



Case Report

Endonasal endoscopic surgery for temporal lobe epilepsy associated with sphenoidal encephalocele

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ABSTRACT

Background: Temporal lobe epilepsy (TLE) associated with temporal lobe encephalocele is rare, and the precise epileptogenic mechanisms and surgical strategies for such cases are still unknown. Although the previous studies have reported good seizure outcomes following chronic subdural electrode recording through invasive craniotomy, only few studies have reported successful epilepsy surgery through endoscopic endonasal lesionectomy.

Case Description: An 18-year-old man developed generalized convulsions at the age of 15 years. Despite treatment with optimal doses of antiepileptic drugs, episodes of speech and reading difficulties were observed 2–3 times per week. Long-term video electroencephalogram (EEG) revealed ictal activities starting from the left anterior temporal region. Magnetic resonance imaging revealed a temporal lobe encephalocele in the left lateral fossa of the sphenoidal sinus (sphenoidal encephalocele). Through the endoscopic endonasal approach, the tip of the encephalocele was exposed. A depth electrode was inserted into the encephalocele, which showed frequent spikes superimposed with high-frequency oscillations (HFOs) suggesting intrinsic epileptogenicity. The encephalocele was resected 8 mm from the tip. Twelve months postoperatively, the patient had no recurrence of seizures on tapering of the medication.

Conclusion: TLE associated with sphenoidal encephalocele could be controlled with endoscopic endonasal lesionectomy, after confirming the high epileptogenicity with analysis of HFOs of intraoperative EEG recorded using an intralesional depth electrode.

Keywords: Depth electrode, Endoscopic endonasal surgery, Epileptogenicity, Sphenoidal encephalocele, Temporal lobe encephalocele

INTRODUCTION

Temporal encephaloceles are rare presentation,^[8,9,13] involving pathological herniation of the temporal lobe through congenital or acquired osseous-dural defects of the skull base or cranial vault.^[13,14] A sphenoidal encephalocele involves either the body or the greater wing of the sphenoid bone.^[14] In a recent study, drug-resistant temporal lobe epilepsy (TLE), in addition to occult or symptomatic cerebrospinal fluid (CSF) fistulas, recurrent meningitis, middle ear

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effusion, and conductive hearing loss, were identified as symptoms of temporal encephalocele.^[12,14] However, the precise epileptogenic mechanism and the surgical strategies to treat temporal encephalocele-related TLE are not fully understood because several patients with rare temporal encephalocele accompanying minute neuroradiological changes tend to be underdiagnosed.^[1,3,12] Herein, we report a case of TLE associated with temporal encephalocele in the lateral fossa of the sphenoid sinus (sphenoidal encephalocele). A lesionectomy was performed through an endoscopic endonasal approach instead of invasive craniotomy, after confirming the high epileptogenicity of the encephalocele with analysis of high-frequency oscillations (HFOs) of intraoperative EEG recorded using a depth electrode in the encephalocele.

CASE DESCRIPTION

An 18-year-old right-handed man was referred to our department for surgical treatment of drug-resistant epilepsy. He had no history of head trauma. He developed generalized seizures at age 15 years. Following treatment with optimal dosage of antiepileptic drugs, including sodium valproate, zonisamide, lacosamide, and clobazam, his generalized seizures were controlled; however, episodes of speech and reading difficulties were observed 2–3 times a week.

The patient was considered normal neurologically. Long-term video electroencephalogram (EEG) demonstrated ictal discharges with low-amplitude, rhythmic, irregular α waves appearing in T3 and T5; these waves demonstrated increasing amplitude, with frequency changing from θ to δ , when the patient experienced a dull sensation that lasted several seconds [Figure 1a].

Magnetic resonance imaging (MRI) revealed a protrusion of the left medial temporal lobe from the middle cranial fossa to the lateral fossa of the sphenoid sinus [Figure 1b]. At that site, a bony defect was also detected on bone-targeted computed tomography (CT) [Figure 1c]. There were no atrophy or signal abnormalities in the left hippocampus. Although ¹⁸F-fluorodeoxyglucose positron emission tomography (FDG-PET) demonstrated decreased FDG accumulation in the left medial temporal area and temporal base area around the lesion [Figure 1d and e], the fusion image of FDG-PET and MRI with fluid-attenuated inversion recovery sequence clearly depicted markedly decreased FDG accumulation in the temporal lobe encephalocele [Figure 1f]. ¹²³I-Iomazenil (IMZ) single-photon emission tomography in the delayed phase demonstrated a decreased IMZ accumulation in the basal area of the left temporal lobe.

Endoscopic endonasal removal of the sphenoidal encephalocele was performed. After opening the sphenoid sinus, posterior ethmoid sinus, and left maxillary sinus from

both the nasal cavities, the posterior wall of the left maxillary sinus was drilled, and the pterygopalatine sac was exposed [Figure 2a]. While flipping the pterygopalatine sac outward, the base of the pterygoid plate and the sphenoid sinus septum connected to it was removed, and the encephalocele divided into two components in the lateral fossa of the left

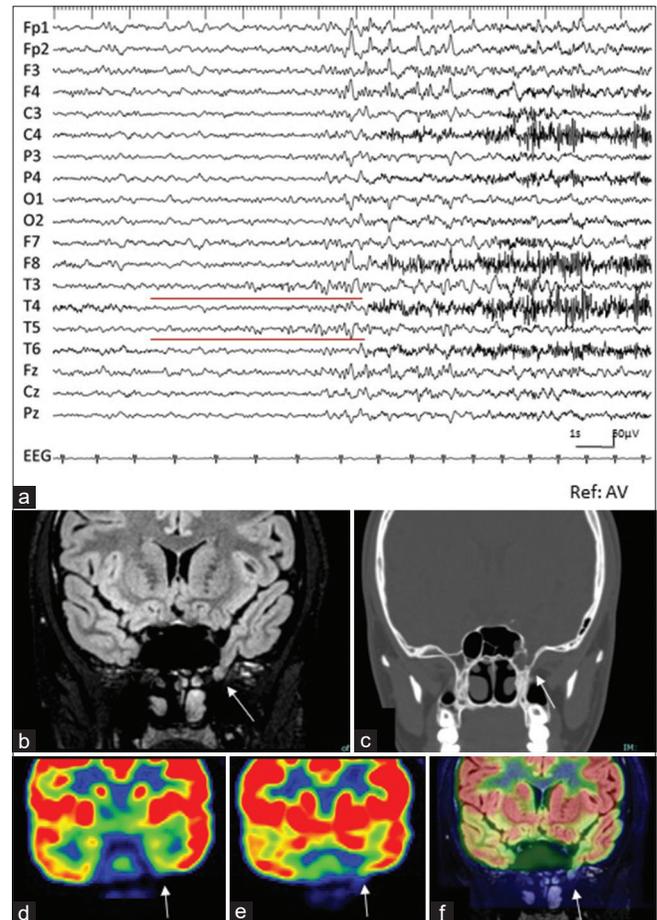


Figure 1: (a) Preoperative electroencephalogram recorded at the start of the habitual seizure (sensitivity: 10 μ V, time constant: 0.1 s, high cut filter: 30 Hz, reference: average). Ictal activities begin with rhythmic alpha activity at T3 and T5 (red line). (b) Coronal view of preoperative magnetic resonance image (MRI) with fluid-attenuated inversion recovery (FLAIR) sequence demonstrates that protrusion of a part of the left temporal lobe is confirmed in the lateral fossa of the sphenoid sinus, indicating sphenoidal encephalocele (Red arrow). (c) Coronal computed tomography (CT) at a comparable level with (b) confirms the bone defect on the left lateral wall of the sphenoid sinus (white arrow). (d and e) Coronal view of ¹⁸F-fluorodeoxyglucose (FDG)-positron emission tomography (PET) image at the hippocampus (d) and a few centimeters behind the sphenoidal encephalocele and at the sphenoidal encephalocele (e). Decreased accumulation of FDG is observed around the left medial and basal temporal area (white arrows). (f) The fusion image of FDG-PET (e) and FLAIR-MRI (b) clearly shows that markedly decreased accumulation of FDG is observed at the tip of the encephalocele (white arrow).

sphenoid sinus was confirmed [Figure 2b]. A depth electrode with three electrode contacts was inserted 1.0 cm into the lateral component of the encephalocele [Figure 2c]. Frequent spikes were recorded [Figure 2d], and the time-frequency analysis revealed superimposing HFOs ranging from 50 Hz to 150 Hz on each spike [Figure 2e]. Piecemeal resection was performed for the encephalocele protruding into the sphenoid sinus [Figure 2f], and subsequent reconstruction was performed with the femoral fascia, fat, nasal septum, and nasal mucosa flap.

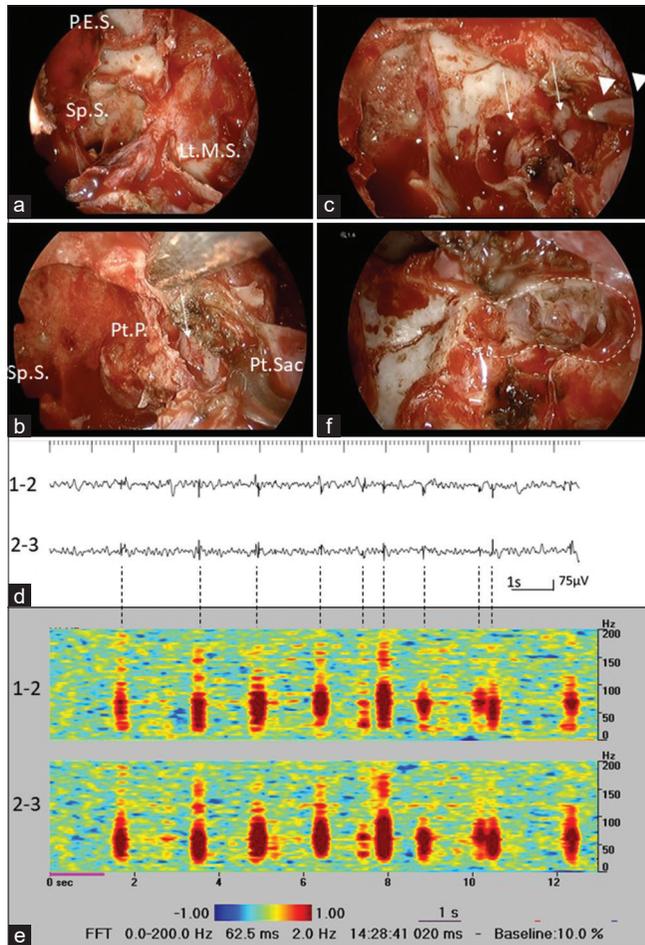


Figure 2: (a) Posterior wall of the left maxillary sinus is drilled, and the pterygopalatine sacs are exposed, after opening the sphenoid sinus (Sp.S.), posterior ethmoid sinus (P.E.S.), and left maxillary sinus (Lt.M.S.) from both nasal cavities. (b) While flipping the pterygopalatine sac (Pt. Sac) outwards, the base of the pterygoid plate (Pt.P.) and the sphenoid sinus septum connected to it is being removed, and encephalocele is confirmed (white arrow). (c) A depth electrode (white arrow heads) is inserted into the encephalocele (white arrow heads). (d) Intraoperative EEG reveals frequent spikes in the encephalocele (bipolar recording, sensitivity: 20 μ V, time constant: 0.1 s, high cut filter: 60 Hz). (e) Time-frequency analysis reveals high-frequency oscillations ranging from 50 Hz to 150 Hz, superimposing to the spikes. (f) The tip of encephalocele is successfully resected (in the white dotted line).

The patient's postoperative course was uneventful, without leakage of CSF. Postoperative MRI confirmed that the sphenoidal encephalocele was resected 8 mm from the tip [Figure 3a]. On postoperative CT, successful repair of the temporal base with nasal septum cartilage was confirmed [Figure 3b]. Histopathological examination of the resected specimen showed cerebral cortical tissue with several reactive astrocytes [Figure 3c]. Twelve months postoperatively, the patient had no seizures, with tapering of medication.

DISCUSSION

Epileptiform discharges around the cortical area of the encephalocele have been previously recorded with intraoperative or chronic electrocorticography using subdural grid electrodes inserted through invasive craniotomy.^[3,12] While this method can depict the epileptogenic lesion over a wide area and its relationship with medial temporal structures, such as the hippocampus, subdural electrodes cannot establish proper contact with the protruded encephalocele itself. Therefore, it remains unknown whether it is better to perform a lesionectomy alone or additional temporal lobe resection including resection of the amygdalo-hippocampus.^[3,6,12]

Due to the recent development in endoscopy, there are increasing surgical reports of temporal encephalocele resection using the endoscopic endonasal approach.^[2,5] However, only three cases of endoscopic endonasal surgery for epileptic temporal lobe encephaloceles have been

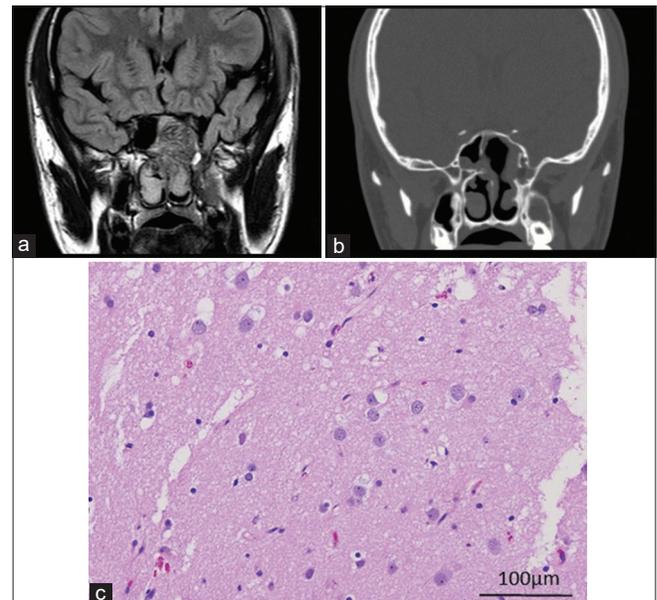


Figure 3: (a) Postoperative MRI confirms that the sphenoidal encephalocele was resected 8 mm from the tip. (b) Postoperative CT shows successful repair of the temporal base with nasal septum cartilage. (c) Histopathological examination shows cerebral cortical tissue with several reactive astrocytes.

reported as per a meeting proceeding.^[11] The most likely reason for this paucity of information is that the epileptogenic mechanism could not be evaluated through this approach. HFOs are a form of brain activity observed on EEG in the frequency range of 80–500 Hz. HFOs can be classified into ripples (80–250 Hz) and fast ripples (250–500 Hz). Recent studies have demonstrated that both ripples and fast ripples can be regarded as new biomarkers of epileptogenesis and ictogenesis.^[4,15] We previously reported on analysis of HFOs of intraoperative EEG recordings from an intralesional depth electrode inserted into the cerebellar ganglioglioma in an infant; the HFOs appeared as ripples, riding on the periodic discharges, and this finding provided supportive evidence for the intrinsic epileptogenicity of cerebellar tumors.^[10] In this case, we used this electrophysiological method through endoscopic endonasal surgery and demonstrated that each spike had superimposing ripples. This finding provides supportive evidence of the intrinsic epileptogenicity of the temporal encephalocele. Moreover, the fusion image of FDG-PET and MRI clearly showed a hypometabolic area at the tip of the encephalocele, while the FDG-PET image alone failed to reveal this finding, probably because of the extracranial localization of the small encephalocele tip. These findings were consistent with our previous observation; in other words, the fusion image of FDG-PET and MRI clearly demonstrated the relationship between epileptogenic lesions and abnormal metabolic areas.^[7]

The drawback of the endoscopic approach was that *en bloc* resection of the encephalocele could not be performed. Because of the small specimens obtained with piecemeal resection in the present case, it was difficult to show the structural abnormality of the cortical lamination. Neurons appeared largely normal, but several reactive astrocytes were observed. Panov *et al.*^[12] previously reported the pathological findings of encephaloceles in six patients the specimens obtained through craniotomy showed mild-to-moderate gliosis.

CONCLUSION

Although our observations reported herein were derived from a single case, temporal lobe encephalocele-related TLE could be controlled with endoscopic endonasal lesionectomy. This involves detecting the high epileptogenicity of the encephalocele by analysis of HFOs of the intraoperative EEG recorded with intralesional depth electrode.

Statement of ethics

This research was conducted ethically in accordance with the World Medical Association Declaration of Helsinki. Written informed consent was obtained from the patient's parents for publication of this case report and any accompanying images.

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Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Abou-Hamden A, Lau M, Fabinyi G, Berkovic S, Jackson GD, Mitchell LA, *et al.* Small temporal pole encephaloceles: A treatable cause of “lesion negative” temporal lobe epilepsy. *Epilepsia* 2010;51:2199-202.
2. Bozkurt G, Turri-Zanoni M, Coden E, Russo F, Elhassan HA, Gallo S, *et al.* Endoscopic endonasal transpterygoid approach to sphenoid sinus lateral recess defects. *J Neurol Surg B Skull Base* 2020;81:553-61.
3. Byrne RW, Smith AP, Roh D, Kanner A. Occult middle fossa encephaloceles in patients with temporal lobe epilepsy. *World Neurosurg* 2010;73:541-6.
4. Cho JR, Joo EY, Koo DL, Hong SC, Hong SB. Clinical utility of interictal high-frequency oscillations recorded with subdural macroelectrodes in partial epilepsy. *J Clin Neurol* 2012;8:22-34.
5. El-Tarabishi MN, Fawaz SA, Sabri SM, El-Sharnobi MM, Sweed A. A modification of endoscopic endonasal approach for management of encephaloceles in sphenoid sinus lateral recess. *Eur Arch Otorhinolaryngol* 2016;273:4305-14.
6. Faulkner HJ, Sandeman DR, Love S, Likeman M, Nunez DA, Lhatoo SD. Epilepsy surgery for refractory epilepsy due to encephalocele: A case report and review of the literature. *Epileptic Disord* 2010;12:160-6.
7. Kikuchi K, Togao O, Yamashita K, Momosaka D, Nakayama T, Kitamura Y, *et al.* Diagnostic accuracy for the epileptogenic zone detection in focal epilepsy could be higher in FDG-PET/MRI than in FDG-PET/CT. *Eur Radiol* 2021;31:2915-22.
8. Lai SY, Kennedy DW, Bolger WE. Sphenoid encephaloceles: Disease management and identification of lesions within the lateral recess of the sphenoid sinus. *Laryngoscope* 2002;112:1800-5.
9. Landreneau FE, Mickey B, Coimbra C. Surgical treatment of cerebrospinal fluid fistulae involving lateral extension of the sphenoid sinus. *Neurosurgery* 1998;42:1101-5.
10. Mukae N, Morioka T, Torio M, Sakai Y, Shimogawa T, Sakata A, *et al.* Periodic discharges with high frequency oscillations recorded from a cerebellar gangliocytoma in an epileptic infant. *Surg Neurol Int* 2021;12:98.

11. Olasunkanmi A, Fisher T, Shin H, Zanation A, Sasaki-Adams D. Endoscopic endonasal approach for resection of middle fossa encephalocele for treatment of refractory temporal lobe epilepsy. *J Neurol Surg B Skull Base* 2016;77:P046.
12. Panov F, Li Y, Chang EF, Knowlton R, Cornes SB. Epilepsy with temporal encephalocele: Characteristics of electrocorticography and surgical outcome. *Epilepsia* 2016;57:e33-8.
13. Wilkins RH, Radtke RA, Burger PC. Spontaneous temporal encephalocele. Case report. *J Neurosurg* 1993;78:492-8.
14. Wind JJ, Caputy AJ, Roberti F. Spontaneous encephaloceles of the temporal lobe. *Neurosurg Focus* 2008;25:E11.
15. Worrell GA, Parish L, Cranstoun SD, Jonas R, Baltuch G, Litt B. High-frequency oscillations and seizure generation in neocortical epilepsy. *Brain* 2004;127:1496-506.

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