Massive Brain Swelling and Death After Cranioplasty: A Systematic Review

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Key words
- Brain edema after cranioplasty
- Cranioplasty complication
- Death after cranioplasty
- Malignant brain swelling after cranioplasty

Abbreviations and Acronyms
AP: Atmospheric pressure
CBF: Cerebral blood flow
CSF: Cerebrospinal fluid
CT: Computed tomography
DC: Decompressive craniectomy
ICP: Intracranial pressure
MBSC: Massive brain swelling after cranioplasty
SSF: Sinking of skin flap
SSSF: Syndrome of sinking skin flap
TBI: Traumatic brain injury

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BACKGROUND: **Although cranioplasty is a common procedure, it may cause a variety of complications. Massive brain swelling after cranioplasty (MBSC) is an unusual complication that has been reported more frequently in recent years. Most of the existing information about this condition is speculative and the cause remains unclear.**

METHODS: **A PubMed and Scopus search adhering to PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) guidelines was performed to include studies reporting patients with MBSC. Different information was analyzed in these cases to describe the characteristics and identify risk factors for MBSC.**

RESULTS: **The search yielded 19 articles with a total of 26 patients. All studies were case reports and small case series. In most patients, preoperative intracranial hypotension and a considerable degree of sinking of skin flap were identified; this was the only constant finding observed in these cases. In addition, we propose a grading system to estimate the degree of preoperative sinking of skin flap and an algorithm with recommendations to decrease the incidence of MBSC.**

CONCLUSIONS: **MBSC is an unusual, highly lethal, and probably underreported condition. The information gathered in this review indicates that MBSC occurs secondary to a cascade of pathologic events triggered by the bone flap implantation. This evidence suggests that the primary pathologic change is a sudden increase in the intracranial pressure acting on a brain chronically exposed to intracranial hypotension.**

INTRODUCTION

Decompressive craniectomy (DC) is frequently performed in cases of refractory intracranial hypertension secondary to several neurologic conditions. DC is usually followed by cranioplasty in those patients who survive the initial injury. It is reported that complications after cranioplasty occur in about a third of cases.[1] In recent years, several studies have reported an unusual complication characterized by a high degree of brain edema occurring after an uneventful cranioplasty. Massive brain swelling after cranioplasty (MBSC) is a condition that entails high mortality the cause of which has not yet been elucidated[2-11]; for these reasons, we consider that a study describing the characteristics of this devastating complication is necessary.

The main purpose of this study was to determine the factors involved in the development of MBSC, as well as identifying measures to prevent the occurrence of this disease.

METHODS

Study Selection

A comprehensive literature search of PubMed and Scopus was performed in accordance with PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) guidelines. The search for publications was undertaken using the following keywords: "death after cranioplasty," "cerebral edema after cranioplasty," "brain edema after cranioplasty," "cranioplasty complications," and "decompressive craniectomy." When different terms were found, these were searched further to find more studies. The search extended to all available English language articles from 1960 to August 2017. Cases were included in the final review only in patients who had clinical and radiologic characteristics of MBSC. Further, the references for all search-selected articles were also reviewed for potential cases. The following studies were excluded: literature reviews, articles not available in full text, articles reporting different diseases from MBSC, articles focusing on different complications of cranioplasty, and articles with incomplete clinical information.

Data Extraction

The following information, if reported, was collected from eligible articles: 1) age, 2) sex, 3) indication for craniectomy, 4) type of craniectomy, 5) presence of sinking

of skin flap (SSF), 6) placement of cerebrospinal fluid (CSF) shunt, 7) craniectomy to cranioplasty time interval, 8) type of cranioplasty material, 9) time interval for manifestation of MBSC and presenting symptoms, 10) postoperative radiologic findings, 11) factors associated with MBSC, 12) treatment, and 13) outcome.

Not all articles provided information about each item; therefore, comparative analysis was limited by the nature of the source data. Statistical analysis was not conducted for this review because comparative analyses could not be performed.

RESULTS

The initial literature search yielded 534 articles. After removal of duplicates, the title and abstract of 366 articles were screened and 321 articles were eliminated based on exclusion criteria. After this initial filter, 45 articles were assessed for eligibility, of which 26 were excluded because of lack of evidence indicating MBSC. Thus, a total of 19 studies encompassing 26 patients were eligible for analysis (**Figure 1**). **Table 1** shows the characteristics of these patients who were identified from previous publications.[2-20]

Several terms were found to define MBSC, including malignant cerebral edema after cranioplasty, hemorrhagic infarction after cranioplasty, death after cranioplasty, sudden death after cranioplasty, reperfusion injury after cranioplasty, pseudohypoxic brain swelling after cranioplasty, massive cerebral swelling after cranioplasty, acute global ischemia after cranioplasty, stroke after cranioplasty, and allergic response to titanium after cranioplasty.

Patient Demographics

The youngest patient reported was 14 years old, and the oldest was 77 years old. The average age was 37.8 years. The largest number of patients were included in the group in their third decade and those older than 60 years (7 patients in each group). Seventy-two percent of patients were male. The indications for craniectomy were traumatic brain injury (TBI) in 52% and stroke in 44% of all patients. In 1 patient, the primary disease was tumoral. Stroke was the commonest indication for DC in the group of patients in old age.

Type of Craniectomy

The most common type of surgery was hemicraniectomy, which was undertaken in 21 patients (81%). In the remaining patients, a bifrontal craniectomy was performed. Hemicraniectomy was performed in every patient with stroke.

Presence of SSF

SSF was observed in every patient in whom the preoperative condition of the craniectomy site was clinically described or a preoperative computed tomography (CT) scan was shown in the article. CT scans were performed at different intervals related to the date of cranioplasty. In 45% of cases, the imaging study was performed in days close to the date of surgery, showing different degrees of cerebral deviation. On the other hand, the diagnosis of different degrees of SSF was performed based only on clinical description in 10 patients.

Presence of CSF Shunts

CSF shunts were implanted in 11 patients (44%). The description of specific details such as type of shunt, time of surgery, or symptoms usually were not mentioned in case descriptions. In addition, only a few studies showed a CT after the shunt was implanted.

Type of Cranioplasty Material

The type of graft material used for cranioplasty was reported in 22 patients. In 50% of cases, an autologous bone flap was used and in the other 50%, different synthetic materials were used, including titanium, methylmethacrylate, and polyetheretherketone.

Timing of Cranioplasty

The shortest time for cranioplasty was 1 month, and the longest interval was 17 months. In 43% of cases, cranioplasty was carried out during the first 3 months after craniectomy; in 13%, the surgery was performed between 3 and 6 months and the operation was performed between 9 and 12 months in 17% of cases; cranioplasty was performed after 12 months in 20% of cases.

Interval for Manifestation and Presenting Symptoms of MBSC

The time interval in which the first symptoms were detected was reported in 12 patients. A time range of 15 minutes to 16 hours was observed, with an average of 3.3 hours. In 5 patients, the symptoms initiated within the first hour after cranioplasty. Regarding the presenting symptoms in these patients, the most common manifestation reported was mydriasis and coma, which were observed in 75% of cases. The presence of seizures was observed in 30% of patients. Other less frequently observed symptoms were tachycardia, arterial hypertension, hypotension, and bradycardia.

Radiologic Findings

In all patients, imaging studies showed a severe grade of diffuse brain edema characterized by effacement of brain sulci and basal cisterns and midline deviation in some cases. In addition, different lesions such as intraparenchymal hemorrhages or ischemic zones were also observed.

Treatment and Outcome

In 65% of cases, some type of craniectomy was performed either by removing the implanted bone flap or performing a contralateral or bilateral craniectomy. In some patients, the craniectomy was associated with another procedure including lobectomy and ventriculostomy. A low percentage of patients were managed only medically with brain antiedema measures. In 4 patients, no treatment was used.

Mortality was observed in 88% of patients despite the treatment already described. The patients who survived remained with some degree of severe disability.

DISCUSSION

Although cranioplasty is commonly performed, it can be associated with several complications, such as infection, bone resorption, and hematomas. Another even rarer complication is MBSC. The pathophysiology of this entity is not completely understood and seems difficult to decipher. Consequently, several hypotheses (e.g., intracranial hypotension, reperfusion injury, hypoxia, autoregulation failure, use of closed suction drains) have been formulated to try to explain the