Neurologic Findings in Infants With Deformational Plagiocephaly

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This study evaluates the neurologic profiles of infants with deformational plagiocephaly. Forty-nine infants with deformational plagiocephaly between the ages of 4 and 13 months (mean age, 8.1 months) are evaluated, along with 50 age-matched control subjects (mean age, 8.1 months). A modified version of the Hammersmith infant neurologic assessment was performed on each infant. A caregiver completed a questionnaire regarding the infant's prematurity, development, and health to date. Results are analyzed using *t* test. There is a statistically significant difference in overall neurologic assessment scores of infants with deformational plagiocephaly vs their healthy peers ($P = .002$). This difference is predominately in tone, whereby infants with deformational plagiocephaly have significantly more abnormal tone than nonplagiocephalic infants ($P = .003$). This abnormality is not one of decreased tone but one of variable tone, deflecting abnormally high and low tone. Infants with deformational plagiocephaly are more likely to have altered tone but not exclusively decreased tone.

Keywords: deformational plagiocephaly; neurodevelopment; plagiocephaly

In 1992, the American Academy of Pediatrics introduced the "Back to Sleep" campaign, recommending that infants should be placed supine to reduce the risk of sudden infant death syndrome.[1] Along with a decrease in deaths due to sudden infant death syndrome after the campaign's introduction, there was a subsequent corresponding increase in the number of infants presenting with deformational plagiocephaly.[2,3] The increase in cases during the past decade has been well documented[2,4,5] and has led to greater investigation into the pathogenesis and associated abnormalities of deformational plagiocephaly.

Historically, deformational plagiocephaly has been considered an abnormality associated with benign craniofacial asymmetry, of primary significance in that it alters appearance. In recent years, however, subtle neurologic abnormalities have been reported to accompany deformational plagiocephaly in infancy and later in life. School-aged

children with deformational plagiocephaly have been shown to be more likely to need special help during primary school in areas such as special education, physical therapy, occupational therapy, and speech therapy.[6] Kordestani et al[7] demonstrated that infants with deformational plagiocephaly showed significant delays in mental and psychomotor development and that none of 110 infants studied showed accelerated development. It has also been shown that mothers of infants diagnosed as having deformational plagiocephaly were more likely to report a perceived developmental delay and lower activity level of their infant than parents of nonplagiocephalic infants.[8] Davis et al[9] demonstrated that infants with deformational plagiocephaly reached motor milestones later compared with their healthy peers. This has led to much speculation surrounding the neurologic implications of deformational plagiocephaly and the role of motor tone in the development of deformational plagiocephaly. Abnormal motor tone, specifically hypotonia, has been lumped into the list of possible causes of deformational plagiocephaly, but no research to date has explored this relationship.[10] The present study evaluated the comprehensive neurologic profiles of infants with deformational plagiocephaly compared with their healthy peers, focusing on development, tone, and posture. We do not endeavor to establish causality between tone and deformational plagiocephaly, nor do we investigate treatment modalities or outcomes; rather, we characterize the neurologic profile of children with deformational plagiocephaly at the time of diagnosis.

Methods

After approval from the Washington University Human Studies Committee, 49 patients with radiographically confirmed deformational plagiocephaly, aged 4 to 13 months, were recruited during a 2-month period. Concurrently, 50 nonplagiocephalic infants (as determined by visual inspection and review of medical history) of similar age were recruited during well-child visits at a nearby pediatrician's office. When available, each child's gestational age, as recorded in their medical record, was noted.

Ages and Stages Questionnaires

The primary parent available completed the age-appropriate Ages and Stages Questionnaires. The Ages and Stages Questionnaires consist of a series of age-appropriate surveys regarding psychomotor development to be completed by parents and caregivers of children aged 4 to 48 months. The series of surveys has been shown to demonstrate excellent interobserver reliability and validity of 85%.[11] Questionnaires were available for only 4-, 6-, 8-, 10-, and 12-month intervals; infants falling between 2 age groups were moved up to the nearest grouping. Each survey contained questions designed to evaluate parental perception of infant communication and gross motor, fine motor, problem solving, and personal-social development. Questionnaires were scored according to Ages and Stages Questionnaires guidelines. A "yes" response received a score of 10 points; "sometimes," 5 points; and "not yet," 0 points. Blank responses were scored by averaging all answered questions in the section. If more than 2 questions in a given section were left unanswered, that section was not scored. Sectional scores were obtained by summing questions in each section. In addition, birth dates, corrected birth dates, and overall health were noted.

Hammersmith Infant Neurologic Assessment

Behavioral state and nonrevised optimality scoring. Each infant was evaluated by a single examiner using a modified version of the Hammersmith infant neurologic assessment for infants aged 2 to 24 months.[12] Variables (Table 1) concerning posture, tone, and behavior were assessed individually and were assigned optimality scores at 12 and 18 months of age as determined by the distribution of frequency of findings in a healthy population in the manner described by Haataja et al.[12,13] Posture and tone variables were scored out of a possible 3 points. A score of 3 corresponded to the finding observed in at least 75% of the healthy population, a score of 2 to that observed in fewer than 25% but in at least 10%, and a score of 1 or 0 to that seen in fewer than 10% of the population, which represents varying degrees of severity, with 0 being the most severe. Behavior variables (social orientation, emotional state, and consciousness) were scored using descriptive scoring rubrics with scales of 1 to 4, 1 to 5,

Table 1. Variables Assessed in 3 Categories

Posture	Head Trunk Arms Legs (supine and sitting) Feet (supine and sitting)
Tone	Scarf sign[a] Passive shoulder resistance[a] Wrist pronation or supination Adductor angle Popliteal angle[a] Ankle dorsiflexion[a] Head lag when pulled to sit Ventral suspension
Behavior	Social orientation Emotional state Consciousness

a. Right and left evaluated and scored independently.

and 1 to 6, respectively, and were included to assess possible confounding due to variations in behavioral state.

Sectional optimality scores were obtained by summing the individual scores for a given section. Global optimality scores were obtained by summing all variables for a given individual. If an individual variable failed to be scored because of infant noncooperation, it was assigned the average of the remaining variables in that section when calculating the sectional and global optimality scores.

Revised tone scoring. The original Hammersmith infant neurologic assessment optimality scoring system rates infants' tone on a scale ranging from normal (seen in > 75% of the population) to abnormal (seen in <10%). To more fully examine the character of the infants' tone, the scoring of the tone section in the Hammersmith infant neurologic assessment was prospectively revised to evaluate the infants' tone as increased, normal, or decreased. In the revised tone scoring, negative scores denote decreased tone, 0 denotes normal tone, and positive scores denote increased tone. The revised tone scoring sheet is given in Table 2. Total revised tone scores were obtained by summing the single variable scores for each patient.

Statistical Analysis

Data were analyzed by calculating and comparing means. Differences in means were tested for statistical significance using *t* test. Analysis was performed using JMP 6.0 (SAS Institute, Cary, North Carolina).

Results

The data for 49 infants with deformational plagiocephaly and 50 age-matched control subjects were analyzed

Table 2. Revised Tone Scoring Used for More In-depth Analysis of Infant Tone

Varable	–2 Low Tone	–1	–0.5	0 Normal	1	2 High Tone
Resistance						
Scarf sign	None	Minimal	–	Moderate	–	Severe
Passive shoulder	–	None	–	Overcome	–	Not overcome
Wrist pronation or supination	–	–	–	None	Overcome	Not overcome
Range						
Adductor angle	–	>170°	150°-170°	80°-150°	–	<80°
Popliteal angle	–	>170°	150°-160°	110°-150°	<90°	<80°
Ankle dorsiflexion	–	< 20°	20°-30°	30°-85°	90°	>90°
Head Lag						
When pulled to sit	Severe	Moderate	–	None	–	–
Back Curvature						
Ventral suspension	Severe	Moderate	–	None	–	–

Table 3. Ages of Cases and Controls

Group	Age, mean (SD), mo	95% Confidence Interval	Gestational Age, mean (SD), wk	95% Confidence Interval	Gestational Age, median, wk
Cases	8.1 (0.3)	7.4-8.8	38.7 (0.3)	38.2-39.3	40
Controls	8.1 (0.5)	7.2-9.0	38.6 (0.2)	38.1-39.1	39
P value	.97	–	.72	–	–

(100% of those who chose to participate). The gestational ages were collected for all case infants and for 48 of 50 control infants. (Two infants had no medical record notation of gestational age.) The mean ages and gestational ages for the 2 groups were almost identical (Table 3).

Ages and Stages Questionnaires