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# Sinking skin syndrome in a decompressive craniectomy series: Clinical and radiological features

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## ABSTRACT

**Background:** The sinking skin syndrome (SSS) is a particular complication after a decompressive craniectomy (DC). It still remains a poorly understood and underestimated entity.

**Methods:** Retrospective case series of craniectomized patients with and without SSS. Clinical and radiological features (DC diameter, shape of craniectomy flap, and midline deviation) were described and relative volumes of intracranial loss were quantified.

**Results:** Twenty-seven patients (63% with SSS). The most common indication for DC was traumatic brain injury: 48.15%. The p50 diameter of DC was 12.8 cm for patients with SSS and 11.1 cm for patients without (Z score = 0.32). DC area was 81.5 $cm^2$ for patients with SSS and 71.43 $cm^2$ for patients without the syndrome (Z score = 0.61). According to the shape of the craniectomy flap, we classified our patients as: «same level» (51.8%), «sunken» (25.9%), and «extracranial herniation» (14.8%). Two patients (7.4%) had paradoxical herniation. Midline deviation was present in 12 (70.6%) patients with SSS. The 3rd ventricle volume average was 1.2 cc for patients with SSS versus 2.35 cc for patients without (Z score = 0.04). About 94.11% of patients (16 out of 17) clearly improved after replacement of the cranial defect.

**Conclusion:** In our series, low 3rd ventricle volumes had a good relation with SSS. The presence of a sunken flap does not guarantee SSS *per se* and we propose the following radiologic description: A = sunken, B = same level, C = extracranial herniation, and D = paradoxical. Replacement of the skull defect is the main treatment.

**Keywords:** Craniectomy, Decompressive, Paradoxical herniation, Sinking flap, Sinking skin syndrome

## INTRODUCTION

The sinking skin syndrome (SSS) or syndrome of the trephined, as first described by Grant and Norcross,[9] is a very particular complication after a decompressive craniectomy (DC). It consists in neurological deterioration believed to be related to the barometric pressure changes over the brain after removing the skull, affecting also cerebrospinal fluid (CSF) and cerebral blood flow (CBF).

Based on an array of case series, many clinical features of this syndrome are described in the literature.[2,3,6,10,13,23] These features can be (1) neurological: headaches, seizures, dizziness, lethargy, aphasia, and hemiplegia; (2) psychological/psychiatric: anxiety, apprehension, insecurity, apathy, and desperation; and (3) physical: discomfort at the craniectomy site. While the clinical signs and symptoms are multifarious, there are common characteristics to this syndrome: symptoms begin weeks or months

after DC, occurrence is independent of the location or type of lesion, and the almost complete resolution after cranioplasty.[2]

To date, many mechanisms have been lucubrated to explain the pathophysiology of this phenomenon. Some authors have suggested brain pulsation due to arterial and venous pressure variations,[7] atmospheric pressure over the cortex,[15] siphon effect created on CSF by the skull defect,[5] and CBF locally disturbances caused by the atmospheric pressure.[24]

From the radiological point of view, the brain of a postcraniectomy patient can undergo one of three following scenarios: (1) the brain is at the same level of the craniectomy flap, (2) the brain is sunken (sunken flap sign), or (3) it can be herniated extracranially. Sometimes, in the context of a sunken brain, a contralateral deviation of the midline can occur giving place a complication called paradoxical herniation. To complicate things further, the SSS can be observed in patients without the sunken flap sign (i.e., extracranial herniation) suggesting that the SSS could be a misnomer.[22]

The main treatment, as mentioned before, is the cranioplasty.[14] While all patients show improvement after replacement of the cranial defect, the time for symptoms to reverse can vary. Some analysis suggests that improvement can be seen even in the first 24 h. The appropriate timing for cranioplasty has not been determined and some literature indicates that there is no direct correlation between improvement and short intervals between craniectomy and cranioplasty.[4,12,16]

## MATERIALS AND METHODS

During the period of 2010–2016, we reviewed retrospectively cases of patients who underwent DC. We collected epidemiological data and images. Two groups were conformed according to the presence or absence of SSS. Initial diagnostic prompting the DC, area, and diameter was described in both groups. In the group of patients with SSS, we also described clinical symptoms and average time from DC to the beginning of symptoms, average time from DC to cranioplasty, and whether they improved afterward.

Radiologically, we described the shape of the craniectomy flap as sunken, in the same level and extracranial herniation. Midline deviation below 5 mm and the presence of paradoxical herniation (defined as deviation of the midline by more than 5 mm away from the craniectomy site) are also described [Figures 1 and 2]. Relative volumes such as intracranial loss, extracranial herniation, and 3rd ventricle volume were calculated by multiplying its surface ($mm^2$, sagittal plane) by its thickness (mm, axial plane) using the reconstruction tools of our DICOM CT software [Figures 3 and 4].

For the statistical analysis, we used Fisher's exact test for categorical variables and using Mann–Whitney's Student's t-test or U-test on numerical ones. The significance level has been set to 0.05 and the statistical package used was Stata V.16 (StataCorp LLC.).

## RESULTS

Through search in our data banks, 98 cases of patients who underwent DC were identified between the 2010 and 2016 period. Of these patients, only 27 had all the relevant clinical information mentioned lines above to conform our series. A graphic describing the process of inclusion/exclusion criteria is shown in Figure 3. With these 27 patients, two groups were conformed: 17 (63%) patients with SSS and 10 (37%) patients without the syndrome.

### Age and sex

The mean age in our series was 48.4 years. In the group of patients with SSS, the median age was 51.7 years, and 53% were female and 47% were male. In the group corresponding to patients without the syndrome, the mean age was 42.9 years and 30% were female and 70% were male [z score = 0.4, Table 1].

### Indications for DC

The most common indication for DC, in general, was traumatic brain injury (TBI) accounting for 48.15% (13 patients), followed by hemorrhagic causes in 18.5% (five patients), ischemic in 14.8%, infection in 11.1%, and tumor in 7.4%. TBI was also the main cause for DC in the group of patients with SSS and the group of patients without the syndrome (53% and 40%, respectively) [Table 1].

### Size and characteristics of DC

The diameter and area of the craniectomy were calculated. For the general population, the p50 diameter of the DC was 12.6 cm and the area of decompression was 78 $cm^2$. In the group of patients with SSS, the p50 was 12.8 cm and for patients without SSS was 11.1 cm (z score = 0.32). We also calculated the area of decompression being 81.5 $cm^2$ for the group of patients with SSS and 71.43 $cm^2$ for the patients without the syndrome (z score = 0.61) [Table 2].

### Clinical features

Of our 27 patients, 17 (62.9%) presented SSS. The most common symptom for patients with SSS was neurologic deficit (82%) followed by psyche changes (47%) [Table 2].

### Radiologic features

As described above, we classified our patients according to the shape of the craniectomy flap as «same level» (14 patients,

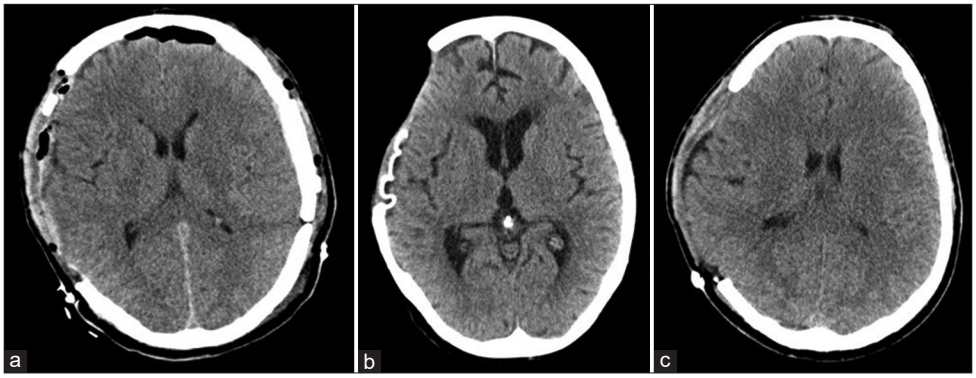


**Figure 1:** Radiological description of the three types of sinking skin syndrome according to computed tomography scan: (a) same level, (b) sunken, and (c) extracranial herniation.

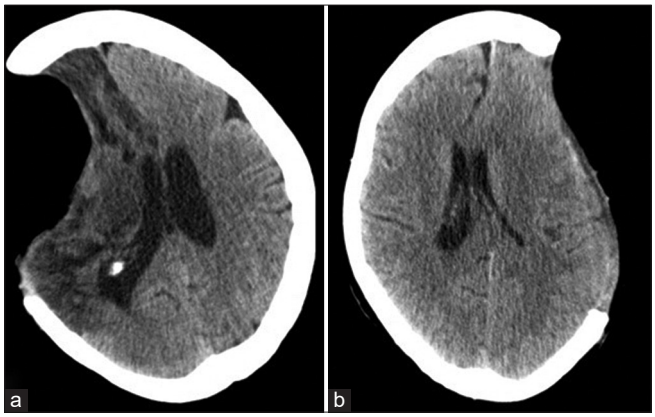


**Figure 2:** (a and b) Paradoxical herniation.

**Table 1:** Demographic characteristics.

| | Sinking skin syndrome | |
|---|---|---|
| | Yes (17) | No (10) |
| Age | 51.7±13.5 | 42.9±15.3 |
| Female | 9 | 3 |
| Male | 8 | 7 |
| Indication for decompressive craniectomy | | |
| Trauma | 9 | 4 |
| Tumor | 1 | 1 |
| Infarction | 2 | 2 |
| Cerebrovascular | 3 | 2 |
| Infection | 2 | 1 |

Fisher's exact=0.963

51.8%), «sunken» (seven patients, 25.9%), and «extracranial herniation» (four patients, 14.8%). Two patients (7.4%) presented the paradoxical herniation phenomena. Midline deviation was present in 12 (70.6%) patients with SSS and 7 (70%) without the syndrome [Table 3].

The sunken flap sign, defined as the observable skin depression at the craniectomy site, was present in six patients in the SSS group (35.2%) and one in the group without the syndrome (10%). The relative sinking volume was calculated in both groups, respectively, 43.76 versus 14.24 cc (z score= 0.5).

**Table 2:** Surgical and clinical characteristics.

| | Sinking skin syndrome | |
|---|---|---|
| | Yes (17) | No (10) |
| Craniectomy size | | |
| Diameter (cm) | 12.8 | 11.1 |
| Area ($cm^2$) | 81.35 | 71.46 |
| Symptoms | | |
| Neurologic deficit | 14/17 82% | – |
| Psyche changes | 8/17 47% | – |
| Level of consciousness | 5/17 29% | – |
| Other* | 3/17 17% | – |
| Average days from DC to SSS | 19 | – |
| Average months from DC to CRX | 2 | 4.15 |
| Average days from DC to improvement | 6.5 (16/17) | – |

DC: Decompressive craniectomy, SSS: Sinking skin syndrome, CRX: Cranioplasty. *Headache, heaviness sensation, etc

**Table 3:** Radiological features.

| | Syndrome of the trephined | |
|---|---|---|
| | Yes (17) | No (10) |
| Shape of the defect | | |
| Sunken | 4 | 3 |
| Same level | 8 | 6 |
| Extracranial herniation | 3 | 1 |
| Paradoxical herniation | 2 | 0 |
| Fisher's exact=0.814 | | |
| Average midline deviation | 0.46 | 0.3 |
| Average 3rd ventricle volume | 1.2 | 2.35 |
| Average loss of intracranial volume | 43.7 (6/17) | 83.12 (2/10) |
| Average herniated extracranial volume | 68.8 (5/17) | 45.4 (4/10) |

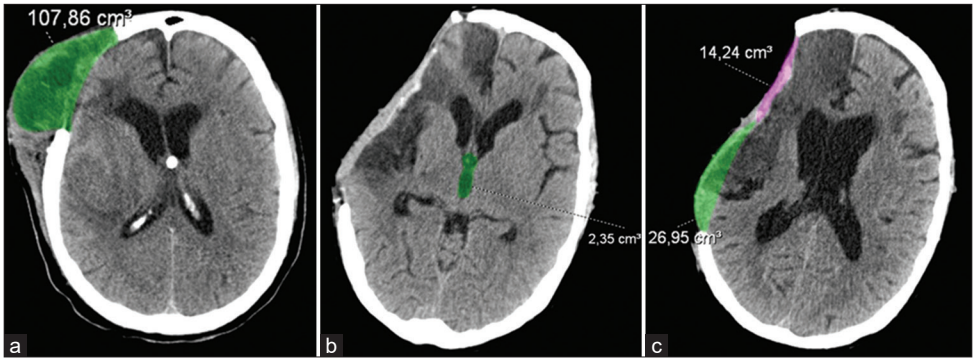


**Figure 3:** Radiological measurements of mentioned relative volumes: extracranial (a), 3rd ventricle (b), and intracranial volume loss (c), Green: Extracranial volume, Pink: Intracranial volume.

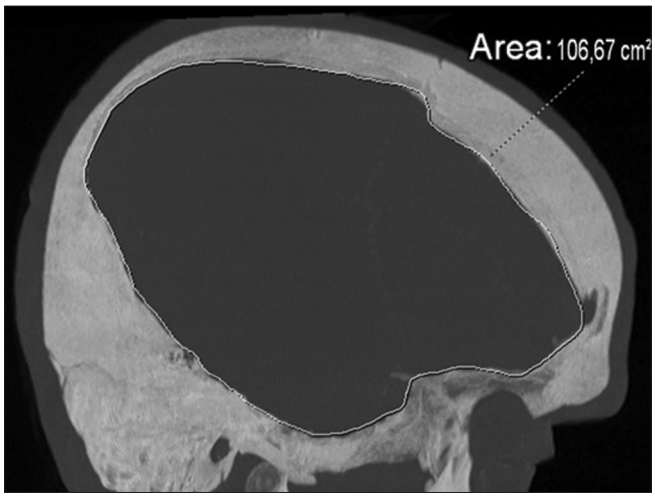


**Figure 4:** Calculation of the decompressive craniectomy area using the reconstruction tools of our DICOM software.

Extracranial herniation was observed in 5 (29.4%) patients with SSS and 2 (20%) in patients without the syndrome. The relative herniated volumes were calculated in both groups also: 68.8 versus 45.4 cc (z score = 0.8), respectively. The presence of paradoxical herniation was confirmed in two patients exclusively in the SSS group.

Finally, the third ventricle volume average in our series was 2.06 cc. In the group of patients with SSS this volume was 1.2 and in the group of patients without the syndrome it was 2.35 cc (z score = 0.04).

### Time from DC to SSS

The average days from the craniectomy to the appearance of the syndrome were 19 days (p25 = 16 and p75 = 22) [Table 3].

### Time from DC to cranioplasty

The average days from the decompression to the cranioplasty were 3 months (p25 = 2 and p75 = 6.3) in the general population. According to our protocols, in our service, we try to replace the defect in the first 3–4 months following decompression whenever it is possible and there is no contraindication. In the group of patients with SSS, the time until cranioplasty was 4.1 months (p50) and for patients without the syndrome was 2 months (z score = 0.33) [Table 3].

### Improvement after cranioplasty

In our series, 94.11% of patients (16 out of 17) clearly improved after replacement of the cranial defect. For improvement, we considered disappearance and/or alleviation of symptoms. The mean time to improvement was 6.5 days (p25 = 4.5 and p75 = 8.5) after cranioplasty. One patient did not improve at all after the procedure [Table 3].

## DISCUSSION

In our series, there were no differences between patients with and without SSS when age and sex were considered. Some studies have suggested a slightly male predominance,[11] which can be explained by the fact that the most common indication for craniectomy is TBI, which is more common in males. In our series, TBI was also the main cause for DC.

In the literature, there are several findings about the increased mean times from DC to cranioplasty (usually ≥9 months) being a factor for higher risk of presenting SSS.[2] In our series, patients with SSS had cranioplasty at 4.1 months since DC and patients without the syndrome had it at 2 months. While the results were not significative in our series, this could be in relation to the underpowered population for the study. This is consistent with the literature and is important to consider that patients with DC have usually more complex indications and it might not be the best idea to replace the bone flap early (i.e., infection, trauma, etc.).

The craniectomy size has been suggested as a factor to be considered. There are no correlations published in

the literature[2,18] but it seems that a craniectomy with an area >100 $cm^2$ could be related with the presence of SSS. In our series, all these patients had craniectomy areas <100 $cm^2$ (p50 = 81.5 $cm^2$; p25 = 55.8, and p75 = 94.5) and still developed the syndrome.

The most common clinical finding was full or some grade of motor weakness (82%). Since motor involvement is one of the most easily recognized signs in this syndrome, some others can be underestimated. In that sense, most authors suggest that cognitive deficit should be actively reported. In our series, almost half of the patients with the SSS (47%) had some kind of cognitive involvement.

As the name suggests, the identification of a sunken flap is related to the presence of SSS. While many reports confirm this association,[5,13] it should be considered that a good percentage of patients with a sunken flap have no symptoms at all. Thus, the presence of a sunken flap does not guarantee SSS.[19,21] These findings are related to the definition used in some series, where a true SST is diagnosed only if symptoms are alleviated after cranioplasty.[22] In our series, 57.14% of patients with a sunken flap had clinical SSS.

Radiological findings are not standardized and there is no radiology-based classification neither, but we can find wholesome descriptions about features of this syndrome. At present, we can only provide broad categories of radiological features based on the morphology of the defect as follows: (a) sunken, (b) same level, (c) extracranial herniation, and (d) paradoxical herniation and we propose that this nomenclature should be used while describing SSS. The use of a single terminology can help to report and describe this underestimated pathology.

Paradoxical herniation has been reported as a rare complication in these patients.[1,17] It is defined as a midline deviation contralateral to the cranial defect. In our series, two patients presented this finding and both had no previous history of CSF drainage as suggested in the literature[1] and both developed SSS. This condition can deteriorate patients to death if not identified and treated.[2,17] Paradoxical herniation and midline deviation might be considered as specifics radiological signs of SSS but it is not the case, since almost 50% of patients with midline shifts larger than 5 mm do not develop the syndrome.[20] We corroborate this result in our series where we had 19 patients with midline deviation and 7 (36.84%) did not presented SSS.

CSF studies in patients after DC can explain some of the incidence of SSS. Besides the well-known variations in the systolic flow velocity due to cranioplasty, there is also the CSF volume loss that can aggravate or promote the appearance of SSS. In postcraniectomized patients, there is evidence that CSF leakage or over drainage can lead to neurological deterioration.[8,25] We used the 3rd ventricle volume measurement and found a statistically difference for patients with SSS who had lower volumes compared with patients without the syndrome (1.2 vs. 2.3 cc, z=0.04) in the same line with the results obtained by Vasung *et al*.[22] When we calculated the relative sinking and relative herniated volumes, we obtained no statistical difference. We believe that this is due to the small sample and larger series with linear model analysis could correct this, since it has been demonstrated the relation between lower relative intracranial CSF volumes and the presence of SSS.[2,22]

Finally, the only treatment for this entity is the cranial defect replacement [Figure 5]. Because barometric parameters inside the skull are altered after a craniectomy, it is logical to think that the only way to correct this is placing the bone back. Although this can sound easy in theory, it is not in practice due to the high variability of diagnosis, complications, and resources. No guidelines are established to prevent this complication and some debate is still present with studies reporting no correlation at all between improvement and early cranioplasty,[4,12,16] and authors suggesting cranioplasty as soon as possible.[12,13,19,21]

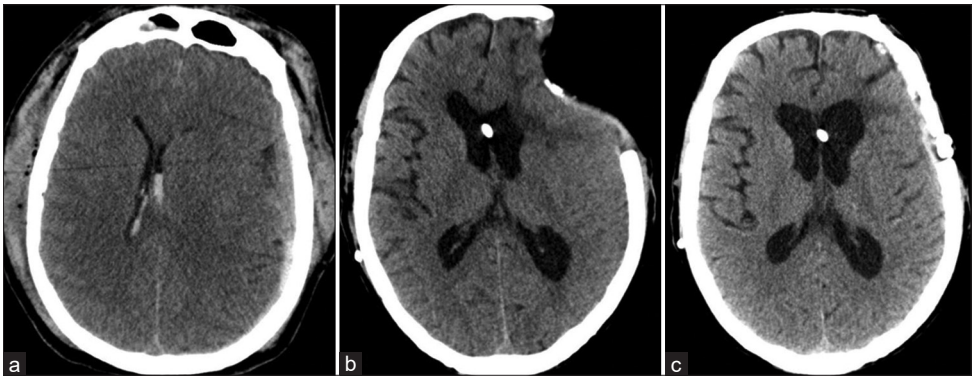


**Figure 5:** Patient with a severe traumatic brain injury and cerebral edema (a) who received a decompressive craniectomy (b). This patient developed sinking skin syndrome with a sunken flap sign and recovered completely after cranioplasty (c).

It is our practice to individualize patients and replace the cranial defect as early as we can considering all variables about diagnosis and probable complications.

## CONCLUSION

The SSS is an underestimated and poorly understood entity not being actively reported nor investigated. While motor weakness is the main feature, cognitive deficit can also appear. Low 3rd ventricle volumes have a good relation with the presence of SSS, while measuring the relative cranial volumes (herniated and loss) could be useful in larger series to predict its development. The presence of a sunken flap does not guarantee SSS and is in the best of interest to establish some kind of classification based on radiological features: A = sunken, B = same level, C = extracranial herniation, and D = paradoxical. Although there is a hot debate about when to replace the cranial defect, we suggest it is vital to consider doing it early on after a DC.

### Declaration of patient consent

Patients' consent not required as patients' identities were not disclosed or compromised.

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Nil.

### Conflicts of interest

There are no conflicts of interest.

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