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Case Report

Intracranial Aneurysm Following Cranial Radiation Therapy

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We report herein a case of a radiation-induced aneurysm. A 69-year-old woman presented with subarachnoid hemorrhage. Eight years previously, she had undergone cranial radiation therapy (total dose of 59.4 Gy) as adjuvant therapy after surgical resection for a chondrosarcoma that was destroying her sphenoid sinus. The patient underwent catheter angiography, which revealed an aneurysm of the anterior communicating artery and luminal narrowing and irregularity in the petrous and lacerum segments of the right internal carotid artery. We attempted surgical clipping of the aneurysm, but there was repeated bleeding. Finally the aneurysm was treated with endovascular trapping. Potentially fatal bleeding also occurred from her internal carotid artery, which had also been irradiated during the previous cranial radiation therapy. We stopped the bleeding with endovascular coil embolization. Because of diffuse vascular changes of the cerebral vessels within irradiated fields, special attention must be paid to their treatment.

Keywords Intracranial aneurysm, Subarachnoid hemorrhage, Radiotherapy

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INTRODUCTION

To date, more than 50 million cancer survivors have been treated with radiotherapy worldwide.[1] It is well documented that radiotherapy can induce a type of vascular disease, termed vasculopathy.[2][3] In addition, it has become increasingly evident that cranial radiation therapy increases the risk of stroke for cancer survivors.[4][5] Although radiation-induced occlusive vasculopathies are well documented, little is known about the characteristics of radiation-induced intracranial aneurysms and their optimal treatment strategy. These events are believed to be a rare complication, and to our knowledge, only 26 cases have been reported in the literature.[6] Herein, we report the case of a woman in whom an anterior communicating artery aneurysm developed eight years after she underwent cranial radiation therapy for a chondrosarcoma, located in the sphenoid sinus.

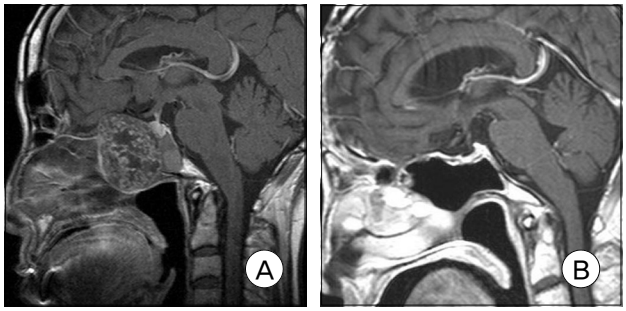

Fig. 1. A 69-year-old woman was found to have a large mass destroying her sphenoid sinus when she lost vision in her right eye eight years prior to current presentation (A). After treatment she was clinically monitored throughout the next eight years with serial Magnetic Resonance Imaging (MRI) and none of the MRI shows any sign of residual or recurrent tumor (B).

Fig. 2. Balloon occlusion test, which was performed before tumor removal, shows a good cross-filling via the anterior communicating and no vascular lesion.

CASE REPORT

History

This 69-year-old woman was found to have a large mass destroying her sphenoid sinus when she lost vision in her right eye eight years prior to current presentation (Fig. 1A). During the surgical removal of tumor, there appeared to be a chance of injuring the internal carotid artery (ICA). We therefore performed preoperative catheter angiography and balloon test occlusion. The anterior communicating artery was widely patent on the balloon test occlusion, and no vascular abnormalities, including aneurysm, were identified by the catheter angiography (Fig. 2). She underwent gross-total resection of the tumor at that time without any adverse event, and a pathological examination confirmed the diagnosis of chondrosarcoma. She subsequently underwent cranial radiation therapy (total dose of 59.4 Gy) as adjuvant treatment. She was clinically monitored throughout the next eight years with serial magnetic resonance imaging (MRI). During this time, she demonstrated no new neurological deficit except slight cognitive decline, and none of the MRIs showed any sign of residual or recurrent tumor (Fig. 1B).

Examination

The patient presented to the emergency room due to severe headache and a slightly drowsy mentality, and computed tomography (CT) revealed intracerebral hemorrhage and subarachnoid hemorrhage (Fig. 3A).

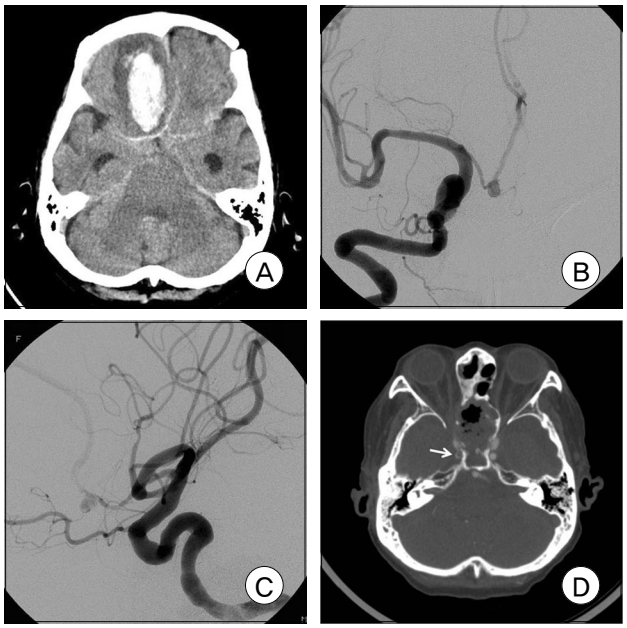

Fig. 3. Brain computed tomography (CT) shows intracerebral and subarachnoid hemorrhage (A). Angiography reveals an aneurysm of the anterior communicating artery (B and C). A source image of CT angiography shows luminal narrowing and irregularity in the petrous and lacerum segments of the right internal carotid artery (D).

The patient underwent catheter angiography, which revealed an aneurysm of the anterior communicating artery and luminal narrowing and irregularity in the petrous and lacerum segments of the right ICA (Fig. 3B, 3C and 3D).

Operation

We suspected that the anterior communicating artery aneurysm had ruptured and attempted endovascular treatment with coil embolization. However, the tortuosity of the bilateral A1 segments of the anterior cerebral arteries (ACAs) and the broad neck of the aneurysm made the endovascular treatment difficult. We stopped the endovascular treatment and surgically clipped the aneurysm. During the microsurgery, the wall of the aneurysm was very rubbery, and the surgical clip slipped from the neck of the aneurysm. After several surgical clipping attempts, the neck of the aneurysm was clipped, and a booster clip was used to reinforce the primary clip. The patient's immediate postoperative neurological state was the

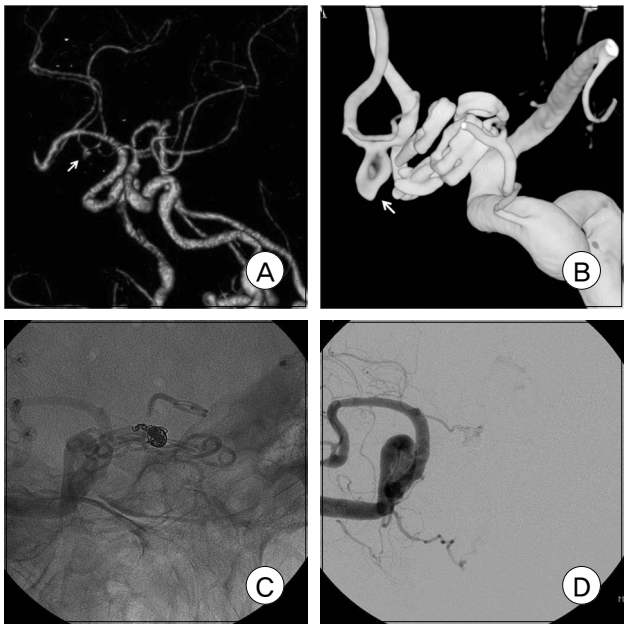

Fig. 4. CT angiography performed ten days after surgery reveals the small, residual neck of the aneurysm (A). Angiography which was performed ten days after clipping shows slippage of clips from the aneurysm (B). Postembolization angiograms (C and D) reveals successful obliteration of the aneurysm.

same as baseline. CT angiography performed ten days after surgery revealed the small, residual neck of the aneurysm, but the patient's state was stationary (Fig. 4A). At 14 days after surgery, the patient's mental state worsened, and a CT revealed rebleeding from the aneurysm; catheter angiography demonstrated that the clips had slipped from the aneurysm (Fig. 4B). We performed emergency surgery using a contra-lateral craniotomy and confirmed that the clips had slipped from the aneurysm; new clips were applied around the aneurysm neck. The patient's neurological state immediately after the 2nd operation was deeply drowsy. However, three days after the 2nd operation, the patient became semicomatose, and CT and catheter angiography revealed rebleeding and the slip of the clips, respectively. After the repeated failure of surgical clipping, we performed endovascular trapping from the right distal A1 to the aneurysm (Fig. 4C). After the procedure, the final catheter angiogram showed complete obliteration of the aneurysm and intact flow of the bilateral A2s (Fig. 4D).

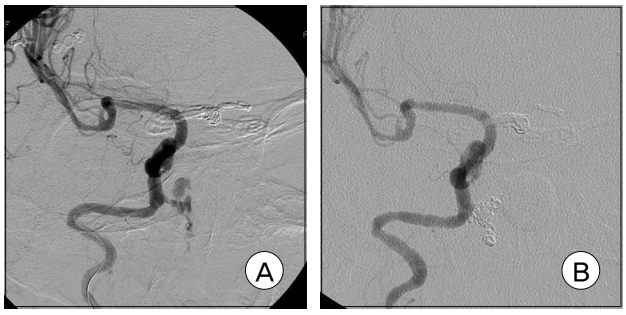

Fig. 5. Angiography shows contrast leakage from the lacerum segment of the right internal carotid artery into the nasal cavity, suggesting carotid blow out syndrome (A). After coil embolization in the ruptured point, no more leakage is noted on angiography (B).

Postoperative Course

After the endovascular treatment, her neurological state was semi-comatose, and we continued conservative management. One month after the endovascular treatment, there was massive bleeding from her nasal cavity, and catheter angiography showed a rupture of the lacerum segment of the right ICA, suggesting carotid blow out syndrome (Fig. 5A). We performed endovascular coil embolization of the ruptured site in the lacerum segment of the right ICA as a life-saving procedure (Fig. 5B). The patient did not recover from the semi-comatose state and was discharged home in a bed-ridden state.

DISCUSSION

Recent studies have proposed that long-term survivors of brain tumor are at risk for late-occurring cerebrovascular accidents and strokes.[5)7)] These studies have suggested that this risk is related to cranial radiation therapy. Radiation-induced aneurysm is rare compared to radiation-induced occlusive vasculopathy. However, a subsequent subarachnoid hemorrhage can be life threatening and often results in severe disability or death.

Our case report described a case of radiation-induced aneurysm. The aneurysm may have occurred by iatrogenic trauma to the artery during the previous surgery. Iatrogenic injury leading to aneurysm for-

mation has been described previously.[8)]

But several features of her aneurysm suggest that it might have been induced by radiation therapy. (1) Its location was similar to that of the tumor and was within the irradiated field. (2) The wall of the aneurysm was very rubbery and differed from that of a typical saccular aneurysm. (3) Catheter angiography performed prior to the cranial radiation therapy demonstrated the absence of an aneurysm. (4) Additionally, there were potentially fatal bleeds from dual lesions within the irradiated field: aneurysm rupture and ICA rupture. These events imply that there were diffuse vascular changes in the cerebral vessels within the irradiated field. Several features of the ICA rupture also indicated that its occurrence may be related to radiotherapy. Its location was also within the irradiated field. The angiographic findings of luminal narrowing and irregularity in the petrous and lacerum segments of the right ICA noted on the catheter angiography when she presented to the emergency room were absent on the catheter angiography performed prior to the cranial radiation therapy. Even though this finding may be atherosclerotic changes due to aging, to our knowledge, there are few reports regarding the spontaneous rupture of this ICA segment resulting only from atherosclerotic change. However, several reports have described suspected common carotid artery vasculopathy and rupture after radiation therapy.[9)10)]

In addition, in this case, it was very difficult to treat the radiation-induced aneurysm with surgical clipping or endovascular coil embolization. Previous reports have proposed that special attention must be paid in the treatment of radiation-induced aneurysms because these aneurysms differ from typical saccular aneurysms by virtue of their shape and location.[11)]

CONCLUSION

In summary, with the increase in the number of survivors of brain tumors treated with cranial radiation therapy, a large number of patients presenting with fatal hemorrhage from radiation induced-aneurysms may also be observed. Because of the diffuse vascular changes in the cerebral vessels within the irradiated field, special attention must be paid to their treatment.

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DISCLOSURE STATEMENT

The authors have no personal financial or institutional interest in any of the materials or devices described in this article.

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