Characteristics of 2733 Cases Diagnosed With Deformational Plagiocephaly and Changes in Risk Factors Over Time

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Objectives: **To describe infant and maternal characteristics among infants with plagiocephaly and to quantify time trends in potential risk factors for plagiocephaly.**

Design: **Case-only study. We described the characteristics of individuals born between 1987 and 2002. We also compared characteristics of individuals born from 1987 through 1990, before the American Academy of Pediatrics 1992 sleep-position recommendations, with those of individuals born from 1991 to 2002.**

Setting: **Children's Craniofacial Center at Children's Hospital and Regional Medical Center in Seattle, Washington.**

Participants: **Subjects included 2733 infants diagnosed with deformational plagiocephaly or brachycephaly before 18 months of age who were born from 1987 to 2002.**

Main Outcome Measure: **Descriptive statistics, odds ratios, and 95% confidence intervals.**

Results: **Among individuals born from 1991 to 2002, 91.6% had occipital-only flattening, 17.2% were brachycephalic, 67.7% were boys, and 9.9% were multiple birth infants. As compared with individuals born from 1987 through 1990, those born from 1991 to 2002 were more apt to be a multiple birth (odds ratio [OR] 3.4, 95% confidence interval [CI]: 0.8, 14.1) and to have a mother $\geq$35 years of age (OR, 3.2; 95% CI, 1.4 to 7.3); they were hospitalized less commonly at birth for 4 or more days (OR, 0.02; 95% CI, 0.01 to 0.06).**

Conclusions: **Several risk factors for plagiocephaly were more common among individuals born after the 1992 American Academy of Pediatrics sleep-position recommendations. These results are consistent with the explanation that supine sleeping modifies the association between such risk factors and plagiocephaly. Further studies with a control group are needed to validate this conclusion.**

KEY WORDS: *plagiocephaly, risk factors, sleep position*

Deformational plagiocephaly (or plagiocephaly) is characterized by an abnormal head shape due to external pressure placed upon the calvaria. Diagnosis usually occurs between 3 and 10 months of age. Prior to 1992, the prevalence of plagiocephaly among infants younger than 1 year of age was reportedly 5% (Clarren et al., 1979). In recent years, craniofacial centers and primary care providers in the United States reported a dramatic increase of up to 600% in referrals for plagiocephaly (Argenta et al., 1996; Kane et al., 1996b; Turk et

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al., 1996). The increase roughly coincides with the 1992 American Academy of Pediatrics (AAP) recommendation to avoid the prone sleep position to reduce the risk of Sudden Infant Death Syndrome (SIDS) (AAP, 1992). Following the 1992 AAP recommendation, the prevalence of supine sleep positioning increased from 13% in 1992 to 70% in 2002 (National Infant Sleep Position Study, 2004). A study conducted in New Zealand, where supine sleep positioning recommendations were first implemented in 1991 suggests that the prevalence of plagiocephaly before 6 months of age is now approximately 10% (Mitchell et al., 1997; Hutchison et al., 2004). Supine sleep positioning is a well-established risk factor for plagiocephaly, and the rapid and large rise in supine sleeping could plausibly account for the sharp increase in the prevalence of plagiocephaly (Kane et al., 1996b; Turk et al., 1996; Hutchison et al., 2003).

That only a fraction of infants who sleep supine develop plagiocephaly, however, suggests that other factors may play a role. For example, an infant constrained *in utero* due to being a twin or macrosomic may have a subtle neck-muscle injury or a flat spot on her head, either of which could potentially predispose her to developing plagiocephaly (Kane et al., 1996a). Other infant, maternal, or delivery characteristics hypothesized to be associated with plagiocephaly include birth injury, prematurity, and congenital anomalies (Clarren, 1981; Hutchison et al., 2003). To the extent that these characteristics are associated with risk of plagiocephaly, some such associations might be potentiated by sleep position. That is, the association might be realized only when infants sleep supine. We conducted a large case-only study to describe birth, maternal demographic, and delivery characteristics among infants diagnosed with positional plagiocephaly between 1987 and 2002. We also sought to quantify time trends in demographic and birth characteristics that are potential risk factors for plagiocephaly, either independently or jointly with the change in sleep position.

Case-only studies (Yang et al., 1997) allow for the indirect estimation of such effect modification between two risk factors when the two risk factors are independent in the general population (Khoury and Flanders, 1996; Albert et al., 2001). If this assumption is met, such an approach provides a more statistically efficient approach to examining effect modification relative to case-control studies, which requires much larger sample sizes for the estimation of first-order associations of equal magnitude (Yang et al., 1997). Independence between putative plagiocephaly risk factors and the likelihood of supine sleeping is a strong assumption that cannot be tested formally without studying a control group of infants without plagiocephaly. Nevertheless, pending confirmation through a study that includes controls, the case-only design allows for the preliminary evaluation of effect modification hypotheses. Specifically in this time-trend analysis, for hypothesized risk factors relatively constant in prevalence over the time period of study, a notable increase in prevalence among cases would be consistent with a role for supine sleeping as a modifier of the association between plagiocephaly and the given risk factor.

METHODS

Case Identification

Infants with plagiocephaly born between 1987 and 2002 in Washington State and diagnosed at the Children's Craniofacial Center at Children's Hospital and Regional Medical Center (Children's Hospital) in Seattle, Washington, before 18 months of age were eligible for the study. Children's Craniofacial Center is a tertiary care center that receives referrals for plagiocephaly from throughout Washington State. It was the primary helmet treatment facility for plagiocephaly in Western Washington during the period of our study. We used the Children's Craniofacial Center registry, a database that has tracked plagiocephaly diagnoses at Children's Craniofacial Center since the early 1980s, to identify plagiocephaly cases. We also queried the Children's Hospital electronic outpatient database, which began in 1994, and inpatient medical records, available since 1981, to identify cases that may have been missed inadvertently by the registry. Although the patients listed in the Children's Craniofacial Center registry carried a specific diagnosis of positional plagiocephaly, infants with plagiocephaly listed in the electronic hospital records could be identified only through the less specific ICD-9 code 754.0, a general diagnosis for ''asymmetry of the skull, face and jaw'' (Hart and Hopkins, 2003). We therefore conducted a chart review on all cases identified from the electronic hospital records to verify diagnosis of plagiocephaly. Our case definition specified that the infant must have been diagnosed with positional, deformational, or nonsynostotic plagiocephaly or brachycephaly by a Children's Craniofacial Center health care provider.

Data Collection on Risk Factors

A list of cases was provided to the Washington State Department of Health (DOH), and these cases were cross-referenced with birth certificate and hospital discharge data by using each infant's name, date of birth, and gender. Birth and death certificate information was linked by the DOH to mother- and infant-related hospitalization data extracted from a database that routinely collected all hospital discharges for inpatient stays at nonfederal hospitals in Washington State (the Comprehensive Hospital Abstract Reporting System, or CHARS) (Parrish et al., 1993; DOH, 2006b). From this data source, we collected birth and delivery characteristics, demographics, and all ICD-9 diagnoses and procedure codes for the birth hospitalization for both the mother and infant. We also obtained hospital discharge data (CHARS) for case infants through the first 12 months of life in order to identify congenital anomalies and health conditions present but not diagnosed at birth. In addition, we obtained Children's Hospital outpatient electronic records from 1994 onward that documented the first 12 months of life.

Analyses

Changes in national sleep-position prevalence were compared graphically to the increase in number of cases diagnosed at our Children's Craniofacial Center (National Infant Sleep Position Study, 2004). Cases were categorized into two groups based on year of birth, those born from 1987 through 1990 and those born from 1991 to 2002. The two groups are divided along the 1992 AAP sleep-position recommendations, with those born before the recommendation most likely sleeping prone and those born after most likely sleeping supine. A decision was made *a priori* to use 1991 as the cutoff for supine sleep positioning even though the AAP recommendations were issued in 1992, because 1991 was the year a widely distributed local newspaper printed the recommendation that parents should place their infants in a nonprone position to avoid SIDS (Guntheroth and Spiers, 1991).

Percentages were used to describe the distribution of select infant birth, maternal demographic, and maternal delivery characteristics of cases across the two time periods. Logistic regression was used to estimate odds ratios (OR) and 95% confidence intervals (CI), comparing case characteristics before (1987 to 1990) and after (1991 to 2002) sleep-position recommendations, adjusting for confounding factors. To enumerate potential confounding factors, we *a priori* identified factors believed associated with both plagiocephaly and a given exposure of interest based on scientific literature and plausibility (Clarren, 1981; Bruneteau and Mulliken, 1992; Kane et al., 1996b; Hutchison et al., 2003). Separate logistic models were generated for each potential risk factor, adjusting for the factors identified *a priori* as potential confounders of that risk factor's association with plagiocephaly (i.e., factors related both to the potential risk factor of interest and to the risk of plagiocephaly but not in the causal pathway) (Koepsell and Weiss, 2003). Unless noted, all variables were missing in fewer than 3% of records. This research was conducted with approval by the Children's Hospital and Regional Medical Center Internal Review Board, Seattle, Washington, and the DOH Institutional Review Board.

Results

The number of cases increased from 41 in 1992 to 484 in 2002 (Fig. 1), a time period in which the number of live births decreased by 1.5% in Washington State (DOH, 2006a). Concomitant with the increase in the number of cases, the national prevalence of supine sleep-positioning prevalence rose from 13% in 1992 to 70% in 2002, whereas prone sleep positioning demonstrated the reverse trend in this same time period (Fig. 1) (National Infant Sleep Position Study, 2004).

Of the 2960 initially eligible cases, 2870 (97%) were linked to a Washington State birth certificate. After excluding 97 subjects who did not meet the case definition, the study population was made up of 2773 cases. Ninety-eight individuals were born from 1987 through 1990, and the remaining 2675 were born from 1991 to 2002. Among those born from 1991 to 2002, after the supine sleep-positioning recommendations, 80.3% were diagnosed with deformational plagiocephaly between 3 and 9 months of age, 17.2% were brachycephalic, 63.6% had moderate or severe flattening, and 88.4% had neck limitations noted in the chart. Sixty-seven percent of case infants born from 1991 to 2002 were boys, 16.5% were less than 37 weeks gestation at birth, 5.8% had an injury at birth, and 21.3% were born with a congenital anomaly.

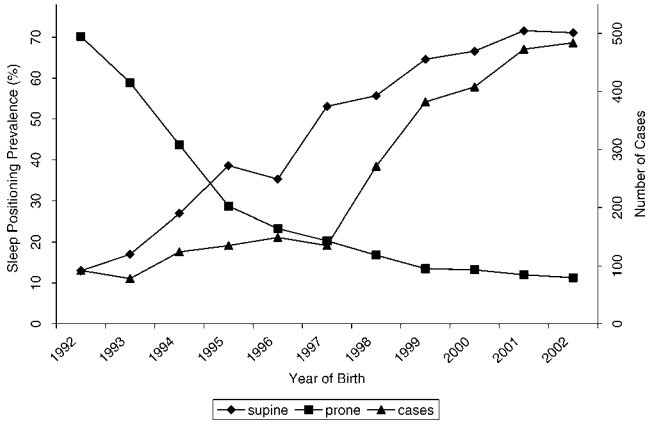

FIGURE 1 Changing sleep-positioning prevalence and number of cases of plagiocephaly.

Comparatively, a greater proportion of case infants born from 1987 to 1990 were diagnosed with plagiocephaly before 3 months of age (17.7% versus 5.5%) compared with infants born from 1991 to 2002. Fewer infants born in the earlier years had occipital-only flattening (34.7% versus 91.6%), were low birth weight (8.2% versus 12.6%), were premature (9.6% versus 16.5%), were a multiple-birth infant (3.1% versus 9.9%), or had a birth hospital stay of 0 to 1 day (6.5% versus 40.6%) as compared with those born in later years (Table 1). Fewer mothers of case infants born from 1987 to 1990 were 35 years or older at the time of birth (9.2% versus 22.7%), primiparous (40.6% versus 53.4%), or had oligohydramnios (1.1% versus 5.6%) relative to mothers of those born from 1991 to 2002. Mothers of infants born from 1987 to 1990 were more often smokers (23.9% versus 11.6%) and more often had a forceps-assisted delivery (19.1% versus 6.2%) relative to mothers of case infants born from 1991 to 2002 (Tables 2 and 3).

For most analyses quantifying time trends in potential risk factors for plagiocephaly, the adjusted estimates did not differ to an important degree from the unadjusted estimate, and so our discussion is limited to the adjusted estimates except where noted. With regard to changes in infant birth characteristics over time, as compared with those born in the earlier period, case infants born from 1991 to 2002 tended to be older at diagnosis (ORs ranged from ~3 to 5 for those 3 months or older as compared with younger than 3 months); to have occipital-only flattening (unadjusted OR, 27.5; 95% CI, 14.1 to 53.6); to be boys (OR, 1.3; 95% CI, 0.9 to 2.0); and to have been a multiple birth (OR, 3.4; 95% CI, 0.8 to 14.1) (Table 1). Being a case infant born from 1991 to 2002 was associated inversely with a gestational age of ≥42 weeks (OR, 0.2; 95%

TABLE 1 Infant Birth Characteristics Among Cases Diagnosed With Plagiocephaly at Seattle Children's Hospital From 1987 to 2002 by Select Year Groups

	1987–1990 (n = 98) %	*1991–2002 (n = 2675) %*	*Unadjusted Odds Ratio (OR)*	*Unadjusted 95% Confidence Interval (CI)*	*Adjusted OR*	*Adjusted 95% CI*
Age at diagnosis (mo)[1,2]						
0–3	17.7	5.5	1.0	Ref	1.0	Ref
3–6	37.5	40.0	3.4	1.9–6.3	3.2	1.5–6.5
6–9	32.3	40.3	4.0	2.2–7.4	4.4	2.1–9.5
9–12	8.3	10.7	4.1	1.7–9.8	3.4	1.3–9.3
12–18	4.2	3.5	2.7	0.9–8.3	5.2	1.1–25.7
Brachycephaly[3,4]						
No	89.2	82.8	1.0	Ref		
Yes	10.8	17.2	1.7	0.8–3.8		
Location of flattening[3]						
Frontal only	16.3	1.6	1.0	Ref		
Occipital only	34.7	91.6	27.5	14.1–53.6		
Occipital and frontal	49.0	6.8	1.4	0.8–2.8		
Severity[3,5]						
Mild	31.2	36.4	1.0	Ref		
Moderate	37.7	46.3	1.0	0.6–1.9		
Severe	31.1	17.3	0.5	0.2–0.9		
Neck involvement (as noted in chart)[3,6]						
No	13.5	11.6	1.0	Ref		
Yes	86.5	88.4	1.2	0.6–2.4		
Male infant[7,8]						
No	38.8	32.3	1.0	Ref	1.0	Ref
Yes	61.2	67.7	1.3	0.9–2.0	1.3	0.8–2.0
Infant birth weight (g)[9–11]						
<2500	8.2	12.6	1.6	0.8–3.4	2.1	0.6–7.1
2500–3999	80.4	76.7	1.0	Ref	1.0	Ref
≥4000	11.3	10.7	1.0	0.5–1.9	0.7	0.4–1.4
Infant gestational age (wk)[9–11]						
≤36	9.6	16.5	1.7	0.9–3.4	2.5	0.8–8.4
37–41	81.9	82.2	1.0	Ref	1.0	Ref
≥42	8.5	1.3	0.2	0.1–0.3	0.2	0.1–0.4
Multiple birth[9,10]						
No	96.9	90.1	1.0	Ref	1.0	Ref
Yes	3.1	9.9	3.5	1.1–11.1	3.4	0.8–14.1
Infant birth injury[7–9,11,12]						
No	86.0	94.2	1.0	Ref	1.0	Ref
Yes	14.0	5.8	0.4	0.2–0.7	0.5	0.3–1.1
Congenital anomaly[9,11]						
No	77.4	78.7	1.0	Ref	1.0	Ref
Yes	22.6	21.3	0.9	0.6–1.5	0.9	0.5–1.5
Length of hospital stay (d)[2,8,9,11,12]						
1	6.5	40.6	1.0	Ref	1.0	Ref
2	50.0	29.9	0.1	0.04–0.2	0.06	0.03–0.16
3	19.6	14.5	0.1	0.05–0.3	0.03	0.01–0.09
≥4	23.9	14.9	0.1	0.04–0.2	0.02	0.01–0.06

Note: Ref = Referent group.

[1] Adjusted estimate accounted for mother's county of residence and race, family income, gestational age, days hospitalized at birth, birth injury, and congenital anomaly.

[2] Variable missing 3% to 6%.

[3] Unadjusted only; no adjustments needed.

[4] Missing brachycephaly information, 8.8%.

[5] Missing severity information, 42.9%.

[6] Missing neck involvement information, 37.1%.

[7] Adjusted estimate accounted for congenital anomaly other than plagiocephaly.

[8] Adjusted estimate accounted for birth weight and gestational age.

[9] Adjusted estimate accounted for maternal age, race, and parity.

[10] Adjusted estimate accounted for household income, mother's marital status, county of residence, maternal smoking in pregnancy, and trimester that prenatal care began.

[11] Adjusted estimate accounted for infant gender and multiple birth.

[12] Adjusted estimate accounted for forcep, vacuum, cesarean section and/or breech or malpresentation delivery, cephalopelvic disproportion, prolonged and/or obstructed labor.

TABLE 2 Maternal Demographic and Socioeconomic Characteristics Among Case Infants Diagnosed With Plagiocephaly at Seattle Children's Hospital Between 1987 and 2002 by Select Year Groups*

	1987–1990 (n = 98) %	*1991–2002 (n = 2675) %*	*Unadjusted OR*	*Unadjusted 95% CI*	*Adjusted OR*	*Adjusted 95% CI*
Mother's age (y)						
<25	25.5	18.1	0.9	0.5–1.5	0.9	0.5–1.8
25–29	34.7	27.2	1.0	Ref	1.0	Ref
30–34	30.6	32.0	1.3	0.8–2.2	1.6	0.9–2.8
≥35	9.2	22.7	3.1	1.5–6.6	3.2	1.4–7.3
Mother's race†						
White	90.8	84.5	1.0	Ref	1.0	Ref
Asian	4.1	7.7	2.0	0.7–5.6	2.8	0.7–11.7
Other	5.1	7.8	1.6	0.7–4.1	1.7	0.6–5.0
Household income‡						
<$30,000	20.2	18.6	1.0	Ref	1.0	Ref
$30–45,000	47.2	44.8	1.0	0.6–1.8	0.9	0.4–1.8
$45–60,000	23.6	26.5	1.2	0.6–2.3	1.1	0.5–2.5
>$60,000	9.0	10.1	1.2	0.5–2.8	0.9	0.3–2.4
Married						
Yes	83.7	82.2	1.0	Ref	1.0	Ref
No	16.3	17.8	1.1	0.6–1.9	2.4	1.1–5.1
Mother's county of residence						
King	58.8	51.0	1.0	Ref	1.0	Ref
Snohomish	8.2	16.9	2.4	1.1–5.0	3.3	1.4–7.4
Pierce	13.4	6.8	0.6	0.3–1.1	1.0	0.5–2.1
Other western Washington	11.3	15.8	1.6	0.8–3.1	3.8	1.5–9.4
Other eastern Washington	8.2	9.5	1.3	0.6–2.8	2.2	0.9–5.4
Mother's parity						
0	40.6	53.4	1.7	1.0–3.1	2.2	1.1–4.2
1	39.6	31.7	1.1	0.6–1.9	1.4	0.7–2.7
≥2	19.8	14.9	1.0	Ref	1.0	Ref
Mother smoked in pregnancy						
No	76.1	88.4	1.0	Ref	1.0	Ref
Yes	23.9	11.6	0.4	0.3–0.7	0.4	0.2–0.7
Trimester prenatal care began§‖						
First	83.7	90.9	1.0	Ref	1.0	Ref
Second	12.0	7.2	0.6	0.3–1.1	0.5	0.2–1.1
Third/No care	4.3	2.0	0.4	0.1–1.2	0.6	0.2–2.3

Note: Ref = Referent group.
* Adjusted estimate accounts for all other variables in the table and multiple birth.
† Variable missing 3% to 6%.
‡ Inflation-adjusted using the Consumer Price Index with 1991 as the referent year.
§ Variable missing 9.9%.
‖ Adjusted estimate also accounts for presence of a congenital anomaly.

CI, 0.1 to 0.4); a birth injury (OR, 0.5; 95% CI, 0.3 to 1.1); and an infant birth-hospital stay of ≥4 days (OR, 0.01; 95% CI, 0.02 to 0.06) compared with case infants born from 1987 to 1990.

Pertaining to shifts in maternal demographic and socioeconomic characteristics, we found that as compared with case infants born from 1987 to 1990, infants with plagiocephaly born from 1991 to 2002 were more apt to have a mother ≥35 years of age at the time of the infant's birth (OR, 3.2; 95% CI, 1.4 to 7.3); to have mothers who were married (OR, 2.4; 95% CI, 1.1 to 5.1); and to have been their mothers' first live birth (OR, 2.2; 95% CI, 1.1 to 4.2). Fewer mothers of case infants born from 1991 to 2002 smoked at the time of the birth than mothers of those born from 1987 to 1990 (OR, 0.4; 95% CI, 0.2 to 0.7). Case infants born in the later years were more commonly referred from northern adjacent Snohomish (OR, 3.3; 95% CI, 1.4 to 7.4) and other western Washington counties (OR, 3.8; 95% CI, 1.5 to 9.4). We noted weaker associations between year of birth and race, family income, and the trimester that prenatal care began; these weaker estimates were also highly variable (Table 2).

Concerning trends in delivery and pregnancy characteristics, mothers who delivered case infants from 1991 to 2002 were more likely than mothers who delivered case infants from 1987 to 1990 to have had a cesarean delivery (OR, 2.0; 95% CI, 1.0 to 4.0) and to have been diagnosed with oligohydramnios during pregnancy (OR, 5.1; 95% CI, 0.7 to 37.3). Case mothers delivering in the later time period were less likely than those who delivered earlier to have had a forceps-assisted delivery (OR, 0.2; 95% CI, 0.1 to 0.4) or cephalopelvic disproportion (OR, 0.4; 95% CI, 0.2 to 0.8). Associations between year of birth and vacuum-assisted delivery, a breech or mal-

TABLE 3 Maternal Delivery Characteristics Among Cases Diagnosed With Plagiocephaly at Seattle Children's Hospital Between 1987 and 2002 by Select Year Groups

	1987–1990 (n = 98) %	*1992–2002 (n = 2675) %*	*Unadjusted OR*	*Unadjusted 95% CI*	*Adjusted OR*	*Adjusted 95% CI*
Forceps-assisted delivery*†‡						
No	80.9	93.8	1.0	Ref	1.0	Ref
Yes	19.1	6.2	0.3	0.2–0.5	0.2	0.1–0.4
Vacuum-assisted delivery*†‡						
No	87.2	88.6	1.0	Ref	1.0	Ref
Yes	12.8	11.4	0.9	0.5–1.6	0.8	0.4–1.5
Cesarean delivery*†‡						
No	81.9	73.8	1.0	Ref	1.0	Ref
Yes	18.1	26.2	1.6	0.9–2.7	2.0	1.0–4.0
Breech/malpresentation*						
No	88.3	86.4	1.0	Ref	1.0	Ref
Yes	11.7	13.6	1.2	0.6–2.3	0.7	0.4–1.4
Oligohydramnios§						
No	98.9	94.4	1.0	Ref	1.0	Ref
Yes	1.1	5.6	5.6	0.8–40.2	5.1	0.7–37.3
Cephalopelvic disproportion*						
No	92.6	95.7	1.0	Ref	1.0	Ref
Yes	7.4	4.3	0.6	0.3–1.2	0.4	0.2–0.8
Prolonged labor*‡						
No	91.5	93.8	1.0	Ref	1.0	Ref
Yes	8.5	6.3	0.7	0.3–1.5	0.6	0.3–1.4
Obstructed labor*‡						
No	94.7	95.2	1.0	Ref	1.0	Ref
Yes	5.3	4.8	0.9	0.4–2.2	1.6	0.5–5.0

Note: Ref = Referent group.
* Adjusted estimate accounts for maternal age, race and parity, and infant gender, birth weight, gestational age, multiple birth, and congenital anomaly.
† Adjusted estimate accounts for prolonged and obstructed labor.
‡ Adjusted estimate accounts for cephalopelvic disproportion and breech/malpresentation delivery.
§ Adjusted estimate accounts for maternal race and infant gestational age, birth weight, and congenital anomaly.

presentation, prolonged labor, and obstructed labor were not strong or were statistically unstable (Table 3).

Discussion

We observed positive trends in both supine sleep positioning and plagiocephaly prevalence that continued through 2002, although the trend began to level off around 2000 (Argenta et al., 1996; Kane et al., 1996b; Turk et al., 1996). Previous studies have demonstrated similar time trends in the relation between sleep position and plagiocephaly but included years only through 1996, just a short while after the AAP issued their initial 1992 sleep-positioning recommendations (Argenta et al., 1996; Kane et al., 1996b; Turk et al., 1996). The concomitant increase in cases of plagiocephaly and in the supine sleep-positioning prevalence is consistent with the evidence that these two factors are related, a relationship supported by case-control studies (Kane et al., 1996b; Hutchison et al., 2003). In this analysis, the interpretation of the relationship depicted in Figure 1 is limited because this is an ecologic comparison, with a measure of sleep position based on national prevalence estimates and not on the cases in the current study (National Infant Sleep Position Study, 2004). There are few, however, if any other factors that could have changed rapidly enough to account for this dramatic increase. Furthermore, we found that case infants born from 1991 to 2002 were 27.5 times as likely as those born from 1987 through 1990 to have occipital-only flattening, which is consistent with the observed shift to supine sleep positioning. Finally, this increase cannot be attributed to an increase in the number of live births in Washington State, which decreased by 1.5% during the period of our study (DOH, 2006a).

We found that several hypothesized risk factors for plagiocephaly were more common among cases occurring after the 1992 AAP recommendations as compared with cases diagnosed prior to 1992. Our adjusted analyses suggest that among plagiocephaly cases, birth after 1990 was associated positively with being a multiple and inversely associated with having a gestational age of ≥42 weeks, a birth injury, or a birth hospital stay of ≥4 days. Mothers whose case infants were born after 1990 were more likely to be ≥35 years of age, to have had no previous live births, or to have had oligohydramnios; and they were less likely to have smoked and to have a forceps-assisted delivery relative to mothers whose case infants were born from 1987 through 1990.

There are at least two possible mechanisms that could have caused the post–sleep recommendation changes in the distribution of birth and maternal characteristics among plagiocephaly cases. First, it is possible that as we had proposed, supine sleeping modifies the association between these risk factors

and plagiocephaly. For example, it may be that being a first-born infant does not influence the risk of plagiocephaly unless the infant is placed supine. That is, the increase in risk caused by being first born could differ depending on the infant's sleep position. In the prone position, facial features and contours may minimize the amount of consistent pressure on the same part of the frontal skull, thereby reducing the risk of plagiocephaly. In this manner, prone sleeping may protect otherwise vulnerable infants such as first-born infants, who may have a predisposing flat spot or a subtle neck injury.

Even if the prevalence of such characteristics stayed relatively constant in the general population over the time period of interest, a causal interaction with supine sleeping combined with the rapid increase in the rate of supine sleeping would make such characteristics appear to increase in prevalence in a sample restricted to cases, which is what we studied (Khoury et al., 1996). Although we had no direct measure of sleep positioning, such possible effect modification between supine sleeping and other putative plagiocephaly risk factors can be assessed indirectly by the magnitude of the temporal association with these other risk factors. The strength of this interpretation depends, however, on the strength of the relationship between the likelihood of supine sleeping and year of diagnosis—which serves in this analysis as a proxy for supine sleeping. Based on annual sleep-positioning prevalence estimates from the National Infant Sleep Study, year of birth strongly correlates with sleep position ($r = .95$) and therefore provides a good proxy for sleep positioning (National Infant Sleep Position Study, 2004). This interpretation also assumes that trending risk factors are unassociated with the likelihood of supine sleeping in the general population (Khoury et al., 1996; Albert et al., 2001). It is questionable whether this assumption holds true. It may be that the probability of sleeping supine is associated with some but not all of the risk factors we examined. For example, it is implausible that oligohydramnios or forceps-assisted delivery is related to how an infant is placed to sleep. However, infants of mothers with high educational attainment and a high family income are more likely to be placed to sleep in the supine position (Corwin et al., 2003). In the latter scenario, the observed increase in these risk factors may be the result of their relation to the supine sleep position. Other potential infant birth and maternal delivery risk factors such as gestational age, birth injury, and breech delivery are unlikely to be related strongly to sleep position. Thus, aside from associations in demographic factors such as maternal race, age, education, and county of residence, this explanation would be unlikely to account for the observed trends.

By using year of birth as a proxy, we have misclassified some infants with regard to their actual sleep positioning. The effect of this misclassification is that our associations may be biased towards the null. To explore this possible bias, we performed sensitivity analyses by reanalyzing the data, limiting the comparison to those born from 1987 to 1991 and those born from 1995 to 2002, excluding the years with the most transition and also likely to have the most misclassification regarding sleep positioning. Our observed estimates for most risk factors of interest were similar (data not shown), suggesting that misclassification during these years did not greatly affect our findings.

A second possible explanation for the observed time trends in this case-only study is that the associations are noncausal and that the prevalence of these apparent risk factors has altered in the general population over the same time period. This may be particularly true for factors related to shifts in obstetric care and fertility practices. Indeed, over the years studied, there were increases among Washington State live births in cesarean deliveries (from 20.4% to 23.3%); multiple births (from 2.1% to 3.0%); and maternal age of ≥35 years of age at the time of the infant's birth (from 8.2% to 14.6%), whereas the proportion of forceps-assisted deliveries decreased (from 7.3% to 1.3%) between 1987 and 2002 (Critchlow and O'Brien, personal communication, 2003). Nationally, the proportion of infants whose birth hospitalization stay was 1 day or less increased from 19.9% to 21.2% between 1990 and 2000 (Kozak et al., 2002). However, it is unlikely that many of the associations we observed are entirely due to these temporal shifts. For example, multiple births in the current case group increased from 3.1% from 1987 through 1990 to 9.9% from 1991 to 2002. This tripling represents a much greater increase than that observed among all live births. The prevalence of births to mothers who were ≥35 years of age increased from 9.2% to 22.7% between 1987 and 2002 among cases but only increased 6.4% in this same time period in the general population (DOH, 2006a). Likewise, among cases the proportion of infants whose birth hospitalization was 1 day or less increased 34.1% but only 1.2% in the general population (Kozak et al., 2002). Similar patterns were observed for cesarean delivery, forceps-assisted deliveries, and oligohydramnios (Critchlow and O'Brien, personal communication, 2003). That these temporal changes are so much greater among a case group than in the general population suggests that these associations are not entirely due to a noncausal increase in the prevalence of these factors over time. However, the extent to which the observed association in our study would remain after accounting for these changes is not possible to determine conclusively without a control group.

One potential limitation of the study reported here is that we ascertained cases only through one hospital, and over the time period of study there may have been changes in referral patterns that could have biased the results (Kane et al., 1996b; Turk et al., 1996). Prior to the AAP recommendations, referrals for plagiocephaly were standard practice but were also extremely uncommon, perhaps because few primary care providers were knowledgeable and experienced at diagnosing this condition. The rarity of the condition meant that pediatricians were likely to refer infants with skull asymmetry to specialists at our institution, due to the importance of differentiating plagiocephaly from craniosynostosis, a much more health-threatening condition. Thus, in the early years of our study, it is likely that most cases of plagiocephaly in Washington State

identified by primary care providers were referred to Children's Craniofacial Center for a confirmatory evaluation.

After the AAP delivered its recommendations, however, and primary care providers became more knowledgeable and able to diagnose plagiocephaly, case ascertainment through Children's Craniofacial Center likely became less complete. Thus, the temporal trends we observed may represent referral biases rather than causal associations. By 1996, many primary care providers were likely proficient in diagnosing plagiocephaly. However, certain parents may have requested referrals to Children's Craniofacial Center (the only location able to provide helmet therapy treatment for plagiocephaly during most of the period of the study) to obtain helmet therapy, a treatment often not covered by insurance. To the extent that infant and maternal characteristics of these parent-driven referrals are different from all plagiocephaly cases born in the same year, this may have caused shifts in case characteristics. This may be particularly true for demographic characteristics such as maternal race, income, and first-born children. Such potential biases are not borne out by the data, however, because corresponding demographic factors such as white maternal race and high family income did not vary to an important degree over the study period. For example, the proportion of case infants born to mothers of white race from 1987 to 2002 declined 6.3% (from 90.8% to 84.5%), whereas a similar 7.0% decrease (from 88.2% to 81.2%) occurred among all live births to white mothers in Washington State between 1987 and 2002 (DOH, 2006a). The proportion of infants born into high income families with an annual income of ≥$60,000 (adjusted for inflation) increased only slightly, from 9.0% (1987 through 1990) to 10.1% (1991 to 2002). There was an increase in the proportion of first-born case infants, which increased from 40.6% to 53.4% between the periods 1987 through 1990 and 1991 to 2002.

The validity of the current results also may be limited by incomplete ascertainment of all cases eligible for our study. Plagiocephaly is typically an outpatient diagnosis. Cases not included in the Craniofacial Center registry may have been missed. In particular, we were unable to identify outpatients born before 1993 who were inadvertently missed by the registry. From 1993 onward, we were able to use an electronic Children's Hospital outpatient database to identify such cases. Using this electronic source, we identified 208 (7.3%) cases not initially included in the registry. Among a subgroup excluding these 208 cases, the crude and adjusted estimates were similar to those for all cases, suggesting that cases missed before 1993 would be unlikely to meaningfully affect the study findings. Though the number of cases in the study population born from 1987 through 1990 (n = 98) was much smaller than in the later time period (n = 2675), most analyses had sufficient numbers even for analyses adjusted for multiple covariates. A further study limitation was that the charts did not consistently report the type, location, or severity of plagiocephaly and contained little information concerning other clinical aspects of the condition, such as ear misalignment or degree of facial asymmetry. Consequently, the data on the clinical characteristics of plagiocephaly were of limited use in our analysis. Other studies have examined clinical characteristics of plagiocephaly in detail (Pople et al., 1996; Mulliken et al., 1999; Littlefield et al., 2004; Graham et al., 2005).

Finally, there are reported inadequacies in the quality and completeness of birth certificate data (Hexter et al., 1990; Hexter and Harris, 1991; Snell et al., 1992; Watkins et al., 1996). In Washington State, this limitation is minimized by linking the birth-hospital discharge data for the mother-infant pair to the birth certificate data. A validity study comparing birth certificate linked to mother-infant birth-hospital discharge data with hospital charts (the gold standard) in Washington State demonstrated that the birth certificate-hospital discharge linked file improved reporting compared with either source alone (Parrish et al., 1993). Underreporting remains a common problem (Parrish et al., 1993), yet for most infant and birth characteristics it is unlikely to have differed systematically over time. Incomplete reporting of this type, if present, could have biased the results toward finding no association. That is, the true temporal associations could have been larger than those we observed.

Conclusion

This study demonstrates time trends in select maternal and infant risk factors for plagiocephaly. These findings are consistent with the hypothesis that sleep position modifies associations between plagiocephaly and demographic and birth characteristics such as multiple births, older maternal age, and maternal parity. That is, an infant's risk of plagiocephaly may be concomitantly influenced by an infant's sleep position and the presence (or absence) of mother's parity, birth injury, and many of the other risk factors discussed here. Alternatively, because we did not include a control group, these temporal shifts simply may reflect changes over time in the prevalence of these factors, though the changes are much larger than expected from temporal trends alone. Although the results, therefore, must be considered preliminary, the increased power of the case-only study (relative to a case-control approach) is a useful tool for screening for potential effect modification. To sort out the possible explanations for the observed time trends, we are conducting a case-control study to further examine risk factors for plagiocephaly.

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