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# Osteoclastic craniectomy for scaphocephaly in infants results in physiological head shapes

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*Introduction:* Sagittal synostosis leading to scaphocephaly is the most common type of craniostenosis being operated. Different treatment options are known, but the optimal treatment method is still controversial. Head growth indicated by measurements of the head's circumference and cephalic index (CI) are valid surrogate parameters for normal head shapes in children. The aim of the study was to analyze if osteoclastic craniectomy (OC) in scaphocephaly children at four to ten months of age results in normal head shapes.

*Patients and methods:* Twenty-seven patients with scaphocephaly underwent OC between 2003 and 2011. The mean patient age at the time of surgery was 6.75 months. The body weight was between 6.1 and 9.3 kg, mean 8.0 kg. The average duration of surgery was 108 minutes. The mean blood loss during the procedure was 168 ml and the mean amount of erythrocyte transfusion was 152 ml. The mean time spent on the ICU was 1.48 days and the mean of total hospital stay was 5.81 days. The operative method is described. During the mean follow-up time of 6.3 years (min 3.8, max 10.4, median 7.1) focus was set on the patient's head growth and cephalic index (CI) following OC. For statistical reason the follow up period was divided into three groups: follow up 2-4 years, 5-7 years and 8-10 years.

*Results:* For all cases the total head growth was 9.5cm (mean) during the follow up period of 6.3 years. Analyzing the mean head growth by bootstrapping analysis, the three observational groups showed a significant increase of the head circumference in all cases being analyzed: group 1 $p=0.003$, group 2 $p=0.005$ and group 3 $p=0.028$ Evaluation of the CI showed a statistically significant change from a pathologic value of 0.67 (mean) preoperatively to a normal value of 0.78 (mean) postoperatively during the follow up analyzing all patients. To precise these findings, the bootstrapping analysis showed in the first period an increase of the mean CI not reaching statistical significance ($p=0.351$). Analyzing the second and third period the CI significantly increased in both groups ($p=0.016$ and $p=0.037$). All patients showed a nearly complete re-ossification during the follow up period. No secondary operation was necessary in any patient of this cohort.

*Conclusion:* As shown in this single-center observational study, the surgical intervention significantly improved the cephalic index and resulted in a symmetric head shape with excellent aesthetic appearance. The results were not dependent on postoperative helmet therapy, and compliance of caregivers. Re-ossification reached 100% within the observation period. According to these data, we recommend osteoclastic craniectomy as the method of choice in infants six to twelve months of age.



## 1. Introduction

Craniostenosis — the premature closure of cranial sutures — results in characteristic skull deformities. The shape of the deformity depends on the suture involved. Previous case reports have shown that some patients with craniostenosis may additionally develop papilledema due to elevated intracranial pressure during their first years of life (Stavrou et al., 1997; Florisson et al., 2010).

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Sagittal synostosis, with an incidence of 1:3000–5000 births, is the most common form of craniostenosis (Thomas et al., 2015). Typically, early sagittal suture closure presents as an elongated head characterized by a narrow and low occiput, a saddle-shaped parietal region, and noticeable frontal bossing (Hubli, 2014). Surgical options range from simple suture excision to total cranial vault remodeling (Renier et al., 1982; McCarthy et al., 1995).

Current surgical methods include osteoclastic craniectomy (OC) with or without open calvarial reconstruction, minimally invasive strip craniectomy with the use of a postoperative molding helmet or spring implantation, cranial distraction, and frontal bone cranioplasty (Binkiewicz-Glinska et al., 2016; Dempsey et al., 2019; Prevost et al., 2019). Different centers favor various procedures, so there remains no consensus on the preferred treatment option.

To add to this debate, we evaluated the intra- and postoperative courses and long-term outcomes following OC for sagittal suture synostosis in children.

The primary aim of our study was to investigate whether OC in 4–10-month-old children with scaphocephaly resulted in normal head shapes.

## 2. Materials and Methods

We analyzed data from 27 pediatric patients who underwent OC between 2003 and 2011. The patients' records and imaging results were retrieved from the institution's electronic archive.

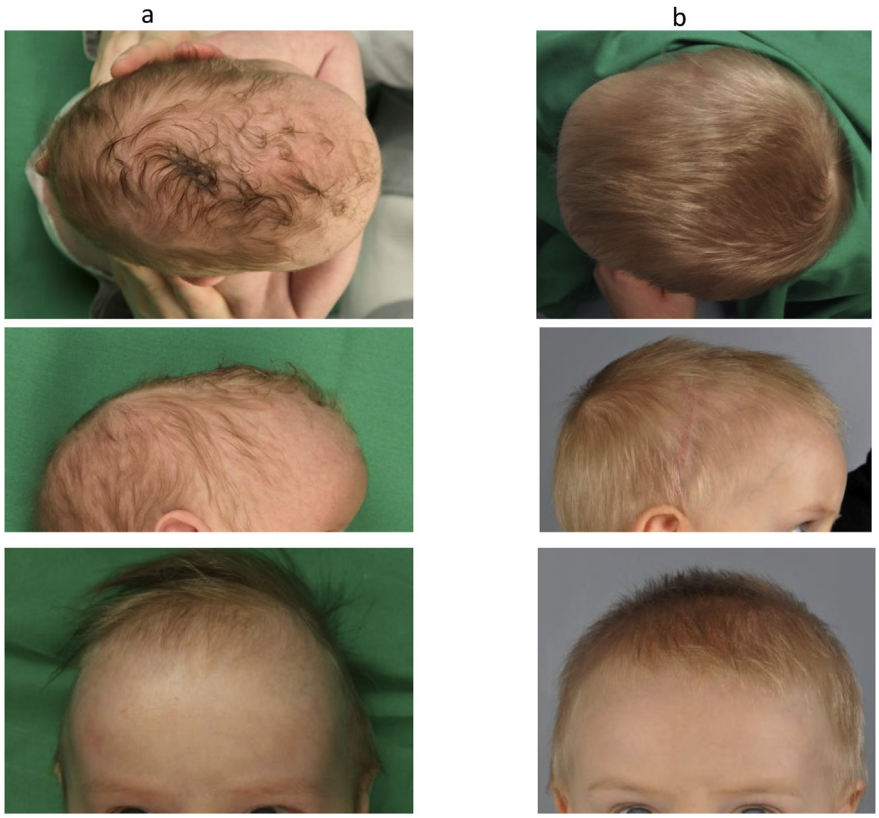
### 2.1. Study population

27 children (24 male and 3 female) were enrolled in this study. All patients presented with the typical clinical appearance of scaphocephaly (Photograph 1a): narrow and low occiput, saddle-shaped parietal region, and noticeable frontal bossing. The diagnosis was confirmed by ultrasound or plane X-ray examination. Fundoscopy was performed routinely to exclude the presence of increased intracranial pressure. All patients underwent OC between 4 and 10 months of age. The surgery was performed by two attending pediatric surgeons at our institution.

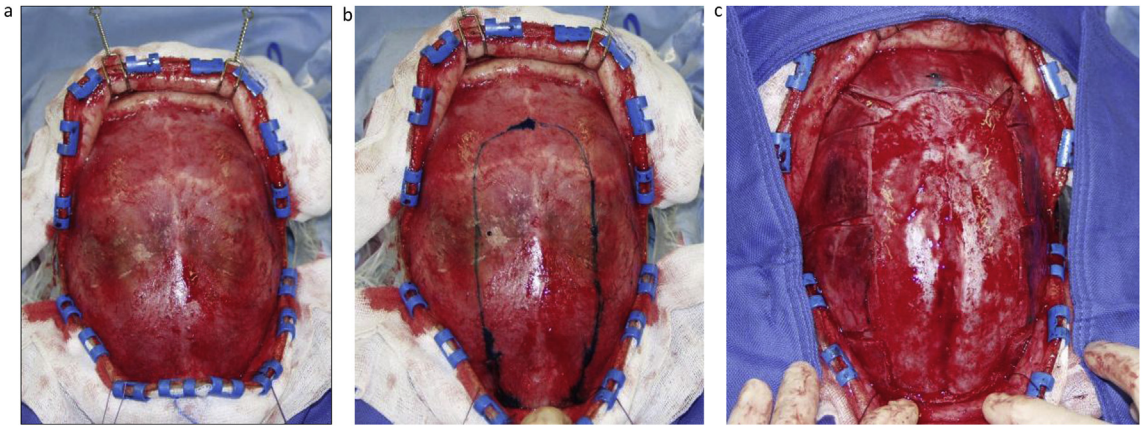
### 2.2. Surgical technique

OC comprises total resection of the calvarial bone margined frontal to the coronary sutures and rostral to the lambdoid sutures, as described by Hassler et al. (1990) (Hassler and Zentner, 1990). The procedure followed a standard protocol. A specialized pediatric anesthesiologist performed general anesthesia and was responsible for intraoperative blood management as well as vasopressor administration. Tranexamic acid was not administered in this cohort. For infection control, prepping was performed by the surgeon, and two pairs of gloves were worn throughout the procedure.

The operation commenced with a curved skin incision from ear to ear. Then the periosteum and skin were dissected from the calvarial bone, first to the front to expose the coronal sutures, then to the occiput to expose the lambdoid sutures (Photograph 2a). The craniotomy margins were set with methylene blue (Photograph 2b). OC was performed, beginning in front of the coronal suture and ending behind the lambdoid suture with V-shaped frontal, temporal, and parietal osteotomies, and with biparietal widening of the skull (Photograph 2c). The periosteum was closed with interrupted sutures and, depending on the amount of bleeding, up to two silicon drains were placed in the subcutaneous plane if required. The subcutis was closed with interrupted sutures, and the




**Photograph 1.** (a) Preoperative aspect of typical scaphocephalus in a 4-month-old patient. (b) Postoperative aesthetic outcome in a patient at the age of 15 months (8 months postoperatively).




**Photographs 2.** (a) After the curved skin incision, the coronary and lambdoid sutures were exposed. (b) Craniotomy margins were indicated with methylene blue. (c) Craniotomy was completed, beginning in front of the coronary sutures and ending behind the lambdoid sutures. V-shaped excisions were made at the frontal, temporal and parietal lobes on each side.

skin was closed with non-absorbable running mattress sutures. The expected blood loss was compensated by erythrocyte transfusion, following international guidelines for pediatric anesthesiology (Stricker et al., 2017).

After the patients regained consciousness following completion of the procedure, they were extubated immediately and transferred to the pediatric intensive care unit (ICU) until at least postoperative day 1. Postoperative analgesia (acetaminophen, dipyrone, and morphine) was administered, and the patients were transferred to the general pediatric surgery ward between postoperative days 1 and 3. Oral feeding was allowed immediately after the procedure, and cefuroxime was administered until the drains were removed. Patients were discharged on postoperative days 4–8, depending on the need for opioids for pain management.

### 2.3. Patient evaluation

Patients were examined in the outpatient clinic at 10 days, 6 weeks, 3 months, and 12 months postoperatively, and yearly thereafter.

The focus of our study during the follow-up period was normal and symmetric head growth. Therefore, at each consultation head circumferences and skull diameters were measured, which are necessary for calculating the CI and the cranial vault asymmetry index (CVAI).

Since the follow-up consultations were not at identical intervals, the children's ages at the consultations differed. We therefore did not compare first or second follow-up measurements but instead compared measurements at specific ages. We grouped the data into measurements taken when the children were 2–4 years, 5–7 years, and 8–10 years of age.

The level of reossification was evaluated by palpation of the skull. X-ray verification of reossification was deliberately avoided in order to reduce radiation exposure. Reossification was set to a level of >95% if no lesion greater than 1 cm was detected upon extensive physical examination. Irregularities of the new calvarial bone were also documented at these consultations.

At one time point — at least 3 years after the procedure — parents were interviewed via questionnaire about their satisfaction with the cosmetic result.

### 2.4. Statistics

Metric variables are described as the minimum, maximum, median, and mean. Statistical differences between groups were tested using Pearson's chi-squared test, with bootstrap analysis also applied.

Statistical analysis was performed using IBM SPSS Statistics 20.0 and Microsoft EXCEL 365. Statistical significance was set at an alpha level of $p \leq 0.05$. All identifying information was irreversibly removed from the data to maintain anonymity.

## 3. Results

Here we present the clinical and aesthetic results for 27 consecutive pediatric patients (24 boys and 3 girls) who underwent surgery for sagittal suture synostosis at our institution. The mean follow-up time was 6.3 years (min 3.8, max 10.4, median 7.1).

The mean patient age at the time of surgery was 6.75 months (min 4.6, max 10.2, median 6.4). The body weight of the patients was between 6.1 kg and 9.3 kg, with a mean of 8.0 kg (median 8.1). The average duration of surgery was 108 min (range 85–145, median 110). The mean blood loss during the procedure (as estimated by the operating surgeon) was 168 ml (min 130, max 200, median 170), and the mean amount of erythrocyte transfusion was 152 ml (min 100, max 180, median 150), as documented in the anesthesia record. Two patients also received fresh plasma. There were no adverse effects. Three patients required intraoperative catecholamines. The mean time spent in the ICU was 1.48 days (min 1, max 3, median 2), and the mean total hospital stay was 5.81 days (min 4, max 8, median 6). The postoperative course was uneventful in all cases, and no wound infection or CSF leakage was detected. A summary of these data is presented in Table 1.

**Table 1**
Patient parameters and perioperative course.

| Parameter | Min | Max | Median | Mean |
|---|---|---|---|---|
| Age at operation (months) | 4.6 | 10.2 | 6.4 | 6.75 |
| Body weight at operation (kg) | 6.1 | 9.3 | 8.1 | 8 |
| Operation time (minutes) | 85 | 145 | 110 | 108 |
| Blood loss (ml) | 130 | 200 | 170 | 168 |
| Transfusion of erythrocytes (ml) | 100 | 180 | 150 | 152 |
| ICU (days) | 1 | 3 | 2 | 1.48 |
| Total hospital stay (days) | 4 | 8 | 6 | 5.81 |
| Follow-up period (years) | 3.8 | 10.4 | 7.1 | 6.3 |

Initiation of reossification was observed at either the 6-week or 3-month follow-up visit using clinical examination, with bone islands first palpable in the temporal region. 22 patients (81.5%) showed nearly complete postoperative reossification (>95%) within 12–24 months. In five patients, the reossification process was slower but reached >95% during the follow-up period. In summary, >95% reossification of the calvaria bone was achieved in all patients during the follow-up period. No artificial bone grafts or reoperations were necessary in any of the cases. Typically, all of the reformed skulls showed bulging fingerprint-like irregularities of the new calvaria bone. These irregularities were recognized and did not disappear during the follow-up period in any of the patients.

### 3.1. Head circumference

For all cases, the mean head growth (increase in circumference) was 9.5 cm during the follow-up period when compared with the preoperative head circumference ($p = 0.002$; Fig. 2).

When mean head growth was analyzed by bootstrap analysis, the three age groups described showed a significant increase in head circumference after surgery in all cases: group 1, $p = 0.003$; group 2, $p = 0.005$; and group 3, $p = 0.028$ (Table 3). No centile shifts in head circumference were detected in any of the patients.

### 3.2. Cephalic index (CI)

Evaluation of CI showed a statistically significant change from a mean preoperative pathologic value of 0.6 (compared to Farkas et al. (2005)) to a mean postoperative value of 0.78 during the follow-up analysis of all patients ($p < 0.001$) (Table 2; Fig. 1).

To verify these findings, we performed a bootstrap analysis. Based on a 1000-time bootstrapping, there was an increase in the mean CI during the first period (group 1, 2–4 years), which did not reach statistical significance ($p = 0.351$) (Table 4). Analysis of the second time period (group 2, 5–7 years), and third time period (group 3, 8–10 years) indicated a significant increase in CI in both cases (group 2, $p = 0.016$; group 3, $p = 0.037$; Table 4). Thus, following OC, the reshaping process of the head seems to start immediately after the operation, with a statistically significant improvement reached 5 years later.

Parents and caregivers reported a high rate of satisfaction: 93.75% perceived an excellent or good outcome; an example is shown in Photograph 1b. 6.25% reported moderate satisfaction with the outcome, because they were unhappy with the visible scar.

### 3.3. Advantages of OC

OC reshapes the head to a normal aesthetic appearance. No molding helmets are necessary following this procedure because the brain itself reshapes the head, and this brain growth is symmetric. The surgery influences the outcome directly, with no dependence on patient or caregiver compliance.

## 4. Discussion

During the first year of life, the brain grows rapidly. Premature closure of the sagittal suture leads to characteristic stigmatizing skull deformities and can increase the risk of developing elevated intracranial pressure. Before the age of 12 months, the sutures that remain open compensate for the closed sutures and prevent intracranial hypertension. After the first year of life, this mechanism is diminished; heads with closed sutures remain smaller than those of healthy children, and the risk of intracranial hypertension becomes eminent.

By the end of the second year of life, the brain has reached 80% of its final size (Zöller, 2003). Early surgical intervention for premature suture closure within the first year of life prevents elevated intracranial pressure and the formation of severe skull deformities (Renier et al., 1982; McCarthy et al., 1995). Craniostenosis repair in children with scaphocephaly normalizes the intracranial volume as well as the shape of the head (Mertens et al., 2017). The periosteum

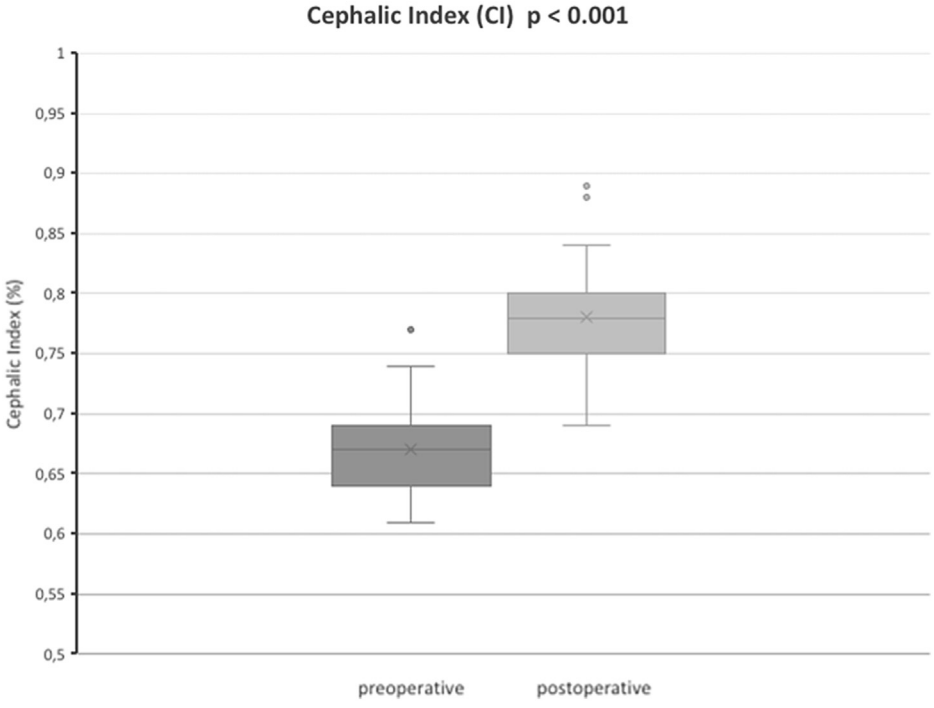



**Fig. 1.** Preoperative versus postoperative cephalic index (CI), $p < 0.001$.

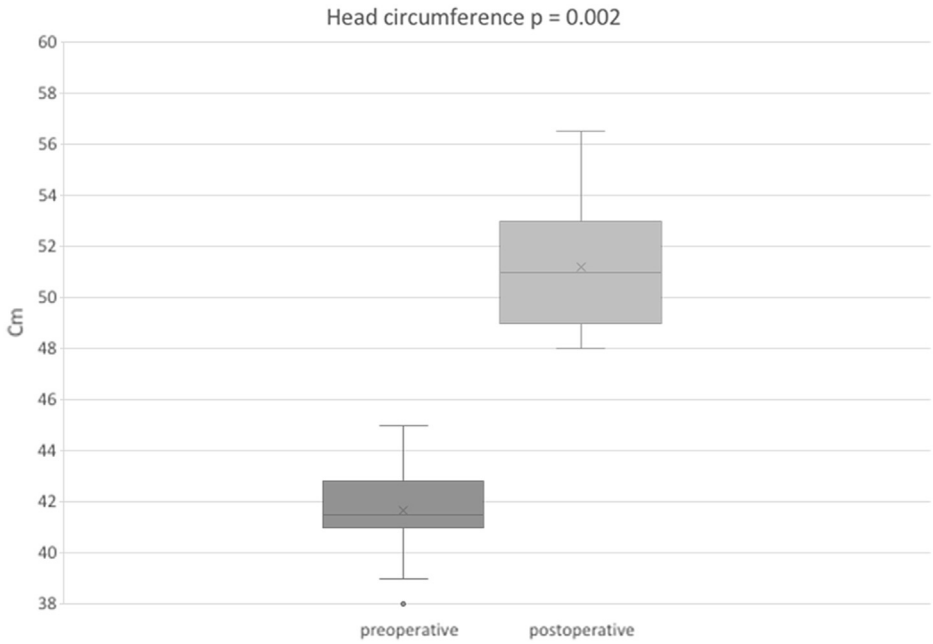



**Fig. 2.** Preoperative versus postoperative head circumference, $p = 0.002$.

and dura mater are anatomical structures that can potentially induce the reossification process.

Head circumferences in our study population showed a significant postoperative increase, with postoperative measurements showing a symmetric head shape. The pathological CI increased to the normal range during the follow-up period. Both parameters (head growth and CI normalization) are reliable indicators for a valid and successful operation in children with scaphocephaly (Mertens et al., 2017; Sharma et al., 2018), and are therefore valid surrogate parameters for assessing craniostenosis repair in children.

**Table 2**
Anthropometric parameters at the end of follow-up, calculated for every patient.

| Parameter | Min | Max | Median | Mean | *p* |
|---|---|---|---|---|---|
| Preoperative cephalic index (CI) | 0.61 | 0.77 | 0.67 | 0.67 | <0.001 |
| Postoperative cephalic index (CI) | 0.69 | 0.89 | 0.78 | 0.78 | <0.001 |
| Cranial vault asymmetry index (CVAI, %) | 0 | 6 | 3 | 2.63 | – |
| Absolute growth of head circumference | 3.8 | 15 | 10 | 9.5 | – |
| Preoperative head circumference (cm) | 38 | 45 | 41.5 | 41.68 | 0.002 |
| Postoperative head circumference (cm) | 48 | 56.5 | 51 | 51.19 | 0.002 |

**Table 3**
Bootstrap analysis of head circumference, groups 1–3.

| | | Head circumference (cm) | Delta, mean (cm) | *n* | *p*-value |
|---|---|---|---|---|---|
| Group 1 (2–4 years) | Prior to intervention<br>Follow-up | 41.2<br>50.2 | 9 | 27 | 0.003 |
| Group 2 (5–7 years) | Prior to intervention<br>Follow-p | 41.5<br>51.5 | 10 | 20 | 0.005 |
| Group 3 (8–10 years) | Prior to intervention<br>Follow-up | 42.2<br>51.76 | 9.66 | 15 | 0.028 |

**Table 4**
Bootstrap analysis of CI, groups 1–3.

| | | Cephalic index (cm) | Delta, mean (cm) | *n* | *p*-value |
|---|---|---|---|---|---|
| Group 1 (2–4 years) | Prior to intervention<br>Follow up | 0.67<br>0.72 | 0.05 | 27 | 0.351 |
| Group 2 (5–7 years) | Prior to intervention<br>Follow up | 0.67<br>0.74 | 0.07 | 20 | 0.016 |
| Group 3 (8–10 years) | Prior to intervention<br>Follow up | 0.67<br>0.78 | 0.11 | 15 | 0.037 |

The results indicated that patients undergoing OC had normal head growth with no significant differences during follow-up when compared with head growth in otherwise healthy children. Likus et al. defined a CI of 81.45 ± 7.06 as the normal range (Likus et al., 2014). The mean postoperative value of the CI in our cohort was 78.1, which is within this range. Our data were consistent with recent publications by Mertens et al. and Sharma et al. (Mertens et al., 2017; Sharma et al., 2018).

With total cranial fault remodeling, the CI improves immediately after surgery. Strikingly, in children who underwent OC, neither the head circumference nor the CI were directly affected postoperatively because of the surgical technique performed. Our data demonstrated that during a mean follow-up period of 6.3 years, both head circumference and CI were normalized. To the best of our knowledge, this is the first study to report this phenomenon.

The mean postoperative CVAI in our series was 2.63, which indicated that a nearly symmetric head shape was obtained (Yoo et al., 2012).

The mean duration of surgery for minimally invasive craniosynostosis repair ranges from 63 min (van Veelen et al., 2018) to 75 min (Chan et al., 2013). Our operation time for OC was somewhat longer, with a mean of 108 min, with duration directly proportional to blood loss, which is an important consideration (Seruya et al., 2012). However, in our opinion, the slightly longer operation time is more than compensated for by the excellent aesthetic appearance and normal head growth after surgery.

In endoscope-assisted interventions, children must wear a modeling helmet for several months. This is a significant burden for the families and affects everyday activities, as recently analyzed by Yan et al. (2018). Yan argues that helmet-based therapies stigmatize children and their families in social settings and present a risk for decubiti and noncompliance (Yan et al., 2018). In 2017, Nguyen et al. analyzed 100 children treated with the endoscopic method and concluded that helmet therapy was the most important factor in the success of that method (Nguyen et al., 2017). Noncompliance jeopardizes the cosmetic results of the procedure. Following OC, the growth of the brain automatically reshapes the head. Because this growth is symmetrical, excellent cosmetic results are certain. The surgery directly influences the outcome, and there is no dependence on patient compliance.

Many authors view the endoscopic technique and helmet therapy as a viable therapeutic option for children up to the age of 3 months (Jimenez and Barone, 1998; Persing, 2004; Jimenez and Barone, 2000). Since most patients — including those seen at our institution — present at a later age, we recommend OC as an excellent option for children who are too old for the endoscope-assisted method.

Freudlsperger et al. published an article on the different therapeutic options for patients with craniostenosis and concluded that the open surgical technique remains the preferred technique in most large centers (Freudlsperger et al., 2015). Another argument in favor of the open procedure was published by Chan et al. who argued that the risk of intraoperative complications during endoscopic intervention is increased because of poor visibility (Chan et al., 2013).

### 4.1. Limitations of the study

The limitations of this study are its retrospective design and the relatively small number of patients owing to the low incidence of this disease. Furthermore, we could not include a control group because it would be unethical to not perform surgery on patients who needed it or to include children who were unable to undergo the procedure for various reasons.

We consciously accepted subjective data concerning the reossification process because we considered radiation protection to be a priority. Therefore, the thickness and quality of the new bone could not be analyzed in this study. 'Black bone' magnetic resonance imaging (MRI) has been described as a potential alternative to radiation-based head scanning studies in infants and children (Eley et al., 2017a, 2017b). For future investigations of OC this non-ionizing method, which avoids the risk of malignancies (Little et al., 2018), could be used to quantify the reossification progress in children.

## 5. Conclusion

This study demonstrated that OC results in symmetric head shapes with normal head growth during the follow-up period. No helmet therapy is necessary, and the outcomes do not depend on caregiver compliance. OC is a feasible and safe treatment option in children aged 4–10 months with scaphocephaly. To date, no long-term data comparing the open and endoscopic approaches have been published. Further investigation with large cohorts will be necessary to draw a final conclusion in this ongoing debate.

### Study design

Retrospective; level of evidence, 3.

### Disclosure of potential conflicts of interest

The authors declare that they have no conflicts of interest.

### Ethical approval

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. This article does not contain any studies with human participants or animals performed by any of the authors.

### Informed consent

Informed consent was obtained from the parents of all individual participants included in the study.

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