiation exposure, thereby causing vessel wall necrosis and vessel occlusion. These led to local blood flow disorders and endovascular pressure increase, which thus resulted in neovascularization and finally angiomatous lesion formation. Meanwhile, because of high permeability and easily bleeding for new blood vessels, the blood oozed to accumulate and form cyst.

Conclusions

GKS is an effective method for intracranial meningiomas. In consideration of angiomatous lesion and delayed cyst formation after GKS, long-term follow-up is necessary for the patients with GKS. Meanwhile GKS should be prudent for patients with intracranial meningiomas in the vicinity superior sagittal sinus.

Disclosure of conflict of interest

None.

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