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ORIGINAL PAPER

Length of synostosis and segmented intracranial volume correlate with age in patients with non-syndromic sagittal synostosis

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Abstract
Purpose The aim of this study is to compare the length of synostosis and segmented intracranial volume (SIV) with age in children with non-syndromic sagittal synostosis.
Methods Thirty-three consecutive patients (22 boys) who had been operated by cranial vault remodeling because of sagittal synostosis were compared retrospectively from 3D-CT imaging data sets obtained from volumetric CT. The mean age of the patients at preoperative CT imaging was 0.49 (range 0.13–1.3) years and at 1-year postoperative imaging 1.8 (range 1.3–3) years. The mean interval between preoperative CT imaging and surgery was 0.25 (range 0–0.8) years. Pearson's correlation and Student's *t* test were used in the statistical analyses.
Results Length of sagittal synostosis correlated positively with age at preoperative CT ($r = 0.688$, $p < 0.01$). Children with total synostosis ($n = 9$) were significantly older (mean age 0.74 vs. 0.4 years, $p < 0.01$) than those with partial synostosis. Of partial synostoses, 9 were located anteriorly, 3 in the middle, and 12 posteriorly. The mean synostosis ratio (synostosis length/total sagittal suture length × 100) was 83%. Preoperative SIV correlated positively with age at preoperative CT ($r = 0.788$, $p < 0.01$), whereas the 1-year postoperative SIV did not correlate with age at operation. The older the child at the time of the operation, the less the percentage SIV increased.
Conclusions Length of sagittal synostosis and SIV increased with age.

Keywords Intracranial volume · Craniosynostosis · Synostosis length · Scaphocephaly

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Introduction

Synostosis of the sagittal suture or scaphocephaly is the most frequent form of craniosynostosis, accounting for 55–60% of all craniosynostoses [1]. Non-syndromic craniosynostosis of the sagittal suture accounts for the majority of all sagittal synostoses [2].

The timing of sagittal suture fusion is variable; it can occur anytime from late first trimester to the early postnatal period [3]. The typical closure pattern in non-syndromic craniosynostosis is that the central part of the sagittal suture fuses first, followed by fusion of either the anterior or the posterior section [1, 4]. According to the fusion sequence, different craniofacial shapes are observed [4]. Typical abnormal head shapes include elongation of the cranial vault, prominent forehead and occiput, variable ridging of the sagittal suture, and increased head circumference.

Sagittal synostosis may lead to changes in intracranial volume (ICV) and intracranial pressure. Findings of previous studies of ICV are conflicting [5–10]. In addition, it is not clear whether age affects length of fusion of the sagittal suture and ICV.

The aim of this study was to compare length of sagittal suture fusion and segmented intracranial volume (SIV) obtained from volumetric CT with age in children with non-syndromic sagittal synostosis.

Patients and methods

The study group consisted of 33 consecutive children (11 girls, 22 boys) with operated non-syndromic isolated sagittal synostosis. All operations were performed by the same senior surgical team at the Cleft Palate and Craniofacial Center, Helsinki University Hospital, during 2007–2012. The surgical procedures aimed to maximally increase ICV by means of cranial vault remodeling. The mean age at surgery was 0.73 (range 0.34–2) years. Six children were operated on after the age of 1 year. As a tertiary referral center, we obtain referrals nationwide. After the patient was referred to our center, corrective surgery was done without delay. However, the patient's age at surgery depended on the age of referral. The need for surgery was based on the combination of radiological and typical clinical findings. Additional examinations such as fundoscopy or intracranial pressure monitoring area not regularly performed at our center.

Preoperative and 1-year postoperative CTs were used in the analysis. The 3D-CT imaging data sets were archived in a form accessible for retrospective analysis. Mean age of the patients at preoperative CT imaging was 0.49 (range 0.13–1.3) years and at 1-year postoperative imaging 1.8 (range 1.3–3) years. The mean interval between preoperative CT imaging and surgery was 0.25 (range 0–0.8) years.

All cranial CTs were obtained using a 64-slice scanner (LightSpeed VCT, GE Medical Systems, Milwaukee, WI, USA) with the following parameters: helical full, 0.5 s rotation time, increment 39.37 mm/rotation (pitch 0.984:1), 100 kV 40 mA and 120 kV 50 mA tube current for those aged less than and over 1.5 years, respectively. Images were reconstructed 0.625-mm thick at 0.312-mm intervals.

The study protocol was approved by Helsinki University Central Hospital. Principles outlined in the Declaration of Helsinki were followed.

Surgical method

An expansive cranial vault remodeling from the forehead to the occiput was performed with the patient placed in a prone position, similarly as described previously by Salyer and Marchac [11]. Bioresorbable polylactide plates and screws were used for fixation after craniectomy and removal and reshaping of the calvarial bones. Thus, an extensive 10- to 20-mm perioperative dead space in the region of the parietal eminences was created between the dura and the fixed bony frame.

Measurement of the sagittal suture/synostosis length, SIV measurement

The ICVs were measured using GE Advantage Workstation 4.4 with VolumeShare. The segmentation workflow was repeated identically for each patient and included steps of manual cut, automated, and density-based segmentation as follows: structures below the skull base and foramen magnum were cut. After bone removal (− 90 to + 155 HU thresholding), all bridges connecting intra- and extracranial soft tissue were cut (erode-open bridges-dilate-close holes). All extracranial soft tissue was then removed using manual cut and auto select tools. Finally, to avoid errors, the segmented volume was verified from axial slices, inspected in VR, and measured.

Statistical analysis

Pearson's correlation and Student's *t* test were used in the statistical analyses.

Results

Length of sagittal synostosis correlated positively with age at preoperative CT ($r = 0.688$, $p < 0.01$, Fig. 1). Children with total synostosis ($n = 9$) were significantly older (mean age 0.74 vs. 0.4 years, $p < 0.01$) than those with partial synostosis. Of partial synostoses, 9 were located anteriorly, 3 in the middle, and 12 posteriorly. All children older than 0.7 years had had total synostosis. The mean synostosis ratio (length of synostosis/total sagittal suture length × 100) was 83% (range 29.7–100).

Preoperative SIV (mean 906.79, range 589–1256 cm^3) correlated positively with age at preoperative CT ($r = 0.788$, $p < 0.01$, Fig. 2). The smallest SIVs were observed in children whose synostosis was located in the middle of the sagittal suture and the largest in those with total synostosis (Fig. 3).

One-year postoperative SIV (mean 1272.9, range 1037–1679 cm^3) did not correlate with age at operation. The older the child at the time of the operation, the less the percentage SIV increased. Age at operation correlated negatively with percentage increase in postoperative SIV ($r = -0.668$, $p < 0.05$).

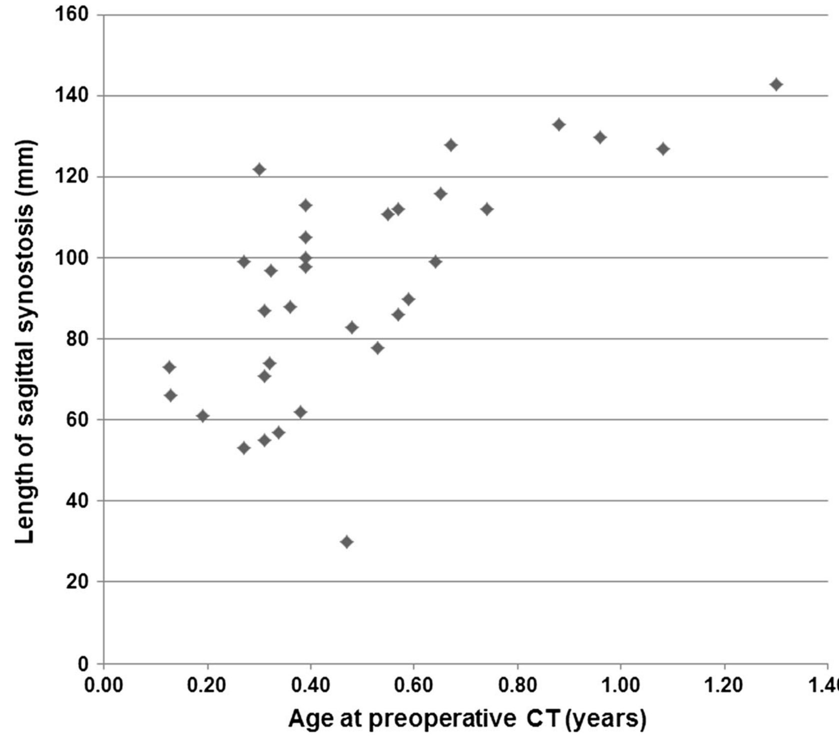
Discussion

We examined the relationship between age and length of synostosis and SIV in patients with single sagittal suture synostosis. Cranial vault volume and length of ossified suture have been used as a means of understanding potential growth restriction and are of concern when considering corrective surgery. Both SIV and length of synostosis were found to increase with age.

Length of sagittal suture synostosis

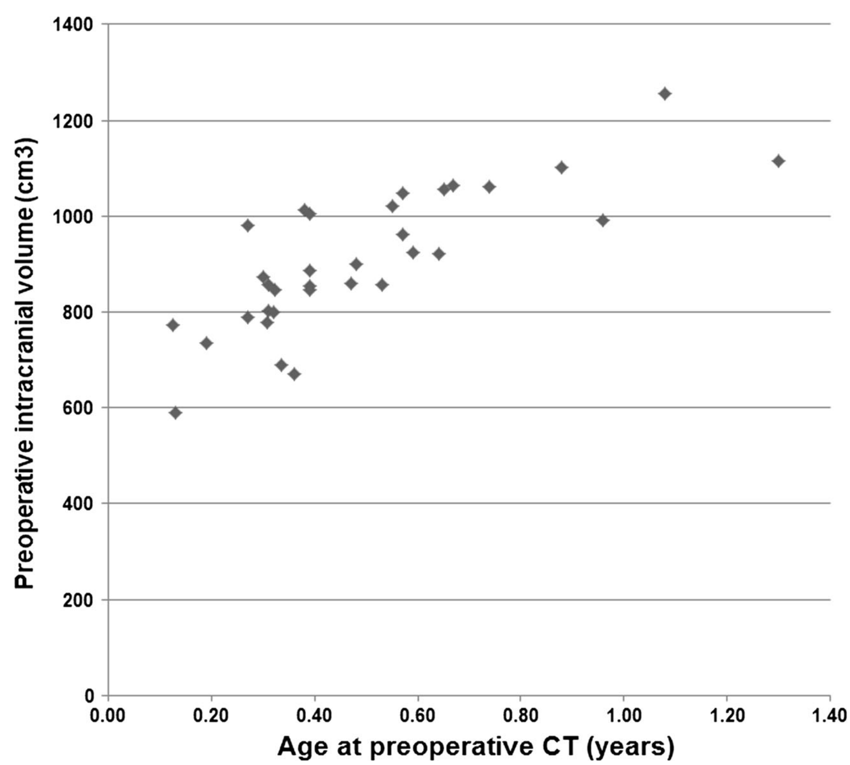
The 3D-CT normative data about ossification of sagittal suture in infants without synostosis are limited. Sim et al. [12]

Fig. 1 Length of sagittal synostosis correlated positively with age at preoperative CT ($r = 0.688$, $p < 0.01$)



evaluated suture closure in 243 infants aged less than 1 year who had undergone 3D-CT scans because of minor head trauma. Initiation of sagittal suture closure before the age of 5 months was not observed. At the age of 12 months, the mean closure rate (length of closed suture line/total length of suture line × 100) was 49.5% [12]. In our study, the closure rates varied from 29.7 to 100%, and the ages at the time of preoperative CTs from 0.13 to 1.13 years. Children with partial central fusions were the youngest ($n = 3$), whereas all those older than 0.7 years ($n = 9$) had total synostosis. According to previous studies, the central section of the sagittal suture appears to be the first to fuse [1, 4]. Then, at least two different

Fig. 2 Preoperative segmented intracranial volume (SIV) correlated positively with age at preoperative CT ($r = 0.788$, $p < 0.01$)



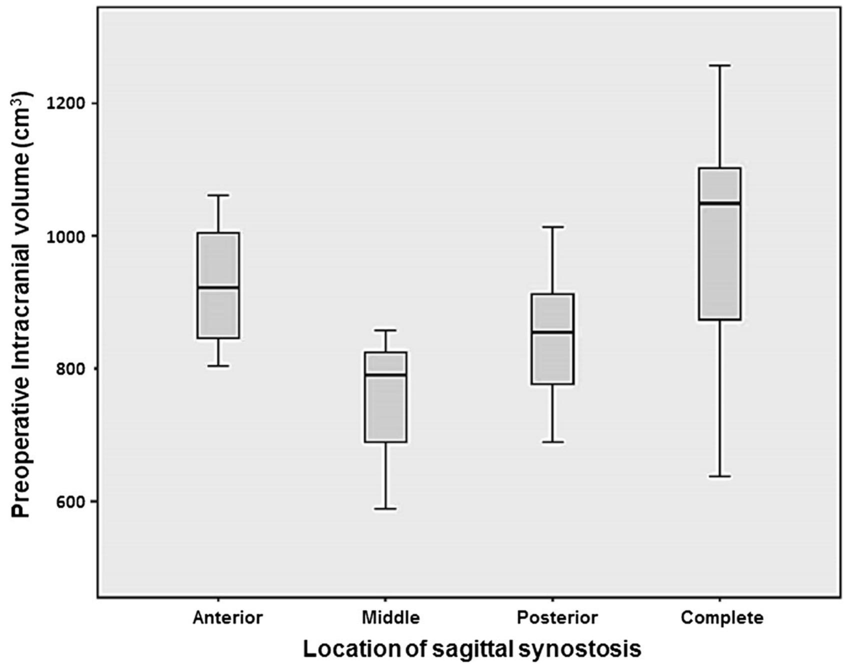


Fig. 3 Preoperative SIV according to location of sagittal synostosis: anterior (n = 9), middle (n = 3), posterior (n = 12), and complete (n = 9)

developmental paths toward complete fusion of the sagittal suture exist; either the anterior section or the posterior section fuses next [4]. A generally accepted idea is that the earlier the synostosis takes place, the greater the effect on skull shape, and conversely, the later the synostosis occurs, the less effect on skull shape. David et al. [13] classified 76 patients according to representative characteristics with sagittal suture in 3D-CTs. Anterior type (24%) featured a transverse retrocoronal band, central type (29%) a heaped sagittal ridge, posterior type (35%) an especially prominent occiput, and complex type (13%) no single dominant characteristic.

Intracranial volume

The literature concerning ICV in sagittal synostosis is controversial. It has been reported to be less, more, or equal to that of age-matched control children [5–10]. Lee et al. [8] studied unoperated patients with sagittal synostosis (n = 46) and found that compared with control males, their ICV was lower during the first 6 months, normal or somewhat greater between 7 and 12 months, and lower at higher ages. Later, Fischer et al. [6] demonstrated with a large material (n = 143) and age- and sex-matched controls that children with sagittal synostosis have normal ICV during the first year of life. The preoperative ICV in children aged less than 180 days was 802 ± 13 and in children older than 180 days 1000 ± 19 [6]. Despite the synostotic suture, other mechanisms, e.g., raised intracranial pressure, can force the skull to grow in excess of normal in these patients [6]. Our study is limited in that no control material was available, and an evaluation of possible growth restriction could not be done. In addition, the material in this preliminary retrospective study was small. However, an important finding was that ICV became larger with age, although length of sagittal synostosis increased.

Surgical method

The goal of the surgical correction of sagittal synostosis is to remodel the cranial vault to allow unimpeded brain growth, ensure normal function, and avoid elevated intracranial pressure. One year after operation, SIV had increased in all patients. The older the child at the time of the operation, the less the percentage SIV increased. According to Heller et al. [7], all patients (n = 24) with non-syndromic isolated sagittal synostosis demonstrated normal volume growth rate postoperatively. The surgical technique may affect the postoperative growth of the skull. For example, placing of the absorbable fixation plates perpendicular to the former synostosis may restrict the increase of width of the skull. Arnaud et al. [14] described secondary coronal synostosis in 10% of patients with sagittal synostosis treated with H-craniectomy. Our technique is different from theirs, and we did not investigate possible secondary coronal synostosis. We used the same technique in all operations, aiming to expand the skull as much as possible. As the immediate ICV gain was not measured, it is not known whether the larger volume in younger patients 1 year after the procedure was gained already at the time of the operation or whether it is the result of more intense growth potential. The longer the synostosis, the more the skull has to be remodeled during the craniosynostosis operation. The timing and technique for optimal surgical treatment are unknown.

SIV measurement

Interpretation of cranial vault volume is often complicated by the lack of standard methods for its measurement and lack of reference material. In addition, the published series are often small. The segmentation of tissue volumes used in this study can be considered a brute force segmentation method, where every crease and ridge of the cranial vault is taken into account. Verification of the segmented volume against the original slice data sets ensures proper registration of the ICV. This work flow is admittedly too complicated and unnecessarily exact for clinical use, but automated segmentation algorithms have shown excellent results [15]. Volume estimates and indices from simple three-plane measurements do not reliably reflect the ICV and are, in our experience, obsolete [16].

Conclusion

Length of sagittal synostosis and SIV increased with age. The older the child at the time of the operation, the less the percentage SIV increased.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

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