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# Plagiocephaly and Brachycephaly in the First Two Years of Life: A Prospective Cohort Study

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**ABSTRACT.** *Objectives.* **Although referrals for nonsynostotic plagiocephaly (NSP) have increased in recent years, the prevalence, natural history, and determinants of the condition have been unclear. The objective of this study was to assess the prevalence and natural history of NSP in normal infants in the first 2 years of life and to identify factors that may contribute to the development of NSP.**

*Methods.* **Two hundred infants were recruited at birth. At 6 weeks, 4 months, 8 months, 12 months, and 2 years, the head circumference shape was digitally photographed, and head shape was quantified using custom-written software. At each age, infants were classified as cases when the cephalic index was ≥93% and/or the oblique cranial length ratio was ≥106%. Neck rotation and a range of infant, infant care, socioeconomic, and obstetric factors were assessed.**

*Results.* **Ninety-six percent of infants were followed to 12 months, and 90.5% were followed to 2 years. Prevalence of plagiocephaly and/or brachycephaly at 6 weeks and 4, 8, 12, and 24 months was 16.0%, 19.7%, 9.2%, 6.8%, and 3.3% respectively. The mean cephalic index by 2 years was 81.6% (range: 72.0%–102.6%); the mean oblique cranial length ratio was 102.6% (range: 100.1%–109.4%). Significant univariate risk factors of NSP at 6 weeks include limited passive neck rotation at birth, preferential head orientation, supine sleep position, and head position not varied when put to sleep. At 4 months, risk factors were male gender, firstborn, limited passive neck rotation at birth, limited active head rotation at 4 months, supine sleeping at birth and 6 weeks, lower activity level, and trying unsuccessfully to vary the head position when putting the infant down to sleep.**

*Conclusions.* **There is a wide range of head shapes in infants, and prevalence of NSP increases to 4 months but diminishes as infants grow older. The majority of cases will have resolved by 2 years of age. Limited head rotation, lower activity levels, and supine sleep position seem to be important determinants.** *Pediatrics* **2004;114: 970–980;** ***plagiocephaly, brachycephaly, anthropometry, cohort studies, infant care, supine position.***

ABBREVIATIONS. NSP, nonsynostotic plagiocephaly; SIDS, sudden infant death syndrome; OCLR, oblique cranial length ratio; PDQ-II, Revised Denver II Prescreening Questionnaire; PAT, Pictorial Assessment of Temperament; OR, odds ratio; CI, confidence interval.

The rising incidence of nonsynostotic plagiocephaly (NSP) has been documented in the literature since 1992 and has been attributed to the adoption of the supine sleep position in accordance with sudden infant death syndrome (SIDS) prevention recommendations.[1] A critical review of the literature in 1998 concluded that the actual prevalence is unknown and that the condition was being recognized more frequently as a result of increased awareness.[2]

Many facets of the plagiocephalic condition are unclear.[3] However, it has been shown that NSP is more likely to occur in boys,[4–7] firstborns,[8,9] premature infants,[10] and those who sleep in the supine position.[10,11] A preferred head orientation[12,13] and limited head rotation[6] may be important determinants. Varying the head position and tummy time seem to be protective, whereas developmental delay and lower activity levels may be associated with cases.[11] Although positional preference has been followed prospectively,[13] no prospective cohort studies of normal infants that have investigated the prevalence of NSP and quantified the development of head shape in the first 2 years have been undertaken.

Strictly speaking, brachycephaly, or a high cephalic index without noticeable skewness of the head, is not true plagiocephaly, which means "oblique head." In NSP, a high cephalic index is sometimes but not always associated with asymmetry. However, as our clinical experience indicates that central occipital flattening is as concerning to parents as the skewed head shape, we have chosen to combine them. It is possible that the mechanisms are the same, the flat area merely corresponding to the preferred resting position, and any associated bossing reflecting different areas of displacement of head volume. Alternatively, it has been postulated that plagiocephaly with asymmetry may more commonly originate from neck muscle dysfunction, whereas brachycephaly results from compression.[14]

We have developed a new head shape measuring technique, HeadsUp, which involves an elastic head circumference band that is photographed digitally from above the head. The photograph is analyzed using a custom-written computer program to obtain measurements to quantify the head shape. The

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method has been used successfully in a pilot study of 60 infants and demonstrated much greater reliability and acceptability compared with measurements obtained using a flexible measuring strip.[15] We have identified cutoff points for both cephalic index and oblique cranial length ratio (OCLR; ie, the ratio of the long cross-diagonal measurement to the shorter cross-diagonal measurement). Beyond these thresholds, central occipital flattening and head shape asymmetry, respectively, are deemed to be abnormal. In practice, we believe that these cutoffs approximate the points at which head deformity becomes visually obvious, particularly where the infant has little hair. The points, 93% for cephalic index and 106% for OCLR, allow for the allocation of the cohort infants to either case or control. We aimed to determine the prevalence, natural history, and risk factors of NSP in the first 2 years of life.

## METHODS

The cohort infants were born at the delivery unit at North Shore Hospital, Auckland, a community maternity unit that deals with low-risk deliveries. On admission to the unit, the mother had the opportunity to opt out of being approached regarding research. The researcher was given a birth list each day, with the names of those who opted out deleted from this list. Selection of every fourth infant on the list yielded a cohort of 238 born between September 2001 and February 2002. Infants with congenital deformities, those who were not domiciled in the Waitemata Health District, those who were planning to move out of the region in the next year, and those who could not be seen in the first week were excluded.

Of the eligible mothers who were invited to participate, 200 (88%) were enrolled in the study (Fig 1). The initial interview was conducted either in the postnatal ward or in the mother's home within the first week after delivery. At this interview, the mother was asked about sociodemographic factors (parents' ages, ethnicity, occupation, and mother's education), obstetric factors (parity, gestation, presentation, method of delivery, length of labor, and multiple birth), and infant details such as date of birth, gender, Apgar scores, and birth measurements. The mother's highest educational level was classified into 1) no qualifications or 1 or more School Certificate (year 11) subjects, 2) sixth form or Bursary qualification (years 12–13), and 3) tertiary education or professional certification. The parents' occupations were rated in accordance with the New Zealand Socio-economic Index of Occupational Status[16] classifications into high, medium, and low socioeconomic status, the highest rating of either parent being used for the classification.

The interviewer assessed the infant's head shape for anything unusual, such as a caput or area of flattening. When possible, when the infant was in a relaxed state, passive head rotation was assessed. This was accomplished by standing behind the supine-lying infant, holding the head between the hands, and gently rotating the head from side to side. Any restriction or tightness in 1 or both directions was noted. The head circumference was measured around the maximum fronto-occipital circumference.

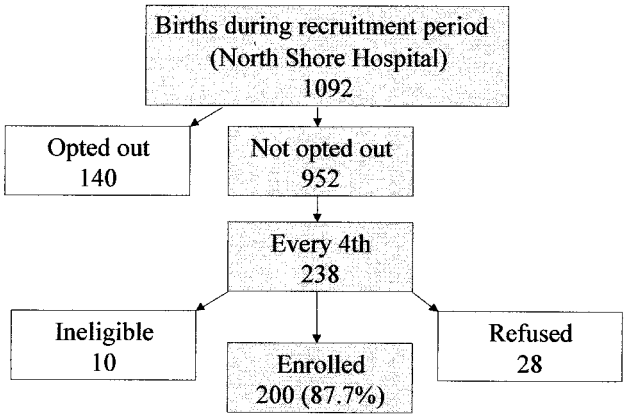


**Fig 1.** Enrollment of subjects.

At 6 weeks, 4 months, 8 months, 12 months, and 24 months, the mother and the infant were visited at home, and a set of digital photographs of the infant's head using the HeadsUp band were taken to document the head shape, as follows. While seated on the mother's knee (or on the floor for older infants), the infant was given 1 or 2 toys to play with if necessary while a close-fitting, nylon stocking–type cap was placed on the head to flatten the hair. The infant's identification was written on a yellow sticker and attached to the stocking cap. A small yellow cape was placed over the shoulders to mask any competing colors in the clothing or surroundings. A soft, elastic, blue headband made of a narrow strip of 7-mm-thick covered neoprene was then placed over the head circumference. On the headband are sliding green ear markers and a red marker to indicate the middle of the nose. The red marker is also a known length (50 mm), which is used to determine scale. After positioning the band and the markers, digital photographs were taken from ~800 mm above the vertex of the head, using a Sony DSC-S50 digital still camera with pivoting LCD viewing screen. Several photographs were taken, and the 3 best were kept for analysis by the HeadsUp computer program. The mean measurements taken from the 3 photographs were used in the final analysis. We did not use the HeadsUp photographic measure on the newborns because of possible birth molding of the head.

The main measure of asymmetry from one side of the head to the other was the OCLR. These lines were taken from points located 40 degrees either side of the posterior midline, obliquely across the head to derived frontozygomatic points in the frontal area of the head circumference. The other important measure used was the cephalic index, a measure of central posterior flattening or brachycephaly, calculated from (head breadth/head length) × 100. Criteria to allocate case definition were made previously using analyses of mean cephalic indices and OCLR measurements obtained from a pilot photograph study (unpublished data). The criteria so defined require that the cephalic index be 93% or above and/or OCLR be 106% or above. Other measurements assessed were head circumference, head area bounded by the blue band, angles of each ear relative to the nose position, and transcranial difference in millimeters.

At 6 weeks, the infant's passive head rotation was assessed as for the newborns. At 4, 8, 12, and 24 months, active head rotation was checked by seating the infant facing outward on the mother's knee, then holding a bright musical toy in front of the infant. The toy was brought around to each side to encourage the infant to follow the toy with the eyes until they were looking across the shoulder, without moving the body around with the head. Any limitation or difficulty in head rotation was recorded.

At each of the follow-up interviews, the mother was asked about such factors as weight, length, and head circumference measurements as recorded in the child's Well Child Record Book (a parent-held community child health nursing record), health problems, hair loss, and the presence of preferential head turning or neck dysfunction. In addition, she was asked about infant care practices such as breastfeeding; pacifier use; sleep position; head varying; total daily duration of tummy time and upright time; bed type; mattress type; underbedding; pillow use; preferred maternal holding positions; and amount of time spent in supine in cots, car seats, bouncers, and other places per day on average.

At the 6-week interview, the mother was given the Revised Denver II Prescreening Questionnaire (PDQ-II)[17] and was asked to go through the items until 3 "no" responses were recorded. At each subsequent interview, she was asked to reassess the previous "no" responses and then to continue the items until 3 "no" responses were recorded again. The 0- to 9-month and 9- to 24-month forms were used. The number of delays and/or cautions for the child's age was recorded. Children with no delays and 1 or no cautions were rated normal. If a child had 1 delay or 2 cautions, then the rating was slightly abnormal, and 2 or more delays or 3 or more cautions were rated abnormal.

Temperament assessment using the Pictorial Assessment of Temperament (PAT)[18] was conducted at 4 months. This consists of a 10-item measure of temperament based on Carey's Revised Infant Temperament Questionnaire and consists of vignettes de-

picting 3 different types of response to 10 different situations, such as getting dressed, waking up, loud noises, etc. The response types are "easy," "average or slow-to-warm-up," and "difficult." A score of 1 is given for "easy" responses, 2 for "average or slow-to-warm-up" responses, and 3 for "difficult" responses, giving a total possible score range of between 10 and 30.

Temperament was also assessed at each age by showing the mother a plastic gauge with a slider on it. At 1 end is a happy face and the words "very settled/easy-going," and at the other end is an unhappy face and the words "very unsettled/difficult." The mother was asked to position the slider to indicate the child's overall temperament at that age. The other side of the gauge is marked with a scale from 0 to 10, and the researcher thus was able to assign a score for the mother's assessment. Activity level at each age was similarly assessed by the mother, with the gauge scoring between "very inactive" (score = 0) and "very active" (score = 10).

The interviewer assessed mattress softness using a subjective rating of soft, medium, or firm. A weight (10.5 g/cm$^2$) on a 6-mm-diameter aluminum plunger gauged in 1-mm increments, which was dropped onto the mattress through a hole in a small circle of wood, also gave an objective assessment of the mattress firmness. If the mattress was extremely hard, then the weight dropped very little, giving a reading close to 0 mm, whereas very soft mattresses gave a higher reading of up to 30 mm. When the mattress was not available, the mother was asked to rate it as soft, medium, or firm.

Baseline data were collected at the first interview. The data were analyzed as a standard case-control analysis using univariate and multivariate logistic regression, using SAS (Release 8.2; SAS Institute, Cary, NC).

All interviews were conducted by the principal investigator, with the exception of some 6-week and 4-month interviews that were done by 1 other trained interviewer. No advice was offered to parents of children who developed a misshapen head shape; however, if at any time a mother became concerned about head shape problems developing in her infant, she was advised to talk to her family doctor or Plunket nurse (community child health nurse) for advice. The Auckland Ethics Committee approved the study.

**TABLE 1.** Cohort Description ($n$ = 200)

| Variable | $n$ (%) |
|---|---|
| Gender | |
| Male | 106 (53.0) |
| Female | 94 (47.0) |
| Parity | |
| Firstborn | 90 (45.0) |
| Later born | 110 (55.0) |
| Multiple birth | |
| Singletons | 199 (99.5) |
| Twin | 1 (0.5) |
| Gestation | |
| <37 wk | 4 (2.0) |
| ≥37 wk | 196 (98.0) |
| Delivery | |
| Normal vaginal | 133 (66.5) |
| Cesarean | 41 (20.5) |
| Assisted vaginal | 26 (13.0) |
| 5-Min Apgar score | |
| <7 | 1 (0.5) |
| ≥7 | 199 (99.5) |
| Maternal age | |
| <25 | 20 (10.0) |
| 25–29 | 38 (19.0) |
| 30–34 | 87 (43.5) |
| 35+ | 55 (27.5) |
| Mother's highest qualification | |
| None or school certificate | 44 (22.3) |
| Sixth form/bursary | 45 (22.9) |
| Tertiary/professional | 108 (54.8) |
| Socioeconomic status | |
| Low | 22 (11.0) |
| Medium | 111 (55.5) |
| High | 67 (33.5) |

## RESULTS

### Cohort Characteristics

The infants who were enrolled in this study were healthy infants who were mostly full term. Ninety-one percent of the initial interviews were completed in the first 36 hours. Of the 200 infants who were enrolled in the study, 100% were seen at 6 weeks, 198 (99%) were seen at 4 months, 196 (98%) were seen at 8 months, 192 (96%) were seen at 12 months, and 181 (90.5%) were followed to 2 years. The characteristics of the cohort are listed in Table 1.

### Head Measurements

Newborn head shape was normal on visual assessment for 63.0% of the cohort. Cone-shaped heads were seen in 13.5% of the cohort; prominent or indented sutures in 12%; and caputs, flat areas, or other "bumps or dents" in 16%.

At the follow-up interviews, there was a wide range for cephalic index and OCLR (Table 2). The widest range for both occurred at 6 weeks, but whereas the maximum OCLR reduced thereafter, the maximum cephalic index was recorded at 12 months. There was also a wide range of ear angle positions seen particularly at the 6-week period, although no difference was detected between cases and control subjects at each age for ear angles or head circumference. Head circumference as measured by tape measure versus that measured by HeadsUp showed a high correlation between the 2 types of measure ($r$ = 0.98) over all ages. At follow-up and using the HeadsUp measure, boys' head circumferences were significantly larger than girls', being ~1 cm larger at each age. There was no difference detected between genders for cephalic index, OCLR, or ear angles.

The mean difference between the transcranial diameters in the plagiocephalic infants was 9.8 mm (SD: 2.0), 11.3 mm (SD: 0.6), 10.3 mm (SD: 0.9), 11.1 mm (SD: 1.2), and 12.0 mm (SD: 2.4), at 6 weeks and 4, 8, 12, and 24 months respectively.

### Prevalence and Natural History

After each follow-up interview, any infant who had a cephalic index of ≥93% and/or an OCLR of ≥106% was classified as a case (see Fig 2). In Fig 2, cases that were defined as normal for the purposes of this study are in the lower left-hand quadrant, brachycephalic cases are in the top left-hand quadrant, plagiocephalic cases are in the lower right-hand quadrant, and those with both are in the top right-hand quadrant.

Using the cutoff points of 93% for cephalic index and 106% for OCLR, the numbers of cases and noncases at each age are illustrated in Figs 3 and 4. Although half of the 6-week cases had resolved by 4 months, 23 new cases had occurred at 4 months. Thereafter, few new cases occurred and the total number of cases started diminishing so that by 12 months, there were only one third as many cases as at 4 months. By the age of 2 years, only 6 (3.3%) children recorded values outside the set parameters: 3 were brachycephalic and 3 were plagiocephalic.

**TABLE 2.** Head Measurements (All Infants)

| Variable | 6 Weeks ($n$ = 200) | | | 4 Months ($n$ = 198) | | | 8 Months ($n$ = 196) | | | 12 Months ($n$ = 192) | | | 2 Years ($n$ = 181) | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | Mean (SD) | Min | Max | Mean (SD) | Min | Max | Mean (SD) | Min | Max | Mean (SD) | Min | Max | Mean (SD) | Min | Max |
| Cephalic index | 83.5 (5.7) | 72.1 | 103.8 | 84.9 (6.2) | 72.5 | 101.3 | 83.8 (5.7) | 73.5 | 103.2 | 82.9 (5.4) | 72.7 | 104.1 | 81.6 (4.8) | 72.0 | 102.6 |
| OCLR | 103.2 (2.5) | 100.2 | 112.2 | 102.9 (2.1) | 100.2 | 109.5 | 102.5 (1.7) | 100.1 | 108.1 | 102.5 (1.7) | 100.2 | 108.8 | 102.6 (1.7) | 100.1 | 109.4 |
| HC | 38.2 (1.3) | 34.1 | 41.6 | 41.7 (1.2) | 38.9 | 44.8 | 44.8 (1.4) | 41.4 | 49.0 | 46.5 (1.4) | 42.8 | 49.6 | 48.6 (1.3) | 45.3 | 51.8 |
| L ear angle | 90.2 (4.6) | 75.7 | 99.5 | 90.6 (3.1) | 82.3 | 98.3 | 89.5 (3.5) | 80.7 | 102.0 | 89.3 (3.1) | 80.0 | 97.3 | 88.1 (3.3) | 80.3 | 97.0 |
| R ear angle | 89.9 (4.6) | 80.5 | 104.7 | 89.5 (3.1) | 81.7 | 97.7 | 90.5 (3.5) | 78.0 | 99.3 | 90.7 (3.1) | 83.0 | 100.0 | 91.9 (3.3) | 83.0 | 99.7 |

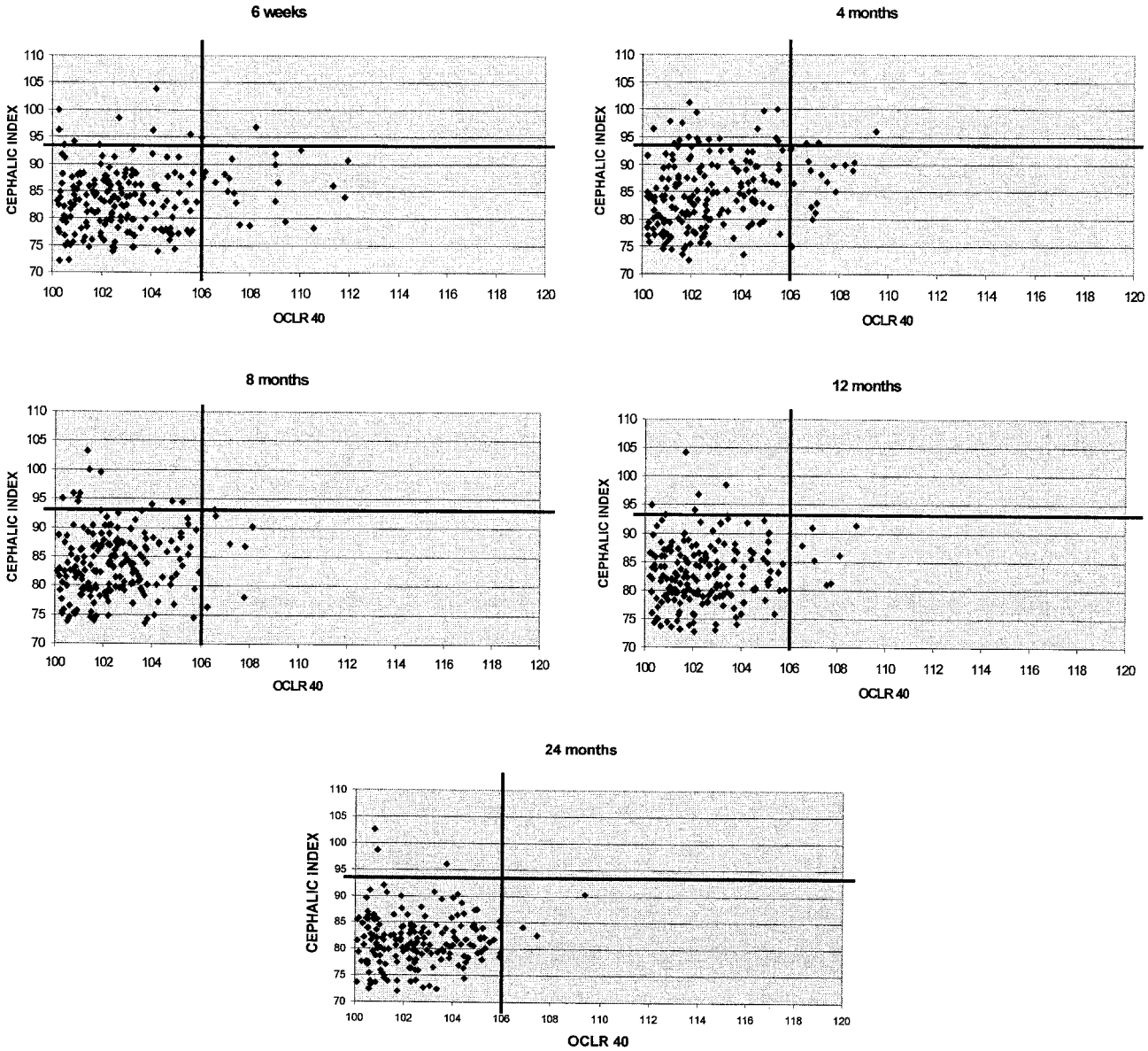


**Fig 2.** Mean cephalic index and OCLR scores for all infants at each age (controls [normal] are in the lower left quadrants of each scatter plot).

One of the 12-month cases was unable to be followed to 2 years.

The overall prevalence rates for the cohort were 16% at 6 weeks, 19.7% at 4 months, 9.2% at 8 months, 6.8% at 12 months, and 3.3% at 24 months. More than twice as many infants were classified as having plagiocephaly alone (OCLR ≥106%) than having brachycephaly alone (cephalic index ≥93%) at 6 weeks, but at both 4 and 8 months, more infants were classified as brachycephalic than plagiocephalic. This

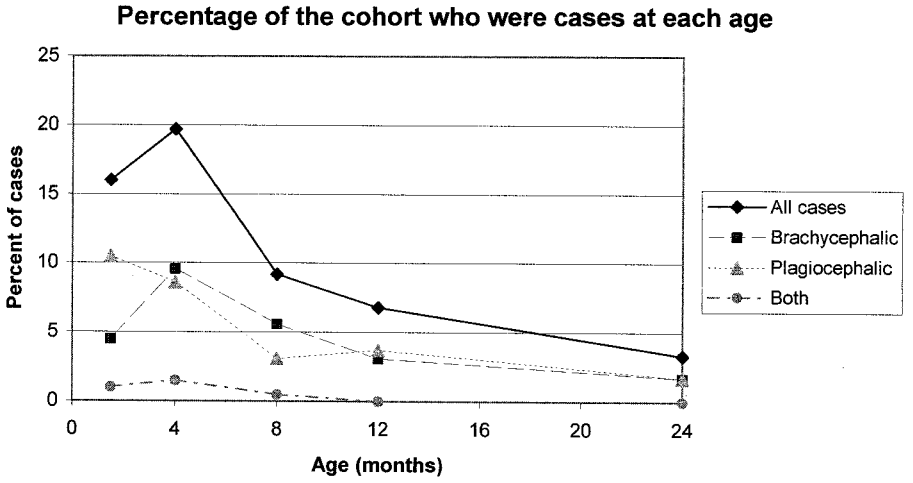


**Fig 3.** Percentage of the cohort who were cases at each age.

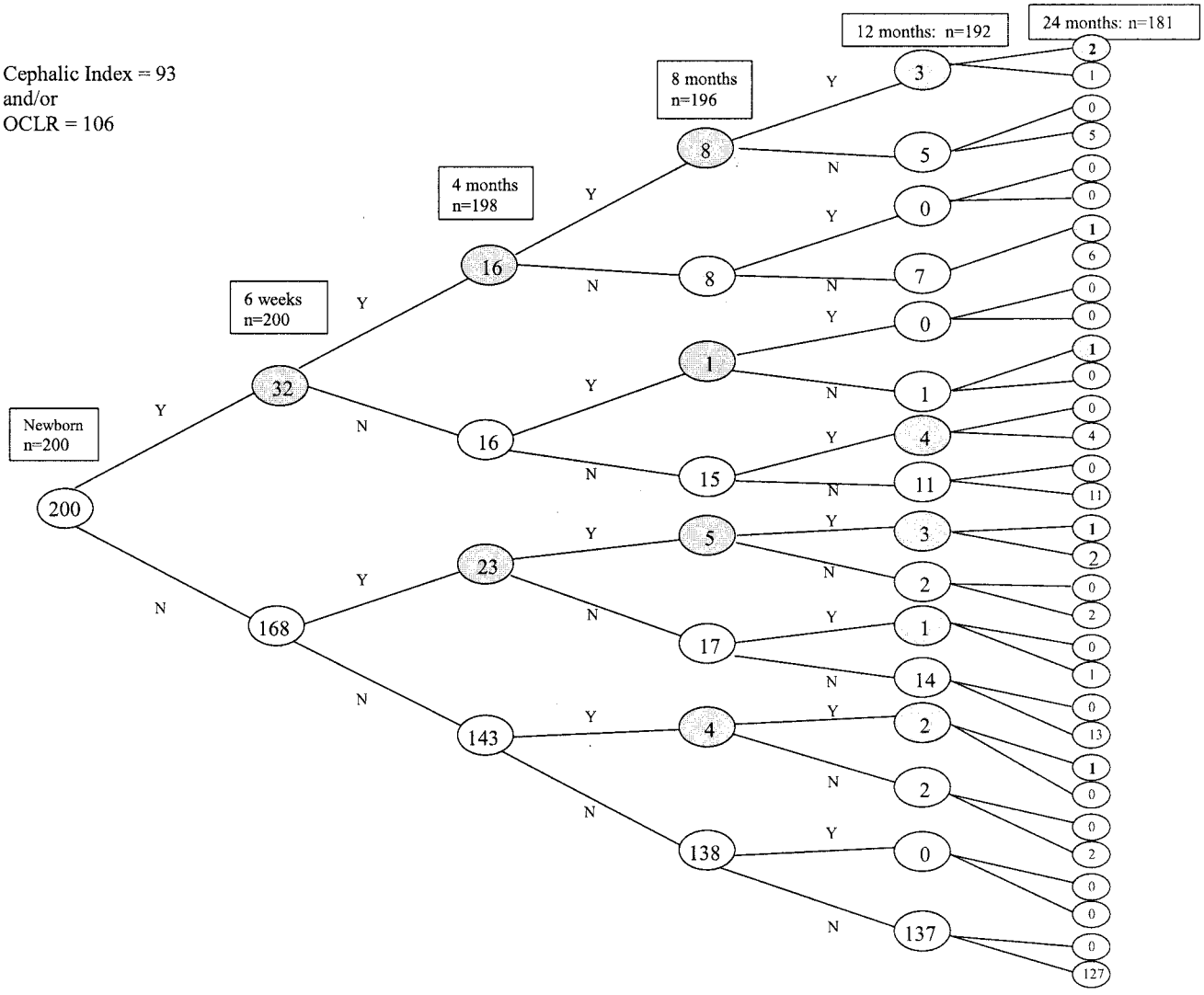


**Fig 4.** Development of plagiocephaly or brachycephaly in the cohort in the first 2 years. (The numbers in shaded areas represent cases. Slight discrepancies in numbers are attributed to small numbers lost to follow-up at 4, 8, 12, and 24 months.)

difference had disappeared by 12 months, with rates for both dropping considerably by that age. In the early months, a few infants were classified as having both plagiocephaly and brachycephaly (Table 3).

Overall, 29.5% of the cohort infants developed either plagiocephaly or brachycephaly or both at some stage during the study observation period (Table 3); however, by the age of 2 years, only 3.3% were still above the classification threshold for abnormality (Table 3). Most cases manifested at 6 weeks or 4

**TABLE 3.** New Cases at Each Age

| Condition | 6 Weeks (*n* [%]) | 4 Months (*n* [%]) | 8 Months (*n* [%]) | 12 Months (*n* [%]) | 2 Years (*n* [%]) | Total (% of original cohort) |
|---|---|---|---|---|---|---|
| Either | 32 (16.0) | 23 (11.6) | 4 (2.0) | 0 (0.0) | 0 (0.0) | 59 (29.5) |
| Plagiocephaly only | 21 (10.5) | 12 (6.1) | 3 (1.5) | 0 (0.0) | 0 (0.0) | 36 (18) |
| Brachycephaly only | 9 (4.5) | 11 (5.6) | 1 (0.5) | 0 (0.0) | 0 (0.0) | 21 (10.5) |
| Both | 2 (1.0) | 0 (0.0) | 0 (0.0) | 0 (0.0) | 0 (0.0) | 2 (1.0) |

months; only 4 new cases had developed at 8 months, and thereafter no infant developed deformation. If the cutoff points were different, then the prevalence of cases at each age would be found to be different. Table 4 shows how prevalence would change if the limits were set at lower or higher levels.

Fifteen case mothers consulted their Plunket nurse or general practitioner regarding their infants' head deformity. One case was also seen by a physiotherapist. Treatment consisted of positioning advice in 9 cases; reassurance only was given in the other 6 cases. Although helmet treatment is available in New Zealand, no case was considered severe enough to be referred for such treatment by their health professional.

Of the 6 2-year cases, there were 3 boys and 3 girls, and 5 were firstborns. All slept supine in the early months. Two of the 3 brachycephalic cases were above the cephalic index cutoff at all ages; the most severe case, a girl born at 36 weeks' gestation, was just below 93% at 6 weeks and thereafter was consistently over 100%. Although her parents had been concerned at 4, 8, and 12 months, by 2 years, they were not particularly worried. Head rotation tests were normal at all ages. In the plagiocephalic cases, 1 case progressively worsened to 8 months, and the OCLR thereafter remained about the same, at ~109%. She showed evidence of head tilt and limited rotation at 4 and 8 months. The mother remarked at 2 years that she still leaned to 1 side when she was tired. This mother was also unconcerned at 2 years. The other 2 plagiocephalic cases' OCLRs tended to vacillate around the cutoffs. One had mild unilateral flattening of the forehead at age 2 years but otherwise looked almost normal. No parent of a 2-year case was concerned about the head shape; 1 control parent expressed mild concern about her son, whose head had improved since 8 months but was still slightly flat in a small area above the band.

**Risk Factors: 6 Weeks**

Owing to the small number of cases at 8, 12, and 24 months, analysis of risk factors was restricted to 6 weeks and 4 months. Six-week cases were significantly more likely to have had a limitation of passive rotation at the newborn interview (crude odds ratio [OR]: 6.17; 95% confidence interval [CI]: 2.03–18.76; Table 5), although no difference was detected in the passive head rotation test performed at 6 weeks. More case mothers reported a preferential head orientation at 6 weeks, although this was of borderline significance.

Ninety-four percent of 6-week cases had been positioned for supine sleep at the newborn interview, compared with 56.5% of control subjects (crude OR: 11.53; 95% CI: 2.67–49.81). By 6 weeks, 81.3% of cases were still sleeping supine, compared with 48.2% of control subjects (crude OR: 4.65; 95% CI: 1.82–11.89). The mothers of cases were less likely to be varying the head position when putting the infant down to sleep (crude OR: 2.75; 95% CI: 1.11–6.82), or they were trying to vary it but were not able to owing to the infant's turning to his or her own preferred position (crude OR: 5.15; 95% CI: 1.90–13.98).

When the total amount of back time was added up, the cases were spending a mean of 19.0 (SD: 4.1) hours a day on their back, compared with control subjects at 14.9 (SD: 6.5) hours ($P < .0001$). Half of the case infants were spending >21 hours a day supine, compared with 23.8% of control infants (crude OR: 3.20; 95% CI: 1.47–6.97), although cases were more likely than control subjects to be spending >1 hour a day upright (crude OR: 2.50; 95% CI: 1.08–5.56). Case infants spent more time than control infants lying in bouncy seats or rockers, although this reached only marginal significance ($P = .08$). Two (6.3%) cases and 3 (1.8%) control subjects had 2 or more developmental delays or 3 or more cautions on the PDQ-II.

**TABLE 4.** Prevalence of Plagiocephaly Using Different Cutoff Criteria

| Cutoff | 6-Week Cases (*n* [%]) | 4-Month Cases (*n* [%]) | 8-Month Cases (*n* [%]) | 12-Month Cases (*n* [%]) | 2-Year Cases (*n* [%]) |
|---|---|---|---|---|---|
| Cephalic index ≥91% and/or OCLR40 ≥105% | 51 (25.5) | 56 (28.0) | 37 (18.9) | 36 (18.7) | 23 (12.7) |
| Cephalic index ≥92% and/or OCLR40 ≥105.5% | 39 (19.5) | 49 (24.7) | 26 (13.3) | 20 (10.4) | 12 (6.6) |
| Cephalic index ≥93% and/or OCLR40 ≥106% | 32 (16.0) | 39 (19.7) | 18 (9.2) | 13 (6.8) | 6 (3.3) |
| Cephalic index ≥94% and/or OCLR40 ≥106% | 30 (15.0) | 36 (18.2) | 18 (9.2) | 12 (6.3) | 6 (3.3) |
| Cephalic index ≥95% and/or OCLR40 ≥107% | 24 (12.0) | 19 (9.6) | 10 (5.1) | 8 (4.2) | 5 (2.8) |

**TABLE 5.** Significant Risk Factors at 6 Weeks

| Variable | Case ($n$ = 32; $n$ [%]) | Control ($n$ = 168; $n$ [%]) | Univariate OR (95% CI, $P$ Value) | Multivariate OR (95% CI, $P$ Value) |
|---|---|---|---|---|
| Limitation of passive rotation - newborn (missing = 16) | | | $\chi^2$ = 12.52, $P$ = .0004 | $\chi^2$ = 11.50, $P$ = .0007 |
| Limited | 7 (25.0) | 8 (5.1) | 6.17 (2.03–18.76) | 9.51 (2.59–34.94) |
| Not limited | 21 (75.0) | 148 (94.9) | 1.00 | 1.00 |
| Reported preferential head orientation (missing = 2) | | | $\chi^2$ = 3.19, $P$ = .07 | |
| Yes | 23 (71.9) | 91 (54.8) | 2.11 (0.92–4.83) | |
| No | 9 (28.1) | 75 (45.2) | 1.00 | |
| Sleep position at newborn interview | | | $P$ < .0001 | |
| Supine only | 30 (93.8) | 95 (56.5) | 11.53 (2.67–49.81) | |
| Nonsupine | 2 (6.2) | 73 (43.5) | 1.00 | |
| Sleep position at 6 wk | | | $\chi^2$ = 11.79, $P$ = .0006 | $\chi^2$ = 9.25, $P$ = .003 |
| Supine only | 26 (81.3) | 81 (48.2) | 4.65 (1.82–11.89) | 5.27 (1.81–15.39) |
| Nonsupine | 6 (18.7) | 87 (51.8) | 1.00 | 1.00 |
| Head position varied | | | $\chi^2$ = 10.94, $P$ = .004 | |
| Yes | 10 (31.2) | 103 (61.3) | 1.00 | |
| No | 12 (37.5) | 45 (26.8) | 2.75 (1.11–6.82) | |
| Tried but unsuccessful | 10 (31.2) | 20 (11.9) | 5.15 (1.90–13.98) | |
| Back time per day | | | $\chi^2$ = 8.57, $P$ = .003 | |
| < 21 h | 16 (50.0) | 128 (76.2) | 1.00 | |
| ≥ 21 h | 16 (50.0) | 40 (23.8) | 3.20 (1.47–6.97) | |
| Upright time per day | | | $\chi^2$ = 4.90, $P$ = .03 | $\chi^2$ = 6.88, $P$ = .009 |
| ≤ 1 h | 9 (28.1) | 83 (49.4) | 1.00 | 1.00 |
| > 1 h | 23 (71.9) | 85 (50.6) | 2.50 (1.08–5.56) | 3.99 (1.42–11.23) |
| Backtime per day (h) | 19.01 (4.13) | 14.89 (6.52) | $P$ < .0001 $\beta$ = 0.14 (0.05–0.22) | |

Factors that were significant at the univariate level all were entered into an initial multivariate model, with the exception of newborn sleep position, which had low numbers in 1 category. The multivariate model was reduced by removing variables that were nonsignificant 1 at a time to ensure they did not affect risk estimates of the other variables. The variables that remained significant in the final multivariate model were newborn passive head rotation (adjusted OR: 9.51; 95% CI: 2.59–34.94), 6-week sleep position (adjusted OR: 5.27; 95% CI: 1.81–15.39), and upright time (adjusted OR: 3.99; 95% CI: 1.42–11.23).

No significant differences were detected between cases and control subjects at 6 weeks for obstetric factors, socioeconomic factors, gender, newborn head circumference, abnormal head shape at the newborn assessment, the presence of hair loss on the back of the head, snoring, activity level, newborn or 6-week weight, length and head circumference, temperament rating, activity level, the amount of reported tummy time per day, breastfeeding, dummy use, the use of positioning aids or pillows, mattress firmness, maternal hand dominance, and the mother's preferred holding position.

### Risk Factors: 4 Months

At 4 months, male gender was of borderline significance (crude OR: 2.03; 95% CI: 0.97–4.22), as was being firstborn (crude OR: 1.82; 95% CI: 0.90–3.70). The 4-month cases were still significantly more likely to have had a limitation of passive head rotation at the newborn interview (crude OR: 7.78; 95% CI: 2.56–23.70), and they were also more likely to have a limitation of active head rotation at the 4-month interview (crude OR: 2.68; 95% CI: 1.23–5.81; Table 6).

In the 4-month cases, having slept supine at 6 weeks was significant (crude OR: 2.33; 95% CI: 1.11–4.93), but sleeping supine at 4 months was not. Similarly, having tried unsuccessfully to vary the head position when putting the infant to sleep at 6 weeks was significant (crude OR: 4.23; 95% CI: 1.74–10.28); however, unsuccessfully varying the head position at 4 months was only marginally significant (crude OR: 2.68; 95% CI: 0.97–7.43). More cases than control subjects had >21 hours of back time at 6 weeks (crude OR: 2.43; 95% CI: 1.47–5.20), but total back time at 4 months was not significantly different. Using a pillow reached marginal significance (crude OR: 2.64; 95% CI: 0.97–7.24).

Although the numbers were small, the 4-month cases were more likely to have had an abnormal result on the 6-week PDQ-II test (crude OR: 18.06; 95% CI: 1.96–166.54). The mothers of 4-month cases were also more likely to report snoring in their infants (crude OR: 5.60; 95% CI: 1.61–19.45) and to report that their infants had low activity levels (crude OR: 3.28; 95% CI: 1.40–7.75). Infants who were cases at 4 months sat alone at a slightly older age than those who were control subjects at 4 months (6.7 months [SD: 0.98] vs 6.4 months [SD: 0.97]; $P$ = .05).

One third of infants scored 15 or less on the PAT test administered at this age, and we categorized these infants as "easy." Case infants were more likely to score in the average to difficult range (≥16) for infant temperament than in the easy range (crude OR: 2.63; 95% CI: 1.09–6.32). However, there was no difference between cases and control subjects for the temperament gauge score assessment by the mothers.

Multivariate analysis of the factors that were significant at the univariate level led to a final multi-

**TABLE 6.** Significant Risk Factors at 4 Months

| Variable | Case ($n$ = 39; $n$ [%]) | Control ($n$ = 161; $n$ [%]) | Univariate OR (95% CI) | Multivariate OR (95% CI) |
|---|---|---|---|---|
| Gender (missing = 2) | | | $\chi^2$ = 3.54, $P$ = .06 | |
| Male | 26 (66.7) | 79 (49.7) | 2.03 (0.97–4.22) | |
| Female | 13 (33.3) | 80 (50.3) | 1.00 | |
| Parity (missing = 2) | | | $\chi^2$ = 2.77, $P$ = .09 | |
| Firstborn | 22 (56.4) | 66 (41.5) | 1.82 (0.90–3.70) | |
| Later born | 17 (43.6) | 93 (58.5) | 1.00 | |
| Limitation of passive rotation - newborn (missing = 16) | | | $\chi^2$ = 13.07, $P$ = .0003 | $\chi^2$ = 8.49, $P$ = .004 |
| Limited | 9 (25.0) | 6 (4.1) | 7.78 (2.56–23.70) | 6.51 (1.85–22.98) |
| Not limited | 27 (75.0) | 140 (95.9) | 1.00 | 1.00 |
| Active rotation at 4 months (missing = 5) | | | $\chi^2$ = 6.20, $P$ = .02 | $\chi^2$ = 5.50, $P$ = .02 |
| Limitation | 14 (35.9) | 27 (17.3) | 2.68 (1.23–5.81) | 3.11 (1.21–8.05) |
| No limitation | 25 (64.1) | 129 (82.7) | 1.00 | 1.00 |
| Sleep position at newborn interview (missing = 2) | | | $\chi^2$ = 3.02, $P$ = .08 | |
| Supine only | 29 (74.4) | 94 (59.1) | 2.01 (0.91–4.40) | |
| Nonsupine | 10 (25.6) | 65 (40.9) | 1.00 | |
| Sleep position at 6 wk (missing = 2) | | | $\chi^2$ = 5.11, $P$ = .02 | |
| Supine only | 27 (69.2) | 78 (49.1) | 2.33 (1.11–4.93) | |
| Nonsupine | 12 (30.8) | 81 (50.9) | 1.00 | |
| Head position varied at 6 wk (missing = 2) | | | $\chi^2$ = 11.28, $P$ = .004 | $\chi^2$ = 9.20, $P$ = .01 |
| Yes | 17 (43.6) | 94 (59.1) | 1.00 | 1.00 |
| No | 9 (23.1) | 48 (30.2) | 1.04 (0.43–2.50) | 1.04 (0.36–3.00) |
| Tried but unsuccessful | 13 (33.3) | 17 (10.7) | 4.23 (1.74–10.28) | 4.28 (1.58–11.59) |
| Head position varied at 4 mo (missing = 2) | | | $\chi^2$ = 4.97, $P$ = .08 | |
| Yes | 9 (23.1) | 46 (28.9) | 1.00 | |
| No | 19 (48.7) | 92 (57.9) | 1.06 (0.44–2.52) | |
| Tried but unsuccessful | 11 (28.2) | 21 (13.2) | 2.68 (0.97–7.43) | |
| Back time per day at 6 wk (missing = 2) | | | $\chi^2$ = 6.05, $P$ = .01 | |
| < 21 hours | 22 (56.4) | 121 (76.1) | 1.00 | |
| ≥ 21 hours | 17 (43.6) | 38 (23.9) | 2.43 (1.47–5.20) | |
| Pillow used (missing = 3) | | | $\chi^2$ = 3.57, p = .06 | |
| Yes | 7 (18.0) | 12 (7.6) | 2.64 (0.97–7.24) | |
| No | 32 (82.0) | 145 (92.4) | 1.00 | |
| PDQ-II delays at 6w (missing = 2) | | | $P$ = .005 (Fisher) | |
| Normal / slightly abnormal | 35 (89.7) | 158 (99.4) | 1.00 | |
| Abnormal: >1 delay or > 2 cautions | 4 (10.3) | 1 (0.6) | 18.06 (1.96–166.54) | |
| Snoring (missing = 2) | | | $\chi^2$ = 7.36, $P$ = .007 | |
| Yes | 6 (15.4) | 5 (3.1) | 5.60 (1.61–19.45) | |
| None/minimal | 33 (84.6) | 154 (96.9) | 1.00 | |
| Activity level at 4m categorised (missing = 2) | | | $\chi^2$ = 7.34, $P$ = .007 | $\chi^2$ = 4.98, $P$ = .03 |
| Low: < 6.5 | 11 (28.2) | 17 (10.7) | 3.28 (1.40–7.75) | 3.28 (1.16–9.29) |
| High: > = 6.5 | 28 (71.8) | 142 (89.3) | 1.00 | 1.00 |
| PAT (temperament test) categorized (missing = 2) | | | $\chi^2$ = 4.63, $P$ = .03 | $\chi^2$ = 5.09, $P$ = .02 |
| Easy: <16 | 7 (17.9) | 58 (36.5) | 1.00 | 1.00 |
| Average-difficult: ≥16 | 32 (82.1) | 101 (63.5) | 2.63 (1.09–6.32) | 3.30 (1.17–9.29) |

variate model in which the following variables were found to be significant: limited passive head rotation at birth (adjusted OR: 6.51; 95% CI: 1.85–22.98), limited active head rotation at 4 months (adjusted OR: 3.11; 95% CI: 1.21–8.05), tried but unable to vary head position at 6 weeks (adjusted OR: 4.28; 95% CI: 1.58–11.59), low activity level at 4 months (adjusted OR: 3.28; 95% CI: 1.16–9.29), and average to difficult rating on PAT test (adjusted OR: 3.30; 95% CI: 1.17–9.29; Table 6).

No significant differences were found in the 4-month cases for obstetric factors; socioeconomic factors; newborn head circumference; abnormal head shape at newborn assessment; weight, length, and head circumference measured at the 3-month well-child check; preferential head orientation; developmental delays either reported or on the PDQ-II; number of cautions or delays on the PDQ-II; sleep position at 4 months; hair loss, tummy time, and back time at 4 months; upright time; bouncinette time; breastfeeding; preferred side of feeding; dummy use; the use of positioning aids; mattress firmness; mother's handedness; and preferred holding position.

## DISCUSSION

Our cohort revealed a wide range of head shape, with cephalic index in the cohort ranging from 72 to 104 and OCLR ranging from 100 to 112 in the first 2 years of life. OCLR range was wide in the early period and reduced over time, whereas cephalic index took longer to develop and subsequently to reduce. The wide range of both cephalic index and OCLR at 6 weeks probably reflects the extreme malleability of the infant cranium in the newborn period. However, it is a matter of conjecture how much of this is attributable to forces acting on the cranium and how much is attributable to genetic tendencies.

The infants in our cohort had wider heads than recorded in an earlier report of normal children. Our mean cephalic indices at 6 weeks and 4, 8, 12, and 24 months were 83.5, 84.9, 83.8, 82.9, and 81.6, respectively. In comparison, mean cephalic indices published in 1977 for 1 month, 4 months, 9 months, 12 months, and 2 years, based on sample sizes of ~40 American infants, were 79.5, 78.2, 78.6, 76.7, and 76.9, respectively.[19] These much lower indices were recorded at a time when prone and side sleeping were frequently the norm. A group of Taiwanese cleft-lip infants were shown to have a cephalic index of 93.0% at 3 months when they slept supine, compared with 82.6 when they slept prone.[20] Ethnic differences may explain some of the variation as there is some evidence of anatomic differences relating to racial origins.[21]

The true prevalence of NSP has been unclear, largely because of disparities in diagnostic criteria and subjective classifications.[2] Quoted prevalence varies between 0.3%, a figure derived from congenital muscular torticollis rates in 1974,[22] to 48% of under-1-year-olds in 1971, based on frontal measurements.[23] Other early studies quoted 5%[24] and 28%,[25] but there were no definitions of plagiocephaly stated. A more recent study of positional preference estimated a plagiocephaly prevalence of 9.9% in all children under the age of 6 months, based on nonquantified visual assessment of asymmetry.[13] Diagnostic disagreements relating to lambdoid synostosis and NSP have also led to confusion in the literature. There is agreement, however, that the prevalence of NSP has increased and is linked with the adoption of the supine sleep position for prevention of SIDS.[1]

However, as we have shown, age affects prevalence rates. Using cutoff points of 93% for cephalic index and 106% for OCLR, the point prevalence of head shape deformity in our cohort was 16% at 6 weeks, 19.7% at 4 months, 9.2% at 8 months, 6.8% at 12 months, and 3.3% at 2 years. Nearly 30% of our cohort exceeded the cutoff levels during the first year, this being the period prevalence for the first year of life in the cohort. It is notable that few new cases appeared at 8 months and no new cases thereafter. Prevalence of either plagiocephaly or brachycephaly or both increased between 6 weeks and 4 months, decreased to one third of the 4-month level by 1 year, and then halved again by 2 years. Thus, 12.8% of 4-month cases were still cases at 2 years, 33.3% of 8-month cases were still cases at 2 years, and 46% of 12-month cases were still cases at 2 years. This is comparable to a Dutch study that showed that 25% of infants who manifested asymmetric head shape between 1 and 6 months still had asymmetry by 2 to 3 years.[13]

There did not seem to be any 1 factor that would have predicted which infants remained in the case group by age 2. However, it is obvious from our data that although there may be some clinical abnormality as the infant grows older, the level of parental concern is extremely low. In speaking to the parents, this seemed to be attributable mostly to hair growth obscuring the abnormality of head shape, in addition to the relative size of the problem appearing smaller as the head grew. The mean difference in transcranial diameters in the cases increased by only 2.2 mm between 6 weeks and 2 years of age. This raises the issue of whether our definition of a case should also take into account head circumference and parental concern. Additional studies are needed to address this issue.

The cases in our cohort were more likely to have been positioned supine for sleep in the first 6 weeks, confirming earlier studies that have shown supine sleeping to be an important determinant for NSP.[6,11,12,26] The total overall time spent supine in the first 6 weeks, both awake and asleep, seems also to be an important predictor. Of interest, the 6-week cases were also having more upright time, and this needs additional investigation to determine a relationship between back and upright time. It is possible that our question was misinterpreted or that upright time is a consequence or predictor of less easy temperament, which showed up later, in the 4-month cases. Although we found no significant difference relating to tummy time in our cohort, it is possible that our small number of cases was not sufficient to establish a relationship; however, we have previously shown that cases are likely to have <5 minutes a day of tummy time at 6 weeks.[11] In light of all of the above, we would still recommend that if infants are sleeping supine, then parents should try to reduce the time that their infants spend on their back during awake time by increasing supervised tummy, upright, and side-lying play time.

Although others[27,28] have believed that antenatal influences are strongly related to the later development of head deformity, we found no effect of obstetric or socioeconomic factors on the development of plagiocephaly in our cohort. However, at both 6 weeks and 4 months, cases were more likely to be those who had an abnormal passive head rotation at the newborn interview, suggesting either an in utero or a birth cause for neck tightness, or else asymmetric neck muscle development in those infants. The numbers were small, and not all infants were tested because to do the test, the infants had to be in a relaxed state, which was not always possible at the time of the interview. This result needs confirming in a larger study. If abnormal head rotation is seen, then physical therapy could be instituted to prevent preferential positioning as a result.

Cases were less active as judged by their mothers at 4 months, and this is in accordance with earlier work in which we have shown that cases had a lower activity level.[11] We hypothesize that less active infants are more likely to remain lying in the same position and thus develop a flat spot if there is a preferred position, particularly if head rotation is inadequate. Detecting differences in developmental delay may need a more comprehensive developmental assessment tool than the PDQ-II. Although there was a small indication of developmental delay at 6 weeks, we found no difference at 4 months. More difficult temperament may also be a factor, although this was significant only on the PAT test and not the overall temperament rating test. The question re-

mains as to whether these factors are causative or resultant of the head deformity.

There are some limitations to this study. Both the prevalence and the severity of the cases are probably conservative as a result of mothers' being highly aware of head shape by participating in the study and possibly taking preventive steps to avoid head flattening. Head shape is a continuous measure ranging from the perfectly symmetrical to the severely abnormal. The cutoff is somewhat arbitrary and was used to carry out logistic regression and to establish ORs for risk factors. The cutoff points of 93% and 106% were derived from a small sample, and prevalence rates would be different given different cutoffs. A receiver-operator characteristic analysis of a large case-control study would be needed to determine optimum cutoff points to resolve this issue. We have opted for a fairly conservative level that we believe adequately reflects visual assessment of abnormality.

Although brachycephaly and plagiocephaly could have been considered as 2 different outcomes, we believed that factors for both were similar and that by excluding brachycephalic cases, they would have been included with the control infants, thus making our estimates of ORs falsely conservative.

Some infants varied in and out of abnormal by being close to the cutoff points, and this in combination with possible measurement error would explain those in the NYNY, YNYN and YNNY categories in Fig 4, although it also possibly reflects that the skull is still malleable and subject to subtle changes in shape. We also recognize that we have small numbers of cases at 6 weeks and 4 months, and therefore our case versus control results need to be treated with caution. Failure to find a significant result may be attributable to sample size. However, excellent retention rates enabled us to track the prevalence through the first 2 years of life, allowing for greater understanding of the dynamics of head shape development as it relates to environmental and other factors and providing reassurance that the majority of cases resolve by 2 years of age.

## CONCLUSIONS

Head shape varied to a great extent in this group of normal infants in the first 2 years of life. The first 4 months seems to be an important time for the initiation of plagiocephaly and brachycephaly. Risk factors are particularly associated with early limitation of head rotation and early resting positions. A limitation in neck function should be checked for in the early weeks and neck motion exercises commenced if necessary to encourage full head turning to both sides. Supine sleeping also plays a major part. Although it is vital that the supine sleep position be maintained for SIDS protection, varying the head position in the first 6 weeks may be important for plagiocephaly prevention. Not being able to achieve this should alert parents to the possibility of limited neck mobility. Infants with lower activity levels may be more susceptible to developing plagiocephaly.

Although the maximum range of head shape deformity was seen at 6 weeks, the greatest point prevalence of plagiocephaly in our cohort was seen at 4 months. Almost 30% of the cohort exceeded the chosen cutoffs for classification of cases at some point in the first 8 months, but most cases improved with time, leaving a point prevalence of NSP of 3.3% at 2 years.

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## THE STATUS SYNDROME

"The alarming message here is that status has become a lethal threat. In the relatively prosperous, industrialized West, Michael Marmot, an epidemiologist at University College, London, writes, 'Where you stand in the social hierarchy is intimately related to your chances of getting ill and your length of life.' And the higher your status, the better your prospects. . . . [A] numbing arsenal of facts and figures serves to show that it is social rank—and not suspiciously similar-sounding factors like income or education—that makes the crucial difference. There's the study of Oscar winners that found they live 4 years longer than their co-stars and fellow nominees, and the fact that with each mile along the subway line from downtown Washington to suburban Montgomery County, MD, life expectancy increases by a year and a half. There is also a mountain of suggestive evidence from primate research: low-status rhesus macaques with heart disease; low-status baboons with soaring cortisol levels and unwholesome amounts of HDL cholesterol. It is not our social position per se that does us in, all this implies, but rather the stress that comes from having less control over our work and lives than people of higher rank. Not that this is exactly news."

Eakin E, review of Marmot M. *The Status Syndrome*. *New York Times Book Review*. August 22, 2004

Noted by JFL, MD

# Plagiocephaly and Brachycephaly in the First Two Years of Life: A Prospective Cohort Study

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