Importance of Early Recognition and Treatment of Deformational Plagiocephaly with Orthotic Cranioplasty

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Objectives: **Infants with positional plagiocephaly often exhibit complex multistructural asymmetries that affect the face and skull base as well as the cranial vault. Dynamic Orthotic Cranioplasty (DOC) was developed as a nonsurgical alternative for the treatment of positional plagiocephaly. The effectiveness of DOC has been discussed elsewhere. The purpose of this study was to assess the influence of factors such as entrance age, treatment time, and initial severity on the effectiveness of correction.**

Methods: **The study sample consisted of 258 children with cranial vault asymmetry (CVA) treated prior to 1 year of age. In addition, 246 patients (92%) exhibited concurrent skull base (SBA) and orbitotragial depth (OTDA) asymmetries. All patients had been diagnosed with nonsynostotic plagiocephaly, did not have other contributing medical conditions, were compliant with DOC protocol, and had complete anthropometric measurements at entrance and exit from treatment.**

Results: **Mean age at start of treatment was 6.5 (±1.9) months (range, 2.8 to 11.0 months), with an average treatment time of 4.1 (±2.2) months. The effects of the treatment variables were analyzed using three-way analysis of variance. As expected, initial severity was significantly associated with the amount of correction (p = .0001). However, treatment time was not significant ($p > .05$). Most importantly, the analysis revealed that, having accounted for initial severity, entrance age had a statistically significant effect [$F(1,254) = 8.36$, p = .0042] on the correction of CVA. Similar results were identified for both the SBA [$F(1,254) = 5.53$, p = .0195] and the OTDA [$F(1,254) = 5.22$, p = .0231] asymmetries.**

Conclusions: **These findings support clinical observations that earlier intervention results in significantly improved treatment of plagiocephaly, independent of the severity of the presenting asymmetries.**

KEY WORDS: *Anthropometrics, clinical outcomes, Dynamic Orthotic Cranioplasty (DOC), positional plagiocephaly*

Positional plagiocephaly refers to an asymmetrical condition of the head arising from extrinsic molding rather than intrinsic synostotic events. The condition is primarily characterized by right or left occipital flattening, with advancement of the ipsilateral ear and prominence of the ipsilateral frontal region. Although an asymmetrical cranial vault is often the most evident feature, infants with positional plagiocephaly frequently exhibit complex multistructural asymmetries that also affect the face and skull base.

These positional deformations result from environmental factors, such as premature birth (Largo and Duc, 1977), restrictive intrauterine environment (Dunn, 1969, 1976; Clarren and Smith, 1977), birth trauma (Clarren et al., 1979), torticollis (Jones, 1969; Watson, 1971; Clarren, 1981; Kawamoto, 1987), sleeping position (Baum and Searls, 1971; Clarren et al., 1979; Clarren, 1981; Bruneteau and Mulliken, 1992; Haung et al., 1995; Huang et al., 1996), lack of full bone mineralization (Baum and Searls, 1971), and neurological deficits (Moss, 1958) and are distinctly different from the temporary deformation that normally occurs during the birthing process. For,

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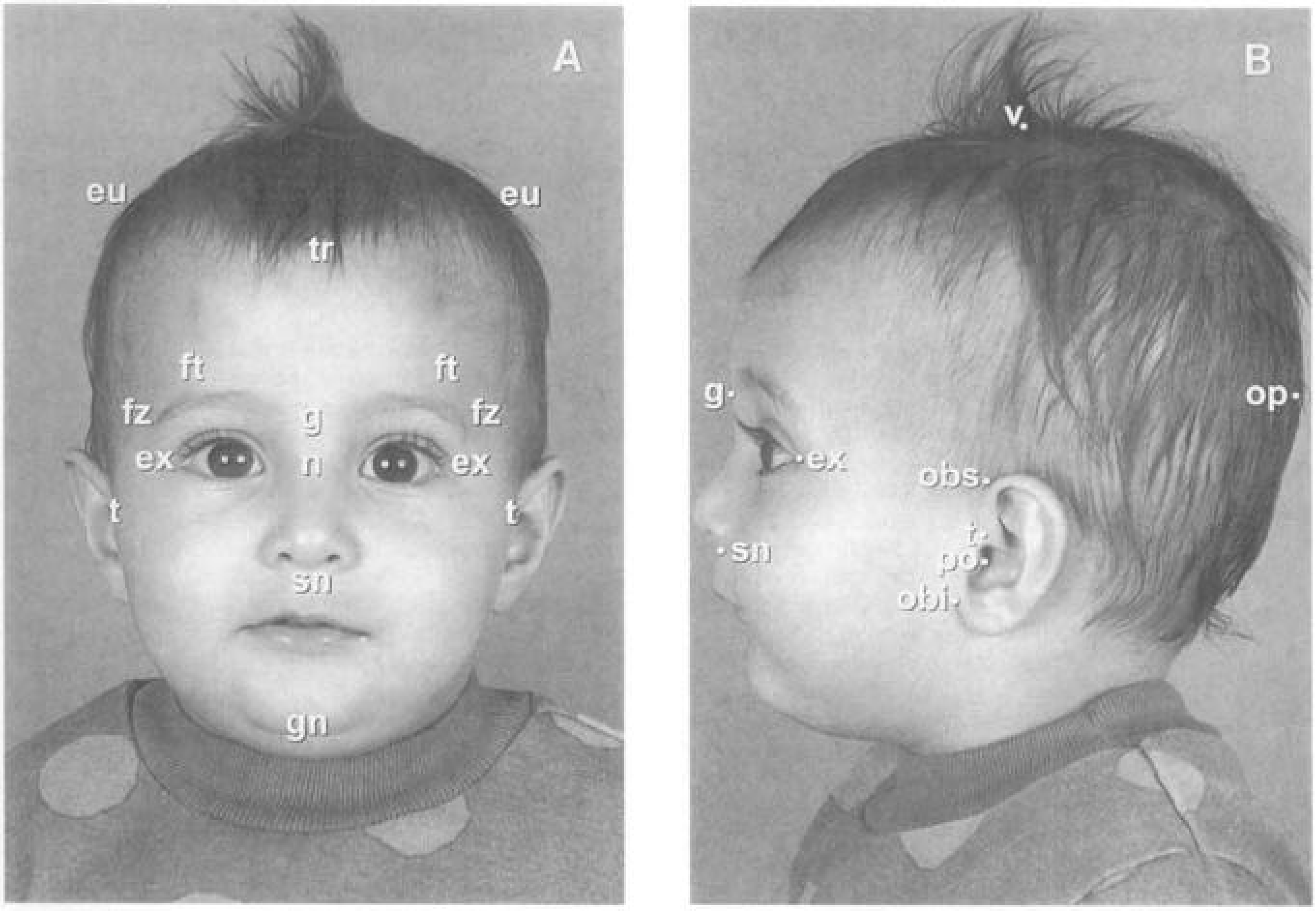

FIGURE 1 **Frontal (A) and lateral (B) views of the key anthropometric landmarks used in the assessment of positional plagiocephaly: eu-eu = maximum cranial breadth; ft-ft = minimum frontal breadth; t-t = cranial base width; g-op = maximum cranial length; fz-fz = supraorbital breadth; fz-g, R, L = supraorbital half-breadth; fz-g-fz, R, L = surface; go-op = head circumference; v-n = anterior head height; v-po = auricular head height; v-gn = total craniofacial head height; fz R-eu L, fz L-eu R = cranial vault asymmetry; sn-t, R, L = morphological face height (skull base asymmetry); ex-t R, L = orbitotragial depth asymmetry.**

although the molding associated with birth gradually resolves in the first 6 weeks of life, the asymmetries associated with positional deformation often persist into adulthood (Danby, 1962; Marsh and Vannier, 1985).

Clinical management of positional plagiocephaly has been as varied as its etiology (e.g., Clarren et al., 1979; Nichter et al., 1986; Pattisapu et al., 1989; Pollack et al., 1997). Our treatment protocol involves nonsurgical management using orthotic cranioplasty. The orthosis is a proprietary thermoplastic construct (Pomatto, 1992) detailed elsewhere (Ripley et al., 1994). We have described this treatment and documented significant reductions of the associated cranial asymmetries by orthotic cranioplasty (Ripley et al., 1994; Littlefield et al., 1998). However, as with any method of intervention, the desired outcome is not always achieved.

In the current study, we examined three factors thought to affect treatment outcome: (1) the initial severity of the asymmetry, (2) the age at which the child enters treatment, and (3) the total treatment time. We focused on these variables because during the course of ongoing program assessment (Ripley et al., 1994; Pomatto et al., 1997; Littlefield et al., 1998), these factors recurred as likely explanations for variable outcomes. The purpose of this study was to evaluate the influence of these factors on the treatment outcome using objective measures and methods.

Materials and Methods

From 1988 to 1995, 756 patients were treated in our clinic using orthotic cranioplasty. Prior to treatment, a detailed medical history was obtained on each patient. The history included information regarding: (1) demographics: sex, race, and date of birth; (2) perinatal—breech, cesarean, forceps, suction, epidural, prematurity, complications, and multiple gestations; and (3) potential etiology—congenital anomalies, *in utero* positioning, sleeping position, torticollis, neck tightness, head tilt, and synostosis. These data, along with clinical notes and the anthropometric measurements described below, were entered into a spreadsheet for storage and analysis.

From start to completion of treatment, anthropometric measurements were taken at 6-week intervals to quantify correction and to clarify the relationship between growth parameters

TABLE 1 Correction of Asymmetry (mean [±SD] in mm)

Parameter	*Pre-treatment*	*Post-treatment*	*Difference (Correction)*	*T*	*Prob.*
Cranial vault asymmetry	8.8 (±3.7)	3.3 (±2.9)	5.5 (±3.2)	27.6	.0001
Skull base asymmetry	6.2 (±2.9)	3.2 (±2.0)	3.0 (±2.4)	20.2	.0001
Orbitotragial depth asymmetry	4.4 (±2.2)	2.4 (±2.7)	2.0 (±1.9)	17.2	.0001

and the response to the orthosis. The anthropometric measurements were obtained by an experienced anthropologist using precision spreading and sliding calipers, along with a linen measuring tape. A total of 14 measurements, as described by Farkas (1981), Hajnis (1972), and Kolar and Salter (1997), were recorded on data forms and entered into a spreadsheet. The most clinically useful measurements describing plagiocephaly are those that define the symmetry or asymmetry of the cranial vault, the skull base, and the upper face (Fig. 1).

Subjects for the current investigation were retrieved from the complete data set based on a strict set of inclusion criteria. Patients included in this analysis: (1) had been treated by orthotic cranioplasty prior to 1 year of age, (2) had been compliant with the treatment protocol, (3) had been diagnosed by the referring physician with plagiocephaly of nonsynostotic origin, (4) did not have any congenital anomalies or other significant medical condition, and (5) had complete anthropometric measurements at entrance and exit from the program that had been obtained by a single experienced anthropologist.

One year of age was chosen as a cut-off based on the physiological consideration that 85% of the postnatal skull growth occurs during the first year of life (Pomerance, 1979). By placing the cut-off at 1 year of age and excluding patients with synostosis or other congenital/medical conditions, we ensured that all subjects in the study had some potential for correction.

Treatment was evaluated using the record of anthropometric measurements. Paired t tests (post- minus pretreatment measures) were used to evaluate the significance of changes in the anthropometric measures, whereas the contributions of the treatment factors (i.e., age at beginning treatment, total treatment time, and initial asymmetry) to final correction were assessed using three-way analysis of variance (ANOVA) with main effects (SAS, 1989). Differences were considered significant if $p < .05$.

Results

The study sample consisted of 258 children meeting the inclusion criteria: 59.3% were male, and 69.9% presented with right plagiocephaly (i.e., flattening of the right parietooccipital region). All patients presented with cranial vault asymmetry (CVA). In addition to CVA, 246 patients (92%) exhibited concurrent skull base (SBA) and orbitotragial depth (OTDA) asymmetries.

Mean age at the start of treatment for the 258 patients in the study was 6.5 (±1.9) months (range, 2.8 to 11.0 months), with 132 of the patients (51.2%) presenting for initial treatment prior to 6 months of age. Average treatment time was 4.1 (±2.2) months (range, 1.3 to 15.5 months), with eight patients (3.1%) requiring two orthoses. Mean pre- and posttreatment measures of asymmetry are presented in Table 1. Mean CVA was reduced from 8.8 to 3.3 mm, where mean SBA decreased from 6.2 to 3.2 mm. Orbitotragial asymmetry was reduced from a pretreatment mean of 4.4 to 2.4 mm following orthotic management. Statistical analyses based on paired comparison of pre- to posttreatment measurements demonstrated significant reductions in asymmetry in all three regions (Table 1), confirming previous findings (i.e., Ripley et al., 1994; Littlefield et al., 1998).

The effects of the treatment variables were assessed using a three-way ANOVA with main effects (Tables 2 through 4). The analysis assessed the combined effects of all treatment variables (i.e., the model), as well as the portion of the variability explained by each treatment effect. As expected, the combined contributions of the three treatment effects (i.e., age at beginning treatment, total treatment time, and initial asymmetry) were significantly associated with the amount of correction ($p = .0001$). The contribution of each effect was evaluated using the Type III sum of squares. In all three regions, the initial severity of the asymmetry was significantly associated with the amount of correction ($p = .0001$). By contrast, having taking into account initial severity and the entrance age, total treatment time did not significantly contribute ($p > .05$) to the amount of correction. More importantly, having accounted for initial severity, we found that entrance age had a statistically significant effect [$F(1,254) = 8.36$, $p = .0042$] on the correction of CVA (Table 2), with similar results identified

TABLE 2 Change in Cranial Vault Asymmetry (CVA)

	Model	*F Value*	*Pr > F*
		68.41	.0001
Source	Type III SS	F value	Pr > F
Age at fitting	48.01	8.36	.0042
Entrance CVA	1065.06	185.44	.0001
Total treatment time	.01	.00	.9657

TABLE 3 Change in Skull Base Asymmetry (SBA)

	Model	*F Value*	*Pr > F*
		68.41	.0001
Source	Type III SS	F value	Pr > F
Age at fitting	14.83	5.53	.0195
Entrance SBA	792.34	295.20	.0001
Total treatment time	10.19	3.80	.0524

TABLE 4 Change in Orbitotragial Depth Asymmetry (OTDA)

	Model	*F Value*	*Pr > F*
		68.41	.0001
Source	Type III SS	F value	Pr > F
Age at fitting	9.61	5.22	.0231
Entrance OTDA	395.33	214.69	.0001
Total treatment time	.95	.52	.4729

for SBA [$F(1,254) = 5.53$, $p = .0195$] (Table 3) and OTDA [$F(1,254) = 5.22$, $p = 0.0231$] (Table 4).

Discussion

We have long realized that the ideal candidate presents in the first 6 months of life. In complex, more severely affected patients, and for those infants who present at an older age, correction is slower. The current findings support clinical observations that earlier intervention results in significantly improved correction of asymmetry. Moreover, since total treatment time did not independently contribute to explaining variability in the amount of correction, these analyses suggest that treatment is more effective in younger children because they have greater potential for growth and not because they are treated longer. Age is important, because correction relies on capturing the rapid growth period of the head to correct the asymmetries. As this window of rapid growth passes, the efficacy of orthotic treatment decreases proportionally as the increasing cranial rigidity gradually diminishes the molding potential.

We have identified two factors that affect our ability to correct positional plagiocephaly. Clearly, the efficacy of the orthotic treatment depends on many factors, most notably the parents' compliance with treatment. However, the importance of the current study cannot be understated. Although the parents of our patients report having noted the abnormal head shape at a mean age of 1.9 months, the average entrance age is 6.5 months. Many of the parents reported that "we had tried repositioning" or "we were waiting for the head to correct on its own." Consequently, treatment is often delayed or even neglected. Unfortunately, clinical experience and anthropometry data suggest that for many of these untreated children, the anticipated spontaneous correction will not occur. For this reason, early recognition and treatment of plagiocephaly are essential to obtaining the optimum clinical outcome.

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