

Determinants of Nonsynostotic Plagiocephaly: A Case-Control Study

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ABSTRACT. *Objective.* There has been a large increase in reported cases of nonsynostotic plagiocephaly in infants since the adoption of supine sleeping recommendations to prevent sudden infant death syndrome. The objective of this study was to identify and quantify the determinants of nonsynostotic plagiocephaly in infants.

Methods. One hundred infants who received a diagnosis of having nonsynostotic plagiocephaly were recruited as case patients and compared with 94 control subjects who were selected from a citywide database of infants. The infants all were aged between 2 and 12 months. Information concerning sociodemographic variables, obstetric factors, infant factors, and infant care practices was obtained by parental interview.

Results. Case patients were significantly more likely to be male (adjusted odds ratio [aOR]: 2.51; 95% confidence interval [CI]: 1.23–5.16), to be a firstborn (aOR: 2.94; 95% CI: 1.46–5.96), and to have been premature (aOR: 3.26; 95% CI: 1.02–10.47). In the first 6 weeks, they were more likely to have been sleeping in the supine position (aOR: 7.02; 95% CI: 2.98–16.53), not to have had the head position varied when put down to sleep (aOR: 7.11; 95% CI: 2.75–18.37), and to have had <5 minutes a day of tummy time (OR: 2.26; 95% CI: 1.03–5.00). Mothers of case patients were more likely to perceive their infants as less active (aOR: 3.23; 95% CI: 1.38–7.56), to have a developmental delay (aOR: 3.32; 95% CI: 1.01–10.85), and to have had a definite preferred head orientation at 6 weeks (aOR: 37.46; 95% CI: 8.44–166.32). Case mothers were more likely to have no or low educational qualifications (aOR: 5.61; 95% CI: 2.02–15.56), although they were more likely to have attended antenatal classes (aOR: 6.61; 95% CI: 1.59–27.47).

Conclusions. Early identification of a preferred head orientation, which may indicate the presence of neck muscle dysfunction, may help prevent the development or further development of nonsynostotic plagiocephaly in infants. Plagiocephaly might also be prevented by varying the head position when putting the very young infant down to sleep and by giving supervised tummy time when awake. *Pediatrics* 2003;112:e316–e322. URL: <http://www.pediatrics.org/cgi/content/full/112/4/e316>; *plagiocephaly, supine position, craniofacial abnormalities, case control studies, infant care.*

ABBREVIATIONS. SIDS, sudden infant death syndrome; NSP, nonsynostotic plagiocephaly; OR, odds ratio; SE, standard error; CI, confidence interval; aOR, adjusted odds ratio.

The reduction in sudden infant death syndrome (SIDS) mortality has been spectacular after the SIDS prevention program, which recommends that infants sleep on their back.^{1–5} However, there seems to have been an associated increase in the incidence of nonsynostotic plagiocephaly (NSP), or deformational occipital plagiocephaly, a deformation or flattening of the back of the head not caused by craniosynostosis. It is likely that there have been both a real increase in NSP and an increase in awareness of the problem.^{6,7}

Although some investigators believe that NSP is only cosmetic and can often be helped with positional changes, many parents are concerned. Consequently, this has the potential to undermine the SIDS prevention program if parents decide to put their infants to sleep in the prone or lateral positions. Supine sleeping in very young infants is 6 times safer than prone sleeping and twice as safe as side sleeping,^{8–10} when infants are at risk of rolling to prone from the side.¹¹ Furthermore, as asymmetric head shapes become more common, there is the potential that plagiocephaly as a result of craniosynostosis may be missed.

We know of no other studies that have examined a wide range of risk factors for plagiocephaly. Clearly, supine sleeping position is implicated,^{12–15} but it is uncertain whether additional factors can affect the risk of developing plagiocephaly.

Advice to parents regarding prevention of NSP has focused on varying the infant's head position while asleep and on giving adequate "tummy time" (supervised prone play) when awake,^{15–19} but to date, there has been little or no evidence to support this advice. The aims of this study were to identify and quantify the determinants of NSP, to distinguish those infants most at risk, and to develop strategies that may help prevent NSP while maintaining the integrity of SIDS prevention advice.

METHODS

One hundred case patients, being consecutive infants between the ages of 2 and 12 months and diagnosed as having NSP, were recruited from the Middlemore Hospital craniofacial clinic and from Auckland pediatric physiotherapy clinics between January and August 2001. The infants had been referred to the clinics by pediatricians, general practitioners, and community child health

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nurses as having severity of NSP greater than that normally dealt with by the referring practitioners. Both visual and anthropometric examinations were used at the clinics to confirm plagiocephaly.

Control infants aged 2 to 12 months were systematically selected from the database of the Auckland region of the Plunket Society, a community child health nursing service. This database represents approximately 90% of all births in the region. Every sixth infant aged between 2 and 12 months was selected, being a total of 200 infants. A letter and information pamphlet were sent to the parents, inviting them to participate in the study. A second mailing to nonresponders, followed by a telephone call by Plunket Society personnel to the remaining nonresponders, was also conducted.

After obtaining informed consent, interviews with the mothers were performed in their homes by the principal investigator or by 1 other trained assistant. Questions related to the infant both at 6 weeks and at the time of interview. The interview questionnaire covered the following variables:

1. Sociodemographic variables: parents' ages, ethnicity, occupation, and mother's education.
2. Obstetric factors: parity, gestation, presentation, method of delivery, length of labor, and multiple birth
3. Infant factors: sex, morphometric measurements (weight, length, and head circumference at birth, 6 weeks, and last visit, taken from the infant's Well Child Health Record book, held by the parent), presence of hair loss on the back of the head, temperament, activity level and developmental delay perceived by the mother, and preferential head orientation
4. Infant care practices: sleep position; head positioning practices; type of bed; type and firmness of mattress, underbedding, and pillows; position and orientation of crib in room; use of mobiles; other resting places such as car seats; total daily time spent in the supine position; daily duration of tummy time; hand dominance of the mother; and parental holding positions and duration

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 (year 12–13), and 3) tertiary education or professional certification. The parents' occupations were rated in accordance with the New Zealand Socioeconomic Index classifications into high, medium, and low socioeconomic status,²⁰ the highest rating of either parent being used for the classification.

The softness of the mattress was assessed by the interviewer using a subjective rating of soft, medium, or firm. A weight (10.5 g/cm²) 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. When the mattress was extremely hard, 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. Where the mattress was not available, the mother was asked to rate it as soft, medium, or firm.

Active head rotation was assessed by seating the infant on the mother's knee, then holding a bright 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 directly looking over each shoulder, without moving the body around with the head. Any limitation in head rotation was recorded, as was any noticeable head tilt.

Statistical analysis was conducted using SAS (Release 8.00; SAS Institute, Cary, NC). Odds ratios (ORs) were estimated by performing univariate and multivariate logistic regression analyses. When the data were distributed normally, *t* tests were performed on continuous variables at the univariate level. Otherwise, a Wilcoxon rank sum test was used.

Variables included in the main multivariate model were sex; age at interview; parity; gestation; sleep position at 6 weeks; and mother's age, ethnicity, and highest qualification (Table 1). Rather than adding other significant variables to the main model, we separately added related variables into groups (obstetric factors and infant care factors) or singly (infant factors), to maintain robustness of the model in light of relatively small numbers. The study received ethical approval from the Auckland Ethics Committee.

RESULTS

One hundred case patients (100% of those invited to participate) took part. Right-sided occipital flattening was seen in 57% of case patients, left-sided in 38%, and bilateral in 5%. Cases were first noticed at a mean (\pm standard error [SE]) of 6.9 \pm 0.46 weeks by a parent (49%), Plunket nurse (24%), or a relative or other (27%). The parents of 72% of the cases believed that the head shape had improved since the condition was first noticed. In 11 (28.9%) of the 38 case patients who had other siblings, including twins, it was reported by the mother that a sibling had also had head asymmetry.

Of the parents of 200 control subjects who were

TABLE 1. Main Multivariate Model

Variable	Case (<i>n</i> [%] or Mean [SE])	Control <i>n</i> (%)	Univariate OR (95% CI <i>P</i> Value)	aOR Main Model or Multivariate (<i>P</i> Value)
Sex			$\chi^2 = 6.58, P = .01$	$\chi^2 = 6.32, P = .01$
Male	71 (71)	50 (53.2)	2.15 (1.19–3.90)	2.51 (1.23–5.16)
Female	29 (29)	44 (46.8)	1.00	1.00
Parity			$\chi^2 = 13.03, P = .0003$	$\chi^2 = 9.01, P = .003$
Firstborn	65 (65.0)	36 (38.7)	2.94 (1.57–5.52)	2.94 (1.46–5.96)
Later born	35 (35.0)	57 (61.2)	1.00	1.00
Gestation			$\chi^2 = 1.91, P = .17$	$\chi^2 = 3.95, P = .05$
<37 wk	15 (15.0)	8 (8.5)	1.90 (0.77–4.71)	3.26 (1.02–10.47)
≥ 37 wk	85 (85.0)	86 (91.5)	1.00	1.00
Sleep position at 6 wk			$\chi^2 = 24.27, P < .0001$	$\chi^2 = 19.86, P < .0001$
Supine	89 (89.0)	52 (55.3)	6.54 (3.10–13.79)	7.02 (2.98–16.53)
Nonsupine	11 (11.0)	42 (44.7)	1.00	1.00
Mother's age			$\chi^2 = 2.66, P = .45$	$\chi^2 = 4.63, P = .20$
<25	9 (9.0)	10 (10.6)	1.23 (0.41–3.74)	1.14 (0.28–4.57)
25–29	16 (16.0)	22 (23.4)	1.00	1.00
30–34	45 (45.0)	33 (35.1)	1.88 (0.86–4.11)	2.26 (0.87–5.87)
35+	30 (30.0)	29 (30.9)	1.42 (0.62–3.23)	2.81 (0.97–8.13)
Mother's highest qualification			$\chi^2 = 4.81, P = .09$	$\chi^2 = 11.46, P = .003$
None or school certificate	28 (28.0)	15 (16.0)	2.26 (1.08–4.73)	5.61 (2.02–15.56)
Sixth form/bursary	25 (25.0)	22 (23.4)	1.38 (0.69–2.75)	2.16 (0.93–5.02)
Tertiary/professional	47 (47.0)	57 (60.6)	1.00	1.00
Age at interview (wk)	25.2 (0.9)	28.0 (0.9)	$P = .02 \beta = -0.0005$ (-0.0001 to -0.0006)	$P = .02 \beta = -0.007$ (-0.001 to -0.13)

sent a postal invitation, 94 (47%) took part. Mothers of control subjects were asked whether they had ever had any concerns about their infant's head shape. Fourteen percent reported moderate or severe flattening, and 18% reported mild flattening. However, none of the control subjects had been referred or treated for plagiocephaly.

At the time of interview, the mean age (\pm SE) of case patients was significantly younger than the control subjects (25.2 ± 0.9 weeks vs 28.2 ± 0.9 weeks; $P = .02$). This was adjusted for in the multivariate model.

Sociodemographic Factors

No significant differences were found between case patients and control subjects for age of parents, ethnicity of parents, occupation, or age mother left school. Educational level was not significant at univariate level, although the point estimate for no or low qualifications was notable in comparison with the highest socioeconomic group. However, at the multivariate level, education level was significant, with an increased risk for the group with no or low qualifications (adjusted OR [aOR]: 5.61; 95% CI: 2.02–15.56; Table 1).

Obstetric Factors

Parity was highly significant, both at the univariate level (crude OR: 2.94; 95% CI: 1.57–5.52) and at the multivariate level (aOR: 2.94; 95% CI: 1.46, 5.96), showing an increased risk for firstborn infants. Other significant obstetric differences at the univariate level were mean length of gestation (\pm SE), with case patients being born earlier (38.2 ± 0.3 weeks vs 39.2 ± 0.2 weeks; $P = .01$), and mean (\pm SE) birth weight (3171 ± 75.9 g vs 3424 ± 73.5 g; $P = .02$). When gestation was categorized in the main multivariate model into preterm (<37 weeks) and term infants, case patients were significantly more likely to be premature (aOR: 3.26; 95% CI: 1.02–10.47). Case patients were also more likely to have had a breech or forceps- or ventouse-assisted delivery, compared with normal or caesarian deliveries (crude OR: 2.48; 95% CI: 1.07–5.74), although this did not remain significant when added to the multivariate model. Regarding antenatal education, more case mothers had

attended antenatal classes for this or a previous infant, compared with control subjects (aOR: 6.61; 95% CI: 1.59–27.47; Table 1).

Obstetric variables that were of borderline significance at the univariate level were abnormal intra-uterine positioning at any time during pregnancy (crude OR: 2.56; 95% CI: 0.95–6.01), abnormal presentation at birth (crude OR: 2.06; 95% CI: 0.96–4.40), longer mean (\pm SE) length of labor (11.5 ± 1.2 hours vs 8.8 ± 0.8 hours; $P = .06$), and an unusual head shape at birth (crude OR: 1.81; 95% CI: 0.93–3.50), as reported by the mother. None of these assumed significance when added to the main multivariate model. The study failed to detect any significant difference between the groups for the mother's age, multiple births, reduced amniotic fluid, uterine abnormality, position in last month of pregnancy, and number of weeks that the head was engaged (Table 2).

Infant Factors

Case patients were more likely to be male (aOR: 2.51; 95% CI: 1.23–5.16), and the mother was more likely to report a perceived developmental delay, mostly being head lag problems or late rolling over (aOR: 3.32; 95% CI: 1.01–10.85). When asked how active their infants were at present, case mothers were more likely than control mothers to report very inactive, inactive, or average levels of activity at present, compared with active to very active levels (aOR: 3.23; 95% CI: 1.38–7.56). This difference was not evident when mothers were asked how active the infant had been at 6 weeks of age (Table 1).

Mothers were also asked whether they had ever noticed their infant having difficulty turning the head. Case patients were more likely to have had a reported difficulty (crude OR: 14.01; 95% CI: 6.61–29.70). When a preferential head orientation at 6 weeks was known, more case patients had a preferred head direction (aOR: 37.46; 95% CI: 8.44–166.32). There was no difference in the direction preferred. When the researcher observed active rotation of the head, more control infants had full range of motion both to the right and to the left (aOR: 20.40; 95% CI: 5.83–71.35; Table 3).

Compared with children without hair loss, infants

TABLE 2. Obstetric Factors

Variable	Case (n [%])	Control (n [%])	Univariate OR (95% CI)	aOR Main Model + Obstetric
Multiple birth (missing = 1)			$\chi^2 = 1.70, P = .19$	$\chi^2 = 1.04, P = .31$
Multiple	12 (12.0)	6 (6.5)	1.98 (0.71–5.50)	0.37 (0.05–2.51)
Singleton	88 (88.0)	87 (98.6)	1.00	1.00
Presentation (missing = 36)			$\chi^2 = 3.45, P = .06$	$\chi^2 = 0.24, P = .63$
OA	58 (69.8)	62 (82.7)	1.00	1.00
Other (OP, breech, transverse)	25 (30.1)	13 (17.3)	2.06 (0.96–4.40)	1.29 (0.46–3.60)
Type of delivery (missing = 1)			$\chi^2 = 4.50, P = .03$	$\chi^2 = 1.54, P = .21$
Normal vaginal/cesarean	79 (79.0)	84 (90.3)	1.00	1.00
Breech, forceps, or ventouse	21 (21.0)	9 (9.7)	2.48 (1.07–5.74)	2.21 (0.63–7.81)
Antenatal class attended			$\chi^2 = 11.61, P = .0007$	$\chi^2 = 6.74, P = .009$
Yes	92 (92.0)	68 (72.3)	2.08, (1.37–3.23)	6.61 (1.59–27.47)
No	8 (8.0)	26 (27.7)	1.00	1.00
Reported head shape at birth			$\chi^2 = 3.02, P = .08$	$\chi^2 = 0.003, P = .96$
Unusual	30 (30.0)	18 (19.1)	1.81 (0.93–3.50)	1.03 (0.34–2.88)
Not noticed, OK	70 (70.0)	76 (80.9)	1.00	1.00

with symmetric hair loss had a lower risk of plagiocephaly (crude OR: 0.31; 95% CI: 0.16–0.61), whereas those with asymmetric hair loss were more likely to have plagiocephaly (crude OR: 3.75; 95% CI: 0.79–17.78), although this was not statistically significant due to small numbers. Factors that failed to show a significant difference between case patients and control subjects were head circumference at birth and at 6 weeks, length at birth, velocity of head circumference growth and weight change from birth to 6 weeks, temperament both at 6 weeks and at interview, activity level at 6 weeks, and postneonatal health problems.

Infant Care Factors

At 6 weeks, case patients were more likely to be sleeping supine (aOR: 7.02; 95% CI: 2.98–16.53). However, at interview, the difference had reversed, with fewer case patients than control subjects sleeping supine (crude OR: 0.37; 95% CI: 0.20–0.65). Five control infants and no case infants were sleeping prone at 6 weeks. All other nonsupine sleepers were in the lateral position or in a combination of lateral and supine positions (Table 1).

When asked whether the head position of the infant, when put down to sleep, had been turned or varied regularly in the first 6 weeks, more case mothers reported not doing so (aOR: 7.11; 95% CI: 2.75–18.37). However, more case parents had tried unsuccessfully to vary the head position but reported that the infant always turned the head back to a preferred position (aOR: 7.38; 95% CI: 2.43–22.42). At the time of interview, case parents were more likely than control parents to be trying to vary the head position (crude OR: 5.88; 95% CI: 3.13–11.11; Table 4).

Although there was no difference detected in whether tummy time was provided, having <5 minutes per day of tummy time at 6 weeks was significant at both the univariate and multivariate levels (aOR: 2.26; 95% CI: 1.03–5.00). Having >20 hours a day of total back time at 6 weeks was also significant at the univariate level (crude OR: 6.44; 95% CI: 3.26–12.74). Total back time included supine sleeping, plus any supine positioning on the floor, couch, car seat, bouncinette, changing table, parent's arms, and

other places. However, this variable was not added to the multivariate model because of intrinsic relationships with supine sleeping and head varying, which reduced the robustness of the model (Table 4).

The mother's preferred holding position at 6 weeks was classified into supine or nonsupine; more case mothers preferred a supine holding position (crude OR: 1.91; 95% CI: 1.07–3.43), although this was not significant at the multivariate level. The case infants spent significantly longer supine while resting the head on the mother's arms at 6 weeks, usually while feeding or cuddling (median of 180 minutes for case patients vs 60 minutes per day for control subjects; $P < .0001$; Table 4).

Analysis of variance was performed to compare the tool measurement and the subjective rating of the infants' mattresses. This showed that the average measurements for depression of the weight increased significantly with the 3 subjective categories (soft, medium, and firm), as assessed by the subjective assessments of the mattresses. A weighted κ analysis yielded a score of 0.65 (95% CI: 0.54–0.76) and hence a fair agreement.

Fewer than half of the mattresses used at 6 weeks were available for measurement by the tool; therefore, these data have been excluded. However, the mattress being used at the time of interview was significantly firmer for case patients than for control subjects at the univariate level but not multivariately (crude OR: 1.99; 95% CI: 1.01–3.93; aOR: 1.91; 95% CI: 0.83–4.43). The type of mattress (foam, innerspring, synthetic fiber, or other) was significantly different for case patients and control subjects at 6 weeks ($P = .01$) but not at time of interview.

Case patients were more likely to be sleeping in a crib rather than a bassinet or other type of bed at 6 weeks (crude OR: 1.90; 95% CI: 1.00–3.59). Although more control subjects than case patients were using pillows at 6 weeks, the numbers were small and no significant difference was detected. There was no difference between case patients and control subjects in the number of hours the infant spent on the primary mattress, a bouncer, or a car seat at 6 weeks. Use of a pacifier and the mother's hand dominance

TABLE 3. Infant Factors

Variable	Case (n [%])	Control (n [%])	Univariate OR (95% CI)	aOR Main Model + Infant Factors
Reported preferential head orientation at 6 wk if known (missing = 16)			$\chi^2 = 29.39, P < .0001$	$\chi^2 = 22.69, P < .0001$
Preference	89 (96.7)	43 (50.0)	29.66 (8.70–101.01)	37.46 (8.44–66.32)
No preference	3 (3.3)	43 (50.0)	1.00	1.00
Activity level now			$\chi^2 = 8.81, P = .002$	$\chi^2 = 7.33, P = .007$
Very active to active	65 (65.0)	79 (84.0)	1.00	1.00
Average to inactive	35 (35.0)	15 (16.0)	2.84 (1.43–5.64)	3.23 (1.38–7.56)
Developmental delay			$\chi^2 = 3.88, P = .05$	$\chi^2 = 3.92, P = .05$
Reported delays	17 (17.0)	7 (7.45)	2.55 (1.00–6.45)	3.32 (1.01–10.85)
No delays	83 (83.0)	87 (92.6)	1.00	1.00
Limitation of head rotation (missing = 11)			$\chi^2 = 31.49, P < .0001$	$\chi^2 = 22.29, P < .0001$
Limitation	51 (52.0)	4 (4.7)	21.97 (7.47–64.65)	20.40 (5.83–71.35)
No limitation	47 (48.0)	81 (95.3)	1.00	1.00
Mattress firmness (missing = 56)			$\chi^2 = 3.91, P = .05$	$\chi^2 = 2.29, P = .13$
Firm (measurement ≤ 10 mm)	46 (60.5)	27 (43.5)	1.99 (1.01–3.93)	1.91 (0.83–4.43)
Softer (measurement > 10 mm)	30 (39.5)	35 (56.5)	1.00	1.00

TABLE 4. Infant Care Factors

Variable	Case (n [%])	Control (n [%])	Univariate OR (95% CI)	aOR Main Model + Infant Care
Head position varied in first 6 wk (when put down to sleep)			$\chi^2 = 35.46, P < .0001$	$\chi^2 = 19.04, P < .0001$
Yes	20 (20.0)	60 (63.8)	1.00	1.00
No	46 (46.0)	22 (23.4)	6.27 (3.06–12.85)	7.11 (2.75–18.37)
Tried—infant moved head back	34 (34.0)	12 (12.8)	8.50 (3.71–19.5)	7.38 (2.43–22.42)
Tummy time at 6 wk			$\chi^2 = 3.80, P = .05$	$\chi^2 = 4.09, P = .04$
<5 min/d	49 (49.0)	33 (35.1)	1.78 (1.00–3.16)	2.26 (1.03–5.00)
≥5 min/d	51 (51.0)	61 (64.9)	1.00	1.00
Mother’s preferred holding position at 6 wk (missing = 3)			$\chi^2 = 4.77, P = .03$	$\chi^2 = 2.26, P = .13$
Infant on back	50 (50.5)	32 (34.8)	1.91 (1.07–3.43)	1.89 (0.83–4.31)
Infant not on back	49 (49.5)	60 (65.2)	1.00	1.00

were not significantly different between case patients and control subjects.

Although more control infants had ever breastfed, this difference was not significant (crude OR: 2.19; 95% CI: 0.85–5.64). Although mean (\pm SE) length of breastfeeding was significantly shorter in case patients (3.4 ± 0.3 months vs 4.6 ± 0.3 months; $P = .002$), length of exclusive breastfeeding was not found to be significant. A Cox regression analysis was performed on breastfeeding length, controlling for case, parity, and mother’s qualification level. Case and parity were not significant, but qualification was highly significant ($P = .0002$); ie, the difference in breastfeeding duration was accounted for by differences in the mothers’ educational levels and not the differences between the case patients and control subjects. Interactions of 6-week sleeping position with firmness of mattress, back time, and head varying all were nonsignificant.

DISCUSSION

Our findings are similar to those reported in other studies showing an approximate 3:2 ratio of right- to left-sided occipital flattening^{7,12,22–24} and a 7-week age at first presentation.^{23,25,26} However, earlier studies have not shown an increased prevalence in siblings, and this is an area that deserves additional investigation, as it may reflect either a genetic predisposition or repetition of infant care practices.

Significant obstetric factors in this study are similar to those reported from other studies. Firstborns and boys were represented strongly among our cases, as in other studies.^{7,14,24,27–31} Although we found shorter gestational length and difficult deliveries to be significant—as has been reported elsewhere^{28,32,33}—fetal positioning, presentation at birth, and length of labor were of borderline significance. A larger study may be needed to detect differences in these factors and others such as multiple births, reduced amniotic fluid, and uterine abnormality.

Preferential head orientation and/or neck dysfunction were strong features of our case group, as in other studies.^{7,13–15,29} It is unclear whether these characteristics contributed to the development of the plagiocephaly or were secondary to the development of a flat spot that became a preferred resting position. Golden et al¹⁴ postulated that supine sleepers do not develop neck and upper-body strength to the same

level as side or prone sleepers. It is likely that an early preferential head orientation, which some^{39–42} consider a normal developmental trait in the first few weeks, in conjunction with low levels of tummy time, high levels of back time, and not varying the head position in the very young infant, may combine to produce a tightness of the neck musculature on one side or a laxness on the other.

As in other reports,^{12,14,15,31} supine sleeping at 6 weeks was strongly related to development of plagiocephaly, both at the univariate and the multivariate levels. By the time of interview, however, only one third of case patients were sleeping supine, probably reflecting attempts at positioning changes to counter head shape asymmetry in the case patients. In younger infants, this has the potential to expose them to less safe sleeping positions and is evidence of undermining of SIDS prevention advice by concerned parents.

It has been postulated that the most prevalent cause of NSP is repeated positioning on the same part of the head, with gravitational forces acting on the soft infant cranium lying on a flat surface.^{31,37} The differences between the case patients and control subjects in head positioning for sleep indicate that varying the head position when laying the child down to sleep in the first 6 weeks may be related to a reduction in the risk of plagiocephaly. It would logically seem that, although unproved, such a practice may in fact be preventive in the development of plagiocephaly. This can be achieved while maintaining the supine sleep position but gently turning the head to opposite sides at alternate sleeps, and is more easily accomplished in the first few weeks, before the infant has developed stronger head control. By the time of interview, most control subjects had stopped this practice, whereas two thirds of case parents were still attempting it, reflecting efforts on the part of case parents to treat the head asymmetry.

Having 5 or more minutes a day of tummy time by 6 weeks of age also seems to be protective. These measures have not previously been demonstrated in normal term infants at home, although one study has shown the protective effect of a regimen of varied sleeping positions, including prone, in preventing dolichocephaly in premature infants in a hospital setting.³⁸

Although no objective measure of developmental

delay was made, more case mothers perceived a delay. Although developmental delay has been linked with NSP,^{15,23,30,33–35} likely delays are a contributory factor in the development of head shape asymmetry. It has been shown that supine sleepers achieve certain milestones later than prone sleepers, although they catch up by 18 months,^{18,36} and it is possible that the reported developmental delays were related to this trend. Although infant temperament, rated between very placid and very unsettled, failed to show a difference between the groups, at the time of interview, the case infants were rated by their mothers as significantly less active than control infants. This difference may be causally related to plagiocephaly and deserves additional consideration.

Case patients were sleeping on firmer mattresses at the time of interview, but this was not significant when other factors were controlled for in the multivariate model. Mattress firmness did not seem to interact with sleep position, head varying, or time spent on the back.

Our case mothers preferred to hold their infants in the supine, rather than the upright, position. Although we did not inquire about the total amount of upright time while awake, it is reasonable to suppose that upright time (eg, upright cuddles on the shoulder) would also confer the benefit of keeping the infant off the occiput and of allowing development of neck strength, although this would need to be confirmed in future studies.

Other studies^{21,30} have found that plagiocephalic infants have a large head circumference. Our case patients and control subjects showed no difference at either the birth or the 6-week measurements. Alopecia was less likely to occur in the case patients, but where it did occur, it was more likely to be asymmetric. Symmetric loss of hair in young infants may be a marker for good head rotation, indicating that the infant is moving the head to varying positions and rubbing the downy newborn hair off in the process.

Of interest is that case mothers were significantly more likely to have attended antenatal education classes. Awareness of SIDS is high in New Zealand, and it is probable that these mothers learned the message of supine sleeping for SIDS prevention. Awareness of NSP was much less, and the preventive strategies were less established.

The level of the mother's educational qualifications was the only socioeconomic variable that reached statistical significance after adjusting for confounders. Although not reported in other studies, this could be a real difference, or it could reflect a sampling error on the part of the control group—ie, more highly educated mothers may have responded to the postal invitation to take part. Another explanation could be that more highly educated mothers may be more likely to intervene in early development of plagiocephaly, and the infants therefore may not need later referral to specialist services.

There were some limitations in this study. Although there was an excellent response to face-to-face recruitment of case patients in plagiocephaly clinics, the selection of community control subjects

by postal invitation led to a lower response rate in the control group. This group may have been mothers who had an interest in head shape development in their infants, as evidenced by 32% reporting head shape concerns about their infants. Our estimates of ORs thus are likely to be conservative.

Another limitation of the study was the retrospective reporting of the 6-week data and estimations of time spent in various places. This may have led to recall bias on the part of the case mothers. Our study was also unable to address such factors as thickness of cranial bones or external hydrocephalus,²¹ which may be important features in determining an infant's propensity to develop NSP. Although the mean age of infants at the time of interview was significantly different by 3 weeks, it seems unlikely that this age difference would have contributed any real difference to the responses between the 2 groups. This variable was controlled for in the multivariate analysis.

CONCLUSIONS

Infants who may be at risk of developing NSP are male, are firstborn, are preterm, sleep supine only, and are those whose mothers are less educated. They are infants who are likely to be less active, to have a developmental delay, and to exhibit a preferential head orientation by 6 weeks. They are likely not to have had the head position varied when being put down to sleep and to have had <5 minutes of tummy time a day in the first 6 weeks.

Antenatal educators, those who provide maternity care, and those involved in monitoring the young infant's health and development should continue to teach the protective benefits of supine sleeping for SIDS prevention, but they should combine this with messages that give parents greater understanding of head shape protection. Parents can help by giving regular periods of supervised prone play from an early age; by holding the infant upright for cuddles; and by not leaving the infant supine for long periods in car seats, on floor blankets, or in bouncers. The head position should be alternated when laying the very young infant down to sleep.

Postural preference and limitation of neck rotation seem to be important factors that may contribute to the development of NSP. A flat spot, a strong preference for turning the head to one side, or difficulty turning the head should be looked for at an early age during routine checks by doctors, midwives, and community child health nurses. If present, then the mother may need to be educated about holding and positioning practices to help address the asymmetry.

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