

A Case of Dermoid Cyst Arising in the Temporal Lobe

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Abstract

Intracranial dermoid cysts are rare congenital lesions that result from abnormal sequestration of ectodermal cells during neural tube formation. These tumors are especially rare in lateral areas such as in the temporal lobe. In this study, we report a case of dermoid cyst located in the right temporal lobe. A 50-year-old man was referred for further treatment of a tumor. CT revealed a low-density mass lesion in the right temporal lobe, with calcification. MRI showed the lesion with high signal intensity on diffusion-weighted imaging, high-low mixed signal intensity on T1-weighted imaging, and iso-high signal mixed intensity on T2-weighted imaging; the capsule was enhanced with gadolinium. Differential diagnosis included dermoid cyst, epidermoid cyst, teratoma, and neurenteric cyst. We decided to perform surgery for the improvement of his symptom, histopathological diagnosis, and radical cure. A right temporal craniotomy was performed, and the tumor was found adherent to the surrounding brain tissue. The tumor was completely removed under subpial dissection. Hair was confirmed in the tumor content. On histopathology, the cyst wall was lined with stratified squamous epithelium, sebaceous glands, small vessel aggregates, and inflammatory infiltrate. Keratinized material and hair were found in the lumen. The patient was discharged 7 days after surgery with no new neurologic deficits. This case was unusual in terms of the effect of gadolinium enhancement on MRI, and the presence of adipose tissue and calcification were useful for diagnosis. It is vital to consider prevention of chemical meningitis due to intrathecal dissemination of the tumor content intraoperatively.

Keywords: dermoid cyst, temporal lobe, MRI

Introduction

Intracranial dermoid cysts are tumors that arise from remnant tissues of the fetal period, contain skin appendages such as sebaceous glands and hair,^{1,2} and are commonly located in the midline of the suprasellar region, cerebellum, and brain stem.^{3,4} Intracranial dermoid cysts are rare, especially in lateral areas such as in the temporal lobe, and account for 0.1% of all brain tumors in Japan.⁵ In this report, we present a surgical case of dermoid cyst of the right temporal lobe and report on preoperative imaging and surgery with some literature review.

Case Report

History and examination

A 50-year-old man, with no past medical history, was referred to our department with a complaint of a headache that started 3 months earlier. At presentation, the patient had no neurological deficit. General physical and systemic examinations were normal.

Plain CT scan of the brain (Fig. 1A) revealed an iso-to-hypo-dense mass lesion in the right temporal lobe with a diameter of 3.0 × 2.8 × 3.0 cm including a small calcified area. On MRI, the tumor appeared as a heterogeneous hyperintense lesion on diffusion weighted imaging (DWI, Fig. 1B) and T2-weighted imaging (T2, Fig. 1C). T1-weighted imaging (T1, Fig. 1D) showed a hypointense lesion with a hyperintense spotty area. In addition, partial enhancement of the capsule was seen in T1 after injection of

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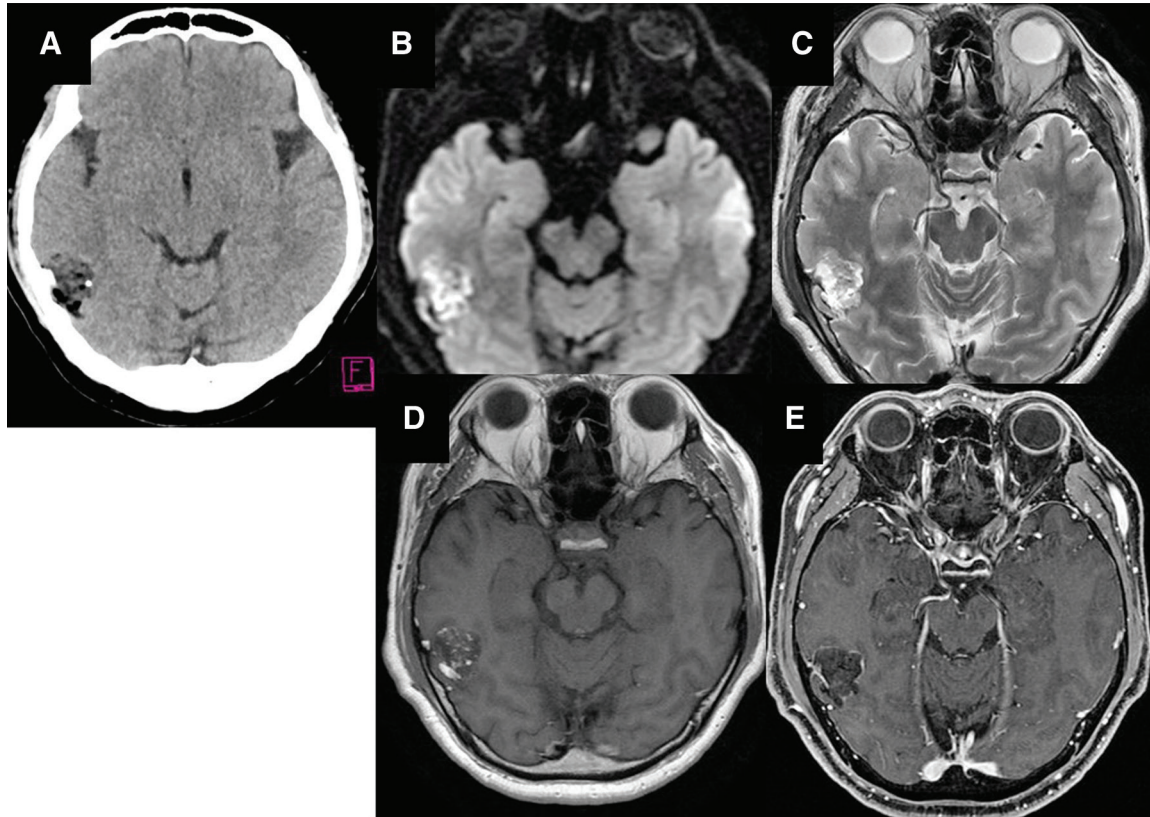


Fig. 1 Preoperative neuroradiological examination. (A) Plain CT showing an iso-to-hypo-dense tumor in the right temporal lobe including a small calcified area. Preoperative MRI showing the tumor in the right temporal lobe. (B and C) The tumor appears as a heterogeneous hyperintense lesion on diffusion-weighted imaging (DWI) (B) and T2-weighted imaging (T2) (C). (D) T1-weighted imaging (T1) showing a hypointense lesion with a hyperintense spotty area. (E) Partial enhancement of the capsule seen on T1 after gadolinium administration (T1Gd+).

gadolinium contrast (Fig. 1E). Based on these imaging findings, the differential diagnosis included epidermoid cyst, neurenteric cyst, teratoma, and infectious disease, in addition to dermoid cyst.

However, because a cyst usually does not take up contrast, and thus is not enhanced by gadolinium, it was difficult to obtain a histopathological diagnosis preoperatively. We decided to perform surgery for the improvement of the symptom, accurate diagnosis, and radical cure.

Surgery and histopathological findings

Gross total resection of the tumor was achieved through a right temporal craniotomy (Fig. 2). There was no adhesion between the dura and the tumor. The tumor was covered with a thick capsule, and the lumen was filled with keratinized material including hair and calcified components. After internal decompression, the capsule was peeled off from the surrounding brain tissue, but the adhesion was severe, and so subpial dissection was performed. Care was taken not to disseminate the tumor contents into the surrounding brain tissue to prevent chemical meningitis.

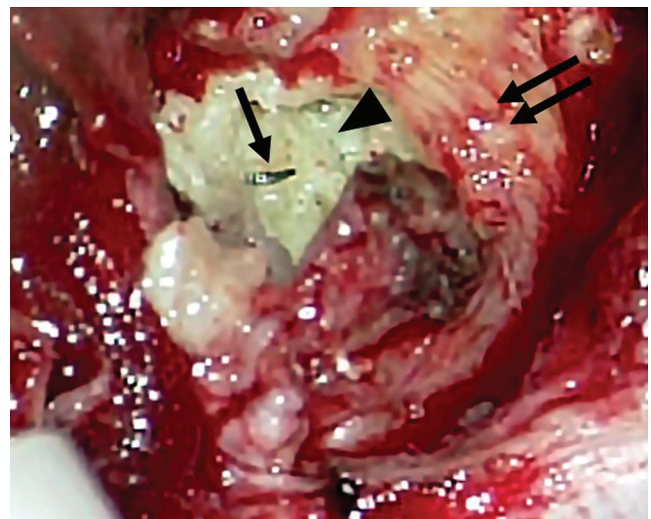


Fig. 2 Operative view during right temporal craniotomy. The lumen is filled with keratinized material (arrow-head) including hair (arrow). The capsule (double arrow) is thick, hard, and elastic.

The cyst wall was composed of squamous epithelium with connective tissue containing small vascular aggregates (Fig. 3A and 3B) and sebaceous glands (Fig. 3A). The cyst lumen was filled with keratin flakes (Fig. 3A). Inflammation was confirmed in the parts of the tumor wall with lymphocyte infiltration, deposition of cholesterol crystals (Fig. 3B), and deposition of hemosiderin (Fig. 3C). Based on these findings, we made a diagnosis of dermoid cyst. There was evidence of peritumoral spread of inflammation.

Postoperative course

The postoperative course was uneventful and the patient was discharged 7 days after surgery with no neurological deficit. Postoperative MRI (Fig. 4A–C) confirmed complete resection of the tumor. The patient did not complain of a headache after discharge.

Discussion

Dermoid cysts are regarded as dysembryogenic tumors arising from ectodermal inclusions of primitive pluripotent cells in the neural tube between the third and the fifth weeks of intrauterine life.^{1,2)} Intracranial dermoid cysts are rare, accounting for 0.1% of brain tumors in Japan.⁵⁾ These often occur at intracranial midline sites, typically around the brain stem and the cerebellar vermis.^{3,4)} The dermoid cysts that occur in the temporal lobe are extremely rare. A slightly high male preponderance has been noted. The prevalent age is wide ranging, but slightly higher in children under 10 years of age and adults in their 30s and 50s.^{5,6)} This case is extremely rare because it was not in the midline but in the temporal lobe. However, the patient was 50 years old and consistent with the predominant age group. To the best of our knowledge, four cases^{7–10)} have been

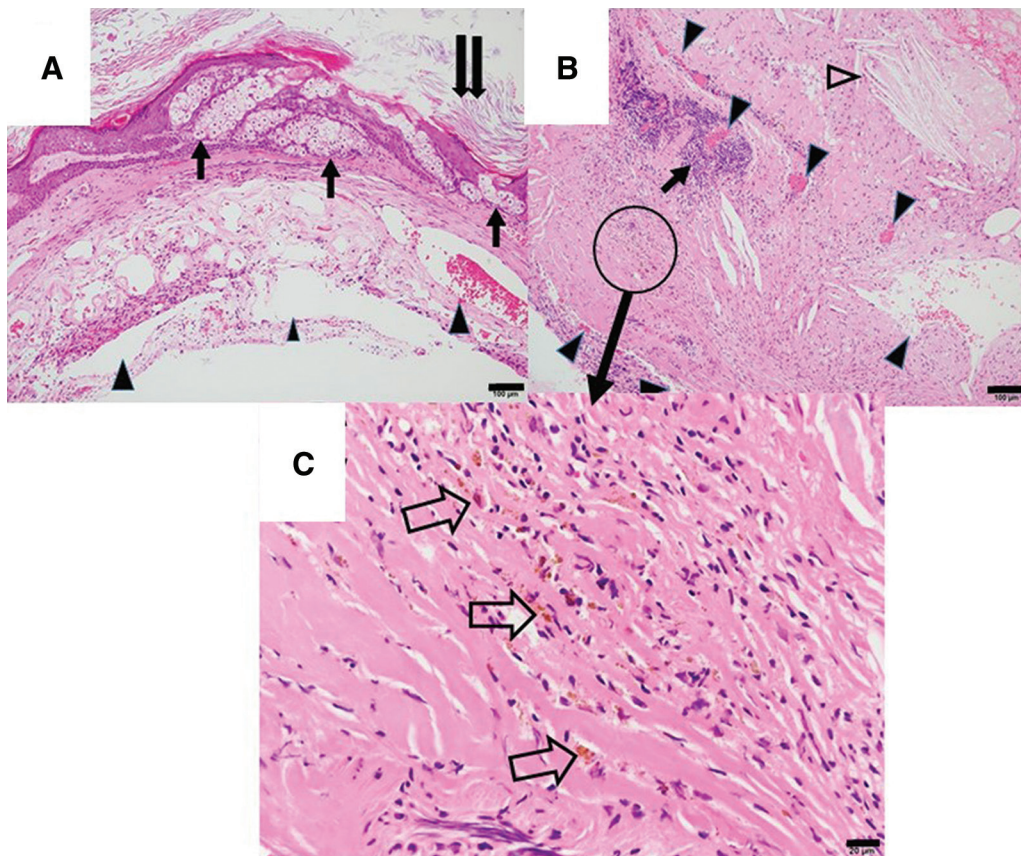


Fig. 3 Hematoxylin-eosin staining (A and B: bar: 100 μ m, C: bar: 20 μ m). (A) The cyst wall is lined with stratified squamous epithelium. The outer layer of the wall is accompanied by connective tissue with vascular aggregates (arrowheads) and sebaceous glands (arrows). The cyst lumen is filled with keratinized material (double arrow). (B) Inflammation is confirmed on parts of the tumor wall with lymphocyte infiltration (arrow), aggregation of small vessels (arrow heads), and cholesterol crystal deposition (white arrowhead). (C) Deposition of hemosiderin is seen (white arrows).

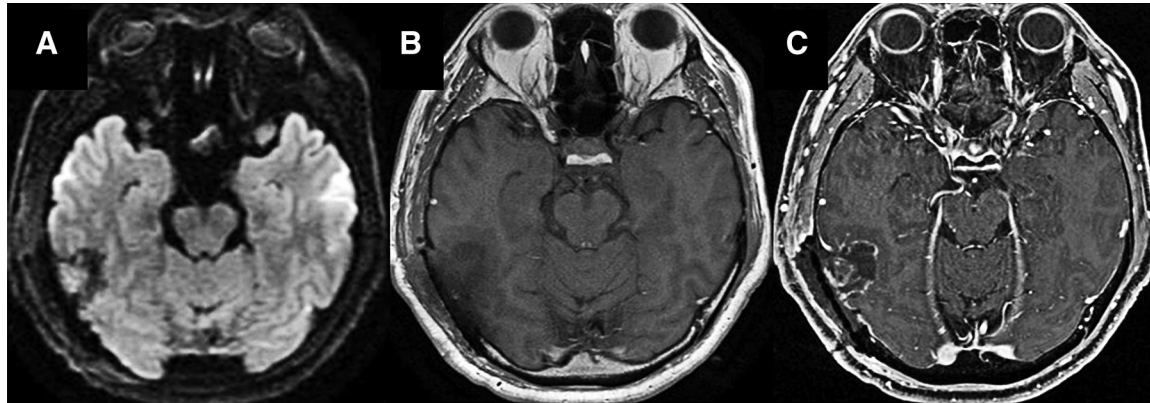


Fig. 4 Postoperative MRI showing total resection of the tumor. (A) DWI. (B) T1. (C) T1Gd(+).

Table 1 Dermoid cyst developing in the temporal lobe

Authors	Age/sex	Size (mm)	Symptom	Therapy	Result
Sugano et al. ¹⁰⁾	15/M	30 × 20 × 40	Seizure, memory disturbance	TR	Good
Velho et al. ⁸⁾	35/F	ND	Seizure, headache	TR	Good
Abderrahmen et al. ⁷⁾	48/F	55 × 40 × 52	Visual disturbance, headache	TR	Good
Mucanj et al. ⁹⁾	52/M	47 × 34 × 30	Seizure, headache	ND	ND
Present case	50/M	30 × 28 × 30	Headache	TR	Good

ND: not described, TR: total resection.

reported about dermoid cyst in temporal lobe. All those cases including our case are summarized in Table 1. The average age of onset is 40 years, younger than the age of all dermoid cyst. In the symptom of all dermoid cyst, headache and seizure occurred in 32.6% and 26.5%,¹¹⁾ respectively. In the cases of temporal lobe, all cases are symptomatic, especially headache occurred in 80% and seizure in 60%. It was indicated that temporal lobe dermoid cysts are prone to symptomatic. The 80% of the cases showed good results due to total resection.

There are many reports on diagnostic imaging of dermoid cysts. Findings vary depending on the tumor content.^{11–13)} Although often visualized as a low-density region on CT, in some cases high-density regions are mixed.^{11,12)} The low-density region is reported to be due to fat and hair components, and the high-density regions are due to calcification, hemorrhagic changes, presence of protein-rich substances and liquefied cholesterol, and calcium deposition due to saponification of fat and keratin components.^{14–16)} The low- and high-density regions appear mixed as a result of the mixture of these contents. MRI shows high signal intensity on T1 and mixed signal intensity on T2 and FLAIR. DWI often reveals high signal intensity and indicates

the presence of fat components.^{11–13)} The heterogeneity of MRI findings is attributed to variable secretions and desquamations of the cyst, that is, the ratio of cholesterol and keratin in the tumor contents.^{11–13)} In this case, high signal intensity area was seen mixed with a low signal intensity area on T1 weighting. We speculated that the tumor component exhibited low signal intensity and the liquid part showed high signal intensity based on the surgical findings. In addition, the tumor capsule showed marked enhancement after gadolinium administration. Regarding the surgical findings, we surmised that the severe adhesion of the tumor to the surrounding brain tissue was due to the spread of inflammation confirmed by the histopathological findings of inflammation and aggregation of blood vessels around the tumor. Contrast enhancement is rare in dermoid cyst,^{12,17)} but has been reported previously.^{18,19)} Sanchez-Mejia et al. noted significant enhancement in their case with intratumoral hemorrhage.¹⁹⁾ This may be related to the hyper-vascularity revealed by gadolinium enhancement. In addition, they described interwoven vascular structures in the surrounding arachnoid space. In our case, vascular aggregation may possibly account for contrast enhancement with gadolinium.

Furthermore, the spread of inflammation around the brain tissue may have also contributed to the contrast enhancement based on our histopathological findings in the tumor wall. We speculate that gadolinium contrast enhancement could provide preoperative information regarding the spread of inflammation to surrounding tissues, and may be effective for surgical planning.

Our case was symptomatic, and the headache resolved after tumor resection. Although the tumor was small, it was presumed to be due to the spread of inflammation to the surrounding brain. This demonstrated that surgical treatment can be effective in symptomatic cases. However, surgical intervention is controversial^{20–22)} in asymptomatic cases. Spontaneous rupture of dermoid cysts may occur, leading to chemical meningitis and hydrocephalus, and surgical treatment should therefore be considered in cases of large tumors.²³⁾ For tumors with lipid-rich content²³⁾ that are located in the supratentorial region,²⁴⁾ rupture may occur, and so it is necessary to consider surgical treatment or ensure close observation. In our case, preoperative diagnosis was difficult based on imaging alone. We considered that accurate histopathological diagnosis was necessary and decided to perform radical curative surgery. It is necessary to aim for total resection to prevent recurrence, but as in this case, some areas with severe adhesions to the surrounding brain tissue had to be dissected under the subpial layer in order to remove the tumor completely. However, in cases where the tumor is located in the cerebellopontine angle, or suprasellar region, complete resection may be difficult because of severe adhesion between the tumor and the cranial nerves, brain stem and vessels. Therefore, excessive resection should be avoided.^{24,25)} Strict postoperative follow-up is required in such cases. In addition, to prevent postoperative chemical meningitis, care should be taken to avoid the dissemination of cyst contents during surgical manipulation.

Conclusions

We have presented a surgical case of dermoid cyst in the temporal lobe. Gadolinium contrast examination may be useful for confirming the spread of inflammation to surrounding brain tissue. At surgery, care should be taken to avoid dissemination to the surrounding brain tissue to prevent chemical meningitis.

Conflicts of Interest Disclosure

The authors have no conflict and interest.

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