Case Reports

Four Cases of Meningeal Hemangiopericytoma Treated with Surgery and Radiotherapy

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We report our experiences of four cases with meningeal hemangiopericytoma treated with surgery and postoperative radiotherapy and survey the literature to elucidate the efficacy of radiotherapy. Patients were treated with surgical resection and 46–52 Gy postoperative radiotherapy. Three patients had local control for 30, 54 and 138 months, respectively and one patient had local recurrence after 49 months. Distant metastases were observed in two patients; one had multiple bone, liver and lung metastases and the other multiple bone and brain metastases. For bone and brain metastases, better tumor control was obtained with palliative radiotherapy and stereotactic radiotherapy. Literature analyses demonstrated that surgery and postoperative radiotherapy of 50 Gy or more resulted in significantly better local control than surgery alone (p = 0.02). Stereotactic radiosurgery was effective for intracranial recurrence or metastasis, especially when the tumor volume was <8 cm³ and >15 Gy at the 50% isodose line was used. Radiotherapy for bone metastases was also effective for palliation.

Key words: meningeal hemangiopericytoma – postoperative radiotherapy – stereotactic radiosurgery

INTRODUCTION

Hemangiopericytoma is a rare tumor that was first described by Schmidt in 1937 and named by Stout and co-workers in 1942 (1,2). This tumor arises from pericapillary cells or pericytes of Zimmerman and can occur anywhere where capillaries are found (1–5). However, hemangiopericytoma develops mainly in the lower extremities, retroperitoneum or pelvis and rarely occurs in the larynx, spleen, meninges or thorax (6–8).

The peak incidence of hemangiopericytoma is in the fourth and fifth decades of life. The natural history of this tumor can be very long. However, it is sometimes a locally aggressive, potentially malignant tumor that produces late local recurrences and distant extraneural metastases, mainly to bone and the lung and liver (9–12). Its incidence of metastasis ranges from 12% (1,2) to 57% (13) and Guthrie et al. demonstrated increasing metastatic frequency, reporting 5-, 10- and 15-year metastasis rates of 13, 33 and 64%, respectively (14).

Meningeal hemangiopericytoma is rare and its incidence is <1% of all central nervous system tumors and 2.4% of meningiomas. Fewer than 100 cases have been reported. There are only a few papers reporting the results of postoperative radiotherapy for unresectable or incompletely excised meningeal hemangiopericytoma (10,12,15,16). Thus, the optimal dose and fractionation for meningeal hemangiopericytoma are not clear.

In this paper, we report our experiences with four cases of meningeal hemangiopericytoma treated with surgery and postoperative radiotherapy and review and analyze the literature in order to elucidate the optimal radiotherapy for meningeal hemangiopericytoma.

CASE REPORTS

CASE 1

In August 1982, a 42-year-old woman was referred to us with headache and brain computed tomography (CT) revealed a tumor 4 cm in diameter in the right parieto-occipital lobe (Fig. 1a). Subtotal tumor resection was performed and histology showed meningeal hemangiopericytoma. Regrowth of the

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residual tumor was observed and total tumor resection was performed in December 1983. Thereafter, the tumor bed was irradiated with 52 Gy in 32 fractions as postoperative radiotherapy.

In June 1991, severe back pain and paresthesia in the right lower extremity appeared and X-ray film of the lumbar spine revealed osteolytic lesions due to regrowth of the tumor in the second lumbar spine. Subtotal tumor resection and internal fixation were performed. Histology proved the metastasis of hemangiopericytoma. As postoperative radiotherapy, we administered 37.5 Gy in 15 fractions to the residual tumor of the second lumbar spine. Regrowth of the tumor of the second lumbar spine occurred in September 1994 and we administered 30 Gy in 12 fractions of reirradiation of the same field.

From January 1994, bone metastases appeared repeatedly and subsequently a total of 11 courses of palliative radiotherapy for bone metastases were performed and good pain relief was achieved in all irradiated sites. The doses of radiotherapy to bone metastases ranged from 25 Gy in five fractions to 50 Gy in 16 fractions. Chest X-ray film showed multiple lung metastases in July 1995 and an abdominal CT scan showed multiple liver metastases in January 1997. No chemotherapy was administered. The patient died of lung metastases in July 1997. Local control of the primary tumor was obtained until her death (Fig. 1b).

CASE 2

In March 1987, a 37-year-old woman was referred to us with headache and paraplegia in the lower extremities. A brain CT scan showed a tumor 6 cm in diameter at the right sphenoid ridge. Subtotal tumor resection was performed and histology showed meningeal hemangiopericytoma (Fig. 2).

In June 1989, a brain CT scan showed regrowth of the tumor and a second operation was performed. In June 1992, a brain CT scan showed tumor regrowth (Fig. 3a) and a third operation for subtotal tumor resection was performed. Subsequently, 50 Gy in 25 fractions to the residual tumor was administered.

From August 1993, brain metastases appeared repeatedly. Four courses of γ-ray stereotactic radiosurgery (gamma-knife) with doses of 20–30 Gy in one fraction at the 50% isodose line and six courses of X-ray stereotactic radiosurgery with doses of 13.2–18.0 Gy in one fraction at the 80% isodose line were carried out. A partial response or stable disease was obtained until her death.

From February 1994, bone metastases appeared repeatedly. Four courses of palliative radiotherapy were performed and...
good pain relief was achieved in all irradiated sites. The doses of radiotherapy ranged 25 Gy in five fractions to 35 Gy in eight fractions.

Local control of the primary tumor was obtained until her death, but the patient died of right pleural metastases in December 1996 (Fig. 3b).

CASE 3
In May 1990, a 37-year-old man was referred to our department with pain and weakness in his left lower extremity. Magnetic resonance imaging (MRI) of the lumbar spine displayed an enhanced tumor in the anterior epidural space of the second lumbar spinal canal. Subtotal resection was performed in August 1990 and the symptoms were relieved. Histology showed meningeal hemangiopericytoma.

In September 1992, MRI of the lumbar spine showed tumor regrowth and a second operation for subtotal tumor resection was performed. We then administered 48 Gy in 24 fractions to the first to fourth lumbar spine. In October 1996, MRI of the lumbar spine revealed local recurrence and additional tumor resections were performed in October 1996, March 1997 and September 1998. At final follow-up in November 2000, the patient was alive in complete paraplegia due to residual tumors in the lumbar spine.

CASE 4
A 68-year-old woman was referred to us with a foot-drop and paresthesia in the bilateral second to fourth fingers in February 1998. MRI of the cervical spine showed an intradural tumor at the sixth cervical spine (Fig. 4a). In March 1998, total tumor resection and laminectomy of the fifth to seventh cervical spine were performed and histology proved meningeal hemangiopericytoma. Postoperative radiotherapy consisting of 46 Gy in 23 fractions at the level from the fifth cervical to first thoracic spine was administered and no local recurrence has occurred for 30 months after treatment (Fig. 4b).

DISCUSSION
POSTOPERATIVE RADIOTHERAPY AND LOCAL RECURRENCE
Some authors have described the advantages of postoperative radiotherapy (12,15,16). Staples et al. reported that local tumor control was achieved in all six cases who received total tumor doses of ≥55 Gy. In their analysis, surgery plus postoperative radiotherapy was superior to surgery alone for relapse-free survival (p = 0.03). However, the number of cases was small and various doses of radiation were used. To elucidate the role of radiotherapy for meningeal hemangiopericytoma, we undertook a literature search for ‘meningeal hemangiopericytoma’ covering the years from 1970 through 2000. However, most of the papers found were not so useful owing to incomplete or undetailed radiation data; data for 53 lesions from 50 patients who were treated with surgery with or without postoperative radiotherapy in 14 reports and an additional four patients from our experience were collected and analyzed (6,12,16–27).

Analyses of local-recurrence-free survival for primary tumors and metastatic tumors that were treated with curative intent were performed using the Kaplan–Meier method, comparing surgery alone and postoperative radiotherapy (Figs 5 and 6). The 10-year local-recurrence-free survival for surgery alone was 20.5% and that for surgery and postoperative radiotherapy of 40 Gy or more was 78.0% (Fig. 5). There was a significant difference between the two groups with the log-rank test (p = 0.005). According to the dose of radiotherapy, it appeared that the addition of radiotherapy at ≥50 Gy resulted in better local control; however, no significant difference was found (Fig. 6, p = 0.167), perhaps owing to the small number of patients.
In our series, three patients (cases 1, 2 and 4) achieved primary tumor control for 12, 4.5 and 2.5 years, respectively, with total or subtotal tumor resection and postoperative radiotherapy consisting of 46–52 Gy in 23–35 fractions. One patient (case 3) experienced local recurrence after 46 months with subtotal tumor resection and postoperative radiotherapy of 48 Gy in 24 fractions.

**EFFECTIVENESS AND RESPONSE TO STEREOTACTIC RADIOSURGERY FOR INTRACRANIAL RECURRENCES OR METASTASES**

In our study, one patient (case 2) was treated with stereotactic radiosurgery for intracranial metastatic disease. Ten courses of stereotactic radiosurgery were performed and the tumor doses were 20–30 Gy in one fraction at the 50% isodose line and 17.8–23.7 Gy in one fraction at the 80% isodose line. A partial response or stable disease was obtained for 39 months until her death.

To evaluate the effectiveness of stereotactic radiosurgery for intracranial recurrences or metastases of hemangiopericytoma, we analyzed 31 lesions from 16 patients who were treated with stereotactic radiosurgery in two reports and our own experience (28,29). Fig. 7 demonstrates tumor responses according to the dose of stereotactic radiosurgery and tumor volume. When the tumor volume was <8 cm³ (<2 cm in diameter) and the irradiation doses were >15 Gy at the 50% isodose line, the response rate was 100%. Follow-up period after radiosurgery ranged from 3 to 40 months (mean: 19.6 months). Circles, complete response; triangles, partial response; ×, no change.

Figure 6. Local-recurrence-free survival of the patients with surgery alone versus surgery with postoperative radiotherapy. There appears to be a better local control with surgery and postoperative radiotherapy of ≥50 Gy, but not significantly. Six patients from one report (20) were excluded because of uncertainty regarding the radiation dose.

Figure 7. Relationship between dose of radiosurgery and tumor volume. When the tumor volume was <8 cm³ and the irradiation doses were >15 Gy at the 50% isodose line, the response rate was 100%. Follow-up period after radiosurgery ranged from 3 to 40 months (mean: 19.6 months). Circles, complete response; triangles, partial response; ×, no change.
lesions had a complete response and the others had partial responses; thus, the response rate was 100%. When the tumor volume was ≥8 cm³ and the irradiation dose was ≥15 Gy at the 50% isodose line, only four of 16 lesions had partial responses, giving a response rate of 25%.

Galanis et al. also reported the effectiveness of the gammaknife for small recurrent disease. They reported on 20 lesions from 10 patients who underwent stereotactic radiosurgery for treatment of recurrent hemangiopericytoma. The marginal tumor doses were 12–18 Gy at the 50% isodose line. All three patients with small tumors (<25 mm in diameter) achieved complete responses and remained disease free for a median follow-up time of 3 years from treatment (9).

PALLIATIVE RADIOTHERAPY FOR BONE METASTASES

In case 1, 25–50 Gy in 5–16 fractions of external irradiation to bone metastases prevented tumor enlargement and provided good pain control for 42 months. In case 2, 25–35 Gy in 5–8 fractions prevented tumor enlargement and gave good pain control for 33 months.

Staples et al. indicated that pain due to bone metastasis was palliated by 12 Gy in four fractions and 30 Gy in 15 fractions (12). Friedman and Egan recommended a dose of up to 30 Gy to be given over 18 days for palliation of metastatic disease (30). Sakata et al. reported that 20–60 Gy in 4–24 fractions relieved pain and prevented pathological fracture due to bone metastasis (17). Effective palliation for bone metastases can be achieved with smaller doses than those required for local tumor control.

CONCLUSION

We have reported four cases of meningeal hemangiopericytoma and a review of the literature. Surgery and postoperative irradiation of ≥50 Gy resulted in significantly better local control than surgery alone. Stereotactic radiosurgery was effective for intracranial recurrences or metastases when the tumor volumes were <8 cm³ and >15 Gy was used at the 50% isodose line. Radiotherapy for bone metastases was also effective for pain relief.

References