

Case Report



A Case of “Sinking Skin Flap Syndrome” in Vegetative State Patient

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Conflict of Interest

The authors have no financial conflicts of interest.

ABSTRACT

Sinking skin flap syndrome is defined by a series of neurological symptoms with skin depression at the site of cranial defect. We experienced neurological improvement in a patient with markedly sunken craniectomy site after ventriculoperitoneal shunt (V-P shunt) clamping operation. A 17-year old female patient was in vegetative state and spastic quadriplegia after traumatic brain injury. She was suffered from frequent vomiting. To evaluate central nervous system problem we checked brain computed tomography which showed that right frontotemporoparietal craniectomy area was markedly sunken and midline was shifting to the left. After V-P shunt clamping operation, craniectomy site was elevated and midline shifting was improved. Vomiting was disappeared. Coma Recovery Scale-revised (CRS-R) score was improved from 3 to 6.

Keywords: Craniectomy; Ventriculoperitoneal shunt; Rehabilitation

INTRODUCTION

“Sinking Skin Flap Syndrome” (SSFS) is a syndrome that can be suspected when a series of neurological symptoms are found along with skin depression at the skull defect.^{1-3,5,7,8,10} Neurological symptoms that may occur include headaches, epilepsy, dizziness, abnormal feelings, numbness, vomiting, changes in consciousness levels, and insomnia.^{1-3,7,8,10} SSFS can occur after a large area of craniectomy after traumatic or spontaneous cerebral hemorrhage, and without cranioplasty for various reasons such as infection and weakness of general condition. We report the diagnosis of SSFS in patients with large cranial defects confirming by the improvement the patient's symptoms like repetitive nausea, vomiting, muscle stiffness, and consciousness through treatment such as ventriculoperitoneal shunt (V-P shunt) clamping operation with a literature review.

CASE REPORT

Written informed consent of the treatment and this report were obtained from guardian of the patient. A 17-year-old female patient was transferred to our hospital due to vegetative mental state, stiff quadriplegia and repeated vomiting. Decompressive craniectomy was done

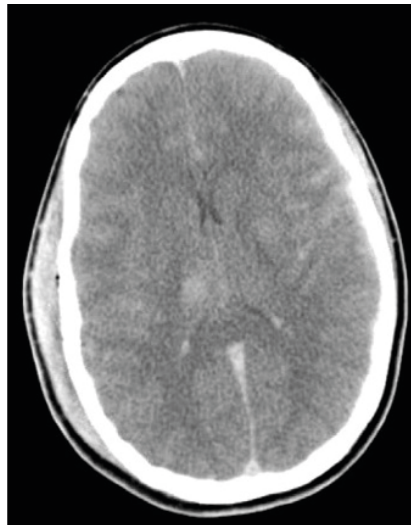


FIGURE 1. At traffic accident, brain computed tomography (CT) showed subarachnoid hemorrhage and brain swelling.

due to traumatic subarachnoid hemorrhage and cerebral edema, which occurred several months ago (**FIGURE 1**). Then, V-P shunt and cranioplasty was done due to hydrocephalus though, after the operation, the brain abscess occurred, and the bone that was implanted during the cranioplasty was removed. At the time of the transfer, the patient's head was in craniectomy state on right frontotemporoparietal area (**FIGURE 2**).

The patient has had difficulty in rehabilitation due to repeated vomiting before being transferred to the hospital, and percutaneous endoscopic gastrostomy (PEG) was performed though, these symptoms did not improve. Nausea and vomiting did not occur in a stable state, but vomiting occurred about 15 minutes after feeding through the PEG tube. The symptoms were also aggravated during wheelchair movement and standing up the tilt table. Increased extensor thrust was observed during wheelchair movement.

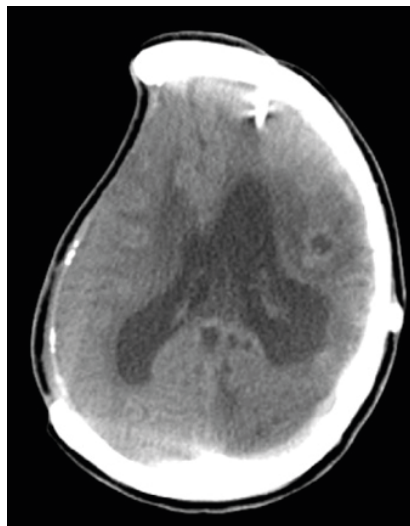


FIGURE 2. After craniectomy and ventriculoperitoneal shunt operation, brain CT showed dilated lateral ventricles and right side large cranial defect. CT: computed tomography.

First of all, blood tests and radiographic examinations were performed to evaluate gastrointestinal problems such as constipation and gastroesophageal reflux disease. However, no specific findings were found, and gastroscopy could not be performed due to bite reflex. Metoclopramide was used to control nausea and vomiting but only showed temporary improvement. Ptosis of eyelid in right side could have suspected the paralysis of oculomotor nerve though, the remaining cranial nerve exam could not be accurately evaluated due to vegetative mental status. Although spontaneous eye opening was possible, no eye contact was observed, and there was no significant reaction except for bite reflex during oral and facial stimulation. Spasticity was measured as grade 1+ on the right upper extremity, 1 on the left upper extremity and 1+ on the left lower extremity on the Modified Ashworth Scale (MAS) grade, deep tendon reflexes were 3+ on both upper and lower extremities. The level of consciousness is evaluated to 8 points on the Glasgow Coma Scale (GCS) (eye opening 4, motor response 3, verbal response 1), persistent vegetative state on the Glasgow Outcome Scale (GOS), and level 2 on the Rancho Los Amigos Levels of Cognitive Function Scale (RLAS). Three points were assessed in the Coma Recovery Scale-revised (CRS-R; **TABLE 1**),⁴⁾ which is used to check the level of consciousness in patients with severe consciousness disorder. Functional activity levels were assessed as fully dependent

TABLE 1. Post-operation CRS-R compared with pre-operation score

Scale	Pre-operation	Post-operation
Auditory function scale		
4. Consistent movement to command		
3. Reproducible movement to command		
2. Localization to sound		
1. Auditory startle		0
0. None	0	
Visual function scale		
5. Object recognition		
4. Object localization: reaching		
3. Visual pursuit		
2. Fixation		
1. Visual startle		
0. None	0	0
Motor function scale		
6. Functional object use		
5. Automatic motor response		
4. Object manipulation		
3. Localization to noxious stimulation		
2. Flexion withdrawal		0
1. Abnormal posturing	0	
0. None		
Oromotor/verbal function scale		
3. Intelligible verbalization		
2. Vocalization/oral movement		
1. Oral reflexive movement	0	0
0. None		
Communication scale		
2. Functional: accurate		
1. Non-functional: intentional		
0. None	0	0
Arousal scale		
3. Attention		
2. Eye opening without stimulation		0
1. Eye opening with stimulation	0	
0. Unarousable		
Total score	3	6

CRS-R: Coma Recovery Scale-revised.



FIGURE 3. After several months, brain CT showed that right craniectomy site was markedly sunken and midline was shifting to the left (about 22 mm) and lateral ventricle was a shrunken state. CT: computed tomography.

with 18 points of Functional Independence Measure (FIM) and 0 points of Korean version of Modified Barthel index (K-MBI). Her blood pressure was 112/80 mmHg and the other vital signs were inconspicuous. All laboratory values were within normal ranges, including electrolytes, serum creatinine, prothrombin time, and the platelet count.

Brain computed tomography (CT) was performed to confirm brain lesions while the symptoms like extensor thrust, vomiting were observed more frequently during wheelchair movement. The brain CT showed significant depression of the right craniectomy site and shifting the midline of the brain by 22 mm to the left (**FIGURE 3**). We suspected of sinking skin flap syndrome and adjust the V-P shunt pressure valve (pressure 13 mmH₂O → 20 mmH₂O). Even though the pressure was increased, there was no significant difference in brain CT, then we closed the valve completely. Therefore, it was deemed that the pressure control valve did not function normally and performed V-P shunt clamping operation under local anesthesia.

After the operation, the brain CT showed a partial correction of the craniectomy site and the movement of the midline shifting corrected to around 6mm (**FIGURE 4**). After surgery, symptoms like vomiting were disappeared in before, during and after the exercise and meals, access to rehabilitation has increased because extensor thrust caused by wheelchair movement also disappeared. Physical examination revealed a visual elevation of the depressed craniectomy site, the ptosis of right eyelid disappeared, opening the eyes symmetrically to the left, improved eye contact, and increased avoidance response during oral and facial stimulation. The range of application of occupational therapy, including stimulation of taste, stimulation of temperature in the mouth and face, has widened.

Spasticity was decreased slightly in both upper and lower extremities as MAS grade 1, and there was no change in deep tendon reflex. There was no significant difference in GCS and RLAS, but in the CRS-R sub-category, eye point was improved by 1 point on auditory function scale without stimulation on arousal scale, and abnormal posturing on motor function scale showed improvement of 1 to 2 points due to flexion withdrawal response. The combined total

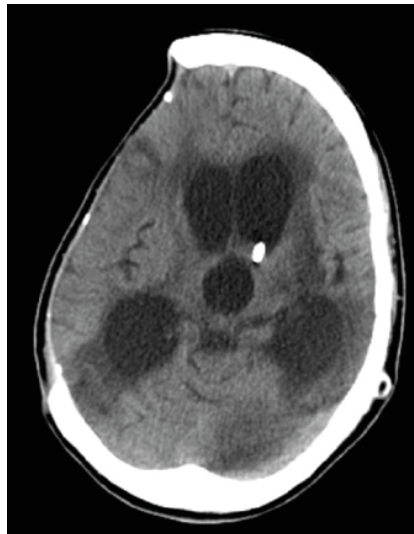


FIGURE 4. After V-P shunt clamping operation, brain CT showed that right craniectomy site was swollen and midline restored (about 6 mm) and mild hydrocephalus.
V-P shunt: ventriculoperitoneal shunt, CT: computed tomography.

scores showed that the overall score improved from 3 to 6 points (**TABLE 1**). Brain CT is to regularly perform to monitor the patient, and the cranioplasty is scheduled.

DISCUSSION

In 1939, Grant et al. defined “Syndrome of the trephined” as epilepsy, headache, dizziness, and emotional changes in patients undergoing craniectomy. In 1945, Gardner reported clinical improvement after cranioplasty in patients with craniectomy, and in 1974 Yamamura et al. used the new term “Sinking Skin Flap Syndrome” to further describe this symptom.¹⁾ SSFS is generally known to occur weeks to months after craniectomy.¹⁰⁾ Symptoms that may occur include headaches, epilepsy, dizziness, abnormal feelings, paralysis, vomiting, changes in consciousness levels, and insomnia.

In our case, the patient's level of consciousness was a vegetative mental state that prevented subjective complaints such as headache, dizziness, abnormal feelings, and insomnia. Only objective symptoms such as vomiting, increased bilateral lower limb stiffness, and extensor thrust were identified. In another hospital, repeated vomiting was regarded only as a symptom of gastrointestinal abnormality, and about 5 months of testing and treatment was done, but failed to show any improvement. SSFS is must be considered to be one of the major factors to consider when presenting persistent vomiting in patients with unconscious after craniectomy.

The most common pathophysiological mechanism of SSFS is the fluid dynamics of cerebrospinal fluid (CSF).²⁾ This hypothesis is a mechanism to explain the symptoms of SSFS due to a decrease in CSF by the bypass of V-P shunt, prolonged dehydration and standing posture.^{3,5)} Also in our case, the patient had undergone V-P shunt, and vomiting worsened in standing postures such as wheelchair movement and tilt table treatment, and related symptoms improved after complete blocking of V-P shunt. This suggests that there is a clear association between hydrodynamic changes in CSF and the development of SSFS. Also in our case, the patient had undergone V-P shunt, and vomiting worsened in standing postures

such as wheelchair movement and tilt table treatment, and related symptoms improved after complete blocking of V-P shunt. This suggests that there is a clear association between hydrodynamic changes in CSF and the development of SSFS. Other pathophysiological mechanisms of SSFS include: 1) direct pressure on the cerebral cortex, 2) effects of changes in atmospheric pressure and brain blood flow, and 3) impairment of brain metabolism.^{1,3,5-8,10)}

For the treatment of SSFS, cranioplasty is considered first.^{1-3,5-8,10)} The extent to which symptoms improve after cranioplasty may vary depending on the time of treatment.¹⁰⁾ Whitfield et al.⁹⁾ reported that symptomatic improvement after cranioplasty at a relatively acute stage of 4–20 weeks, but did not show significant improvement when treated after a chronic phase of more than 3 years.¹⁰⁾ Another treatment for SSFS, Han et al. proposed a method for adjusting the pressure control valve of the V-P shunt.⁵⁾ Our patient attempted V-P shunt and cranioplasty for hydrocephalus 4 months after the injury in the previous hospital, but the brain abscess occurred and the craniectomy was done again. After 6 months after the injury, symptoms like vomiting were observed, and about 11 months after the symptoms occurred SSFS was diagnosed. Since this patient was passive for reoperation due to a negative experience with previous cranioplasty, we attempted to adjust another treatment method of V-P shunt pressure regulating valve to improve the symptoms. In cases where cranioplasty is difficult to consider, such as in this patient, adjustment of the V-P shunt pressure control valve may be a good alternative.

We diagnosed SSFS through neurological symptoms like vomiting, increased spasticity and the results of brain CT in patients with vegetative mental state in chronic phase. And we report this case with a review of the literature that we have experienced increased access to rehabilitation therapy by the treatment of SSFS.

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