



Case Report

# Resection of a lateral supratentorial endodermal cyst complicated by postoperative seizures: A case report

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## ABSTRACT

**Background:** Endodermal cysts are uncommon cystic lesions usually located at the ventral aspects of the spine. A lateral supratentorial location of such cysts is extremely rare. A unique case of a lateral supratentorial endodermal cyst that required surgical intervention due to uncus herniation, complicated with postoperative seizures, is presented.

**Case Description:** A 48-year-old man presented with transient motor aphasia and diplopia. Magnetic resonance imaging showed a cystic lesion occupying the left frontal and temporal convexity with midline shift and uncus herniation. Cyst resection was performed, and cyst contents with mucous-like components were aspirated. Histopathological examination showed an endodermal cyst. The patient showed no neurological deficits immediately after surgery but developed tonic-clonic seizures 9 h after surgery. Sedation and intubation were required to control the seizures. After administering multiple antiepileptic drugs, he was extubated on the 5<sup>th</sup> day after surgery. He was discharged home in a month with mild impairment in dexterity of his right hand.

**Conclusion:** Surgical intervention for endodermal cysts can be complicated by postoperative seizures caused by chemical irritation of brain cortex due to spillage of cyst contents. It is important to irrigate the cyst wall very well intraoperatively and pay attention not to spill the cyst fluid to unaffected locations. Preoperative administration of antiepileptic drugs should also be considered if endodermal cysts, not simple arachnoid cysts, are suspected preoperatively.

**Keywords:** Endodermal cyst, Neurenteric cyst, Postoperative seizure, Status epilepticus

## INTRODUCTION

Endodermal cysts are rare, benign, cystic lesions that commonly arise at the ventral aspects of the spine. There have been reports of intracranial lesions, but they tend to be localized in the midline in the infratentorial compartment, and lateral supratentorial endodermal cysts are even rarer. Total or subtotal cyst resection leads to satisfactory postsurgical outcomes, but perioperative complications such as seizures sometimes occur. A unique case of a surgically treated lateral supratentorial endodermal cyst followed by postoperative status epilepticus is presented.

## CASE PRESENTATION

### Patient history

The patient was a 48-year-old man with no significant medical history who presented with transient motor aphasia. On neurological examination, diplopia was noted on the right lateral gaze, which suggested partial left oculomotor nerve palsy, with no dysfunction of pupillary reflexes. Head magnetic resonance imaging (MRI) showed a large supratentorial cystic lesion that occupied the left temporal and frontal convexity, with a 10-mm midline shift and uncal herniation. The cyst contents showed low intensity on T1-weighted images [Figure 1a] and high intensity on T2-weighted images [Figure 1b], with no diffusion restriction. The cyst fluid showed a cerebrospinal fluid (CSF)-like pattern in most sequences, other than in fluid-attenuated inversion recovery images, where the content showed slightly higher intensity than regular CSF [Figure 1c]. There was a mucous-like nodule with isointensity on T1-weighted images and mild high intensity on T2-weighted images. The lesion was not enhanced the following gadolinium injection.

It was concluded that the mass effect from the cyst caused uncal herniation [Figure 1d], resulting in partial left oculomotor palsy, as well as seizures manifesting as transient aphasia, and thus, it was decided to perform cyst resection.

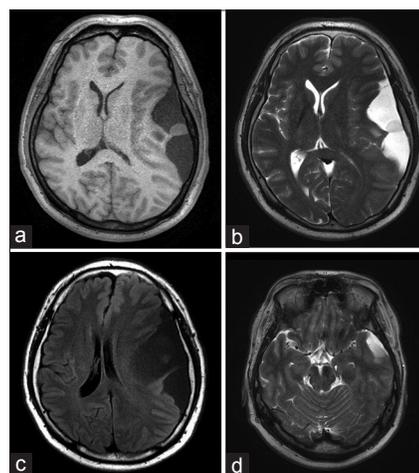
### Surgery

Under general anesthesia, a left frontotemporal craniotomy was performed. As the dura mater was opened, a large extra-axial cyst was seen, covered with a thin whitish membrane and containing white turbid fluid [Figure 2]. After the cyst wall was opened, cyst contents with mucous-like components were aspirated. Most of the cyst membranes were resected, but some part of the membrane that adhered to the brain surface strongly was left. The cyst wall was sent for histopathological examination, which showed that the lesion was endodermal in origin. No obvious intraoperative complications were noted. The patient awoke from the anesthesia promptly, was extubated in the operating room, and was then sent to an intensive care unit.

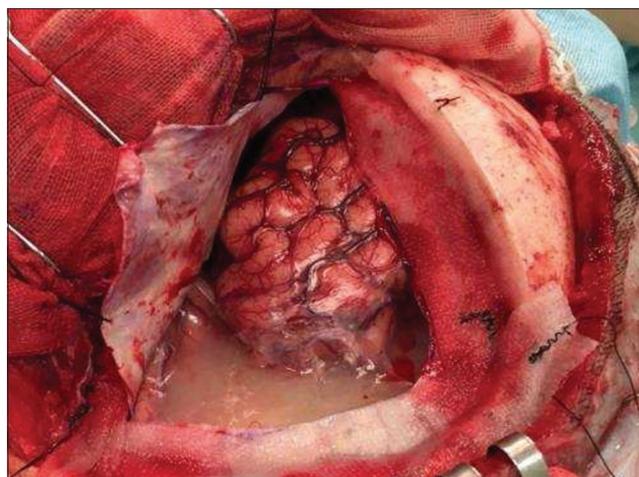
### Postoperative course

The patient did not demonstrate any neurological deficits immediately after surgery. However, he experienced a tonic-clonic seizure 9 h after surgery. Diazepam and levetiracetam (LEV) were immediately administered, but frequent complex partial seizures still occurred. Fosphenytoin (f-PHT) was also administered, and he was intubated and sedated with propofol. Lumbar puncture was performed on postoperative day 1, and the obtained CSF was whitish and opaque, with the initial pressure as high as 330 mmH<sub>2</sub>O. The cell count was

11/uL, glucose was 68 mg/dL, and protein was 46.6 mg/dL. The electroencephalogram showed theta bursts localized in bilateral frontal lobes. Status epilepticus caused by chemical meningitis was diagnosed. The seizure was finally satisfactorily controlled with LEV 4000 mg, f-PHT 7.5 mg/kg, lacosamide 200 mg, and propofol 0.3 mg/kg/h, with minimal simple partial seizures affecting the right hand. Propofol was stopped on the 5<sup>th</sup> day, and the patient was extubated. Postoperative course shown above is organized as a timeline [Figure 3]. Mild right hemiplegia and motor aphasia persisted, which gradually improved with rehabilitation. On postoperative MRI performed 2 weeks after the surgery, the



**Figure 1:** Preoperative magnetic resonance imaging showing T1-weighted (a), T2-weighted (b), and fluid-attenuated inversion recovery (c) images. The cyst content demonstrates slightly higher intensity than regular cerebrospinal fluid on fluid-attenuated inversion recovery images, containing a mucous-like nodule with isointensity on T1WI and mild high intensity on T2WI. Uncal herniation is observed at the level of the mesencephalon (d).

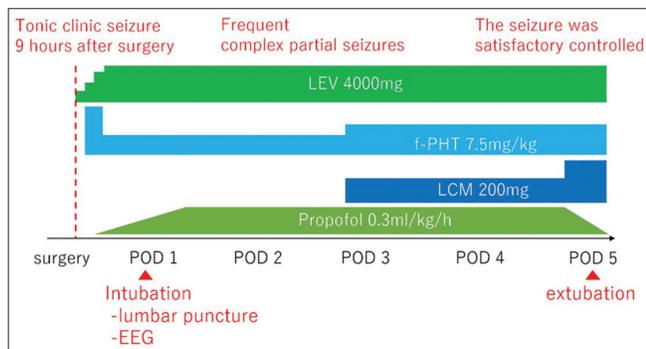


**Figure 2:** Intraoperative presentation of the cyst. The cyst is covered by a thin whitish membrane and contains white turbid fluid.

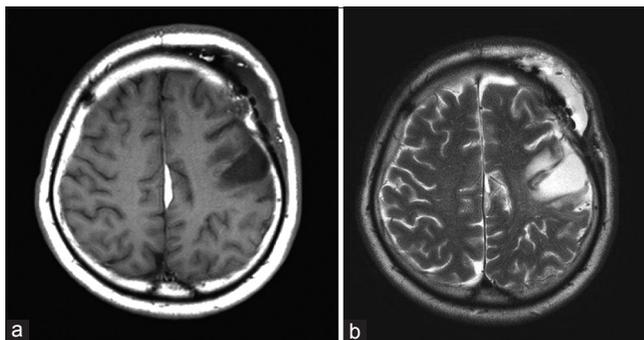
cyst shrank dramatically with satisfactory decompression [Figure 4]. The patient continued rehabilitation and was discharged home in 1 month. Mild impairment in dexterity of his right hand remained. No recurrence was noted on computed tomography 2 months after the surgery.

### Pathological findings

On histopathological examination, the cyst wall contained pseudostratified ciliated columnar epithelium with a basement membrane on hematoxylin and eosin staining [Figure 5a]. A few goblet cells were also present on staining with periodic acid-Schiff [Figure 5b], mucicarmine, and Alcian blue stains. The presence of secretory granules, basement membrane, and cilia suggested an endodermal origin. Ectodermal markers such as S-100 and GFAP were negative. Epithelial markers such as cytokeratin CAM5.2 and epithelial membrane antigen were positive, which supported the diagnosis of an endodermal cyst. Carcinoembryonic antigen (CEA) was also positive in the cytoplasm in a few epithelial cells [Figure 5c].



**Figure 3:** Postoperative course is organized as a timeline. The patient experienced a tonic-clinic seizure 9 h after surgery. Lumbar puncture and electroencephalogram were performed on postoperative day 1. The seizure was finally controlled with levetiracetam 4000 mg, Fosphenytoin 7.5 mg/kg, Lacosamide 200 mg, and propofol 0.3 mg/kg/h.

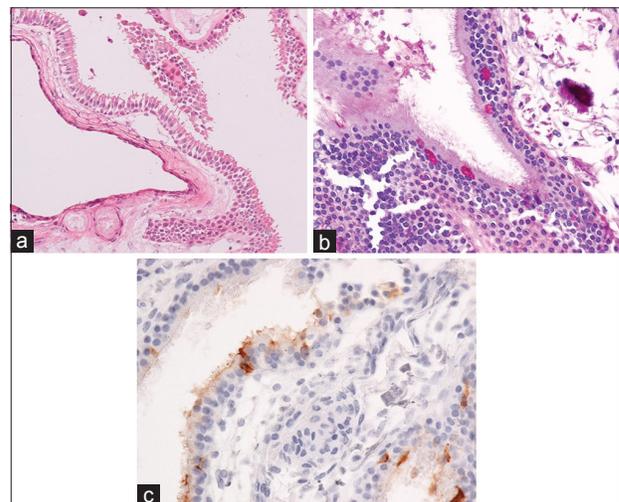


**Figure 4:** (a and b) Postoperative magnetic resonance imaging shows cyst shrinkage and disappearance of the mass effect.

### DISCUSSION

Endodermal cysts often occur at the ventral aspects of the spine, especially at the lower cervical and upper thoracic levels. Intracranial lesions are rare, and they have usually been reported in the infratentorial compartments such as the posterior fossa or craniocervical junction, especially in the midline. A lateral supratentorial location is extremely rare for endodermal cysts.

The pathogenesis of endodermal cysts is not yet fully understood. There have been three major hypotheses regarding the origins of endodermal cysts. The predominantly supported hypothesis is that they originate from the neurenteric canal,<sup>[4]</sup> an embryonic structure that forms a temporary link connecting the yolk sac and the amniotic cavity in the 3<sup>rd</sup> week of development. It temporarily connects neuroectoderm and endoderm, which forms the foregut and respiratory buds. The canal normally disappears with the completion of a notochord, but it can transform into an endodermal cyst when it fails to dissolve. The midline location of the neurenteric canal may account for endodermal cysts' midline preference, but lateral supratentorial lesions cannot be explained by this theory. Furthermore, invagination of endodermal cells terminates at the level of the clivus in the process of gastrulation, which cannot account for the supratentorial location of these cysts.<sup>[8]</sup> The second hypothesis is that endodermal cysts originate from a remnant Seesel's pouch, a transient diverticulum formed at the oropharyngeal membrane in the 3<sup>rd</sup> week of development. The pouch is known to be the origin of Rathke's cleft cysts and colloid cysts, which share immunohistochemical characteristics



**Figure 5:** Histopathological features of the cyst wall. Hematoxylin and eosin staining shows pseudostratified ciliated columnar epithelium with a basement membrane (a). The presence of goblet cells is shown by periodic acid-Schiff staining (b). Carcinoembryonic antigen is also positive in the cytoplasm in a few epithelial cells, suggesting carcinoembryonic antigen secretory capacity (c).

with endodermal cysts.<sup>[7]</sup> This theory still cannot explain the lateral location of lesions. The third hypothesis is that the cyst is generated from multipotent endodermal cells that migrate along the neuroectoderm.<sup>[2]</sup> In this theory, the neurenteric canal is only a channel of migration of precursor cyst cells, and this may explain their lateral location in the cranium. It is not fully understood how endodermal cysts develop, and why a few cases are located laterally remains unclear.

Endodermal cysts are often misdiagnosed as arachnoid cysts due to the CSF-like intensity of their contents on MRI. In some cases, the cyst fluid shows a slightly higher intensity than CSF on T1-weighted images, reflecting the protein-rich nature of the cyst contents. Some previous studies showed a nodule with high intensity on T2WI, surrounded by fluid, as in the present case.<sup>[3]</sup> This may be an important preoperative diagnostic key for distinguishing endodermal cysts from the more common arachnoid cysts.

There is no established treatment strategy for lateral supratentorial endodermal cysts due to their rarity. Surgical resection is thought to lead to satisfactory outcomes with a low recurrence rate. Góes *et al.* collected 45 cases of endodermal cysts that were supratentorial in origin.<sup>[5]</sup> Of them, 43 underwent craniotomy and cyst resection, and only two of them showed recurrence. Although seizure is a common preoperative symptom, occurring in 35.5%, only two patients experienced postoperative seizures.<sup>[5,9]</sup> One explanation for these complications is that cyst fluid contains digestive enzymes of gastric cells, and thus, spillage of the cyst contents directly stimulates the adjacent cerebral cortex, provoking postoperative seizures. Akimoto *et al.* reported that the cyst fluid contained a CEA level as high as 1400 ng/ml, exceeding the upper limit of measurement.<sup>[1]</sup> CEA is a glycoprotein known to be elevated in the tissues of some types of malignant tumors such as colorectal and lung cancers, but it is also present in the normal mucosal epithelium of the fetal colorectal system.<sup>[6]</sup> In their report, CEA was positive in epithelial cells, as in the present case, suggesting CEA secretory capacity.

In the present case, intractable seizures continued for 3 days after surgery, probably due to chemical irritation to the brain. To prevent these complications, we suggest that it is important to irrigate the cyst wall very well and pay attention to not spill the whitish cloudy fluid onto unaffected areas. Preoperative administration of antiepileptic drugs can also be considered.

## CONCLUSION

Surgical intervention for endodermal cysts can be complicated by postoperative seizures caused by chemical irritation of brain cortex due to spillage of cyst contents. Careful irrigation of the cyst wall and preoperative administration of antiepileptic drugs should be considered if endodermal cysts are suspected preoperatively.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

## Conflicts of interest

There are no conflicts of interest.

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