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Original Article

Impact of extra-axial cerebrospinal fluid collection in frontal morphology after surgical treatment of scaphocephaly

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Abstract

Background: Infants with sagittal suture synostosis often present a pathologic dilatation of subarachnoid spaces. The impact of such subarachnoid spaces' enlargement in the morphology of the skull, especially on the forehead and on the surgical outcome, was analyzed.

Methods: Children less than 6 months of age undergoing a surgical correction of the scaphocephaly with Renier's H technique between 2003 and 2008 were included in the study. In these patients, preoperative and postoperative fronto-occipital diameter (FOD), biparietal diameter (BPD), temporal width (TW), and naso-frontal angle (NFA) were measured. Cranial index (CI) and the difference between preoperative and postoperative CI (ΔCI) were calculated. Preoperative cranio-cortical width (CCW) was measured to analyze the subarachnoid spaces' volumes. The children here considered were then divided into two groups: Group 1 with CCW within normal estimated value corrected for age and Group 2 with CCW larger than estimated normal value.

Results: About 159 children were enrolled (72.3% male). CCW was larger than expected in 95 children (59.8%). A positive correlation between CCW and BPD ($P \leq 0.001$) and a negative correlation between CCW and NFA ($P \leq 0.001$) were found. When comparing the two groups, the mean age at preoperative computed tomography (CT) scan was 121 days in Group 1 and 110 days in Group 2. The mean age at operation was 130 days in Group 1 and 123 in Group 2. The mean age at postoperative examination (RX or CT scan) was 53.4 months in Group 1 and 51.8 months in Group 2. Preoperatively, the mean BPD, TW, and CI were significantly larger in Group 2 ($P \leq 0.01$), whereas the NFA was significantly narrower ($P = 0.03$). Postoperative analysis showed that ΔCI was statistically different between the two groups (Group 1: 10%,

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Group 2: 7%; $P < 0.04$). The duration of follow-up period ranged between 19 and 129 months.

Conclusion: Two main subtypes of forehead of infants with scaphocephaly may be distinguished. Indeed, the morphology of the forehead differs when a pathologic subarachnoid spaces' enlargement is present preoperatively and it also affects the postoperative evolution. Such observation highlights the importance of evaluating whether subarachnoid spaces are enlarged when planning a surgical correction in isolated sagittal suture synostosis.

Key Words: Craniostenosis, isolated, nonsyndromic, outcome, prognosis, sagittal suture, surgical correction, results

INTRODUCTION

Scaphocephaly by sagittal suture synostosis is the most common form of isolated craniosynostosis, accounting for 40%–60% of cases of craniosynostosis.[8,9,11] The skull is typically reduced in its biparietal length and elongated in the anteroposterior diameter, with frontal bossing and prominence of occipital pole. There is a pronounced reduction in cephalic index and an increase in the head circumference.[14]

The deformation may raise an aesthetic concern, and in some of patients an increased intracranial pressure (ICP) may be observed.[2,13,31] Alterations in cognitive development and speech delay have been reported with minor language anomalies found in up to 30%–40% of the cases.[4]

There is a strong consensus on the time of surgery, with best functional and cosmetic results achieved within the first 6 months of life.[24] In fact, the importance of an early surgery has been reiterate in literature since the 50s,[1,24,28,32] given the ability of growing brain to change the cranial morphology after surgery.[23,25] Moreover, studies concerning the mental development demonstrated a better IQ in patients surgically treated at an early age.[2]

When performed, neuroimaging investigations may find some associated features such as copper beaten skull, fingerprinting, and altered anatomy in venous drainage.[3,19,26] In infants with isolated scaphocephaly, an enlargement of the subarachnoid spaces is commonly observed especially in the frontal region and less frequently in the occipital region.[12,18,20,21,34] Does this increased volume of the subarachnoid spaces have an impact in the typical feature of the forehead (frontal bossing) frequently noticed at presentation? Does it have an impact on the results of the surgical correction of the scaphocephaly?

To answer these questions, we analyzed the surgical results obtained in the surgical management of scaphocephaly at the pediatric Neurosurgical Department at Necker Enfants Malades in Paris.

PATIENTS AND METHODS

We retrospectively reviewed the medical files and imaging studies of patients with isolated sagittal synostosis treated at the Hôpital Necker Enfants Malades between 2003 and 2008 operated on with the same surgical technique ("Renier's H technique")[11] [Figure 1] before 6 months of age. Children addressed to our Center with a digitized skull computed tomography (CT) scan were included.

The following selection criteria were adopted: toddlers younger than 6 months of life and isolated sagittal suture synostosis, without intracranial anomalies other than an enlargement of subarachnoid spaces. Children with other cranio-facial malformations, multisutural synostosis, or brain anomalies were excluded. All patients operated on with a different surgical technique were excluded as well.

Data collection

For each patient, CT scan and cranial X-rays were collected. The following pre- and postoperative parameters were measured: naso-frontal angle (NFA), fronto-occipital

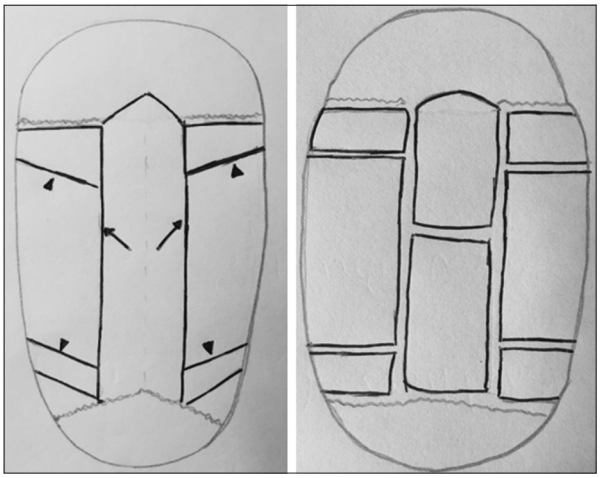

Figure 1: Left. Osteotomies in Renier's H technique. 4 cm-sagittal strip on the midline is generally performed (*arrow*) with four triangular retrocoronal wedges (two retrocoronal and two prelambdoid-*head of arrows*). Right. sagittal strip is removed and then shortened to adapt to new fronto-occipital diameter. Lateral bone flaps are "green-stick" fractured at the base

diameter (FOD), biparietal diameter (BPD), and temporal width (TW) as a bipterional diameter (TW). All these parameters were calculated with Carestream Software® on preoperative CT scan.

FOD, BPD, and TW were measured in axial slices from one inner table to the other inner table and the NFA in the sagittal reconstruction of CT [Figure 2]. The same parameters were then measured on CT scan or X-ray image performed during follow-up. Cranial index (CI) is calculated as the ratio between BPD and FOD, and the difference between preoperative and postoperative CI in each group (defined as ΔCI) was then calculated.

The dimension of subarachnoid spaces was calculated as cranio-cortical width (CCW), which is the widest vertical distance between brain surface and calvarium. The classical threshold of 4 mm adopted as physiological parameter according to Libicher *et al.*[21] was corrected for age following the formula 2.32845 + 0.208036 × (age) × 0.003709 × (age) according to Lam *et al.*[20]

The patients were then divided into two groups: the first group (Group 1) was composed of infants with an enlargement of subarachnoid spaces that are smaller or equal than the threshold corrected for age ("physiological" enlargement) and the second group (Group 2) was composed of infants with a greater enlargement ("pathological" enlargement).

Statistical analysis

Data were collected using Microsoft Excel 2010®, and statistical analysis was performed with SPSS® and JMPPro 9 ® software. The *t*-test was used to compare the preoperative and postoperative measurements between groups. Kruskal–Wallis test was used to compare ΔCI. A significant level of $P < 0.05$ was considered for all analyses.

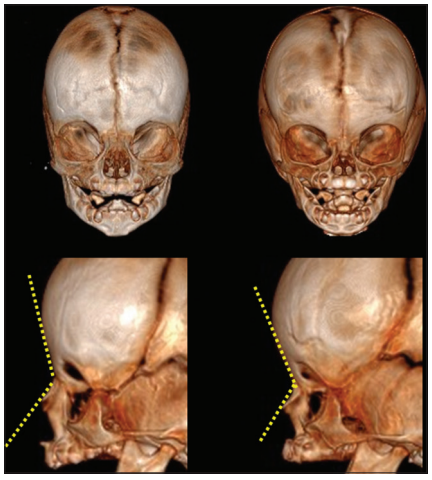

Figure 2: Patient from Group 1 (*left*) and from Group 2 (*right*). The first one present a more narrow biparietal diameter (*upper*) with a large naso-frontal angle (*lower*). Patient from Group 2 shows a larger transverse diameter and small naso-frontal angle

RESULTS

Data are summarized in Table 1.

Population

Seventeen among the 176 patients who met the selection criteria had some incomplete imaging studies or were lost at follow-up; thus, finally 159 children were considered in this study.

There was a clear male predominance (115 patients, 72.3%) as expected. The median age at CT scan was 115 days (min. 82 days, max. 164 days). The median age at operation was 125.5 days (min. 82 days, max. 177 days). No perioperative differences were found during surgery within the two groups. In particular, no dural tears were recorded in this series in either group. The follow-up period ranged between 19 and 129 months.

According to the enlargement of subarachnoid spaces, 64 patients (40.2%) were in Group 1 ("physiological") and 95 patients (59.8%) in Group 2 (pathological) [Figure 3].

The two groups were comparable in terms of age at surgery and age at CT examination as well as age at postoperative control. The mean age at intervention was 130 days in Group 1 and 123 days in Group 2. The mean age at preoperative CT scan was 121 days in Group 1 and 111 days in Group 2. The mean age at postoperative RX or CT scan was 53.4 and 51.8 months for Groups 1 and 2, respectively.

Measurements

When considering the overall series, a positive correlation between CCW and BPD was found, $R^2 = 0.086$,

Table 1: Synopsis of the data

	Group 1 (64 pts)	Group 2 (95 pts)	*P*
Sex and age data			
Gender	49M, 15 F	66 M,29 F	
Mean age at intervention	130	123	
m.a. at preop. CT-scan	121 days	111 days	
m.a. at postop XR or CT scan	53.4 mo.	51.8 mo	
Preoperative features			
BPD	9.6 cm	9.9 cm	0.009
TW	8.2 cm	8.5 cm	0.008
NFA	130.42°	125.68°	0.03
FOD	15.4 cm	15.2 cm	0.19
CI	62%	65%	0.002
Postoperative features			
BPD	13.5 cm	13.4 cm	0.69
TW	10.5 cm	10.8 cm	0.22
NFA	131.4°	130°	0.15
FOD	18.9 cm	18.6 cm	0.28
CI	72%	72%	0.17
ΔCI	10	7	0.034

P = 0.0002) [Figure 4a]. Conversely, between CCW and NFA, there was a negative correlation: R^2 = 0.0944, P = 0.0001 [Figure 4b].

When distinguishing the two groups, the mean preoperative BPD and the TW were significantly larger in Group 2 (9.9 vs 9.6 mm, P = 0.009 for BPD and 8.5 vs 8.2 mm, P = 0.008 for TW, for Groups 1 and 2, respectively). The mean NFA was significantly narrower in children of Group 2 (130.42° vs 125.68°, P = 0.03).

Group 1 exhibited larger FOD, but this result was not statistically significant (15.4 vs 15.2 mm, P = 0.19). CI was 62% in Group 1 and 65% in Group 2 (P = 0.002).

No differences between all postoperative measurements were observed between the two groups (13.5 vs 13.4 mm for BPD, 18.9 vs 18.6 mm for FOD, 131.4° vs 130° for NFA, 72% vs 72% for CI, each P > 0.1). ΔCI in Group 1 was 10, and in Group 2 it was 7. The difference between ΔCI was statistically significant (P = 0.034).

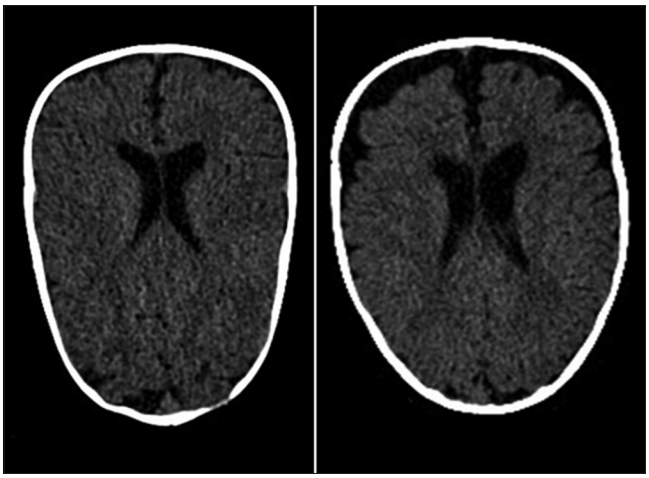

Figure 3: Patient from Group 1 (*left*) and from Group 2 (*right*). In Group 1 subarachnoid spaces are normal for age. In Group 2 there is a pathologic subarachnoid space enlargement

DISCUSSION

Subarachnoid spaces vary widely during the first 2 years of life. An enlargement of subarachnoid spaces is considered as physiologic in the first months of life. It increases from birth up to about 7 months of age;[20] after this period, a gradual decline is generally observed.[18] Ultrasonographic brain studies performed in a population of 278 full-term children demonstrated the particular ascending–descending trend of subarachnoid spaces' dimensions with a peak of enlargement around the seventh month of life followed by a progressive reduction.[20]

Subarachnoid spaces can be measured as CCW – the vertical distance between brain surface and calvarium, inter-hemispheric width (IHW), and sinocortical width (SCW) on a coronal plane.[20] No consensus exists concerning the upper limits of the "normal" measurements because of physiological changes during the first months of life. In the literature, the subarachnoid spaces are considered as normal up to 4 or to 10 mm for CCW. The upper limit of SCW ranges from 2 to 10 mm and for IHW from 6 to 8.5 mm.[12,20,34] However, a recent statistical analysis performed on 89 healthy infants proposed an upper limit of 4 mm for "physiological" CCW based on 95th percentile.[12] Lam *et al.*[20] further defined the "physiological" upper limit of CCW according to age. We thus used this age-corrected cut-off value in this work; patients were classified into two subgroups depending on the extension of subarachnoid spaces' enlargement.

A pathologic enlargement of subarachnoid spaces is considered as common in sagittal craniosynostosis. Our findings are consistent with the literature where up to 70% of sagittal synostosis presents this abnormal condition.[15,22]

A mechanism of venous compression has been postulated in the literature concerning such enlargement of

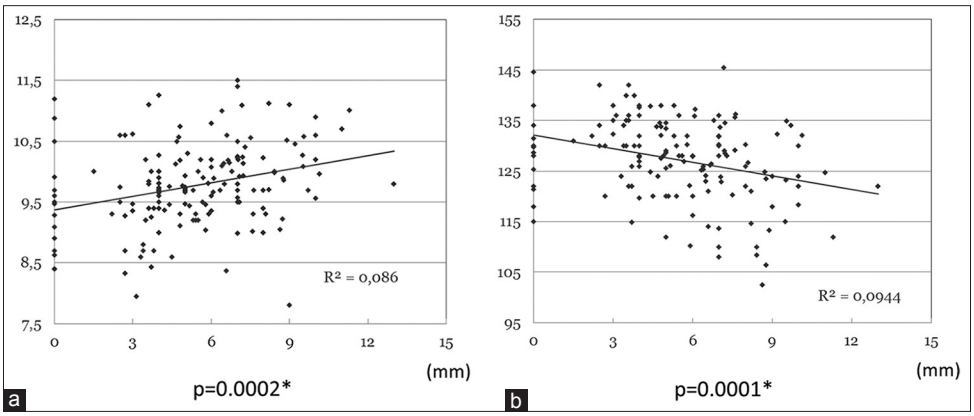

Figure 4: (a) Positive correlation between CCW and biparietal diameter, R^2=0.086, p=0.0002. (b) Negative correlation between CCW and NFA, R^2=0.0944, p=0.0001

subarachnoid spaces: the compression of sagittal sinus would result in a consequent alteration of cerebrospinal fluid (CSF) absorption.[15] Chadduck *et al*. who studied the distribution of subarachnoid spaces (SS) enlargement[7] contested the theory of impaired CSF absorption because such mechanism should also determine a ventricular enlargement and an uniform dilatation of extra-axial cerebrospinal spaces, whereas dilatation of subarachnoid spaces is mainly frontal in scaphocephaly. A hydrodynamic mechanism was hypothesized: the transmission of brain pulsation through the fluid-filled and dilated subarachnoid space could generate the compensatory skull growth. It should be considered as a factor able to change the cranial morphology. To our knowledge, no studies were performed in literature concerning the role of subarachnoid space enlargement in modifying the cranial shape and morphological outcome after surgical treatment for sagittal suture synostosis. However, the understanding of the origin of pericerebral dilatation lies outside of the purpose of our study.

Our analysis shows that the presence of an enlargement of the subarachnoid spaces is associated with a modification of the shape of the forehead. Correction of the shape and bulging of the forehead is indeed one of the most challenging problems in surgical management of scaphocephaly. The amelioration of forehead aspect can be obtained with the sole growing brain potential within sixth month of life,[11] but it often requires a more extensive and complex treatment in old patients.[27,30] Depending on the age of patients at surgery, the subtype of sagittal synostosis, and the main pathologic aspect of the skull, several surgical techniques have been propounded to treat this form of craniosynostosis:[10] small linear suturectomies or craniotomies,[29] simple and modified pi-procedure,[5,16] spring-assisted cranioplasty,[33] extensive calvarial vault remodeling,[17] and endoscopic strip craniectomy.[6]

In this series, all the patients in both groups were operated on with the same surgical technique, that is, "Renier's H technique," that is, a 4-cm strip craniectomy from bregma to lambda and temporal/parietal bone flap osteotomies.[11] It allows a good exposition of the whole cranial vault with an immediate shortening of FOD and enlarging the transverse diameters.

The homogeneity of population is also demonstrated by the mean age at intervention (123 in the second group and 130 in the first group). These elements also minimize the physiological difference in enlargement of subarachnoid spaces related to age.

Given the uniformity of population, two types of foreheads could be distinguished in scaphocephaly.

In the first group of sagittal synostosis, there is a predominant narrow bitemporal and biparietal width, a long FOD, and a large NFA. In this group, the CI is particularly low and the form can be defined cosmetically "severe." Subarachnoid spaces are virtual or comparable to normal children.

Conversely, children in the second group present a large and prominent frontal bossing, with a narrow NFA. These features are associated with dilated subarachnoid spaces. In these infants, the CI is milder than in the first group.

These findings suggest that despite a common anatomical closure of the sagittal suture, the so-called "compensatory growth" of the skull differs with two main forms: one with large subarachnoid spaces and prominent forehead and another with small subarachnoid spaces and severe alteration in CI, thus the brain may be differently affected in children with scaphocephaly according to these two subtypes.

These two subtypes respond differently to the same surgery. Patients without a pathologic enlargement of subarachnoid spaces (Group 1) exhibit only a minor improvement of the forehead with a non-significative amelioration in the postoperative NFA, whereas patients in Group 2 show a major modification in NFA. These data also suggest that it is possible to try and "foresee" how the forehead might respond to a craniofacial retrocoronal procedure and to determine in which subgroup of patients with isolated sagittal suture synostosis the aspect of the forehead might not change significantly. However, the final measurements are similar in the two groups showing that H technique can be used in both subtypes.

The main limit of this study remains the analysis of the evolution of the subarachnoid spaces. No CT or magnetic resonance imaging was performed routinely in the postoperative period in the population herein studied; for such a reason, it is not possible to assess the evolution of the enlargement of subarachnoid spaces after surgery. Because no ICP recording was systematically performed, it is not possible to associate one or other CSF pattern with an actual risk of developing a raised ICP.

Further prospective studies are needed to analyze the impact of surgical correction on subarachnoid spaces.

CONCLUSION

A pathologic subarachnoid space enlargement is found in two-thirds of scaphocephalies. The presence of such extra-axial CSF collection in these patients has a significant cosmetic impact on the preoperative morphology of the forehead and on the response to surgical correction.

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Conflicts of interest
There are no conflicts of interest.

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