Nonconvulsive status epilepticus due to pneumocephalus after suprasellar arachnoid cyst fenestration with transsphenoidal surgery: illustrative case

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BACKGROUND Nonconvulsive status epilepticus (NCSE) requires prompt diagnosis and treatment, particularly after neurosurgical procedures for cerebral damage. Here, the authors reported an extremely rare case of suprasellar arachnoid cyst presenting with NCSE after cyst fenestration with transsphenoidal surgery, which was associated with pneumocephalus.

OBSERVATIONS A 61-year-old man presented with visual impairment and was diagnosed with a suprasellar arachnoid cyst on magnetic resonance imaging (MRI). The patient received cyst fenestration with endonasal transsphenoidal surgery. His visual symptoms improved immediately after the operation; however, on postoperative day 3, semicoma appeared and was prolonged. The patient was diagnosed with NCSE due to pneumocephalus based on MRI and electroencephalography (EEG) findings. The administration of antiepileptic drugs (AEDs) improved his clinical symptoms and the abnormal findings on MRI and EEG.

LESSONS This is the first case of NCSE with pneumocephalus after transsphenoidal surgery for a suprasellar arachnoid cyst. Pneumocephalus due to cerebrospinal fluid leakage can cause NCSE. Arterial spin labeling perfusion imaging and diffusion-weighted imaging are as useful for differentially diagnosing NCSE as EEG and AED tests.

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KEYWORDS nonconvulsive status epilepticus; postoperative pneumocephalus; endoscopic endonasal transsphenoidal surgery; diffusion-weighted imaging; arterial spin labeling; suprasellar arachnoid cyst

Postoperative nonconvulsive status epilepticus (NCSE) has been well documented to occur in 2.8%–15% of patients undergoing neurosurgical procedures, such as tumor resections, clippings, and surgeries for traumatic diseases.1–4 Most cases have cerebral parenchymal damage directly incurred by surgical morbidity or the primary lesion.5,6 However, the incidence of NCSE without direct neuronal damage has not been clarified yet. Particularly, NCSE as a complication of transsphenoidal surgery has not been reported. Only a few cases with seizure episodes have been referred after transsphenoidal surgery for intrasellar lesions.7 Here, we report the first case of prolonged severe unconsciousness after pneumocephalus due to endoscopic endonasal transsphenoidal surgery (eTSS) and discuss a potential etiology of NCSE.

Illustrative Case
A 61-year-old man was admitted to an outpatient ophthalmological clinic because of visual impairment for 3 months. The patient has well-controlled hypertension and dyslipidemia and no previous seizure episodes. Neurological examinations showed bilateral low visual acuity (20/40) and bitemporal hemianopsia. Endocrinological examination showed mild hypothyroidism and hypogonadism. Brain
magnetic resonance imaging (MRI) demonstrated a suprasellar cystic lesion compressing the optic chiasm (Fig. 1). From these findings, the patient was diagnosed with a symptomatic suprasellar arachnoid cyst.

Operative Procedure
The patient received cyst fenestration using an endonasal transphenoidal approach. The sellar floor was sufficiently fenestrated, and the dura mater was incised to expose the cyst wall. The cyst contents were colorless and transparent. The cyst wall was partially resected and communicated with the subarachnoid space above the saddle.

After cyst fenestration, the cyst shrank. The sellar floor was reconstructed in a multilayered fashion using dural closure and a nasal mucosa to prevent postoperative cerebrospinal fluid (CSF) leakage.

Postoperative Course
The patient’s bitemporal hemianopsia improved after surgery. On postoperative day 1, computed tomography revealed cyst shrinkage and chiasmal decompression (Fig. 2). Pneumocephalus was found in the bilateral frontal lobe, associated with intraoperative CSF leakage. The postoperative course was uneventful. However, on postoperative day 3, the patient experienced acute consciousness disturbance, with a Glasgow Coma Scale score of 6 (E1V1M4). Emergency MRI showed no hemorrhage or infarction, and slightly high signals were observed in the bilateral frontal cortex on diffusion-weighted imaging (DWI) and arterial spin labeling perfusion imaging (ASL-PI) (Fig. 3). Because the patient had been in a semicoma for 48 hours, electroencephalography (EEG) was performed to identify the subclinical seizure state, appearing generalized delta–theta activity without spike or sharp waves (Fig. 4A). We suspected NCSE and administered 5 mg of diazepam intravenously. His consciousness rapidly improved, and he could obey simple verbal commands after that. We diagnosed NCSE using the modified Salzburg Consensus Criteria. Levetiracetam was initiated at a dose of 1,000 mg/day. Because his consciousness did not fully recover after levetiracetam administration, we began clonazepam at a dose of 1 mg/day as an add-on therapy. His consciousness gradually recovered, and on postoperative day 15, the patient became alert. Brain MRI showed improvement of the abnormal hyperperfusion lesion in the bilateral frontal cortex on ASL-PI (Fig. 5). EEG appeared normal to a posterior dominant alpha activity without any epileptiform discharges (Fig. 4B). The patient was subsequently discharged without any neurological deficits or seizure recurrence.

Discussion
Observations
Here, we report the first case of postoperative NCSE during the subacute period after eTSS for a suprasellar arachnoid cyst, which was diagnosed using EEG, antiepileptic drug (AED) tests, and MRI.

Although EEG and AED tests are generally used to diagnose NCSE, MRI sequences, such as ASL-PI and DWI, are also helpful in diagnosing NCSE and confirming the effectiveness of treatment. In the patient in this case report, the findings on ASL-PI and DWI changed during the period in which the patient was unconscious.

FIG. 1. Preoperative axial (A), coronal (B), and sagittal (C) T2-weighted MRI demonstrating a suprasellar cystic lesion compressing the optic chiasm (arrows).

FIG. 2. Postoperative axial (A), coronal (B), and sagittal (C) computed tomography showing arachnoid cyst shrinkage and the dominance of pneumocephalus in the bilateral frontal convexity.
and improved as the patient recovered. Recent studies have reported the sensitivity of ASL-PI and DWI in diagnosing NCSE. Specifically, the findings of ASL-PI hyperperfusion with cortical hyperintensity on DWI indicate the epileptogenic lesion and the related activated cortex. These changes suggest that ictal hyperperfusion is associated with increased glucose and oxygen demand. Subsequently, cytotoxic edema in the epileptic cortical neurons, which results from the disturbance of metabolism and circulation due to prolonged epileptic activity, is secondarily detected on DWI. These MRI findings in patients with acute status epilepticus are transient; their complete disappearance on both ASL-PI and DWI was confirmed several days after the start of treatment. These findings are useful for differentially diagnosing NCSE.

Regarding the pathogenesis of postoperative NCSE without cerebral damage, we hypothesize that pneumocephalus due to CSF leakage can be implicated in the subduction of the brain, which leads to mechanical stretching of the cerebral surface and cortical vein. Therefore, regional circulation changes and functional failure of the brain may cause NCSE. NCSE occurring because of intracranial hypotension as a consequence of spinal surgery has been reported, and up-mentioned mechanisms, including traction on cortical structures, an increase in the cerebral blood flow, and cortical irritation secondary to subdural hygromas, were suggested.

Therefore, preventing pneumocephalus due to CSF leakage during surgery is necessary. In the patient in this case report, we performed complete closure of the sellar floor using multilayered techniques. In this patient, rhinorrhea was not observed after surgery. To prevent NCSE, we must consider modifying the surgical procedures to replace air with fluid during dural closure. In the postoperative period, we may keep a patient on bed rest for a longer period to avoid cortical vein traction due to intracranial air movement or consider performing an additional burr hole surgery to refill the air cavity with saline, especially for patients with larger pneumocephalus after sphenoidal surgery.

Lessons

This case illustrates an extremely rare cause of NCSE with pneumocephalus after eTSS for a suprasellar arachnoid cyst.

FIG. 3. On MRI performed on postoperative day 3 during consciousness disturbance, axial ASL-PI (A) showed hyperperfusion and axial DWI (B) showed high-intensity areas at the bilateral frontal lobe (arrows) located around the pneumocephalus area.

FIG. 4. A: EEG when the patient was in a semicoma, demonstrating generalized delta and theta activities (3.5–6 Hz). No epileptiform discharges, such as spike or sharp waves, were observed. B: EEG after recovery of consciousness, revealing normal background activity (10–11 Hz) without any epileptiform discharge.
Pneumocephalus due to CSF leakage can cause NCSE, and ASL-PI and DWI are as useful as EEG and AED tests in diagnosing NCSE. To date, this is the first observation, and better awareness of this association may improve recognition of this symptom; consequently, treatment may be initiated early.

A limitation of this case report is the lack of analysis with long-term continuous EEG, which is recommended to diagnose NCSE accordingly, treatment may be initiated early.

References
1. Kikuta Y, Kubota Y, Nakamoto H, Chernov M, Kawamata T. Nonconvulsive status epilepticus after surgery for ruptured intracranial aneurysms: incidence, associated factors, and impact on the outcome. Clin Neurol Neurosurg. 2021;200:106298.
2. Morioka T, Sayama T, Mukae N, et al. Nonconvulsive status epilepticus during perioperative period of cerebrovascular surgery. Neurol Med Chir (Tokyo). 2011;51(3):171–179.
3. Dennis LJ, Claassen J, Hirsch LJ, Emerson RG, Connolly ES, Mayer SA. Nonconvulsive status epilepticus after subarachnoid hemorrhage. Neurosurgery. 2002;51(5):1136–1144.
4. Little AS, Kerrigan JR, McDougall CG, et al. Nonconvulsive status epilepticus in patients suffering spontaneous subarachnoid hemorrhage. J Neurosurg. 2007;106(6):805–811.
5. Matsubara S, Sato S, Kodama T, et al. Nonconvulsive status epilepticus in acute intracerebral hemorrhage. Stroke. 2016;47(7):1759–1761.
6. Sayegh ET, Fakunjead S, Oh T, Bloch O, Parsa AT. Anticonvulsant prophylaxis for brain tumor surgery: determining the current best available evidence. J Neurosurg. 2014;121(5):1139–1147.
7. Koirala S, Shrestha BK, Lohani S, Bishokarma S, Devkota UP. Postoperative complications of transsphenoidal pituitary adenectomy: a single institution based experience. Kathmandu Univ Med J. 2019;17(66):123–125.
8. Trinka E, Leitinger M. Which EEG patterns in coma are nonconvulsive status epilepticus? Epilepsy Behav. 2019;49:203–222.
9. Leitinger M, Trinka E, Gardella E, et al. Diagnostic accuracy of the Salzburg EEG criteria for non-convulsive status epilepticus: a retrospective study. Lancet Neurol. 2016;15(10):1054–1062.
10. Flacke S, Wüllner U, Keller E, Hamzei F, Urbach H. Reversible changes in echo planar perfusion- and diffusion-weighted MRI in status epilepticus. Neuroradiology. 2000;42(2):92–95.
11. Szabo K, Poeppel A, Pohlmann-Eden B, et al. Diffusion-weighted and perfusion MRI demonstrates parenchymal changes in complex partial status epilepticus. Brain. 2005;128(Pt 6):1369–1376.
12. Shimogawa T, Morioka T, Sayama T, et al. The initial use of arterial spin labeling perfusion and diffusion-weighted magnetic resonance images in the diagnosis of nonconvulsive partial status epilepticus. Epilepsy Res. 2017;129:162–173.
13. Kanazawa Y, Morioka T, Arakawa S, Furuta Y, Nakanishi A, Kitazono T. Nonconvulsive partial status epilepticus mimicking recurrent infarction revealed by diffusion-weighted and arterial spin labeling perfusion magnetic resonance images. J Stroke Cerebrovasc Dis. 2015;24(4):731–738.
14. Gelisse P, Genton P, Crespel A, Lefevre PH. Will MRI replace the EEG for the diagnosis of nonconvulsive status epilepticus, especially focal? Rev Neurol (Paris). 2021;177(4):359–369.
15. Hedna VS, Kumar A, Miller B, et al. Intracranial hypotension masquerading as nonconvulsive status epilepticus: report of 3 cases. J Neurosurg. 2014;120(3):624–627.
16. Gilmour GS, Scott J, Couillard P. Leaking the diagnosis: a case of nonconvulsive status epilepticus due to intracranial hypotension. Neurocrit Care. 2019;31(3):562–566.
17. You CG, Zheng XS. Postoperative pneumocephalus increases the recurrence rate of chronic subdural hematoma. Clin Neurol Neurosurg. 2018;166:56–60.
18. Herman ST, Abend NS, Black TP, et al. Consensus statement on continuous EEG in critically ill adults and children, part I: indications. J Clin Neurophysiol. 2015;32(2):87–95.

Disclosures
The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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Conception and design: Kawataki, Saito, Ogiwara, Kinouchi. Acquisition of data: Kagami, Ogiwara, Hanihara, Kinouchi. Analysis and interpretation of data: Kagami, Kinouchi. Drafting the article: Kawataki, Kagami, Hanihara, Kinouchi. Critically revising the article: Kawataki, Kagami, Ogiwara, Kazama, Kinouchi. Reviewed submitted version of manuscript: Kawataki, Kinouchi. Approved the final version of the manuscript on behalf of all authors: Kawataki. Administrative/technical/material support: Kinouchi. Study supervision: Kawataki, Ogiwara, Kinouchi.

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FIG. 5. On postoperative days 6 (A), 11 (B), and 20 (C), axial ASL-PI showed that the hyperperfusion area at the bifrontal lobe had improved.