

A canine case with cystic meningioma showing miraculous reduction of the cystic lesion

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ABSTRACT. A 12-year-old spayed female Labrador retriever was presented with forebrain signs. Brain MRI revealed a huge cystic lesion with the thickened falx in the frontal region. The brain parenchyma surrounding the lesion showed significant signs of a mass effect and also increased intracranial pressure. However, the dog suddenly became lucid after about two weeks, and an MRI scan one month after the initial study revealed a dramatically shrunken cystic lesion. The dog survived for over a year until it was euthanized for other reasons, and the brain lesion was diagnosed as a cystic meningioma histologically. To the authors' knowledge, this is the first report that described the reduction of the cystic lesion of a cystic meningioma in dogs.

KEY WORDS: dog, cystic meningioma, MRI

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Meningiomas are the most common primary brain tumor in dogs: the incidence of canine cystic meningioma has been reported to be 12.5–32% [12, 18]. It is not uncommon for a cystic lesion to be larger than a tumor [5, 15]. Sometimes, increased intracranial pressure (ICP) is caused by cyst expansion rather than tumoral growth. We report and discuss a rare case of cystic meningioma that showed a significant reduction of the cystic component, resulting in long-term survival. To the authors' knowledge, no reports have described the reduction of the cystic lesion associated with a meningioma either in veterinary medicine or in human medicine.

A 12-year-old spayed female Labrador retriever was presented to a private clinic with cluster seizures, circling towards the left and decreased mental status (day 0). The owner reported that the dog had one seizure 2 months previously. CBC, serum chemistry, urine analysis, thoracic and abdominal radiographs, heart and abdominal ultrasound, and ECG were within normal range. A neurological examination found depression, left circling and decreased postural reactions of the right side, which suggested left forebrain dysfunction.

To confirm the lesion, an initial MRI scan was performed using a 0.4-tesla (T) system (day 10). The MRI revealed a large cystic lesion showing hyperintensity on T2-weighted

images (T2W), and hypointensities on T1-weighted (T1W) and fluid-attenuated inversion recovery (FLAIR) images extending from the left parafalx olfactory bulb to the frontal region that somewhat extended to the right (Fig. 1). The left side of the cerebral falx was included within the cystic lesion, and was slightly thickened and hyperintense on T2W, isointense on T1W and clearly enhanced on Gadolinium-enhanced T1W (Gd-T1W). The outer wall of the cystic lesion was not enhanced. The maximum diameter of the cyst, including the parenchymal lesion of the falx, was 2.0 cm × 2.2 cm × 4.2 cm. The cerebral parenchyma surrounding the lesion showed a significant mass effect and other increased ICP findings consisting of cerebral sulci effacement, displacement of the lamina quadrigemina, deformation of the interthalamic adhesion, rostral cerebellar contour flattening, third ventricle compression and suprapineal recess compression; however, perilesional edema and brain herniation were not observed [3]. These MR findings reminded a cystic meningioma as a top of the differential diagnoses that included subarachnoid cyst, brain abscess and other tumors with cystic lesion. Based on the clinical findings and brain MRI, there was concern about the risk of brain herniation, so emergency surgical decompression was recommended. Although the owner wanted the surgery to be done as soon as possible, there was a 1-month wait until it could be performed at the university teaching hospital. During this waiting period, the dog was medically managed with phenobarbital (Phenobal; Daiichi-Sankyo, Tokyo, Japan) (1.25 mg/kg, BID), zonisamide (Excegran; Dainippon-Sumitomo, Osaka, Japan) (4.1 mg/kg, BID), isosorbide (Isobide; Kowa, Tokyo, Japan) (0.5 ml/kg, BID) and prednisolone (Predonine; Shionogi, Osaka, Japan) (0.3 mg/kg, SID).

On the day of the operation (day 38), the surgeon expected

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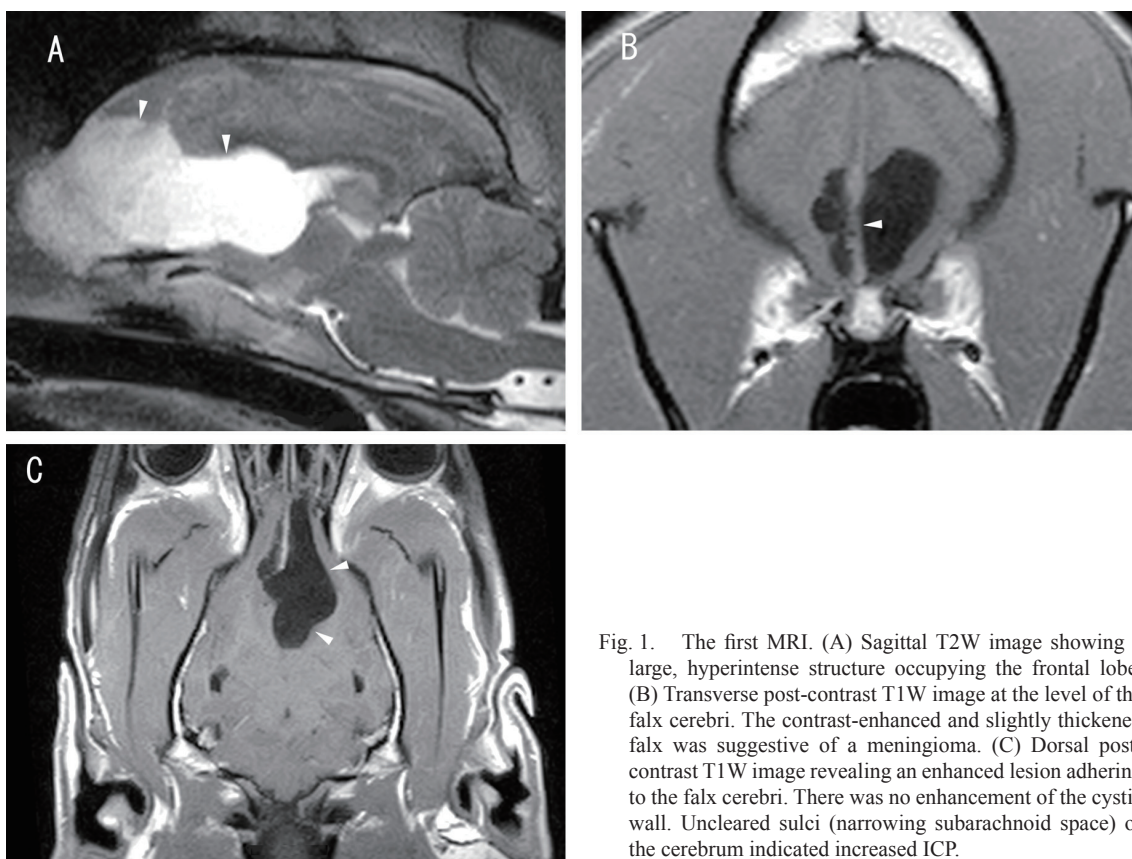


Fig. 1. The first MRI. (A) Sagittal T2W image showing a large, hyperintense structure occupying the frontal lobe. (B) Transverse post-contrast T1W image at the level of the falx cerebri. The contrast-enhanced and slightly thickened falx was suggestive of a meningioma. (C) Dorsal post-contrast T1W image revealing an enhanced lesion adhering to the falx cerebri. There was no enhancement of the cystic wall. Uncleared sulci (narrowing subarachnoid space) of the cerebrum indicated increased ICP.

that the dog might be unable to walk or stand normally and would have a decreased level of consciousness due to increased ICP, but the dog walked herself into the consultation room and was alert. On neurological examination, the dog showed a decreased menace response in the right eye and a delayed proprioceptive positioning in the right pelvic limbs, but everything else was unremarkable. The owner had said that the dog had suddenly become lucid about two weeks previously.

On the same day, a second MRI scan was performed using a 3.0-T system for preoperative evaluation. Although there was no change in the contrast-enhancing lesion of the falx, the cystic lesion was 1.2 cm × 2.1 cm × 3.5 cm, indicating a significant reduction in volume. There was no signal change on the T2W and T1W images; however, the signal intensity of the cyst was increased on FLAIR images (i.e., unsuppressed fluid signal), suggesting inflammation or hemorrhage. In addition, the increased ICP findings observed on the first MRI had disappeared. Because of the improvements in clinical signs and in MRI findings, the surgery was cancelled, and the dog was continued on medical therapy. Hydroxyurea (Hydrea; Bristol-Myers, Tokyo, Japan) (10 mg/kg, SID) was added to the current regimen of medications stated above (from day 40).

Subsequently, orofacial focal seizures and generalized seizures were observed several times, but there was no progression of neurological abnormalities. A follow-up (third)

MRI scan was performed 45 days after hydroxyurea was started (day 85) using a 0.4-T system. The cyst continued to decrease, reaching a final diameter of 1.0 cm × 1.9 cm × 2.2 cm. In addition, the signal intensity of the cyst on FLAIR images had recovered to hypointensity.

The dog lived for more than a year without any obvious neurological progression, but she developed perianal adenocarcinoma. Because it became difficult for the dog to defecate, it was euthanized on day 544. Just before the dog was euthanized, a final MRI scan (0.4-T) was performed with the owner's consent. As compared with the third scan, there was no significant change in the size of the tumor or that of the cyst (Fig. 2). The autopsy was performed after euthanasia.

Histological examination of the brain revealed that the cystic lesion was connected to the falx and consisted of proliferating tumor cells in the left frontal region. The thickened falx and cystic wall, which comprised proliferating multilayered (stratified) meningeal epithelial cells, led to the diagnosis of meningotheial meningioma, cystic type. Because the wall of an arachnoid cyst is generally composed of monolayer epithelium, we judged that this multilayered wall was a neoplastic cyst. The cystic lesions were supposed to dehiscence to the midline region (Fig. 3).

Meningiomas account for 33–49% of primary brain tumors in dog [17]. In addition, Labrador retrievers are predisposed to developing brain tumors [2, 4, 18]. Although the cyst, which seemed to be the cause of the clinical signs (in-

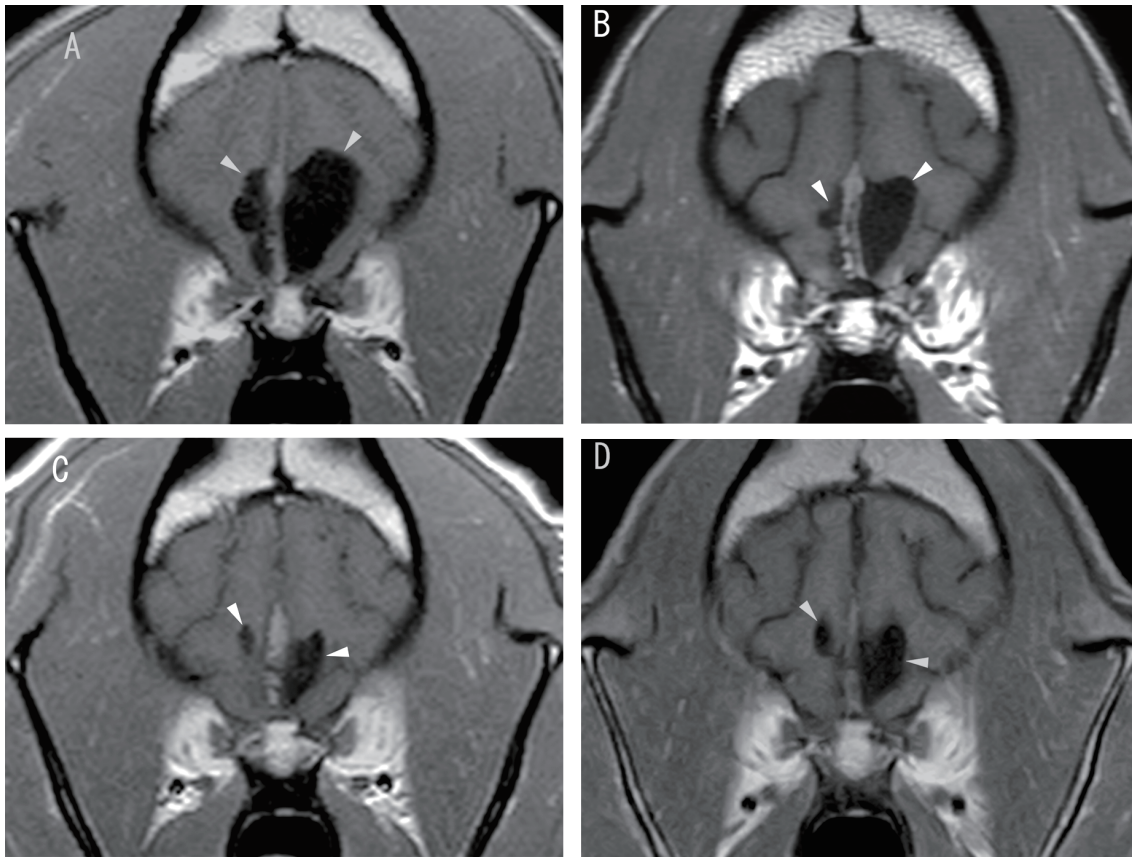


Fig. 2. Comparison with the time course of the MRI findings. Transverse post-contrast T1W images at the level of the falx cerebri: (A) first (day 10); (B) second (day 38); (C) third (day 85); and (D) fourth (day 544). Comparing images A and B, there was a significant reduction in the cystic lesion and spreading into the subarachnoid spaces. There was no obvious change between images C and D.

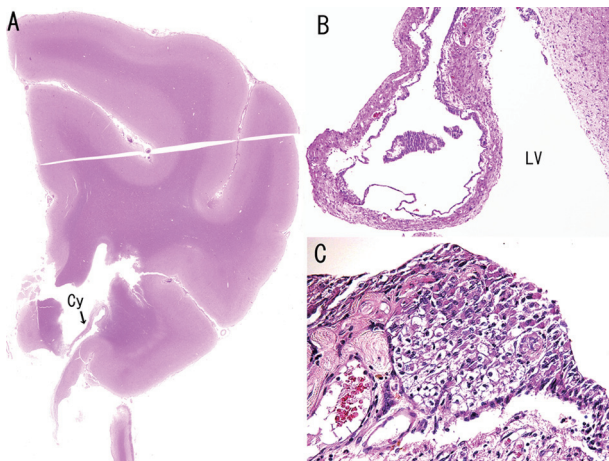


Fig. 3. Histopathological appearance of the cystic lesion. (A) The over view of the frontal cortex with cystic lesion (Cy, Arrow), H&E, $\times 10$. (B) The cystic tumor was not directly connected with the lateral ventricle (LV), H&E, $\times 200$. (C) Tumor cells of the cystic lesions were mostly polygonal-shaped with wide, clear to eosinophilic cytoplasm, H&E, $\times 400$. Their nuclei were round to oval-shaped with indistinct nucleoli.

creased ICP), was significantly larger than the parenchymal component, a cystic meningioma was clinically diagnosed based on the MRI findings. Nauta *et al.* [13] classified cystic formation associated with meningiomas into four types: those located centrally within the tumor, such that the cyst surrounded by macroscopic tumor throughout (type I); those located peripherally within the tumor, but still wholly within the margins of the tumor, such that there is a microscopically visible attenuated rim of tumor cells along the peripheral margins of the cyst (type II); those within the adjacent brain, such that they actually lie within the adjacent brain rather than within the tumor itself (type III); and those at the interface of the tumor and brain, as a loculation of CSF in the subarachnoid space, such that they do not appear within either the tumor or the brain itself (type IV). Because the cyst wall was not enhanced by gadolinium in MR imaging, the tumor in this case was thought to be classified as type III or IV [5, 7, 9, 10, 16, 20]. However, the histopathological diagnosis was type II.

On the first MRI, the brain parenchyma surrounding the lesion showed a mass effect, and there were findings of increased ICP. On the second MRI, significant reduction of the cystic lesion had occurred even though the tumoral

parenchyma (i.e., enhanced falx) was not changed obviously, and increased ICP findings were no longer observed. Furthermore, the dog had improved clinically. Although it is undeniable that clinical improvement was caused by medication (i.e., isosorbide and prednisolone), because the owner reported that the symptoms improved suddenly, it is unlikely that the medications alleviated them. It is likely that the cyst had ruptured miraculously due to for some reason.

Typically, excessive expansion of cystic lesions in the forebrain may increase ICP and might lead to transtentorial herniation. In the present case, however, we hypothesized that the cyst wall was too thin and vulnerable to such a high ICP and ruptured before a transtentorial herniation occurred. The unsuppressed intensity of the intracystic fluid on FLAIR in the second MRI, which may be indicative of minor inflammatory/hemorrhage due to rupture, supports this hypothesis. Because the intracystic fluid probably leaked into the subarachnoid space from the hiatus, the dog could survive. In addition, the present case had a larger cystic volume than mass volume, causing a brain herniation as reported previously [19]. Therefore, if the cystic lesion of the present case was parenchymatous, it would undoubtedly have caused a transtentorial herniation. On the other hand, many reports have described the spontaneous reduction of an arachnoid cyst in human medicine [6, 11, 14]. These reports suggest that physiologic transient elevation of ICP (breath hold, cough or sports activities, etc.), minor head trauma, meningitis and hemorrhage may cause spontaneous rupture of the arachnoid cyst [6]. Therefore, although the present case was classified as cystic meningioma type II, the possibility that similar events had occurred cannot be ruled out. However, because there is no history of trauma or other findings, it is most likely that the cyst had ruptured miraculously due to extremely high ICP.

Because the clinical behavior of meningiomas progresses slowly and the survival duration tends to be longer as compared with other tumors [1, 8], cyst reduction may bring about longer survival like as observed in the present case. However, the circumstances of this case are thought to be extremely rare, and the authors think that surgical resection or decompression is still the first choice for the treatment of cystic meningiomas.

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