Long-term aesthetic results of frontoorbital correction for frontal plagiocephaly

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Object. Long-term aesthetic results of craniofacial surgery for frontal plagiocephaly were evaluated by two observers who used a scoring system based on deformities typical for this type of craniosynostosis.

Methods. In this retrospective study, pre- and postoperative photographs of 59 patients were scored for items typical of frontal plagiocephaly: shape of the forehead, orbital dystopia, and temporal depression. Each item was quantified as normal or absent (0 points), a mild deformity (1 point) or a severe deformity (2 points).

Preoperatively, the most obvious deformity was the shape of the forehead, whereas postoperatively temporal retrusion was the main deformity. There was a good overall correction of the presenting deformities, with a decline in the mean score from 3.7 to 0.86. There was a statistically significant weak correlation between pre- and postoperative scores for orbital dystopia alone. This finding indicates that the severity of the initial disease is not a major contributor to the final result. Furthermore, the surgical outcome seemed to be stable over time and was not influenced by the timing of surgery if it took place when the children were between 6 and 15 months of age.

Conclusions. Early craniofacial correction for frontal plagiocephaly results in a stable, acceptable aesthetic outcome on which the initial deformity has little effect. The most common associated craniofacial characteristics are corrected well to very well when surgery is performed within the first 6 to 15 months of life. The main focus of the deformity in both the short and long term remains the temporal depression, and thus it requires extra attention during surgery.

Key Words • craniosynostosis • plagiocephaly • surgical treatment • outcome • pediatric neurosurgery

Craniosynostosis results from the premature (pre- or postnatal) fusion of one or more cranial sutures and occurs in approximately 1:2000–2500 live births. During embryogenesis, the coronal suture first appears at 16 weeks’ gestation. By 18 weeks, it has been established along its entire length, coinciding with the period during which synostosis develops. Frontal plagiocephaly accounts for approximately 30% of all cases of single-suture craniosynostosis treated at our center.

In unilateral coronal synostosis, growth in the AP direction across the affected coronal suture is restricted, resulting in frontal plagiocephaly. Clinical features of frontal plagiocephaly include retrusion of the supraorbital rim and frontal bone (with compensatory bossing of the contralateral side of the forehead), orbital dystopia, and temporal depression. Surgical intervention is directed at correction of the asymmetrical forehead and supraorbital bar, with an advancement of the bone at the affected side. Preferably, this procedure is undertaken before the patient reaches 1 year of age.

The results of craniosynostosis operations have been evaluated by using a variety of methods such as CT, computed tomography, and anthropometric analysis, all of which provide accurate and reproducible information. These methods are time consuming, however, and their data-gathering function can involve additional exposure to radiation. Therefore, we developed a simple scoring system that relies on photographs taken routinely. In the present study of long-term aesthetic results obtained at the craniofacial center in Rotterdam, a panel of clinicians assessed typical facial features in patients with frontal plagiocephaly, both pre- and postoperatively.

Clinical Material and Methods

Patient Characteristics

Case notes of patients with frontal plagiocephaly who were treated surgically at our department between 1972 and 2002 were reviewed. Patients were selected for the panel assessment if the following criteria were met: 1) follow up took place at the Sophia Children’s Hospital; 2) standardized AP and lateral preoperative photographs were available; and 3) standardized AP and a long-term series of lateral postoperative photographs were available (minimum 12 months postoperative).

A total of 139 patients with nonsyndromic frontal plagio-
griocephaly underwent surgery at our unit. Eleven patients did not have a complete medical chart, 34 patients did not fulfill the first criterion, six patients did not fulfill the second, and 23 patients did not have a photograph that was taken at least 12 months after the operation. Six patients underwent surgery in which a different technique was used. Altogether, 59 patients were included in the study. The postoperative follow-up period ranged from 12 months to 16.5 years (mean 7 years, 10 months).

Surgical Technique

One surgeon (J.M.V.) performed the surgery in most of the patients. Although some variation in technique exists between surgeons, in general the approach used in this procedure is similar. A bicoronal incision is made and the forehead flap is created at the subgaleal level. An anteriorly based periosteal flap is created, thus exposing the supraorbital rims, the base of the nasal bone, and the superior part of the lateral orbital walls. Both frontal bones are removed in one piece, after which the entire supraorbital bar is excised. The supraorbital bar is reshaped by advancing the synostotic side together with some backwards positioning of the contralateral side. The forehead is split in the midline and the resulting bone pieces are adjusted and replaced in the most optimal position. The supraorbital bar and frontal bones are fixated with either stainless-steel wires used for cerclage or, more recently, with Vicryl sutures.

Evaluation of the Photographs

Two panel members evaluated the AP and lateral photographs independently. The pre- and postoperative photographs were randomly assessed. Neither panelist performed surgery in any of the children. One of the panelists was a junior plastic surgeon and had significant experience in assessing craniofacial abnormalities; the other panelist was a medical student. The junior plastic surgeon explained only the characteristics of nonoperated frontal plagiocephaly to the medical student so as not to influence the way in which the student assessed the photographs. The photographs of the patients were evaluated for three items: shape of the forehead, orbital dystopia, and temporal depression. Scores of 0 (normal), 1 (moderate deformity), and 2 (severe deformity) were assigned for each item. For each set of photographs (one AP and two lateral) a maximum of six points could be given, signifying the most severe presentation of frontal plagiocephaly. Each member scored the photographs on two separate occasions. A third score was given in case both observers did not agree, the difference between the total scores could be given, signifying the most severe presentation of frontal plagiocephaly. Each member scored the photographs on two separate occasions. A third score was given in case the second score was different from the first, resulting in one conclusive score per observer for each of the three items and for each of the photographs.

To assess the level of agreement between observers, the kappa statistic was used. The same method was used to assess intraobserver agreement. Both pre- and postoperative photographs were included in this assessment.

Six goals were set for this study: 1) determine the aesthetic outcome of the operation by comparing pre- and postoperative photographs; 2) identify the most obvious deformities both pre- and postoperatively; 3) determine the influence of the severity of the preoperative deformity on aesthetic outcome; 4) establish the effect of age at operation on outcome; 5) evaluate the stability of surgical results; and 6) study the effect of surgical experience on outcome.

The preoperative photograph and the most recent photograph of a patient were used for the evaluation of Goals 1 through 4 and 6. To evaluate the stability of the surgical result (Goal 5), the first and most recent postoperative photographs of all patients were mixed together and scored randomly. The photographs were not organized by patient or by postoperative interval.

Statistical analysis was conducted using commercially available software (SPSS version 10; SPSS Inc., Chicago, IL). For each of the three evaluated items of each set of photographs, the average of the individual scores given by the two observers was calculated and used in further statistical evaluation. The Spearman rank correlation was used to assess monotonic relationships between pre- and postoperative scores of the evaluated items and to assess the relationship between scores of the patients’ postoperative outcomes and the age at which the operation was performed. In this assessment we included only children who underwent surgery at 6 to 15 months of age; the number of older children was too small for inclusion. The Spearman rank correlation was also used to assess the relationship between the assessed items and year of surgery. To establish the stability of the surgical results obtained, the Wilcoxon signed-rank test was used to compare the first and last postoperative scores of a patient given by the observers.

Results

The kappa statistic for the level of agreement between the two observers for shape of the forehead, orbital dystopia, and temporal depression was 0.65, 0.82, and 0.68, respectively (p < 0.001). Although no absolute guideline can be given for what constitutes poor, fair, or good reliability, according to Landis and Koch these figures can be taken to represent good to very good reliability. Intraobserver agreement was also assessed using the kappa statistic. The reliability for all scores given by each observer was 0.64. According to Landis and Koch, this represents good reliability.

The two panel members agreed on the total preoperative appearance grade in 63% of the cases, and they agreed in 68% on postoperative appearance. In cases in which the observers did not agree, the difference between the total scores was in almost all cases limited to one point.

Aesthetic Outcome

Preoperatively, the mean scores for the items evaluated were 1.05 for temporal depression, 1.18 for orbital dystopia, and 1.47 for shape of the forehead. Postoperatively, the mean scores for the same three items were 0.32, 0.31, and 0.25, respectively (Table 1). Overall, the total mean preoperative score dropped from 3.7 to 0.88 postoperatively (Fig. 1).

The scores for all three items decreased after surgery, with the shape of the forehead having the largest decrease (mean 1.22 points). The mean decreases in orbital dystopia and temporal depression scores were 0.86 and 0.73, respectively.

Most Obvious Deformity Pre- and Postoperatively

Before surgery, the highest score was given for the shape of the forehead, which constituted the most obvious initial deformity. After correction, the most obvious residual deformities were temporal depression and orbital dystopia.
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<table>
<thead>
<tr>
<th>Item</th>
<th>Preop</th>
<th>Postop</th>
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<tbody>
<tr>
<td>temporal retrusion</td>
<td>1.05 (0.05)</td>
<td>0.32 (0.06)</td>
</tr>
<tr>
<td>orbital dystopia</td>
<td>1.18 (0.07)</td>
<td>0.31 (0.07)</td>
</tr>
<tr>
<td>shape of the forehead</td>
<td>1.47 (0.06)</td>
<td>0.25 (0.04)</td>
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* Data are expressed as means (standard error of the means).

Influence of Severity of Initial Deformity

There was no correlation between the pre- and postoperative scores regarding the shape of the forehead (r = 0.051) and temporal depression (r = 0.024), and only a weak significant positive correlation was found for orbital dystopia (r = 0.143, p < 0.05). This result indicates that the outcomes for the three items were minimally influenced, if at all, by the severity of the deformity preoperatively.

Effect of Age at Surgery

The patients underwent surgery at a mean age of 17 months (range 3–87 months), with 61% undergoing surgery between the ages of 6 and 15 months. According to the Spearman rank correlation, our data indicate that there was no significant correlation between a child’s postoperative appearance and the age in months at which the operation was performed within this 9-month interval (−0.09 < r < 0.12).

Stability of Surgical Result

The Wilcoxon signed-rank test is used to assess systematic median differences in a variable measured on two occasions. This test was used to evaluate the stability of the surgical result by assessing the postoperative differences between the first score given after surgery and the last one. A minor descending trend was seen in the scores (indicating clinical improvement) for shape of the forehead and orbital dystopia during follow up, while scores for temporal depression rose slightly. The results for orbital dystopia and temporal depression were statistically significant.

Effect of Surgical Experience

Using the Spearman rank correlation, a weak but significant negative correlation (r = −0.427, p < 0.05) was found between temporal depression and year of surgery, suggesting that better results were achieved with the increasing experience of the surgeon. For shape of the forehead and orbital dystopia, no significant correlations (r = 0.075 and 0.012, respectively) were found.

Discussion

Several methods have been developed for the analysis of the results of craniofacial reconstruction for craniostenosis. Posnick, et al., quantified surgical results for trigonocephaly and scaphocephaly by using CT scans; however, the exposure to radiation and the need for sedation to obtain a CT scan are drawbacks of this method. Hansen, et al., used anthropometric assessment to compare three techniques for surgical correction in 22 children with synostotic frontal plagiocephaly. These researchers found residual retrusion of the supraorbital rim on the affected side. McCarthy, et al., analyzed the condition of 57 patients with synostotic frontal plagiocephaly and categorized their results using the Whitaker classification system. They found complete correction of vertical orbital dystopia in 12 patients, improvement in 14, and no change in six of the 32 patients who were available for postoperative analysis. These investigators did not assess temporal depression, however, which is a typical deformity for this type of craniostenosis.

In the present study, we used standardized photographs (taken by our professional photographer for research and clinical follow up) to develop a reliable scoring system to evaluate postoperative results based on typical deformities for frontal plagiocephaly. The advantage of our technique in comparison to other proposed methods lies in the fact that photographs were taken routinely at regular intervals (when the patient was 2, 4, 6, 9, 12, 15, and 18 years of age); therefore, these data are readily available and no exposure to radiation is involved. Furthermore, this method is easily reproducible and is not time consuming. It could therefore be a good tool to use in creating benchmarks of aesthetic outcomes at various craniofacial centers.

Although bird’s- and worm’s-eye photographic views are the optimal ones for assessing symmetry of the forehead, we did not include them in our assessment. These views were readily available for the preoperative period, but views documenting the postoperative period were available for only a few of our patients. To ensure a fair assessment, we left them out of our study. A theoretical drawback of using a two-dimensional photograph is its exaggeration of deformities in comparison with the three-dimensional impression that is given in real life. Yet a photographic assessment does provide a more accurate representation than a mirror image, which is also two-dimensional.

Our data show that surgical correction of frontal plagiocephaly produces considerable improvement in all characteristic features. Preoperatively, the most obvious deformity was the shape of the forehead, whereas the most obvious postoperative deformity was situated at the temporal region and orbital area (Figs. 2–4). Possible explanations for the temporal deformity are either a restriction of growth at this area, a lesser volume of the temporal muscle as a result of...
its detachment during surgery, or inadequate correction during the surgical procedure. Although surgery does not include correction of orbital dystopia, release of the fronto-supraorbital region appears to allow normalization of the horizontal position of the orbits.

In contrast to findings reported in other studies, the severity of the initial deformity was apparently not a major contributor to the postoperative outcome. In our study, no significant correlation was found between pre- and postoperative scores for shape of the forehead and temporal regions. Only for orbital dystopia was a weak significant positive correlation between pre- and postoperative scores found, suggesting that patients who present with obvious orbital dystopia probably have some residual deformity postoperatively. Furthermore, there was no evidence in our series that the outcomes were influenced by the patient’s age at surgery, within the limits of 6 to 15 months.

We found minor changes in the postoperative scores during follow up, and although these results were statistically significant, they are too small to be of any clinical importance. We can therefore conclude that our results remained stable over time. Consequently (and in accordance with the studies by Friede, et al., and Jünger, et al.), the most logical explanation for the residual deformity at the temporal region seems to be inadequate correction during surgery. Furthermore, we found a weak but significant negative correlation between temporal depression and year of surgery; this indicates that better results were obtained as the surgeon became more experienced.

**Conclusions**

Early craniofacial correction for frontal plagiocephaly results in a stable, acceptable aesthetic result on which the initial deformity has little effect. The most commonly associated craniofacial characteristics are corrected well to very well if surgery is performed within the first 6 to 15 months of the child’s life. The primary focus of deformity in the
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short and long term remains temporal depression, and thus it requires extra attention during surgery.

References


**Fig. 4.** Preoperative (upper) and short- (center) and long-term (lower) postoperative follow-up photographs of a patient who experienced improvement in the temporal region but retained a minor residual deformity. Preoperative scores: shape of the forehead, 2; orbital dystopia, 2; temporal retrusion, 2. Postoperative short-term scores: shape of the forehead, 0; orbital dystopia, 1; temporal retrusion, 0. Postoperative long-term scores: shape of the forehead, 0; orbital dystopia, 0; temporal retrusion, 1. The patient was 7.5 months of age at surgery, and the long-term follow-up photograph was taken 5.5 years postoperatively. Short-term postoperative photographs were taken within 1 year of surgery.

**References**


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