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症　　例

Malignant Meningioma with Repeated Multiple Recurrence A Case Report

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Abstract

A case of malignant meningioma with repeated multiple recurrence was reported. A 22 year-old male first underwent the total removal of left parieto-occipital convexity meningioma in 1965. Although he had lived an uneventful life after the operation, he recognized motor weakness of the left lower limb in April 1985 when he was 41 years old and CT revealed a large tumor in the parieto-occipital parasagittal region. He underwent the total removal of the tumor and cranioplasty on May 23, 1985. After the second operation, repeated recurrence of multiple tumors was seen, which were in the frontal, parietal and occipital convexities, parasagittal regions and falx. He underwent further operations on January 23, 1986, December 11, 1986, March 30, 1987 and July 20, 1987 in addition to the first and second ones. Histological study on every operation indicated malignant meningioma with mitosis, hypercellularity and necrosis. Though radiotherapy (56 Gy whole brain irradiation) was conducted after the sixth oepration, multiple tumors recurred and clinical symptoms and signs deteriorated gradually and he finally died September 9, 1989. In malignant meningioma such as our case, early aggressive radiotherapy and chemotherapy should be considered besides radical operations.

Introduction

Though meningioma is generally benign tumor, there are some cases of malignant meningioma, in which rapid recurrence after resection, infiltration into the brain parenchyma or that from the cranium into the scalp as well as distant extracranial metastasis are seen[4,5,9]. We had a case of malignant meningioma with repeated recurrence of multiple tumors within a short time period. In this paper we discuss our case referring to some literatures.

Key words: Malignant menigioma, Multiple recurrence, Operation, Radiotherapy, Chemotherapy.
索引語：悪性髄膜腫，多発性再発，手術，放射線療法，化学療法.
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Case report

(1) The first admission (22 years old)

A 22 year-old left handed male came to our hospital with chief complaints of decreased visual acuity and motor weakness of the right lower limb. Either he or his family had had no significant anamneses including that of von Recklinghausen's disease. He recognized decreased visual acuity of both eyes two years ago and motor weakness of the right lower limb ten months ago. He was hospitalized on November 25, 1965. Neurological fingings on admission were bilateral papilledema and the right hemiparesis. The left common carotid angiography indicated some signs of compression of the vessels by a tumor such as anterior and downward displacement of the sylvian triangle and the square shift of the anterior cerebral artery, but no remarkable tumor stain was recognized (Fig. 1a). The left parieto-occipital craniotomy and total removal of the convexity meningioma was performed on December 6. The histological study of the tumor revealed malignant meningioma with hypercellularity, pleomorphism and mitosis (Fig. 1b). After the operation, right hemiparesis had gradually improved and he was discharged on December 28. He had been administered anticonvulsants as an outpatient but he stopped attendance after a certain period since he had no symptoms.

(2) The second admission (41 years old)

He recognized motor weakness of the left lower limb in April 1985 and was hospitalized on May 13, 1985. Neurological findings on admission were motor weakness of the left lower limb, bilateral optic atrophy and the narrowing of visual field. The enhanced CT revealed a large high density mass in the bilateral parieto-occipital parasagittal region (Fig. 2a). The bilateral parieto-occipital

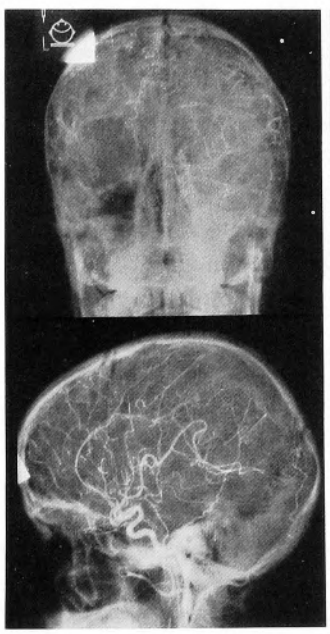

Fig. 1a

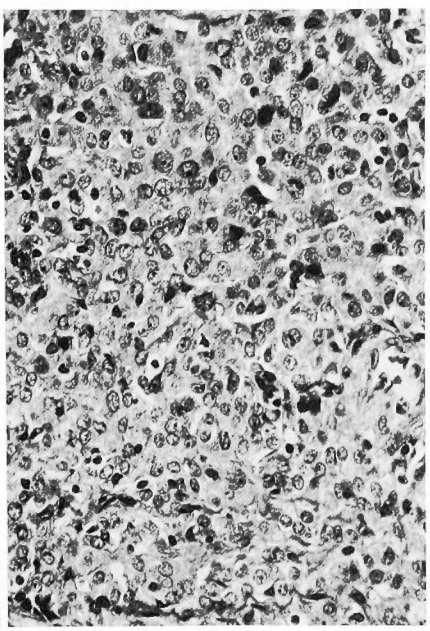

Fig. 1b

Fig. 1a Left common carotid angiography. Anterior and downward displacement of the sylvian triangle and square shift of the anterior cerebral artery are recognized but no remarkable tumor stain is seen.

Fig. 1b Histological findings at the first operation. Tumor cells with various sizes of ovoid or irregular shaped nuclei and large cytoplasm proliferate, and hypercellularity, pleomorphism and mitosis are seen. (×200)

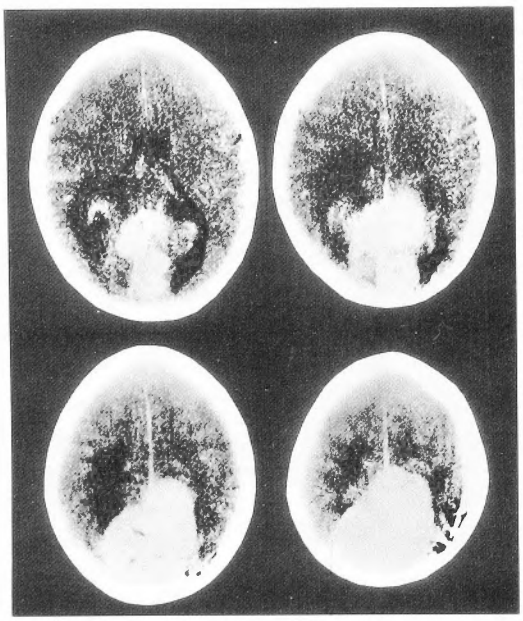

Fig. 2a

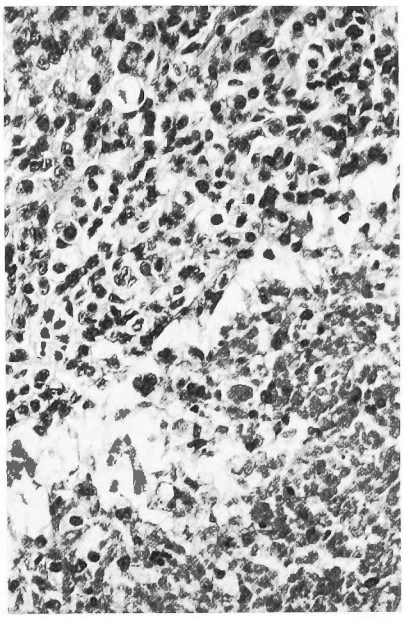

Fig. 2b

Fig. 2a Enhanced CT before the second operation. A large high density mass is seen in the bilateral parieto-occipital parasagittal region.

Fig. 2b Histological findings at the second operation. Hypercellularity, pleomorphism, mitosis and necrosis are seen. (×200)

craniotomy, total removal of parasagittal meningioma and the cranioplasty were performed on May 23. The histological study of the tumor revealed malignant meningioma with hypercellularity, pleomorphism, mitosis and necrosis (Fig. 2b). After the operation, motor weakness of the left lower limb had gradually improved and he was discharged on June 21.

(3) The third admission (42 years old)

As he had developed motor weakness and convulsion of the left lower limb since November 1986, he was hospitalized on January 13, 1986. Neurological findings on admission were motor weakness and sensory disturbance of the left lower limb and left Babinski sign. The enhanced CT revealed multiple tumors in the right parietal parasagittal region, on the left side of the parietal falx and on the left side of the occipital falx (Fig. 3a). The parieto-occipital craniotomy, total removal of the tumors was performed on January 23. The histological study revealed malignant meningioma and all the tumors resected from the different regions had the identical histological findings. He was discharged on April 7 with slight motor weakness of the left lower limb.

(4) The fourth admission (43 years old)

As CT performed in November revealed the recurrence of tumors, he was hospitalized again on December 1. Neurological findings on admission were motor weakness and sensory diturbance of the left lower limb and left Babinski sign. The enhanced CT revealed multiple tumors on either side of the parietal falx and falco-tentorial region (Fig. 3b). The bilateral parieto-occipital craniotomy, total removal of the tumors was performed on December 11. The histological study indicated malignant meningioma and all the tumors resected from the different regions had the identical histological findings. After operation there were no changes observed in the neurological findings and he was

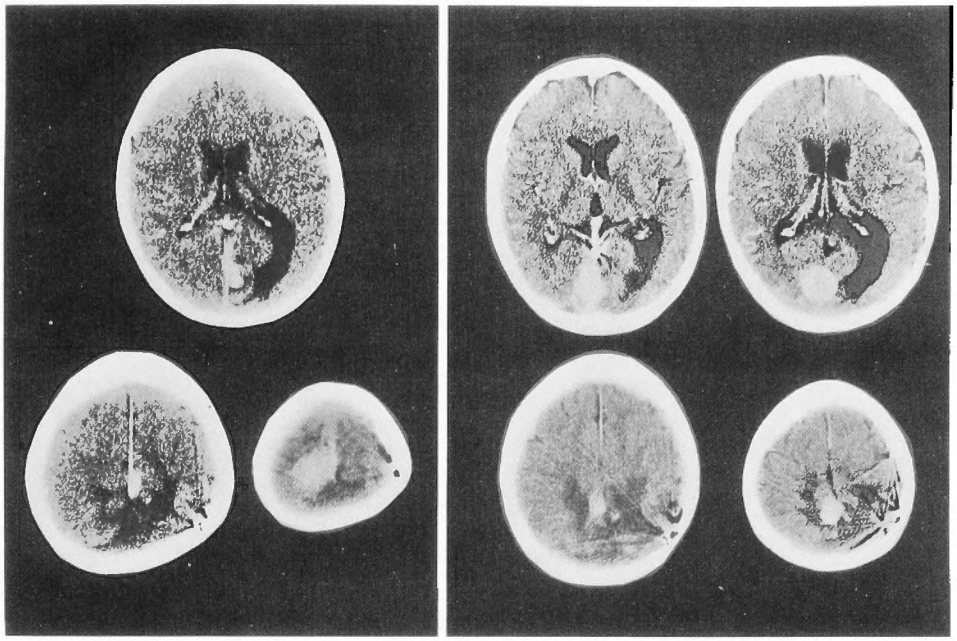

Fig. 3a Fig. 3b

Fig. 3a Enhanced CT before the third operation. Tumors are seen on the right side of the parietal parasagittal region, on the left side of the parietal falx and on the left side of the occipital falx.

Fig. 3b Enhanced CT before the fourth operation. Multiple tumors are seen on either side of the parietal falx and falco-tentorial region.

discharged on December 27.

(5) The fifth admission (43 years old)

As the motor weakness of the left lower limb had gradually been aggravated and the seizure had appeared since the middle of January 1987, he was hospitalized again on March 20. Neurological findings on admission were motor weakness and sensory disturbance of the left lower limb and left Babinski sign. The enhanced CT revealed multiple tumors in the right frontal convexity and in the parietal and occipital falx (Fig. 4a). The fronto-parieto-occipital craniotomy, total removal of the tumors was performed on March 30. The histological study revealed malignant meningioma with infiltration into brain parenchyma, hypercellularity, pleomorphism and mitosis, and all the tumors resected from the different regions had the identical histological findings (Fig. 4b). Just after the operation, he developed motor weakness of the right lower limb and aggravating motor weakness of the left lower limb, because of the disturbance of venous return due to the injury of bridging veins and superior sagittal sinus during operation. The motor weakness was once improved by rehabilitation but was aggravated again in June. The enhanced CT conducted in July revealed multiple tumors in the tentorium, the occipital convexity and parietal falx (Fig. 5). The parieto-occipital craniotomy, total removal of the tumors was performed on July 20. The histological study indicated malignant meningioma and all the tumors resected from the different regions had the identical histological findings. He had undergone the radiotherapy (56 Gy whole brain irradiation) since July 31 and was discharged on April 7, 1988.

(6) The sixth admission (45 years old)

As he had developed generalized convulsion since October 1988, he was hospitalized on

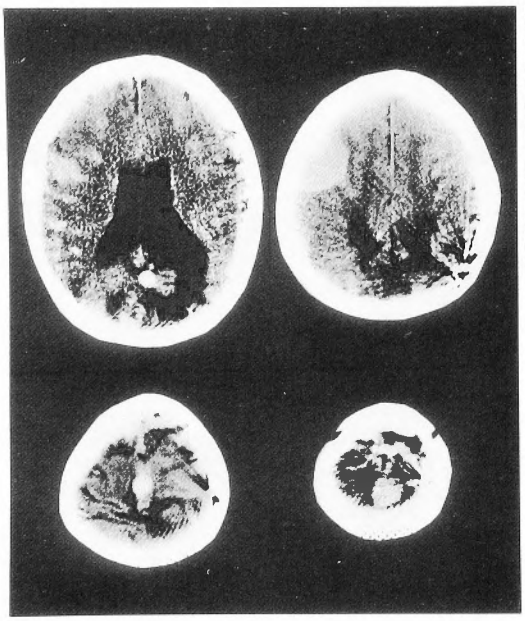

Fig. 4a

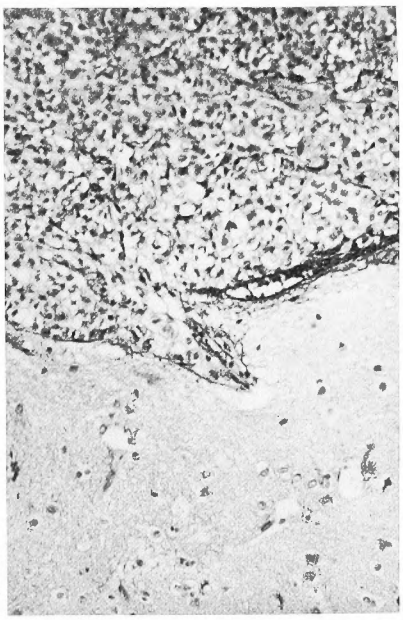

Fig. 4b

Fig. 4a Enhanced CT before the fifth operation. Multiple tumors are seen in the right frontal convexity, the parietal and occipital falx.

Fig. 4b Histological findings at the fifth operation. Infiltration of tumor cells with hypercellularity, pleomorphism, and mitosis into the brain parenchyma is seen. (×100)

November 24. Neurological findings on admission were motor weakness and sensory disturbance of both lower limbs, bilateral Babinski sign. The enhanced CT revealed multiple tumors in the right frontal convexity the left occipital convexity and on either side of the parietal falx (Fig. 6a). The fronto-parieto-occipital craniotomy, total removal of tumors was performed on January 18, 1989. The tumor in the left occipital convexity infiltrated into the bone and further the scalp. The histological study revealed malignant meningioma and all the tumors resected from the different regions had the identical histological findings (Fig. 6b). There were no changes observed in the neurological findings and he was discharged on April 16.

(7) The seventh admission (45 years old)

As he had developed motor aphasia, motor weakness of the left upper limb and urinary incontinence since the end of June, he was hospitalized on July 5. Neurological findings on admission were motor and sensory aphasia, motor weakness of both lower limbs and left upper limb and bilateral Babinski sign. The enhanced CT revealed a large tumor in the right frontal convexity and tumors in bilateral occipital convexity and on the left side of the parietal falx and revealed the midline shift (Fig. 7). No radical or aggressive treatment such as an operation was performed this time at the request of his family and only conservative treatment was conducted. The motor weakness of the left upper limb and both lower limbs was gradually aggravated. He became unconscious and died from cerebral herniation on September 9, 1989. Autopsy was not conducted.

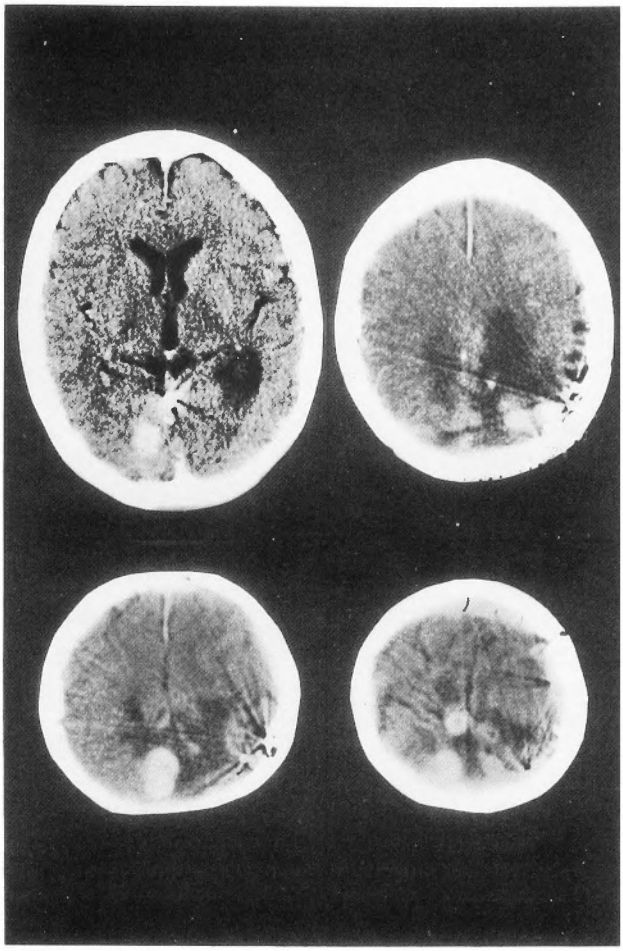

Fig. 5 Enhanced CT before the sixth operation. Multiple tumors are seen in the tentorium and the occipital convexity and parietal falx.

Discussion

Though meningioma is generally benign tumor, there are some cases of malignant meningioma, which has malignant features histologically and develops infiltration from cranium into scalp, distant extracranial matastasis, repeated recurrences in a short time period and rapid growth[4,5,9]. In our case the histological findings indicated malignant features such as hypercellularity, pleomorphism, mitosis, necrosis and infiltration into the cerebral parenchyma. Alvarez et al.[2] listed the following histological findings as the criteria of the malignant meningioma: (1) high mitotic rate, (2) high cellularity, (3) necrosis, (4) infiltration of the underlying brain, (5) poor differentiation and (6) distant blood-borne metastasis. They called meningioma with high mitotic rate and at least one of the other features atypical meningioma. Since distant metastasis was not proved in our case, it may be an example of the atypical meningioma as Alvarez et al. proposed. But we think this case may be classified as malignant meningioma from the clinical features such as repeated multiple recurrence and rapid growth in a short time period, infiltration from the cranium into the scalp and

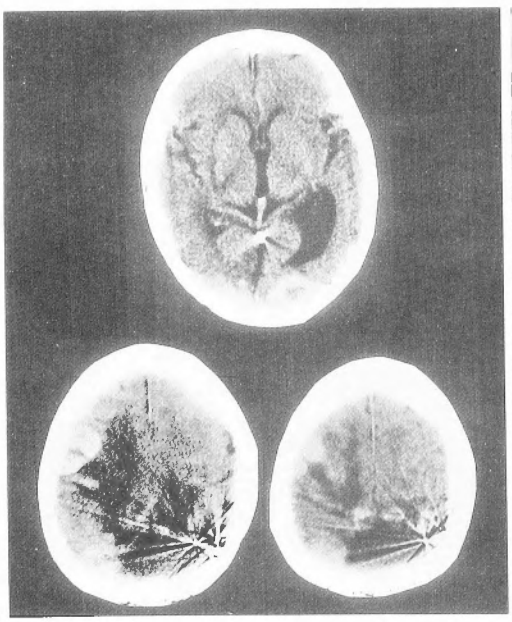

Fig. 6a

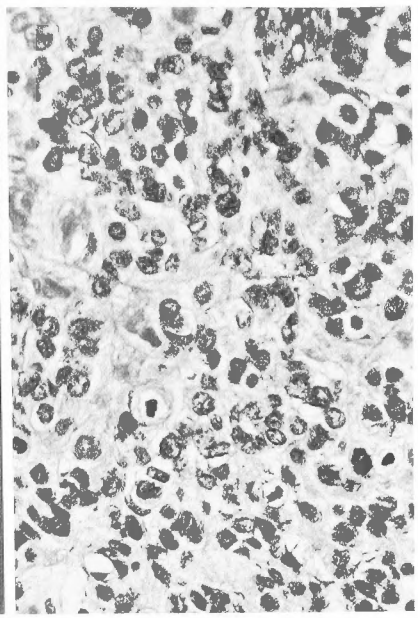

Fig. 6b

Fig. 6a Enhanced CT before the seventh operation. Multiple tumors are seen in the right frontal convexity, the left occipital convexity and on either side of the parietal falx.

Fig. 6b Histological findings at the seventh operation. Hypercellularity, pleomorphism and mitosis are seen. (×200)

histological findings.

LEVIN et al.[6] stated three possible causes of multiple meningioma: (1) existence of multicentric foci, (2) transmission of tumor cells via CSF pathways and (3) venous transmission. ABTAHI[1] classified twelve cases of multiple meningioma into three groups; (1) first group: multiple meningioma with neurofibromatosis (3 cases) (2) second group: multiple meningioma alone without neurofibromatosis (5 cases) (3) third group: Multiple tumors were first recognized at the second or third recurrence operation (4 cases). Our case was not associated with neurofibromatosis. The tumor on the second operation might be occurred originally since twenty years had elapsed between the first and the second operation and the tumor sites at the first and second operations were different. However, multiple tumors were repeatedly recurred in a short time period after the second operation. Tumors were recurred in sites distant from as well as adjacent to the operated regions. The histological findings of multiple tumors were identical. From these findings, possible causes of multiple recurrence of the tumors in our case may be that tumor cells are disseminated to the periphery of the lesions by the operative procedure, transmission of tumor cells via CSF pathways and venous transmission due to infiltration into the sinus.

Effectiveness of radiotherapy against meningioma was reported in literature but it is still controversial[3,7,8,10,12]. We also tried radiotherapy in this case but only managed to elongate a time period before the recurrence and could not prevent the recurrence.

As for chemotherapy to meningioma, YAMADA et al.[11] reported that they performed intracarotid administration of adriamycin against extracranially infiltrating malignant meningioma and the tumor completely disappeared. They reported that though the patient died from hemorrhagic in-

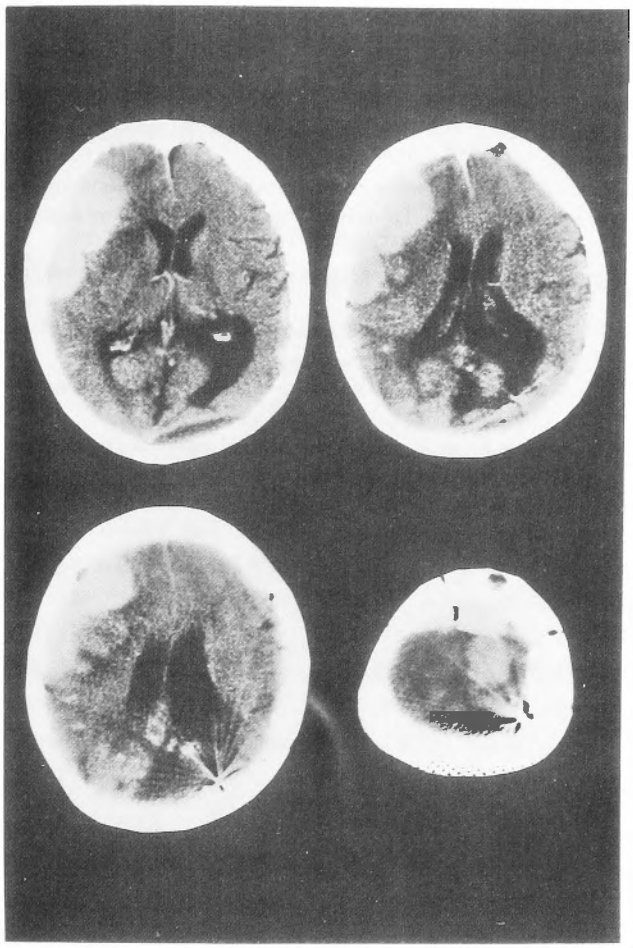

Fig. 7 Enhanced CT on the seventh admission. Multiple tumors are seen in the right frontal convexity, in the bilateral occipital convexity and on the left side of the parietal falx. The midline shift is also recognized.

farction of both lungs and congestive pulmonary edema which were considered to be the side effect of adriamycin, no tumors were found on autopsy. We think that chemotherapy is a method worth trying against malignant meningioma such as our case with repeated recurrence despite the radical operations and radiotherapy as long as we fully take care of the side effects.

Since complete cure of the malignant meningioma such as our case is not easy, the greatest care should be taken at the operation to aim at the total removal of the tumor, not to disseminate the tumor cells by operative procedure and to resect the dura near the attachment of the tumor as wide as possible. From the early stage, aggressive treatment with radiotherapy and chemotherapy should be considered in addition to the radical operation.

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和文抄録

多発性再発を繰り返した悪性髄膜腫の1例

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多発性再発を繰り返した悪性髄膜腫の1例を報告した．症例は，1965年，22歳の時，左頭頂後頭部弓隆部の髄膜腫の全摘出を受けた男性である．その後経過良好であったが，1985年4月，41歳の時，左下肢の運動麻痺をきたし，CT で頭頂後頭部傍矢状洞部に大きな腫瘍を認め，5月23日，腫瘍全摘出と頭蓋形成術を受けた．2回目の手術後，前頭部，頭頂部，後頭部弓隆部，傍矢状洞部，大脳鎌，小脳テントに，多発性に繰り返し腫瘍が再発し，4回，計6回の手術を受けた．腫瘍の組織学的所見は，いずれの手術でも，細胞分裂，壊死，正常脳への浸潤，高細胞密度を示す悪性髄膜腫であった．6回目の手術後，56 Gy の放射線療法を受けたが，多発性の腫瘍の再発を認め，臨床症状が徐々に悪化し，1989年9月9日死亡した．本例のような悪性髄膜腫の治療は，手術的に腫瘍を摘出するのみならず，早期から積極的に放射線療法，化学療法を試みるべきであると考える．