Spontaneously Ruptured Craniopharyngioma Cyst Without Meningitic Symptoms
—Two Case Reports—

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Abstract

Two cases of spontaneous rupture of cystic craniopharyngioma without chemical meningitis are described. A 70-year-old woman complained of headache and visual field disturbance in July 1993. The tumor was extirpated in November 1993 and again in December 1996. After regular periodic follow-up evaluation, she was hospitalized for reoperation because of expansion of the cyst on magnetic resonance (MR) imaging in November 1998. However, preoperative MR imaging taken 8 weeks later revealed spontaneous reduction of the cyst. A 69-year-old woman noticed deteriorating vision and bitemporal hemianopsia in November 1998. The tumor was removed in December 1998, and 50.4 Gy postoperative radiotherapy was administered. MR imaging in May 2000 demonstrated an enlargement of the cyst, so she was hospitalized again for operation. However, preoperative MR imaging taken 7 weeks later showed spontaneous reduction of the cyst. Neither of the cases of cyst rupture were accompanied by symptoms of chemical meningitis. The signal intensity of the tumors on T1-weighted MR imaging declined after cyst reduction. Thereafter, the cysts increased in size again at 7 months and 5 months. Regular follow-up on MR imaging is necessary, since the cyst size can increase rapidly, even after spontaneous rupture.

Key words: spontaneous rupture, craniopharyngioma, cystic brain tumor, meningitis

Introduction

Suprasellar cystic tumors occasionally show spontaneous remission in patients with dermoid cyst, craniopharyngioma, and Rathke's cleft cyst. Spontaneous remission of suprasellar cystic tumors is considered to be due to rupture of the cyst. Most such cases of spontaneous rupture are accompanied by chemical meningitis. Only 15 reports of spontaneous cyst shrinkage due to rupture associated with craniopharyngioma are available, and many recent reports do not discuss subsequent surgical treatment.4,7–9) Spontaneous rupture of craniopharyngioma cysts without chemical meningitis are extremely rare, and the natural history after spontaneous rupture is unclear. We report two cases of spontaneous rupture of cystic craniopharyngioma without signs of chemical meningitis, followed by an increase in the cyst size after a short period of reduction.

Case Reports

Case 1: A 70-year-old woman complained of morning headache and progressive visual disturbance in July 1993. She had no significant medical or family histories. Computed tomography (CT) and magnetic resonance (MR) imaging identified suprasellar tumor. Neurological and endocrinological examinations found only bitemporal upper quadrant hemianopsia. Head CT showed a suprasellar low density cystic lesion with partially calcified thin walls. MR imaging showed the cyst as low and high signal intensities on the T1- and T2-weighted images, respectively, and the left side of the cyst wall was enhanced by gadolinium-diethylenetriaminepenta-acetic acid (Gd-DTPA). The preoperative diagnosis was craniopharyngioma or dermoid cyst. The tumor cyst was evacuated, and the tube of an Ommaya’s reservoir was inserted through the partially removed wall. Her visual field and acuity improved, and MR imaging showed the cyst had disappeared.

She was hospitalized again with complaints of visual disorder and recurrence of the cyst in Decem-
Fig. 1 Case 1. A: T1-weighted magnetic resonance (MR) image taken during cyst expansion showing a suprasellar cystic mass lesion measuring $23 \times 18 \times 22$ mm. The signal intensity of the cyst is higher than that of the cerebrospinal fluid. B: T1-weighted MR image taken during cyst reduction showing the tumor measuring $16 \times 12 \times 18$ mm. The signal intensity of the cyst has decreased compared with before reduction.

Fig. 2 Case 2. A: T1-weighted magnetic resonance (MR) image taken during cyst expansion showing a suprasellar cystic mass lesion measuring $30 \times 23 \times 30$ mm. The signal intensity of the cyst is higher than that of the cerebrospinal fluid. B: T1-weighted MR image taken during cyst reduction showing the tumor measuring $26 \times 22 \times 12$ mm. The signal intensity of the cyst has decreased compared with before reduction.

Spontaneous Ruptured Craniopharyngioma Cyst

Case 1: A 53-year-old man had noticed progressive blurred vision since November 1996. The MR imaging findings were similar to those at the onset. The Ommaya’s reservoir had become obstructed, so was removed together with the tumor through a transcranial approach.

Her visual field deteriorated and MR imaging indicated expansion of the cyst to $23 \times 18 \times 22$ mm in November 1998 (Fig. 1A). A third operation was scheduled. However, preoperative MR imaging taken 8 weeks after the last MR imaging revealed definite involution of the cyst to $16 \times 12 \times 18$ mm and a decrease of signal intensity on the T1-weighted image (Fig. 1B). Spontaneous rupture of the cyst was considered, although no meningitic symptoms were observed around the involution. The operation was canceled. Her visual field had slightly recovered before leaving the hospital.

MR imaging suggested slight expansion of the cyst without any change of signal intensity in August 1999, 7 months after spontaneous rupture. The cyst had expanded distinctly to $22 \times 18 \times 20$ mm in January 2000, and the signal intensity had also increased again on T1-weighted imaging. The patient noticed deterioration of her visual acuity, and the tumor was removed almost completely through a transcranial approach in February 2000. We could not detect any distinct rupture point of the cyst during the surgery. The cyst fluid was a transparent light yellow liquid without cholesterol crystals. The final histological diagnosis was craniopharyngioma with adamantinomatous pattern. Postoperative radiotherapy was not carried out, because MR imaging demonstrated no mass lesion after the surgery.

Case 2: A 69-year-old woman had noticed progressive blurred vision since November 1998. She had no particular family and past medical histories. Neurological examination on admission revealed bitemporal hemianopsia, but no endocrine abnormality. Head CT showed a cystic mass lesion with partial calcification. MR imaging showed a cystic lesion in the suprasellar area, with the content appearing as slightly high and high signal intensity on the T1- and T2-weighted images, respectively. The cyst wall appeared as iso-signal intensity with enhancement by Gd-DTPA. Cystic craniopharyngioma was suspected. The tumor including the cyst wall was removed through a transcranial approach. The histological diagnosis was craniopharyngioma with adamantinomatous pattern. Postoperative radiotherapy was performed with a total dosage of 50.4 Gy.

MR imaging confirmed recurrence of the cyst in May 2000. The cyst had expanded to $30 \times 23 \times 30$ mm (Fig. 2A). Her neurological symptoms showed no change, but she was hospitalized for surgery. However, preoperative MR imaging taken 7 weeks later showed the cyst had reduced to $26 \times 22 \times 12$ mm with decreased signal intensity on the T1-weighted image (Fig. 2B). These findings indicated spontaneous rupture of the cyst. No meningitic symptom was recognized, so we did not examine her cerebrospinal fluid. The operation was postponed.

Five months after the rupture, MR imaging showed the cyst had enlarged again with increased
signal intensity on the T1-weighted image. Reoperation was performed, and an indwelling tube of an Ommaya’s reservoir was implanted for drainage. We could not determine the distinct rupture point during the surgery, although there were some thin portions of the cyst wall. The cyst content was a transparent light yellow liquid without cholesterol crystals. Histological examination indicated typical craniopharyngioma with adamantinomatous pattern.

Discussion

The mechanism of rupture may depend on weakness of the cyst wall caused by cyst expansion inducing degeneration of the cyst wall.5) The presence of thin cyst walls was confirmed at the first operation in our two cases. In addition, before and after spontaneous rupture, the signal intensity of the cyst decreased to similar to that of the cerebrospinal fluid on T1-weighted MR imaging. Considering these findings, we infer that spontaneous rupture of the cyst resulted in the observed cyst reduction.

Chemical meningitis is supposed to be caused by the cholesterol contained in the cyst fluid of craniopharyngioma, resulting in headache, fever, nuchal rigidity, and consciousness disorder.2,7) In addition, rupture of the cyst may cause cerebral infarction following vasospasm.8) However, the quantity of cholesterol produced by cultured craniopharyngiomas varies greatly.3) A case of craniopharyngioma with low cholesterol content has also been reported.1) Therefore, the possibility of chemical meningitis is low if the craniopharyngioma has little cholesterol content.4,9) Unfortunately, MR imaging cannot provide an estimate of the cholesterol level, because protein concentrations contribute more to the signal intensity than lipid concentrations on the T1-weighted image.6) However, we consider that the cholesterol levels in our two cases were low, so the rupture of the cyst did not result in meningitic symptoms. The cyst content of our cases was a transparent light yellow liquid without cholesterol crystals, although the signal intensity of the cyst content had increased on the T1-weighted image. Spontaneous rupture of craniopharyngioma without chemical meningitis may become more commonly detected by improved methods of MR imaging and CT.

The reported follow-up periods are limited to the period of the spontaneous rupture. Few case reports described the natural course after the rupture. In our two cases, MR imaging performed 7 and 5 months after the shrinkage indicated increased signal intensity on the T1-weighted image and enlargement of the cysts. Reoperation was required for both patients. Careful continued observation is necessary even after spontaneous shrinkage of the cyst.

References


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