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ORIGINAL ARTICLE

Visualization of skull remodeling and re-ossification after reconstructive surgery and mosaic-like cranioplasty in sagittal synostosis and deformational brachycephaly[1]

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Abstract

Introduction Extended vertex craniectomy in sagittal synostosis (SS) and transposition craniectomy in severe deformational brachycephaly (DB) combined with mosaic-like cranioplasty (M-LC) have been performed in 17 and 24 infants from 2001 to 2003. The hitherto not well-known mechanisms of remodeling and effectiveness of M-LC is assessed by X-ray and anthropometry.

Methods Follow-ups included skull radiograms preoperatively and 3 and 15 months postoperatively, which were analyzed by craniometry according to Haas, long-term anthropometry, and clinical follow-up till mean age of 7.6 and 7.4 years. Analysis included the following: time course of cephalic indexes (CI), sizes of distances (breadth, length, height) and modulus, and mean deviation of distances and modulus from the normal age- and sex-dependent values; evaluation of re-ossification of the operative defects covered by M-L C.

Results CI in SS is normalized in early follow-up with stabilization thereafter; CI of DB is gradually normalized till late follow-up. Remodeling occurs in both disorders by active and passive mechanisms: increased growth of distances with preoperative minus and decreased growth of distances with surplus. The latter mechanism adds more to the postoperative remodeling. M-LC leads to concentric and final complete re-ossification of the defects.

Discussion Significant remodeling of the skull vault is observed in both disorders by the demonstrated time course and mechanisms. M-LC does not hinder early remodeling and guarantees re-ossification of the defects.

Keywords Scaphocephy/postural brachycephaly · Surgery · X-ray craniometry/anthropometry · Long-term skull transformation

Different surgical procedures are performed in SS whereas DB is mostly a domain of molding helmets. The former can be divided into complete reconstructions [1, 2] and procedures with partial correction leaving preconditions for postsurgical spontaneous or remodeling by external forces [3–9]. Young infants have two advantages with regard to spontaneous remodeling: rapid skull growth and spontaneous re-ossification after gross removal of the cranial bone. Drawbacks of wide resections are possible residual bone defects.

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The aim of this study is to describe the mechanisms and extent of spontaneous remodeling after surgery and visualize re-ossification of the cranium after vertex craniectomy in SS and transposition craniectomy in severe non-sutural DB combined with M-LC, as well as to prove the usefulness of plain skull X-ray in this context.

Patients and methods

Seventeen infants with SS and 24 with severe DB (CI ≥ 93 [10]) have been operated in 2001–2003 Their characteristics, follow-up, and methods of surgery are shown in Table 1.

[1] Part of the paper presented at the Swiss Congress of Radiology, Congress Center Basel, Switzerland, June 4–6, 2015. Wolf R. and Kaiser G.L. Visualization of skull remodeling by plain skull X-ray imaging in long-term follow-up after cranioplasty for synostosis and other disorders—radiological and clinical prospective study

Table 1 Characteristics of cohort 01–03

Type of cohort and disorder	Cohort 01–03 SS	Cohort 01–03 DB
Patients	$N = 17$	$N = 24$
X-ray CI	67.9 ± 3.9 $N = 15/16$	98.2 ± 5.1 $N = 19/20$
Age at surgery	4.5 ± 1.2	7.3 ± 4.1
Type of surgery	Vertex craniectomy with mosaic-like cranioplasty (M-LC)	Transposition craniectomy with mosaic-like cranioplasty (M-LC)
Follow-up time		
X-ray	≥ 1.5 years	≥ 1.5 years
Clinical	7.6 ± 2.3 years	7.4 ± 3.3 years

The large defects after craniectomy are covered with small pieces (≤ 1 cm of diameter) obtained from the removed bones. This method has been introduced in 2001 ([11] and Fig. 1)

Plain A–P and lateral skull X-rays were performed preoperatively and prospectively 3 and 15 months postoperatively combined with clinical examination and anthropometry. The latter tools were continued at 2–3 yearly intervals until school age.

Clinical length, breadth, and CI are measured according to Farkas and Kolar et al. [12, 13]. The X-rays were used to copy the outlines of skull on transparent paper and measure breadth, length, and height and calculate CI and modulus according to Haas [14], as well as to describe the time course of skull shape and re-ossification at the site of M-LC.

The data were analyzed as follows: (1) time course of CI and comparison with clinical CI. (2) time course of the distances, modulus (sum of distances/3 [14]), and their absolute increase (the known radiological enlargement + 10%, + 16% [14]) was used for correction of the attained distances to real size. (3) Following description of the position of all individual values in relation to the age- and sex-dependent values and calculation of their mean deviation from the median of normal values [14] during the observed time, multiple line plots were constructed.

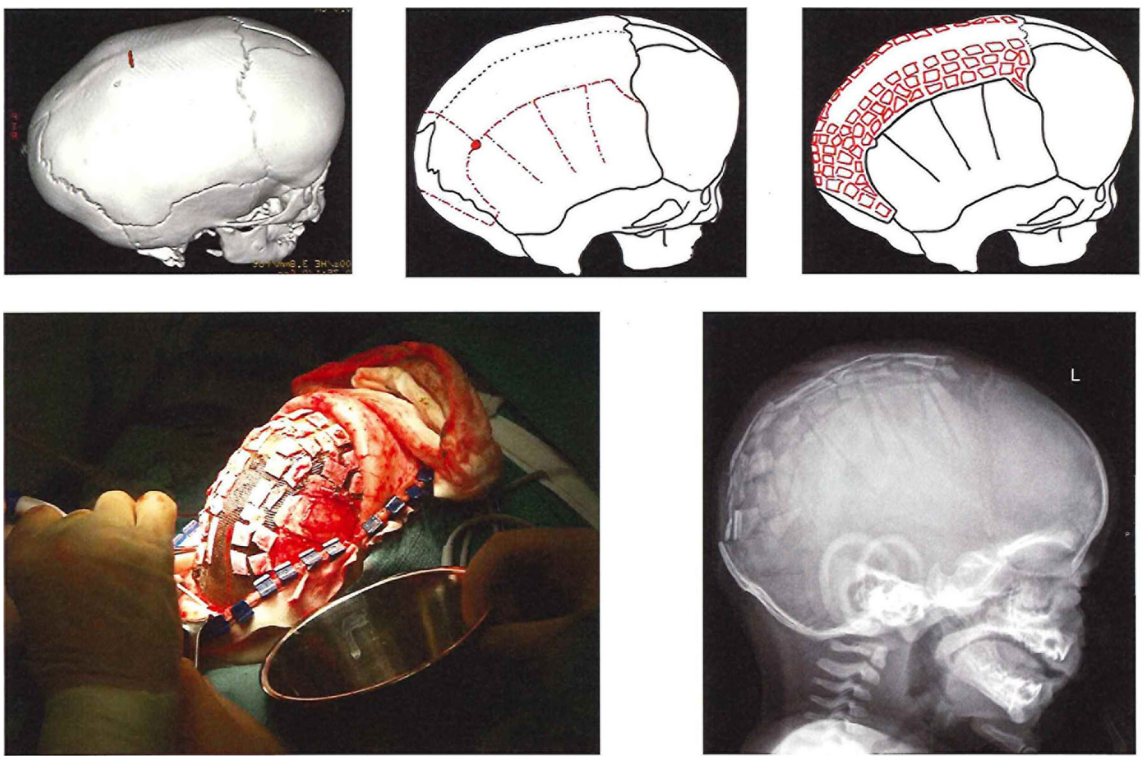

Fig. 1 At the top from left to right: the extension of vertex craniectomy including the bulging back of the skull and the principle of M-LC; at the bottom from left to right: the ongoing M-LC which covers the large defects and a lateral plain skull X-ray shortly after surgery. The bone pieces should be positioned close to each other including the site of former sagittal suture (designed by courtesy of V. Oesch)

Table 2 Pre- and postoperative radiological and clinical CIs in SS of cohort 01–03

X-ray CI	Preoperatively	3 months	1.5 years Postoperatively	Mean age 5.10 years	WSRT/Pt-test[1]/[2], [1]/[3], and [1]/[4] < 0.001
$N = 17/16$	68.1 ± 3.6^1	76.8 ± 4.2^2	$76.5. \pm 3.6^3$	78.75 ± 3.2^4	Pt-test[2]/[3] and [3]/[4] ns
Clinical CI	Preoperatively	3 months	1.5 years Postoperatively	Mean age 7.6 years	Pt-test[1]/[2], [1]/[3], and [1]/[4] < 0.001
$N = 17/15$	66.4 ± 4.3^1	77.6 ± 5.4^2	77.6 ± 3.2^3	78.0 ± 4.3^4	Pt-test[2]/[3] and [1]/[4] ns

$p < 0.05$ significant

Statistical analysis

Sigma Stat® Version 3.5 for analysis of quantitative data.

Graphics program Paint Version 1703, Snagit Editor 13.1.3, Microsoft Office Professional Plus 2016.

Results

The mean preoperative radiological CI of SS is abnormally low (Table 2). Normal range is 72.7–87.7 at 2–6 months [14]. At the first follow-up, the mean CIs have changed significantly and do not change thereafter. The mean radiological CIs are not significantly different from the clinical (*t* test $p > 0.05$) (Fig. 2).

The pre- and postoperative radiological and clinical CIs of DB are before surgery high (Table 3). Normal range of CI is 72.7–87.7 at 2–6 months [14]). At the first and second follow-ups, mean CIs have diminished stepwise and significantly. The mean radiological CIs are not significantly different from the clinical CIs (*t* test $p > 0.05$) (Fig. 2).

The mean dimensions have grown significantly in each interval. Three months postoperatively, breadth and height have grown significantly more than length. The mean increase of length is not significantly different from that of breadth at the second follow-up (Table 4).

Three months postoperatively, growth of length is significantly larger than breadth and height and it remains so at the second follow-up (Table 5). The mean increases of growth of the first and second periods are statistically not different in all four dimensions. The means of the three distances are before surgery close to each other; thereafter, length is clearly larger than the other two.

The time course of mean deviation of distances and modulus from the normal X-ray data are depicted in Fig. 3 for both pathologies.

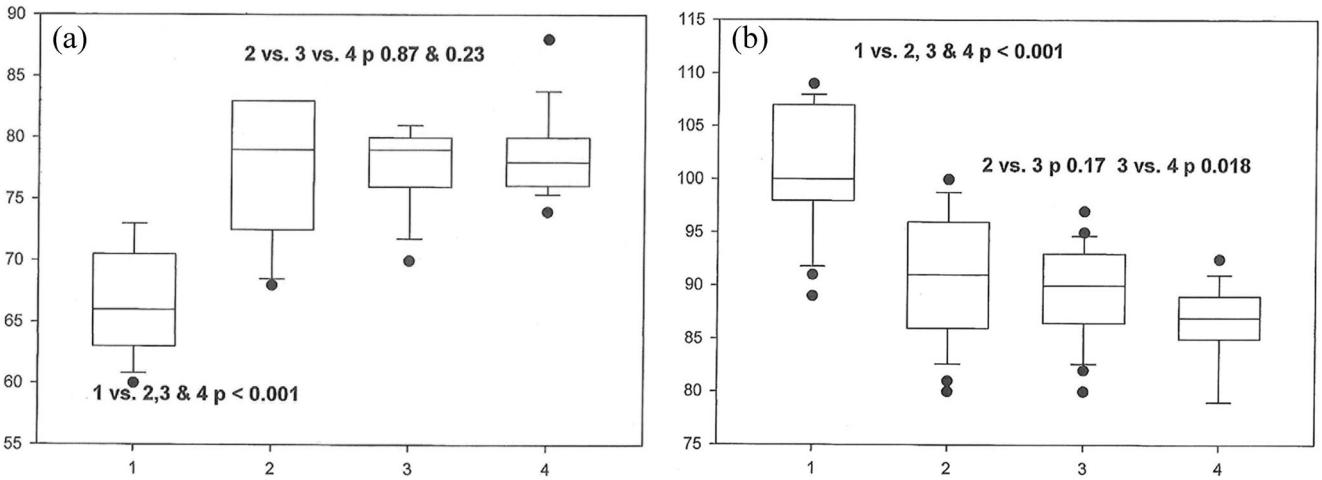

Fig. 2 **a** Boxplot of pre- and postoperative clinical CIs in SS. Depicted are median, 1SD, and 95% CI at median ages of 3 months preoperatively, at median ages of 8 and 22 months, and mean age of 7.6 years postoperatively. All mean postoperative values do not significantly differ from each other. The lower limit of 95% CI is 75.5 at late clinical follow-up (normal range 74–80 for boys and 73–79 for girls [15]. **b** Boxplot of pre- and postoperative clinical CIs in DB. Depicted are the same parameters as in Fig. 2a but preoperatively at median ages of 5, 10, and 23.25 months and mean age of 7.4 years postoperatively. All mean postoperative values are significantly different from the preoperative but they are less different from the first to second early follow-up clinically in contrast to the X-ray values ($p < 0.001$).The upper limit of 95% CI is 91 at late clinical follow-up (normal upper limit < 93 [16] and current normative values 86–88 in populations of supine sleeping children [17])

Table 3 Pre- and postoperative radiological and clinical CIs in DB of cohort 01–03

X-ray CI	Preoperatively	3 months	1.5 years Postoperatively	Mean age 9.11 years	Pt-test$^{1/2}$ < 0.001
$N = 20/19$	98.0 ± 4.6^{1}	91.2 ± 5.0^{2}	88.1 ± 3.2^{3}	89.0 ± 6.3	Pt-test$^{2/3}$ < 0.001
Clinical CI	Preoperatively	3 months	1.5 years Postoperatively	Mean age 7.4 years	Pt-test$^{1/2}$, $^{1/3}$, and $^{1/4}$ $p < 0.001$
$N = 20/19$	101.2 ± 5.9^{1}	90.8 ± 5.5^{2}	89.7 ± 4.2^{3}	86.3 ± 3.9^{4}	Pt-test$^{2/3}$ and $^{3/4}$ ns and 0.018

$p < 0.05$ significant

Table 4 Time course of distances and modulus and their increase in SS cohort 01–03

Time course	Distance in cm Mean, 1SD, and median values			Increase of distance in cm Mean and median values	
	Preoperative	Follow-up 1	Follow-up 2	1st change	2nd change
Median age	3	8	22 months	8	22 months
Breadth *B*	9.5^{1} 9.5 ± 0.65	11.3^{2} 11.3 ± 0.45	12.25^{3}12.3 ± 0.4	1.8^{1} 1.75 ± 0.5	0.85^{1} 0.85 ± 0.3
Length *L*	14.0^{1} 14.2 ± 0.85	14.8^{2} 14.8 ± 0.65	15.9^{3} 15.9 ± 0.8	0.7^{2} 0.7 ± 0.9	1.1^{2} 1.15 ± 0.65
Height *H*	10.2^{1} 10.2 ± 1.05	12.1^{2} 12.1 ± 0.6	12.6^{3} 12.8 ± 0.8	1.9^{3} 1.75 ± 0.95	0.5^{3} 0.5 ± 0.5
Modulus *M*	11.15^{1} 11.3 ± 0.7	12.7^{2} 12.7 ± 0.4	13.6^{3} 13.75 ± 0.6	1.7^{4} 1.5 ± 0.7	1.2^{4} 1.2 ± 0.3
Statistics $N = 17/16$	$^{1/2}$, $^{2/3}$, and $^{1/3}$ *p t* test (WSR test) < 0.001			$^{1/2}$ and $^{3/2}$ WRS and *t* test 0.001 1st vs. 2nd change *B*, *H*, and *M* *p t* test < 0.001 and 0.003, *L* ns	$^{1/2}$ and $^{1/3}$ *t* test ns

$p < 0.05$ significant

Table 5 Time course of the distances and the modulus and their increase in DB of cohort 01–03

Time course	Distance in cm Mean and median values			Increase of distance in cm Mean and median values	
	Preoperative	Follow-up 1	Follow-up 2	Follow-up Period 1	Follow-up Period 2
Median age	5	10	23.25 months	10	23.25 months
Breadth *B*	12.0^{1} 12.2 ± 0.9	12.6^{2} 12.7 ± 0.65	13.1^{3} 13.3 ± 0.5	0.5^{1} 0.5 ± 0.7	0.5^{1} 0.5 ± 0.4
Length *L*	12.2^{1} 12.35 ± 0.9	13.7^{2} 13.7 ± 0.8	15.1^{3} 15.1 ± 0.5	1.45^{2} 1.45 ± 0.6	1.3^{2} 1.35 ± 0.5
Height *H*	11.5^{1} 11.7 ± 0.9	11.8^{2} 11.95 ± 0.7	12.5^{3} 12.5 ± 0.4	0.25^{3} 0.3 ± 0.9	0.6^{3} 0.55 ± 0.6
Modulus *M*	11.9^{1} 11.95 ± 0.8	12.7^{2} 12.75 ± 0.6	13.55^{3}13.55 ± 0.4	0.8^{4} 0.75 ± 0.55	0.8^{4} 0.9 ± 0.3
Statistics $N = 20/19$	$^{1/2}$, $^{2/3}$, and $^{1/3}$ *p t* test < 0.001 except for $^{1/2}$ *H* ns B^{1} vs. L^{1} and H^{1} *t* test ns B^{3} vs. L^{3} and H^{3} and L^{3} vs. H^{3} *t* test < 0.001			$^{1/2}$ and $^{3/2}$ *t* test < 0.001 $^{1/1}$, $^{2/2}$, $^{3/3}$, and $^{4/4}$ *t* test ns	$^{1/2}$ and $^{3/2}$ *t* test < 0.001/0.002

$p < 0.05$ significant

a

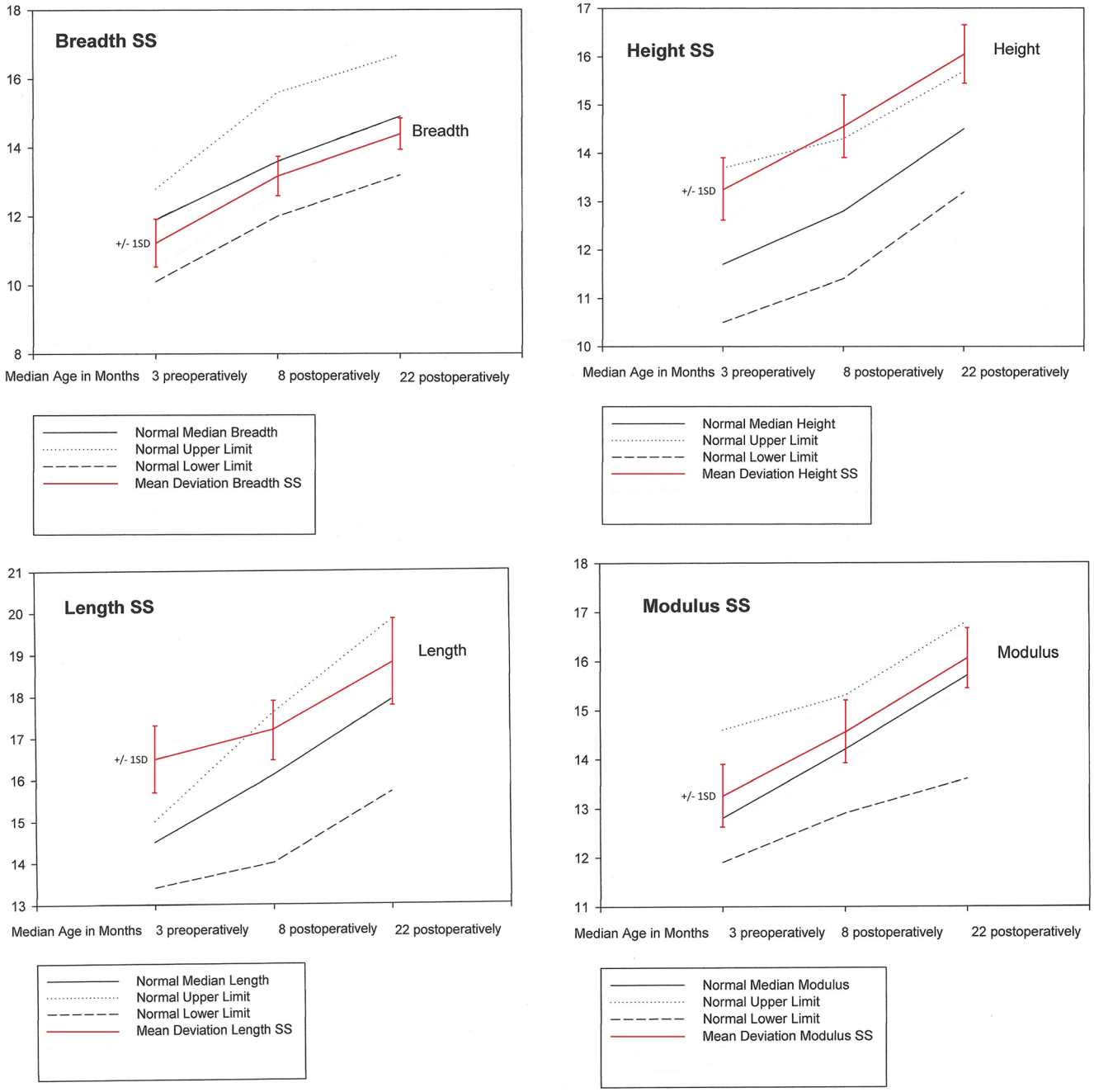

Fig. 3 **a** The time course of mean deviation ± 1SD of the distances and modulus of SS from the normal X-ray median and range [14]. The mean deviation of breadth approaches in the first period the normal median ± 1SD is finally positioned in the upper part below the normal median. The mean deviation of length ± 1SD enters in the first follow-up period the upper half of normal range. Height has grown initially stronger and thereafter less than the normal median. Nevertheless, the mean deviation is finally somewhat above the upper limit in contrast to preoperatively. The deviation of modulus (± SD) remains in the lowest area above the normal median (the calculation of the mean deviation ± 1SD from the normal values [14] available from the author). **b** The time course of mean deviation ± 1SD of the distances and modulus of DB from the normal X-ray median and range [14]. Mean deviation of breadth is before surgery above the normal range and reaches the upper half of the normal range. Growth of breadth and height has diminished in both periods compared with the normal median and influenced the modulus; in contrast to length, in which the negative mean deviation was unexpectedly small and reached the normal median already at the first follow-up

b

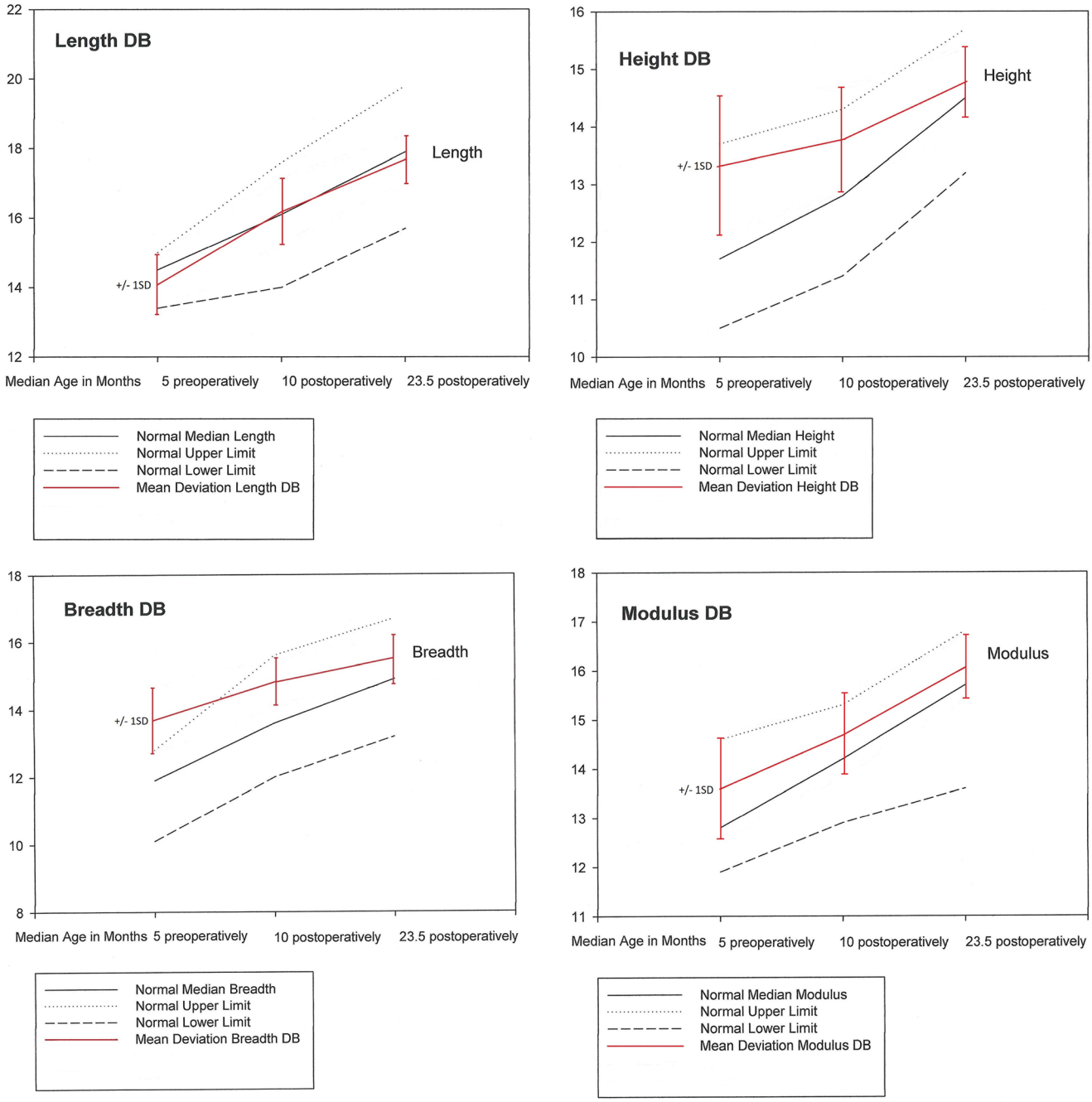

Fig. 3 (continued)

Comparison of the serial X-rays demonstrated normalization and amelioration of shape and proportion of the skull at the second early follow-up (Figs. 4 and 5).

Mosaic-like cranioplasty The single bone pieces become larger with less clear margins at the first and fuse with the adjacent pieces at the second early radiological follow-up (Fig. 6).

Residual bone defects have not been observed at the second early X-ray follow-up and clinically at late follow-up in cohort 01–03 except for the first patient who needed secondary cranioplasty. Following this experience, the site of former SS was also closed with bone pieces.

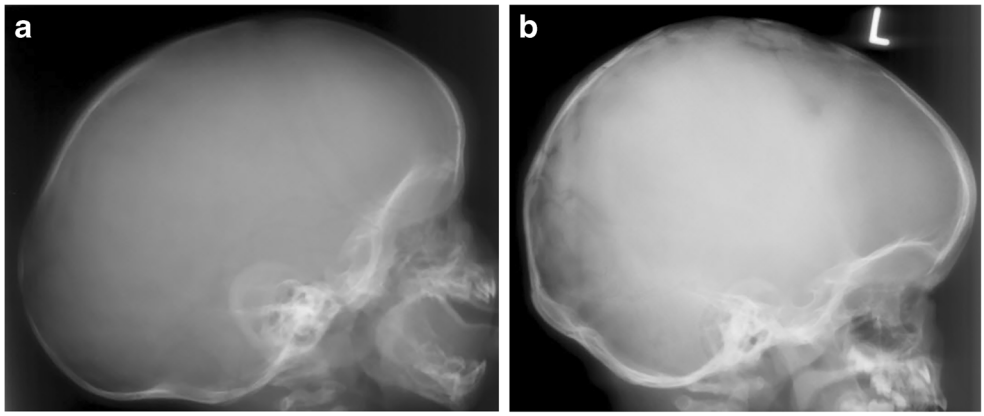

Fig. 4 **a**, **b** Lateral skull X-rays before surgery (at 4 months) in SS and at the second early follow-up (at 24 months). Normalization of skull shape after craniectomy, no large defects after M-LC

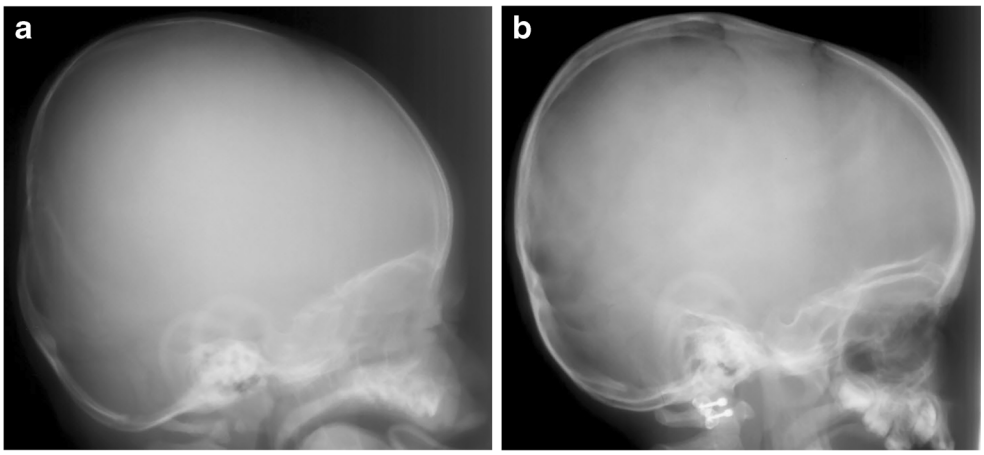

Fig. 5 **a**, **b** Lateral skull X-rays before surgery (at 5 months) in DB and at the second early follow-up (at 24.5 months). Distinct amelioration of skull shape after craniectomy, no large defects after M-LC

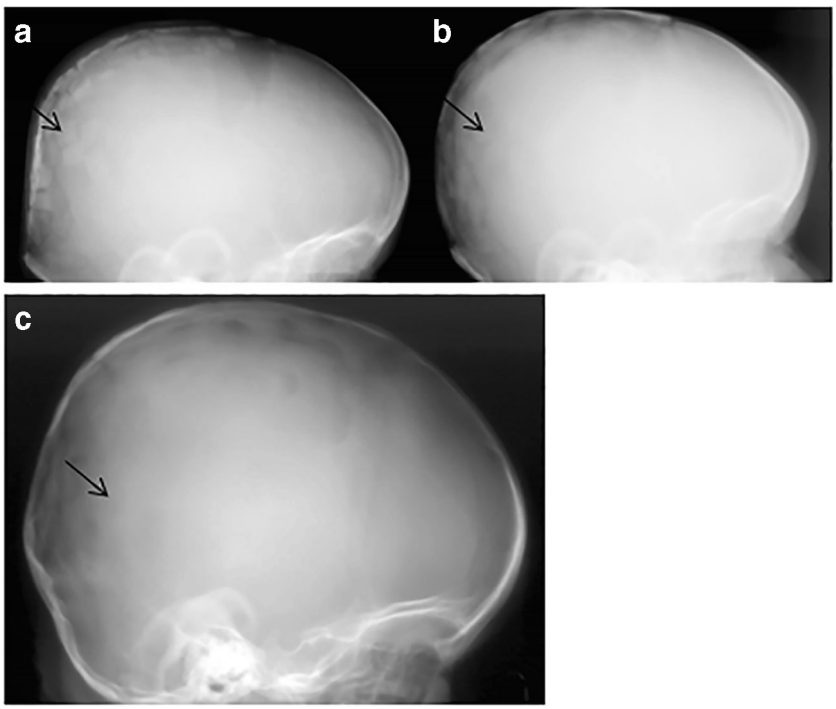

Fig. 6 **a**–**c** From the left to right at the top and at the bottom: the single mosaic-like bone pieces are distinctly recognizable shortly after cranioplasty. The real size of breadth of a single piece is 6.75 mm, becomes larger (8.1 mm) with smoother margins at the first and fuses with the adjacent pieces at the second early radiological follow-up (8.55 mm)

Discussion

First information about the extent of remodeling after surgery of SS and severe DB is attained by the time course of CI. It shows significant amelioration postoperatively in both pathologies and no radiological worsening at early and if the anthropometric CI data are included at late follow-up.

The mean increase of radiological cephalic index was 8.7% at the second early and of the clinical 11.6% at late follow-up in SS. The former value is somewhat smaller than that of Heller et al. which revealed 10.6% worked out by CT scans [1]. Agrawal et al. [18] have already demonstrated similar long-term anthropometric outcome in infants with SS who had characteristics comparable to our group. However, they quoted a slight decrease in CI after the initial 6-month period of improvement. Similar long-term results have been reported recently by Thomas et al. [19] after skull remodeling with no significant long-term relapse in contrast to strip craniectomy with significantly lower values of CI.

Metzler et al. [2] performed total cranial vault remodeling for SS at 8–10 months of age. Corresponding values of CI value were significantly lower at late follow-up than early after surgery. Thwin et al. [20] who compared strip craniectomy with total cranial remodeling and performed a meta-analysis stated that current data imply neither procedure offers a clear long-term advantage over the other.

Interestingly, *remodeling is different in DB* as an example of a cranial deformity without synostosis from that of SS: Correction of CI occurs in severe DB stepwise at early follow-ups on X-rays and later on clinically within several years, whereas definitive CI is attained in SS mainly 3 months postoperatively. The somewhat less favorable results at the second early follow-up in DB can be explained by the older age at surgery and different surgical technique. Nevertheless, the mean decrease of X-ray CI by 10.1% (that is not significantly different from the amount of increase in SS) is followed by a final long-term anthropometric decrease by 14.9% which is superior to the mean values of 2.9–5.1% after orthotic helmet therapy of DB in older and younger infants reported by Graham et al. [17] and the results of Teichgraeber et al. who stated that DB did not normalize in contrast to plagiocephaly [21]. Steinberg et al. [22] reported complete correction of posterior positional cranial deformity (cranial ratio < 0.85) by helmet therapy in 95% of 1531 infants of whom one fifth and fourth had isolated DB.

In spite of doubts about the reliability of two-dimensional CI as quantitative description of skull deformities and their outcome [15, 23], van Lindert et al. stated that cephalic index is still the most accurate and most easily available value in SS to date [24] and regression of the CI indicates a tendency to recapitulate the primary deformity of SS (5).

The size of the distances and modulus and their changes in the two early follow-ups provide more information about the mechanism and quantity of growth leading to skull remodeling. Their means increase significantly in both disorders. *Overall restriction of growth by surgery is therefore unlikely.*

The growth spurt of breadth and height in SS which has been triggered by surgery occurs only in the first early period. Thereafter, growth of distances is smaller as before except for length with constant increase. However, increase of breadth is not significantly different from that of length what explains the maintenance of the earlier attained normal CI. Although the same is true vice versa for growth of length in the first early period after surgery for DB, remodeling is different in the second early period: All distances and the modulus grow continuously with length at a higher level which is now significantly larger than the remainder. *The described extraordinary changes of growth can be interpreted as mechanisms to correct either SS or posterior DB*.

Remodeling in the two early postoperative periods is *even more precisely assessed*, if the mean of age- and sex-dependent deviations of all individual data are calculated, compared with the available data of Haas [14] and finally visualized by multiple line plots. Almost one third of the values of breadth and almost all of length fall in SS before surgery below and above the normal range respectively. This smaller deficit in breadth than surplus in length is consistent with the results of Kolar et al. [23] who quoted that reduced cephalic index reflects increased cranial length more than reduced width. Wilbrand et al. [25] who compared the anthropometric data of 69 infants with natural SS mostly of age 4–9 months with normative age- and sex-dependent percentiles showed that breadth was in 29% below the 10th and length in > 90% above the 90th percentile. Postoperatively, almost all our patients fell in the normal range.

By contrast, more than half of the values of breadth and height are prior surgery above the normal range in DB and only less than 10% of the values of length are initially below the normal range; the supposed deficit of length in DB is therefore only relative. After surgery, almost all data fall into the normal range.

These data demonstrate that *two mechanisms of remodeling of the skull exist*: A passive mechanism which slows down growth as in length of SS and breadth and height of DB and an active mechanism which stimulates growth of breadth and height in SS and length in DB. Except for height in SS, the active mechanism is much smaller than the passive. Both mechanisms are mainly effective in the first early follow-up period in SS and in both periods in DB. The insight into remodeling is useful if similar procedures as ours are performed which expected postsurgical remodeling. *Maximum postoperative remodeling in SS can be expected only in the first 3 months*; optimal time for surgery is before 6 months of age because maximum growth spurt of the skull occurs at this period of time and possible surplus of growth becomes smaller thereafter. The

overwhelming growth of height in the first early period is a drawback of vertex craniectomy but it may be useful because major deficit of height occurs in natural SS (23).

Radiographs show that the re-implanted bone pieces are re-vascularized, initiate concentric re-ossification within 15 months, and prevent bony defects. This procedure is less time-consuming than using particulate skull bone graft at the time of fronto-orbital advancement [26]. Plain skull X-rays *are no longer required in the follow-up of our patients* provided expert anthropometry and equal measurement of height such as the ear-to-ear perimeter distance [15] are performed, because reliable results of the applied techniques occur in a defined period of time and anthropometric CIs are not significantly different from those attained by X-rays [25].

Plain X-rays can be discussed immediately before surgery and at puberty; they can be useful in suspected SS without scaphocephaly [27], lambdoid synostosis, and for re-synostosis and secondary synostosis of other sutures [28].

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Compliance with ethical standards

Conflict of interest No financial support or benefits were given to the author from any source related to the scientific work reported in this article.

The author got the permission to perform the study "Visualization of Skull Remodeling and Re-ossification after Reconstructive Surgery and M-LC in Sagittal Synostosis and Severe Posterior DB (VSRRRSSDB)" and to publish it from the Cantonal Commission of Ethics Berne of Swiss Ethics on May 2, 2017.

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