

Sinking Skin Flap Syndrome and Paradoxical Herniation After Hemicraniectomy for Malignant Hemispheric Infarction

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Background and Purpose—“Sinking skin flap” (SSF) syndrome is a rare complication after large craniectomy that may progress to “paradoxical” herniation as a consequence of atmospheric pressure exceeding intracranial pressure. The prevalence and characteristics of SSF syndrome after hemicraniectomy for malignant infarction of the middle cerebral artery are not well known.

Methods—We analyzed a prospective cohort of 27 patients who underwent hemicraniectomy for malignant middle cerebral artery infarction. All had a clinical and brain imaging follow-up at 3 months and were followed until cranioplasty.

Results—Three of 27 patients (11%) had, at 3 to 5 months posthemicraniectomy, SSF syndrome with severe orthostatic headache as the main symptom. In addition, 4 patients (15%) had radiological SSF syndrome but no clinical symptoms except partial seizures in one. Patients with SSF syndrome had a smaller surface of craniectomy (76.2 cm² versus 88.7 cm², $P=0.05$) and a tendency toward larger infarct volume, an older age, and a longer delay to cranioplasty than those without this syndrome.

Conclusions—SSF syndrome either clinically symptomatic or asymptomatic affects one fourth of patients 3 to 5 months after hemicraniectomy for malignant middle cerebral artery infarction. It should be diagnosed as early as possible to avoid progression to a paradoxical herniation. (*Stroke*. 2010;41:560-562.)

Key Words: complications ■ surgery ■ hemicraniectomy ■ malignant cerebral artery infarction ■ herniation

The syndrome of the “trephined” or the “sinking skin flap” (SSF) syndrome is a rare complication after a large skull bone defect.¹ It consists of a sunken skin above the bone defect with neurological symptoms such as severe headaches, mental changes, focal deficits, or seizures.^{1,2} The SSF may progress to “paradoxical herniation” as a consequence of the atmospheric pressure exceeding intracranial pressure and may eventually lead to coma and death.

The objective of our study was to determine, in a prospective cohort of malignant infarction of the middle cerebral artery (MCA), the prevalence and characteristics of SSF syndrome and of any radiological sunken skin flap without symptoms after hemicraniectomy.

Materials and Methods

Patients

All patients randomized in the surgical arm of DEcompressive Craniectomy In MALignant middle cerebral artery infarcts (DECIMAL), a trial that compared medical treatment and hemicraniectomy in malignant MCA infarction as well as all consecutive patients who

had hemicraniectomy for a malignant MCA infarction in our stroke center after the end of DECIMAL inclusion but according to the same criteria, were considered.³

All surviving patients had at the 3-month follow-up a clinical and brain imaging evaluation (axial and/or coronal fluid-attenuated inversion recovery MRI and/or axial and/or coronal CT scan) and were then followed every 3 months for 1 year. The timing and procedure of cranioplasty were left to the discretion of the neurosurgeon. The study was approved by an institutional ethics committee, and the patient or a close relative gave informed consent.

Radiological Evaluation

The following measures were performed by the same neuroradiologist (J.-P.G.): (1) the maximum horizontal surface of the skull was estimated either on axial CT scan or axial fluid-attenuated inversion recovery MRI using the following formula: $\pi/4 \times A \times B$ in which “A” was the maximum distance from the anterior and posterior inner tables of the skull and “B/2” the half-depth to the inner skull surface opposite to the bone flap measured at the midpoint of “A”; (2) the whole surface of craniectomy was estimated either on coronal reformation and axial plane CT scan or axial and coronal fluid-attenuated inversion recovery MRI using the following formula: $\pi/4 \times C \times D$ in which “C” was the maximum vertical diameter and

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Table 1. Patients' Characteristics

	Patients With SSF Syndrome				Patients With Asymptomatic Radiological SSF		
	Patient 1	Patient 2	Patient 3	Patient 4	Patient 5	Patient 6	Patient 7
Gender/age, years	Male/47	Male/45	Male/53	Male/54	Female/39	Male/54	Female/50
Infarction of the dominant hemisphere	No	No	No	Yes	Yes	Yes	Yes
Baseline diffusion-weighted imaging infarct volume, cm ³	220	300	155	350	320	220	153
Time from craniectomy to symptoms	5 months	4 months	3 months	5 months
Orthostatic symptoms	Headache, vomiting, drowsiness, bilateral Babinski	Headache, vomiting	Headache, vomiting
Other symptoms	Partial motor seizures	Partial motor seizures
SSF	Severe	Severe	Moderate	Severe	Moderate	Moderate	Moderate
Subfalcine herniation	Yes	Yes	No	Yes	No	No	No
Midbrain compression	Yes	Yes	No	No	No	No	No
Emergency treatment and response	Supine position, normal saline (9%)/complete resolution	Supine position, 1.5 L/day of oral hydration/partial resolution	Supine position, normal saline (9%)/complete resolution

“D” the maximum horizontal diameter of the bone defect; and (3) radiological SSF syndrome was defined by any anterior or posterior negative skin depression below a horizontal line drawn from both anterior and posterior outer tables of the bone defect at any of the following 2 levels: just above the plane of the thalamus (“thalamus” level) and just above the lateral ventricles (“ventricles” level). The measures were performed at 3 months and, in the case of delayed cranioplasty, at the last brain imaging when available.

Statistics

To evaluate the associations between SSF syndrome and baseline clinical and radiological parameters, we used a Fisher test for categorical variables and an analysis of variance for continuous variables.

Results

A total of 40 patients were considered. Among the 31 surviving patients, one was excluded because of brain abscess and 3 because of missing imaging data. Thus, a total of 27 patients (15 men, 12 women; mean±SD age, 43.6±9.3 years; range, 22 to 55 years) were analyzed.

During follow-up, 3 patients had SSF syndrome with severe orthostatic headache (Table 1). In one patient, orthostatic headache progressed in a few days to confusion, drowsiness, bilateral Babinski sign, subfalcine herniation, and midbrain compression (Figure). One additional patient had

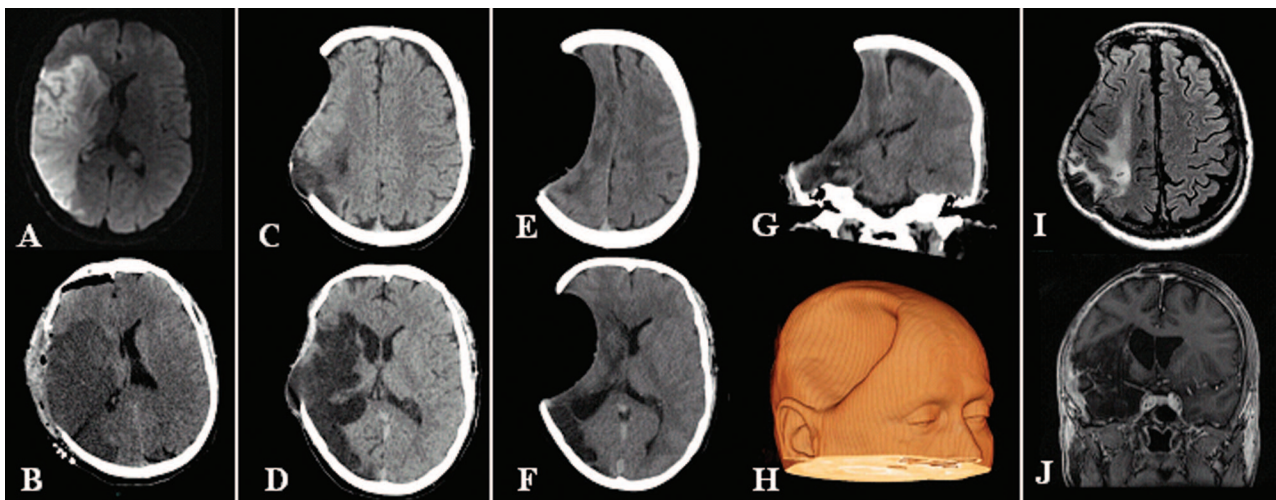


Figure. Brain imaging of Patient 1: (A) axial diffusion-weighted imaging: right MCA infarction; (B) axial CT scan 24 hours later: large craniectomy with slight midline shift; (C–D) axial CT scan at 3 months: right hemispheric hypodensity at the thalamus level (D) and moderate anterior skin depression at the ventricles level (C); (E–G) axial CT scan at the time of clinical signs of brain herniation: sunken skin flap and severe compression of the right hemisphere with subfalcine herniation; (H) CT scan, 3-dimensional volume: profound sunken skin flap; (I) axial fluid-attenuated inversion recovery and (J) coronal T1-weighted image after 48 hours of supine position and intravenous hydration: complete resolution of the sunken skin flap.

Table 2. Comparison Between Patients With (n=7) and Without (n=20) SSF Syndrome

	SSF Syndrome (Symptomatic or Clinically Asymptomatic)		P Value
	No (n=20 Patients)	Yes (n=7 Patients)	
Mean age (SD)	41.8 (9.7)	49.0 (5.5)	0.08
Women, %	83.3	16.7	0.41
Men, %	66.7	33.3	
Mean of baseline diffusion-weighted imaging infarct volume (SD), cm ³	213.2 (66.3)	242.9 (80.3)	0.34
Mean craniectomy surface (SD), cm ²	88.6 (12.9)	76.25 (15.7)	0.05
Mean of the maximum horizontal surface of the skull (SD), cm ²	165.27 (17.36)	155.27 (29.19)	0.29
Mean ratio of the total surface of craniectomy to the maximum horizontal surface of the skull (SD), %	0.54 (0.10)	0.51 (0.17)	0.57
Mean time to cranioplasty (SD), days	198 (98)	250 (116)	0.26

radiological SSF and had 2 short episodes of partial seizures. Three other patients, all with severe global aphasia, had at 3 months radiological SSF but no orthostatic symptoms. None had a precipitating factor such as lumbar puncture.

Supine position with the head turned to the side of the craniectomy and intravenous fluid administration rapidly resolved orthostatic symptoms in 2 patients. One patient remained with a sunken skin flap for several weeks, because hemodynamic measures were not possible because of heart failure. However, after cranioplasty, the headaches completely resolved and the patient was able to move his hemiplegic arm against gravity, which was not possible before the operation.

When comparing patients without (n=20) and those with SSF syndrome either clinically symptomatic or asymptomatic (n=7), we found in patients with this syndrome a significantly smaller craniectomy surface (76.2 cm² versus 88.7 cm², $P=0.05$) and a nonsignificant tendency toward a larger baseline infarct volume, an older age, and a longer delay to cranioplasty (Table 2).

Discussion

In this prospective cohort of 27 patients who had hemicraniectomy for malignant MCA infarction, 3 patients (11%) had at 3 to 5 months after the stroke an SSF syndrome with severe orthostatic headache as the main symptom. Among them, one patient deteriorated to paradoxical herniation. We also found that 4 patients (15%) had radiological SSF but no clinical symptoms except partial seizures in one. However, in these patients, subtle neurological symptoms may not have been appreciated because of the aphasia and the motor deficit.

Neurological symptoms such as headaches, mental changes, and language or motor deficits have been reported in SSF syndrome. These symptoms were regressive after recumbency or epidural blood patch and resolved completely after cranioplasty as was the case in our patients.⁴ It is notable that in one of our patients, even motor deficit improved after cranioplasty. This finding suggests that in malignant MCA infarction, SSF syndrome may delay neurological recovery. Increase in cerebral blood flow after cranioplasty has been reported, suggesting that decreased cerebral blood flow may be one of the underlying mechanisms by which SSF syndrome causes focal deficits.^{5,6}

In our study, we found that patients with SSF syndrome have a significantly smaller surface of craniectomy and a tendency to be older and to have a larger baseline infarct volume. One hypothesis would be that more severe poststroke brain atrophy contributes to a decrease in intracranial volume and subsequently to a decrease in intracranial pressure in an open skull. In addition, smaller size of the craniectomy may lead to secondary injuries because of inadequate hemispheric decompression and finally to a larger hemispheric lesion.² However, these findings have to be confirmed in a larger cohort of patients.

After hemicraniectomy for malignant hemispheric infarction, the best time for cranioplasty is not well known. Considering our data, it may be justifiable to replace the bone defect during the first 2 to 3 months poststroke. In addition, SSF syndrome should also be considered as a delayed complication of hemicraniectomy in the evaluation of its long-term benefit.

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Disclosures

None.

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