

# Neurodevelopmental Delays in Children with Deformational Plagiocephaly

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**Background:** The purpose of this study was to determine whether, in fact, infants with deformational plagiocephaly, or plagiocephaly without synostosis, demonstrated cognitive and psychomotor developmental delays when compared with a standardized population. Through this study, we chose to expand upon our earlier findings from 2001 on patients with deformational plagiocephaly.

**Methods:** The study population includes a total of 110 consecutive patients, prospectively followed then retrospectively reviewed. Each infant was assessed using the Bayley Scales of Infant Development-II scoring system. The developmental analysis was categorized as either mental or psychomotor using the mental developmental index or the psychomotor developmental index, respectively. These infants were subcategorized into four groups: accelerated, normal, mild, or severely delayed. The groups were then compared with a standardized Bayley's age-matched population, using chi-square test goodness-of-fit tests.

**Results:** Infants with deformational plagiocephaly were found to have significantly different psychomotor development indexes and mental developmental indexes when compared with the standardized population ( $p < 0.0001$ ;  $p < 0.0001$ ). With regards to the mental developmental index scores, none of the infants with deformational plagiocephaly were accelerated, 90 percent were normal, 7 percent were mildly delayed, and 3 percent were severely delayed. With regards to the psychomotor development index scores, none of infants were accelerated, 74 percent were normal, 19 percent were mildly delayed, and 7 percent were severely delayed.

**Conclusions:** This study indicates that before any intervention, infants with deformational plagiocephaly show significant delays in both mental and psychomotor development. Also of particular note is that no child with deformational plagiocephaly showed accelerated development. (*Plast. Reconstr. Surg.* 117: 207, 2006.)

Debate continues to rage over the significance and cause of craniosynostosis.<sup>1-10</sup> Unfortunately, the phenomenon of deformational plagiocephaly seems to have been forgotten somewhat in this debate. Most researches have presumed that plagiocephaly without synostosis is not associated with any developmental delays. However, data have been presented hinting that this notion may be incorrect. Miller and Clarren<sup>11</sup> performed a review of 254 patients with deformational plagiocephaly and found that 39.7 percent of those patients required special education and assistance. Similarly, in assessing motor development, Davis et

al.<sup>12</sup> found that children with deformational plagiocephaly had poor motor tone and required muscle training and development. These findings have an important effect on the general population as the prevalence of deformational plagiocephaly has increased dramatically since the 1992 sudden infant death syndrome campaign of the American Association of Pediatricians.<sup>13</sup> In 1992, in an attempt to avert the increasing numbers of sudden infant death syndrome, the American Association of Pediatricians recommended that children be placed supine while sleeping. This had immediate effect both in decreasing the number of sudden infant death syndrome cases and in increasing the number of infants with deformational plagiocephaly. Argenta et al.<sup>14</sup> noted this increase in prevalence in their study in 1996. This was corroborated by the results of Kane et al.<sup>15</sup> In 2001, in an effort to further our understanding of this phenomenon and its effects, we presented our initial findings based on 42 infants with defor-

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mational plagiocephaly and demonstrated that a significant number of these patients were affected developmentally, with delays both in mental and psychomotor function. To better understand and highlight these findings, we continued our study in a prospective manner and now present a larger population of 110 consecutive patients with deformational plagiocephaly.

## PATIENTS AND METHODS

### Patients

#### Study Population

One hundred ten consecutive infants with radiographically confirmed deformational plagiocephaly without synostosis were enrolled in this prospective study between 1997 and 2003. The mean ages of the patients studied were 0.69 years of age (range, 3 months, 7 days to 10 months, 17 days) at the time of initial evaluation. There were a total of 69 boys and 41 girls.

A smaller population of 63 patients was retrospectively studied with particular emphasis on available confounding data points. The data collected were low birthweight (less than 5.5 pounds at time of birth)<sup>16</sup>; prematurity (less than 37 weeks at time of birth)<sup>17,18</sup>; mothers' exposure to alcohol, cocaine, or recreational drugs; family history of congenital defects; multiple gestations; type of delivery (vaginal or cesarean delivery); early illness and/or intensive care unit admission, failure to thrive, congenital defects (i.e., cleft lip or cleft palate), syndromic conditions (i.e., Pierre-Robin sequence or VATER syndrome (vertebral/vascular abnormalities, anal atresia, tracheoesophageal fistula, esophageal atresia, and renal abnormalities), and torticollis. This information was obtained by evaluating patient charts and birth records retrospectively. If the data were not found, the patient's family was contacted by members of the research team using telephone or mail and the data were then sequestered. Once contacted, a series of questions was asked of the family members and the most complete data were collected. For 47 patients, this method was not successful and, therefore, the patients were excluded from detailed analysis.

Of this small population of 63 patients for whom confounding variable data were available, a smaller group of 23 infants also was studied. This smaller group of infants was without any abnormalities or any confounding variables.

#### Neurodevelopment Assessment

In the neurodevelopment assessment, variability was kept to a minimum as the same licensed

psychologist and the same professional counselor from the Child Study Center and the Department of Pediatrics at the University of Oklahoma Health Sciences Center assessed all of the enrolled patients. The infants were assessed using the Bayley Scales of Infant Development-II, a standardized measure that provides indices of development; the mental development index; and psychomotor development index. It is designed for use with infants from 1 month to 42 months of age. The mental development index assesses cognitive, language, and personal-social abilities while the psychomotor development index assesses fine and gross motor skills. These indexes have a mean of 100, with an SD of 15 and 16, respectively.<sup>2</sup> An infant's score is derived by comparing the infant's performance to a standardized same-age sample.

Four constructs have been identified in the Bayley Scales evaluation: cognitive, language, motor, and personal/social. For the infants in this study, there was a focus on early language skills, problem-solving abilities, imitation skills, fine motor abilities (such as grasping objects), and gross motor skills. Because of this focus, administration of the Bayley Scales required particular skills and expertise in the areas of infant and child development. The evaluation had to be conducted by an expert with training in the administration and interpretation of comprehensive and developmental skill measurements.

The Bayley Scales evaluation took approximately 1½ hours and the assessments were conducted with the infants' caregivers present. The cognitive portion of the test was administered with the child seated on the caregiver's lap at a table and/or lying on the floor. The motor portion was conducted with the child on the floor. If the child was walking, several items were completed in a physical therapy motor room.

Once the Bayley Scales evaluation was completed, each infant was given a motor development index and a psychomotor development index score. Both standard means were based on a score of 100. For the mental development index, each score was categorized as accelerated if the score was greater than 115 (SD, > +1); normal if the score was between 85 and 115 (SD, -1 to +1); mildly delayed if the score was between 70 and 84 (SD, -2 to -1); and severely delayed if the score was 69 or less (SD, < -2). For the psychomotor development index, each score was categorized as accelerated if the score was greater than 116 (SD, > +1); normal if the score was between 84 and 116 (SD, -1 to +1); mildly delayed if the score was

between 68 and 83 (SD,  $-2$  to  $-1$ ); and severely delayed if the score was 67 or less (SD,  $< -2$ ).

The Bayley Scales evaluation was administered before any type of intervention (i.e., molding helmet therapy) was instituted. For premature infants, standard age adjustments were made in accordance with the Bayley Scales evaluation up to 24 months of age during testing.

## Methods

Once neurodevelopmental assessment was complete and scores were calculated, the children were categorized into one of four groups, as listed above. The frequency distributions of the four groups (accelerated, normal, mildly delayed, and severely delayed) were then compared with the expected frequency derived from the Bayley Scales standardized sample.

### Statistical Methods

#### Chi-Square Test

An exact chi-square goodness-of-fit test was used to compare the distribution of the developmental scores in the larger study population and the Bayley Scales evaluation standardized sample. An exact chi-square goodness-of-fit test was also used to compare the distributions in the smaller population of 63 patients for which confounding data points were available. The chi-square apparatus was again used to analyze the diagnosticity of the sample population of 23 infants in whom all confounding data points were negative.

#### Multivariate Exploration of Confounding Factors

With regards to the smaller population of 63 patients, a subcategory analysis was applied to all confounding factors. All confounding measures were dichotomously coded as “yes = 1” (confound present) and “no = 0” responses (excluding sex, male patients = 1, female patients = 0) and subjected to two exploratory approaches. In addition, quantitative values of low birthweight were assessed for finer measurement relationships. The first exploratory approach predicted severity of each Bayley Scales measure separately using a stepwise selection

procedure in a multiple-regression analysis. The second approach examined the multivariate prediction of both the mental development index and the psychomotor development index using a fully specified linear canonical regression equation. Canonical correlation analysis creates  $m$  orthogonal factors (where  $m$  is the smallest number of variables on either side of the multivariate equation) by creating  $m$  linear combinations of the dependent and independent variables. The combinations are derived such that the correlation among pairs of summed factors (i.e., a summed dependent and independent factor) is maximized. (For a basic applied summary, see Tabachnick & Fidell, 2001; for more detailed information, see Johnson & Wichern, 1998.) All exploratory procedures were run using SAS version 8.1 for Windows. The stepwise selection regressions were performed using the SAS PROC REG procedure with entry and removal criteria set at  $p = 0.15$ . The canonical analysis was run within SAS PROC CANCORR.

## RESULTS

A total of 110 infants, 69 boys and 41 girls, were studied. A mental development index and psychomotor development index score was tabulated for each of the patients, for a total of 220 data points. When the mental development index and psychomotor development index scores were compared with the scores of a standard sample population, the scores for the patients with deformational plagiocephaly were found to be significantly different ( $p < 0.0001$ ). When the mental development index scores were examined, no patients were found to be in the accelerated group, 99 patients (90 percent) were in the normal group, eight patients (7 percent) were in the moderately delayed group, and three patients (3 percent) were in the severely delayed group. Table 1 shows the expected frequency of patients for each category group. When the psychomotor development index scores were examined, again no patients were found to be in the accelerated group,

**Table 1.** MDI Scores for Deformational Plagiocephaly Infants\*

MDI Score Groups	Distribution Expected (%)	Deformational Plagiocephaly (n)	Deformational Plagiocephaly (%)
Accelerated	16.5	0	0
Normal	68.7	99	90
Mild delay	12.5	8	7
Severe delay	2.3	3	3
Total	100	110	100
$p$		0.0001	

MDI, mental development index.

\* $n = 110$ .

81 patients (74 percent) were normal, 21 patients (19 percent) were moderately delayed, and eight patients (7 percent) were severely delayed. Table 2 shows the expected frequency of patients for each category group. The sample population had an expected frequency of 14.8 percent of infants with accelerated development. In patients with deformational plagiocephaly, none were accelerated. Also, while in the sample population there was an expected frequency of 11 percent moderate delay, and an additional 1.6 percent of severe delay, in the deformational plagiocephaly group studied, 26 percent were moderately or severely delayed. Both of these findings were statistically significant.

Of the 110 patients, more specific data were available on a small population of 63 patients, 40 boys and 23 girls (Table 3). In this small group, mental development index and psychomotor development index scores also were compared with the Bayley Scales of Infant Development-II standardized sample and, again, were found to be significantly different ( $p < 0.0001$ ; Tables 4 and 5). When the mental development index scores of this small population of infants were examined, no patients were found to be in the accelerated group, 57 patients (91 percent) were in the normal group, four patients (6 percent) were in the moderately delayed group, and two patients (3 percent) were in the severely delayed group. Table 4 shows the expected frequency of patients for each category group. When the psychomotor development index scores of this small population were examined, again, no patients were found to be in the accelerated group, 47 patients (75 percent) were normal, 12 patients (19 percent) were moderately delayed, and four patients (6 percent) were severely delayed. Table 5 shows the expected frequency for each category group.

In the population analyzed, the mental development index and psychomotor development index score frequencies were statistically significant. The mental development index sample popula-

tion had an expected frequency of 16.5 percent of infants with accelerated development. In the study population, no infant showed accelerated development. Similarly, the psychomotor development index sample population had an expected frequency of 14.8 percent of infants with accelerated development. In the deformational plagiocephaly patients, none were accelerated. Also, while in the sample population there was an expected frequency of 11 percent moderate delay, and an additional 1.6 percent of severe delay, in the deformational plagiocephaly group studied, 25 percent were moderately or severely delayed. Both of these findings were statistically significant.

In a smaller population of 23 infants, 15 boys and eight girls, all confounding factors were negative. No abnormalities were seen in any of the possible confounding data points collected. Interestingly, in this smaller population of plagiocephaly patients in whom all confounders were negative, the mental development index and psychomotor development index score frequencies compared favorably to the expected frequencies (Tables 6 and 7). When the mental development index scores were examined, no patients were found to be in the accelerated group, 21 patients (91.3 percent) were in the normal group, two patients (8.7 percent) were in the moderately delayed group, and none were in the severely delayed group. When the psychomotor development index scores were examined, again, no patients were found to be in the accelerated group, 20 patients (87 percent) were in the normal group, three patients (13 percent) were in the moderately delayed group, and none were severely delayed. The mental development index sample population had an expected frequency of 85.2 percent of infants with normal or accelerated development. In the deformational plagiocephaly patients with no confounding variables, the frequency was 91.3 percent. Similarly, the psychomotor development index sample population had an expected frequency of 87.4 percent of infants with normal or accelerated development. In the smaller population of 23 patients, a

**Table 2.** PDI Scores for Deformational Plagiocephaly Infants\*

PDI Score Groups	Distribution Expected (%)	Deformational Plagiocephaly (n)	Deformational Plagiocephaly (%)
Accelerated	14.8	0	0
Normal	72.6	81	74
Mild delay	11	21	19
Severe delay	1.6	8	7
Total	100	110	100
<i>p</i>		0.0001	

MDI, mental development index; PDI, psychomotor development index.

\* $n = 110$ .

**Table 3.** MDI and PDI Scores for Smaller Population of Deformational Plagiocephaly Infants\* with Confounding Variables

Patient	MDI	PDI	Sex	Low Birth Weight	Premature Birth	Cesarean Delivery	Multiple Gestations	CD	SC	FH	Torticollis	Sick/ICU	FTT	Mother Alcohol/Drug Use
1	93	81	M	N	N	N	N	N	N	N	N	N	N	N
2	100	91	M	N	Y	N	N	N	N	N	Y	Y	Y	Y
3	101	97	M	N	N	Y	N	N	N	N	N	N	N	Y
4	102	97	M	N	N	N	N	N	N	N	N	N	N	N
5	88	97	M	N	N	Y	N	N	N	N	N	N	N	N
6	91	94	M	N	N	N	N	N	N	N	Y	Y	N	N
7	101	107	F	N	Y	N	N	N	N	N	N	N	N	N
8	100	103	F	N	N	Y	N	N	N	N	Y	Y	N	N
9	92	79	M	N	N	Y	N	N	N	N	N	N	N	N
10	98	103	M	N	N	N	N	N	N	N	N	N	N	N
11	88	61	M	Y	N	Y	Y	Y	N	N	Y	Y	N	N
12	87	88	M	N	N	N	N	N	N	N	N	N	N	N
13	101	108	M	N	N	Y	N	N	N	N	Y	Y	N	N
14	85	83	M	N	Y	N	N	Y	N	N	N	N	N	N
15	89	60	M	N	N	Y	N	N	N	N	N	N	N	N
16	93	87	F	Y	N	Y	N	N	N	N	N	Y	N	N
17	94	100	F	N	N	N	N	N	N	N	N	N	N	N
18	97	102	M	N	Y	N	N	N	N	N	N	N	N	N
19	102	91	M	N	N	N	N	N	N	N	N	N	N	N
20	99	114	F	N	N	N	N	N	N	N	N	N	N	N
21	50	67	F	Y	Y	N	N	Y	N	Y	N	Y	N	N
22	101	92	F	Y	N	Y	Y	N	N	N	N	N	N	N
23	99	101	F	N	N	N	N	N	N	N	N	N	N	N
24	102	83	F	N	N	Y	N	N	N	N	N	N	N	N
25	101	91	M	N	N	Y	N	N	N	N	N	N	N	N
26	98	97	M	N	N	N	N	N	N	N	N	N	N	N
27	96	88	F	N	Y	N	N	N	N	N	N	N	N	N
28	96	97	M	N	N	N	N	N	N	N	N	N	N	N
29	96	80	F	Y	N	N	N	N	N	N	N	N	N	N
30	84	93	F	N	N	Y	N	N	N	Y	N	N	N	N
31	89	92	M	N	N	Y	N	N	N	N	N	N	N	N
32	96	95	F	N	N	N	N	N	N	N	N	N	N	N
33	94	88	M	Y	Y	Y	Y	N	N	N	Y	Y	N	N
34	88	81	M	N	Y	N	N	N	N	N	N	N	N	N
35	101	90	M	N	N	N	N	N	N	N	N	N	N	N
36	109	101	F	N	N	N	N	N	N	N	N	N	N	N
37	90	79	F	N	Y	N	N	N	N	N	N	N	N	N
38	101	88	M	N	N	N	N	N	N	N	Y	Y	N	N
39	56	62	M	N	N	N	N	N	N	N	N	Y	N	N
40	91	94	M	N	N	N	N	N	N	N	N	N	N	N
41	78	70	M	N	N	N	N	N	N	N	N	N	N	N
42	94	88	M	N	N	N	N	N	N	N	N	N	N	N
43	94	104	F	N	Y	N	N	Y	N	N	N	N	N	N
44	89	104	F	N	N	N	N	N	N	N	N	N	N	N
45	105	102	M	N	N	Y	N	N	N	N	N	N	N	N
46	91	91	M	N	N	Y	N	N	N	N	Y	N	N	N
47	97	95	M	N	N	Y	N	N	N	N	Y	N	N	N
48	93	101	M	N	Y	N	N	N	N	N	N	N	N	N
49	88	76	M	N	N	Y	N	Y	N	N	N	N	N	N
50	85	79	M	N	N	Y	Y	N	N	Y	N	N	N	N
51	81	75	M	N	N	Y	Y	N	N	Y	Y	N	N	N
52	96	88	M	N	N	N	N	N	N	N	N	N	N	N
53	109	85	M	N	N	Y	N	N	N	Y	N	N	N	N
54	98	94	F	N	N	N	N	N	N	N	Y	Y	N	N
55	93	102	F	N	N	N	N	N	N	N	N	N	N	N
56	82	69	F	N	N	N	N	N	N	N	N	N	N	N
57	93	88	M	N	Y	Y	N	N	N	N	N	N	N	N
58	98	86	M	N	N	N	N	N	N	N	N	N	N	N
59	89	88	M	N	N	N	N	N	N	N	N	N	N	N
60	93	97	F	N	N	Y	N	N	N	N	N	N	N	N
61	95	90	F	N	N	Y	Y	N	N	Y	N	N	N	N
62	101	90	F	Y	N	Y	Y	N	N	Y	N	N	N	N
63	98	98	M	N	N	N	N	N	N	N	N	N	N	N

MDI, mental development index; PDI, psychomotor development index; CD, congenital defects; SC, syndromic conditions; FH, family history of congenital defects; ICU, intensive care unit admission; FTT, failure to thrive.

\*n = 63.

**Table 4.** MDI Scores for Small Population of Deformational Plagiocephaly Infants\* with Available Confounding Data Points

MDI Score Groups	Distribution Expected (%)	Deformational Plagiocephaly (n)	Deformational Plagiocephaly (%)
Accelerated	16.5	0	0
Normal	68.7	57	91
Mild delay	12.5	4	6
Severe delay	2.3	2	3
Total	100	63	100
<i>p</i>		0.0001	

PDI, psychomotor development index.  
\**n* = 63.

**Table 5.** PDI Scores for Small Population of Deformational Plagiocephaly Infants\* with Available Confounding Data Points

PDI Score Groups	Distribution Expected (%)	Deformational Plagiocephaly (n)	Deformational Plagiocephaly (%)
Accelerated	14.8	0	0
Normal	72.6	47	75
Mild delay	11	12	19
Severe delay	1.6	4	6
Total	100	63	100
<i>p</i>		0.0001	

PDI, psychomotor development index.  
\**n* = 63.

**Table 6.** MDI Scores for Smaller Population of Deformational Plagiocephaly Infants\* with no Abnormalities (Negative Confounders)

MDI Score Groups	Distribution Expected (%)	Deformational Plagiocephaly (n)	Deformational Plagiocephaly (%)
Accelerated	16.5	0	0
Normal	68.7	21	91.3
Mild delay	12.5	2	8.7
Severe delay	2.3	0	0
Total	100	23	100
<i>p</i>		0.09	

MDI, mental development index.  
\**n* = 23.

frequency of 87 percent of infants showed normal or accelerated development.

**Confounding Factors**

The data available on this smaller population of 63 infants were scrutinized in regards to possible confounding factors, such as low birthweight, prematurity, mother’s alcohol and/or drug use in the prenatal period, family history of congenital disease, multiple gestations, type of delivery, early perinatal sickness and/or intensive care unit admission, failure to thrive, congenital defects, syndromic conditions, or torticollis. The complete data set is presented in Table 3.

**Sex**

A total of 63 infants were studied, 40 boys and 23 girls. The average age at the initial time of evaluation was 0.67 years of age (range, 3 months, 7 days to 10 months, 16 days).

**Low Birthweight**

Eight of the 63 infants (12.7 percent) were born with a weight of less than 5.5 pounds (range, 3 to 5.4 pounds; average, 4.4 pounds). Two were boys and six were girls. Of these eight, four needed intensive care unit admission and early care, but all survived. None had subsequent failure to thrive. In retrospective review, none of the infants’

**Table 7.** PDI Scores for Smaller Population of Deformational Plagiocephaly Infants with no Abnormalities (Negative Confounders)

PDI Score Groups	Distribution Expected (%)	Deformational Plagiocephaly (n)	Deformational Plagiocephaly (%)
Accelerated	14.8	0	0
Normal	72.6	20	87
Mild delay	11	3	13
Severe delay	1.6	0	0
Total	100	23	100
<i>p</i>		0.18	

PDI, psychomotor development index.  
*n* = 23.

mothers had perinatal exposure to drugs or alcohol. Three of these six had a congenital defect or defects detected at birth, one of which was familial in nature.

### Prematurity

Twelve of 63 infants (19.1 percent) were premature (born before 37 weeks of gestation). Seven were boys and five were girls. Of the 12, two had low birthweight, one of which was the result of a multiple gestation. Three needed intensive care unit admission, with one having failure to thrive subsequently. This one male infant had a severe degree of exposure to alcohol and drugs through the mother in the prenatal period. Interestingly, this patient also had torticollis.

Three of the 12 infants had congenital defects detected at the time of birth. None had any syndromic presentations. One infant had symphalangism that was previously noted in her family.

### Alcohol or Drug Use by the Mother

Two of the 63 infants (3.2 percent) were identified as having mothers who had exposure to drugs and alcohol during the prenatal period. Both were male. This prenatal exposure was corroborated by bloodwork at the time of birth that was positive for cocaine in the bloodstreams of these infants. Neither of these infants had low birthweight and neither was premature. Neither infant had any evidence of any congenital disease nor any syndromic presentation. One infant did, however, have significant difficulty after birth and required extended admission and care in the intensive care unit. He subsequently suffered from failure to thrive.

### Family History of Congenital Defects

Seven of 63 infants (11.2 percent) had a family history of deformational plagiocephaly. Three were male and four were female. Of the seven,

four infants were twins, both of whom had deformational plagiocephaly. The other three infants had various immediate family members with a history of an abnormally shaped head at birth. Six of the seven had cesarean delivery. Of the seven total infants, two had low birthweight while only one was premature. One infant presented with a congenital defect. This same infant showed early difficulty and required additional intensive care. None of the infants presented with any syndromic conditions and none had any subsequent failure to thrive.

### Multiple Gestations

Seven of 63 infants (11.2 percent) were the result of multiple gestations. All seven infants were had cesarean delivery. One was premature by dates. Four of the seven were low in birthweight, as would be expected for a multiple-gestational child. One infant, a male, had multiple congenital defects including a tracheoesophageal fistula, a duodenal atresia, and hypospadias. No underlying syndromic presentation was detected, however. The patient underwent several corrective surgical procedures. Of the seven, two had early sicknesses requiring additional intensive care. None had failure to thrive or maternal drug or alcohol exposure.

### Route of Delivery (Vaginal or Cesarean Delivery)

Twenty-five of 63 infants (39.7 percent) had cesarean delivery. Seventeen were male and eight were female. Five infants had low birthweight at time of delivery, one of whom was premature. An additional infant delivered by cesarean was preterm, but he was of regular weight. Of the total 25 infants, seven were the result of multiple gestations. Two showed congenital defects. None of the 25 had syndromic presentations. Six had a family history of deformational plagiocephaly, while one had a history of maternal drug exposure. While

five of the 25 had need for early care, none had any subsequent difficulty or failure to thrive.

### Early Perinatal Sickness/Intensive Care Unit Admission

Eleven of 63 infants (17.5 percent) needed early perinatal care and intensive care unit admission. Four were female and seven were male. Four of these infants had low birthweight, less than 5.5 pounds; two were premature. Another two infants had evidence of congenital disease. One of these two had a difficult presentation with a tracheoesophageal fistula and duodenal atresia (mentioned previously) requiring more rigorous care and surgical correction. This child went on to do well. Of the eleven, only one had failure to thrive and he had positive cocaine exposure through his mother.

### Failure to Thrive

Only one of 63 infants (1.6 percent) in our study group had failure to thrive. He was born at 36 weeks of gestation, with a birthweight of 7 pounds. He had no evidence of any congenital or syndromic diseases. He did have torticollis. He had positive cocaine blood tests. He went on to have an extended intensive care unit stay with subsequent failure to thrive. At a recent 5-year follow-up, he appears to meet the weight and height criterion for his age. Unfortunately, no psychomotor development index or mental development index follow-up is available since the adoptive parents do not wish to have the child undergo any further testing.

### Congenital Defects

Five of 63 infants (8 percent) had congenital deformities. One had a tracheoesophageal fistula, duodenal atresia, and hypospadias; one had hypospadias only; one had symphalangism; one had mandibular hypoplasia; and one had cryptorchidism. Three were male and two were female. Three had low birthweight. Two had intensive care unit admissions but none had failure to thrive. None had prenatal exposure to drugs or alcohol

through their mother. One infant did have a family history of congenital disorders. None had syndromic presentations.

### Syndromic Diseases

None of the patients observed in the smaller group had a syndromic presentation. In the larger group of 110 infants, a female infant presented with VATER syndrome. By report, her family apparently had a history of congenital disorders. This could not be substantiated or refuted. Unfortunately, other details about her prenatal care, her birth, and her perinatal care were not obtainable since she was quickly adopted. For this reason, she was excluded from the subcategory analysis.

### Torticollis

Eleven of 63 patients (17.5 percent) had evidence of torticollis. Nine were male and two were female. Two had low birthweight and two were premature. Seven of the 11 had cesarean delivery. Two of these were multiple gestations. One had a congenital presentation. Eight of the 11 needed intensive care. Of these eight, one had failure to thrive but also had a history of maternal drug exposure.

### Multivariate Analysis of Confounding Factors

The stepwise selection method for prediction of the mental development index severity chose a four-variable model that included congenital defects, family history of congenital defects, early sickness (requiring intensive care unit admission), and evidence of torticollis. Parameter estimates and model statistics are presented in Table 8. The full model accounted for nearly 29 percent of the total sample variance in the mental development index,  $F(4,58) = 5.88$ ,  $p < 0.01$ . The model prediction of psychomotor development index demonstrated similar success. Final selection left five predictors: low birthweight, premature birth status, congenital defects, family history, and sex, that, altogether, accounted for 30 percent of the psychomotor development index variance

**Table 8.** Model Estimates for MDI Stepwise Regression

Predictor	DF	Estimate	SE	t	p
Intercept	1	95.25	1.29	74.01	< 0.01
Congenital defect	1	-10.27	4.09	-2.51	0.01
Family history	1	-7.09	3.43	-2.06	0.04
Torticollis	1	8.99	3.84	2.34	0.02
ICU admission	1	-10.92	3.90	-2.80	< 0.01

MDI, mental development index; ICU, intensive care unit

**Table 9.** Model Estimates for PDI Stepwise Regression

Predictor	DF	Estimate	SE	t	p
Intercept	1	73.71	7.75	9.51	< 0.01
Low birthweight	1	3.15	1.08	2.91	< 0.01
Premature birth	1	5.62	3.65	1.54	0.13
Congenital defect	1	-12.11	5.03	2.41	0.02
Family history	1	-6.39	4.22	1.52	0.14
Sex*	1	-8.81	2.88	3.05	< 0.01

PDI, psychomotor development index.

\*Sex effect is estimated for male participants; it represents the expected differences in male patients (compared with female patients) in mean psychomotor development index after controlling other model variates.

( $F(5,57) = 4.79, p < 0.01$ ). Model estimates are listed in Table IX. Interestingly, of all the confounding factors used, the presence of torticollis seems to be positively correlated with a higher mental development index. This is more than likely an artifact due to sampling error.

Due to the shared variance between the two low-birthweight variables, only the quantitative measure was kept for the fully specified canonical regression. The first canonical factor solution was statistically significant, ( $F(22,100) = 1.99, p = 0.01$ ), with an overall correlation estimate of 0.62 (the second factor was not significant,  $p > 0.20$ , and will not be discussed).

Our canonical factor significance tests should be judged with a moderate degree of skepticism given the normality and homoscedasticity assumptions inherent in this inferential technique. Our inclusion of ordinal variates may have substantially affected the difference between actual and nominal alphas. Descriptive correlational information, on the other hand, is unaffected by these assumption violations.

The dependent factor(s) heavily favored the mental development index measurement ( $r = 0.96$ ) but still correlated substantially with psychomotor development index ( $r = 0.81$ ) (both measures were already highly correlated,  $r = 0.61$ ). Given this predominance toward the mental development index measurement, not surprisingly, three of the five most substantially weighted predictors were intensive care unit admission, family history, and congenital defects (Table 10, for standardized weight estimates). The addition of two major psychomotor development index predictors (Table 9), sex and low-birthweight, completed this list of five. With the exception of sex, raw measurements of these same variables gave the highest correlations with the dependent canonical factor (Table 10). The independent canonical factor correlated highly with both mental development index and psychomotor development index, accounting for 36 percent and 26 percent of their variances, respectively.

## DISCUSSION

We prospectively collected data on 110 infants with radiographically proven plagiocephaly without synostosis. We conducted mental and psychomotor testing on these infants early in their care. We then followed these patients as closely as we could over time. Our initial analysis of their motor and psychomotor testing showed the population to have significant delays. We came to this conclusion by giving infants mental development index and psychomotor development index scores and comparing these scores to the Bayley Scales of Infant Development-II standardized samples. We found that the study population did not meet the expected frequencies. In the sample population, we expected to see mental development index and psychomotor development index score frequencies in the accelerated range (16.5 percent and 14.8 percent, respectively). We saw none. In the psychomotor development index sample population, we expected a frequency of 12.6 percent of moderately delayed and severely delayed pa-

**Table 10.** Standardized Weights for and Correlations with First Canonical Factors

	Standardized Weights	Correlations with Opposite Factor
Dependent factor		
MDI	0.737	0.96
PDI	0.361	0.81
Independent factor		
ICU admission	-0.57	-0.37
Family history	-0.54	-0.42
Sex*	-0.51	-0.16
Low birthweight	0.46	0.40
Congenital defect	-0.46	-0.61
Torticollis	0.40	0.08
Cesarean delivery	0.25	0.03
Multiple gestation	0.18	-0.19
Premature birth	0.14	-0.19
Prenatal drug exposure	0.11	0.20
Failure to thrive	0.09	0.11

MDI, mental development index; PDI, psychomotor development index; ICU, intensive care unit.

\*Sex effect is estimated for male participants; it represents the expected male differences (compared with females) in mean psychomotor development index after controlling other model variates.

tients. We saw a frequency of 29 percent, more than two times what was expected. Each of these findings was then scrutinized using a chi-square test and was found to be statistically significant ( $p < 0.0001$ ). Patients with plagiocephaly without synostosis did worse and were developmentally delayed. To check the validity of our sample population, a smaller group of 23 patients, in which confounding variable data were available and, more importantly, no abnormalities were noted, was selected. The mental and psychomotor developmental scores and frequencies were tabulated and found to be comparable to those expected in the general population. These scores also bring to light that children without any of the confounding factors described above do not have an increased incidence of developmental delays despite having a deformational plagiocephaly.

The exact underlying physiology and cause for our findings are not understood and are beyond the scope of this discussion. However, in an effort to highlight associations or confounding factors, much more specific data were collected in a retrospective fashion on these infants. Data on only 63 patients could be collected completely. For this reason, 47 infants were excluded from the subcategory analysis. The group of 63 infants was compared with the larger population of 110 infants in their psychomotor development index and mental development index scores and population frequencies and was found to be representative.

Using multivariate analysis, we found a high degree of correlation between the confounding variables studied and the mental development index and psychomotor development index scores, respectively. These correlations were statistically significant. Given the similar findings between the stepwise and the canonical procedures of analysis, the confounding variables chosen for investigation appear to account for the severity variation of *both* mental and psychomotor development in our smaller sample. The five most consistent predictors of this variation were sex, low birthweight, family history, congenital defects, and early sickness/intensive care unit admission (Table 10). Based upon these findings, we initiated inquiries into common practices in our intensive care units, since the intensive care unit/early sickness variable was most weighted of all of the confounding variables. We found that the common practice now in the intensive care unit care for preterm and premature infants is careful cardiac and respiratory monitoring. In practical terms, this translated into more cardiac and respiratory monitoring devices. Logistically, the presence of additional mon-

itoring wires and machines prevented parents from lifting their infants from the cribs to hold them. They were more likely to allow them to lay in one position or another for long periods of time. This restriction in head positioning also applied to the nursing teams who were noted inadvertently not to lift the infants as often, only adding to the time that the infants' heads and positions were kept stationary. This pattern of behavior, both on the part of parents and nursing teams, most likely has a direct correlation with the increased incidence of deformational plagiocephaly in the intensive care unit infants. We have begun efforts to change this pattern of behavior.

When the initial data from our institution was published in 2001, criticisms were made about the validity of our findings. This larger study only further substantiates our original data. Dr. Persing made an astute criticism<sup>19</sup> noting that our findings may be due to our exclusive use of the Bayley Scales of Infant Development-II evaluation for our analysis of development in these infants. We agree that the battery of tests such as the Wechsler Intelligence Scale for Children, Wide Range Achievement Test, and the Vineland Test may be better in some respects in categorizing development. However, these tests are designed for older children. The Wechsler Intelligence Scale for children is used only with children 2½ years of age and older. The Wide Range Achievement Test is used for children older than five. This battery of tests, unfortunately, does not apply to our infant population. In our institution, the Bayley Scales evaluation has been used with much success and our examiners are quite proficient in their abilities. Moreover, the Bayley Scales evaluation continues to be the test of choice on infants, our study population. We do realize that more testing is needed on these children. This is an effort we are undertaking currently.

A second valid criticism, that our findings may be true but that our follow-up may be too short, was expressed. This criticism is valid in that a large number of infants seen with deformational plagiocephaly are lost in long-term follow-up after helmet therapy or after parents are taught to alternate the children's sleeping position and habits. A precipitous drop of 40 to 50 percent is seen in the number of children who return to the clinic for the 2-year follow-up. This could simply be a reflection of the parents reasoning that since the child's skull no longer looks abnormal, there is nothing further about which to be concerned. Unfortunately, this may not be true. As we compile long-term follow-up mental and psychomotor data on these patients, we may be able to better determine if this is accurate.

This is the main reason for the exclusion of 47 infants from our original study population of 110.

Our data show a statistical association between plagiocephaly without synostosis and mental and psychomotor delay. This is an unexpected finding with dramatic implications. At this time, we cannot find with any certainty why there is an association between deformational plagiocephaly and developmental delay. More than likely, this is due to some underlying physiology present in patients with deformational plagiocephaly. It is possible that the malformed calvaria restricts the growth of the cortex, blunting the gyri and sulci. This, then, may have a direct effect on cortical development, either directly or through dural signaling. Cortical blunting has been demonstrated using magnetic resonance imaging in patients with craniosynostosis and is a well-documented phenomenon.<sup>20</sup> It is possible that a similar cause is at work in patients with deformational plagiocephaly. What effect these cortical abnormalities may have on mental and psychomotor development is unknown. Several authors have attempted to elucidate this relationship. None has come forth with a clear answer. Kapp-Simon et al.<sup>21</sup> studied a group of infants with craniosynostosis and found that the severity of anatomic craniofacial deformity was unrelated to mental development. In 1998, Kapp-Simon et al., however, found a separate study population of craniosynostotic patients to have a higher rate of learning disabilities. They attributed these abnormal findings to sample size error. Speltz et al.<sup>22</sup> studied nonsyndromic sagittal synostotic infants and found no patients with mental development index scores in the mentally retarded or borderline range of intellectual function. This contradicts our findings. However, all of the studies used for comparison were completed on infants with craniosynostosis. This may not be the origin at hand with patients with deformational plagiocephaly. Again, the relationship between deformational plagiocephaly and developmental delay appears to be undefined.

As more data are collected, should a strong causal relationship be found to exist between deformational plagiocephaly and developmental delay, interventions need to be planned. To start with, we advocate a modification of the positioning of the infants' head position every few hours during sleep in an attempt to balance pressures placed on the skull. We have begun this effort in the intensive care units. We have tried to educate the parents to change the position of the bed or the child's toys so that the infant preferentially looks away and moves his or her head from the flattened side. We have tried placing a foam wedge or a

rolled towel underneath the flattened area in an attempt to keep the pressure off the flattened occipital region. We also advocate helmet therapy should the skull deformation be clinically significant. Helmet therapy is instituted and kept in place until 1 year of age, at which time the skull begins to harden and external compression therapy no longer has any effect. We currently are compiling the post-intervention data to see if in fact helmet therapy has a beneficial effect in reducing the developmental delays in these children.

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