Article

Risk Factors for Deformational Plagiocephaly at Birth and at 7 Weeks of Age: A Prospective Cohort Study

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Abstract

OBJECTIVE. The purpose of this work was to identify risk factors for deformational plagiocephaly within 48 hours of birth and at 7 weeks of age.

PATIENTS AND METHODS. This was a prospective cohort study in which 380 healthy neonates born at term in Bernhoven Hospital in Veghel were followed at birth and at 7 weeks of age. Data regarding obstetrics, sociodemographics, asymmetry of the skull, anthropometrics, motor development, positioning, and care factors related to potentially provoking deformational plagiocephaly were gathered, with special interest for putative risk factors. The main outcome measure at birth and at 7 weeks of age was deformational plagiocephaly, assessed using the plagiocephalometry parameter oblique diameter difference index, a ratio variable, calculated as the longest divided by the shortest oblique diameter of the skull $\times$ 100%. A cutoff point of $\geq$104% was used to indicate severe deformational plagiocephaly.

RESULTS. Only in 9 of 23 children who presented deformational plagiocephaly at birth was deformational plagiocephaly present at follow-up, whereas in 75 other children, deformational plagiocephaly developed between birth and follow-up. At birth, 3 of 14 putative risk factors were associated with severe flattening of the skull: gender, birth rank, and brachycephaly. At 7 weeks of age, 8 of 28 putative risk factors were associated with severe flattening: gender, birth rank, head position when sleeping, position on chest of drawers, method of feeding, positioning during bottle-feeding, and tummy time when awake. Early achievement of motor milestones was a protective factor for developing deformational plagiocephaly. Deformational plagiocephaly at birth was not a predictor for deformational plagiocephaly at 7 weeks of age. There was no significant relation between supine sleeping and deformational plagiocephaly.

CONCLUSIONS. Three determinants were associated with an increased risk of deformational plagiocephaly at birth: male gender, first-born birth rank, and brachycephaly. Eight factors were associated with an increased risk of deformational plagiocephaly at 7 weeks of age: male gender, first-born birth rank, positional preference when sleeping, head to the same side on chest of drawers, only bottle feeding, positioning to the same side during bottle feeding, tummy time when awake <3 times per day, and slow achievement of motor milestones. This study supports the hypothesis that specific nursing habits, as well as motor development and positional preference, are primarily associated with the development of deformational plagiocephaly. Earlier achievement of motor milestones probably protects the child from developing deformational plagiocephaly. Implementation of practices based on this new evidence of preventing and diminishing deformational plagiocephaly in child health care centers is very important.

Key Words:

asymmetry in infancy

cohort study

deformational plagiocephaly

motor development

positional preference

physical therapy

Deformational plagiocephaly (DP) refers to a condition in which the infant's head and possibly the face are deformed as a result of prenatal and/or postnatal external molding forces to the malleable and growing cranium.[1-3] This often leads to an asymmetric cranium, ear misalignment, and facial asymmetry.[3-5] The prevalence of DP in children below the age of 6 months of age varies between 13% at birth,[6] 16% at 6 weeks of age,[7] and 19.7% at 4 months of age.[7] The prevalence and development of DP within the first 6 weeks after birth and the associations between DP at birth and DP later on have never been explored in detail. In the literature, DP is attributed to a restrictive intrauterine

environment, premature birth, assisted vaginal delivery, prolonged labor, unusual birth position, multiple birth, and primiparity.[3,6,8,9] Male gender, nonvarying head position when asleep, supine (sleeping) position, positional preference, developmental delay, and lower activity level have all been described as risk factors,[7,10-12] whereas placing the child in the prone position when awake for 5 minutes a day seems to be a protective factor.[12,13] Many clinicians consider DP to be a minor and purely cosmetic condition.[14] Although an association has been found between DP and auditory processing disorders,[15] mandibular asymmetry,[16] and strabismus,[17] causality has never been established.[12,14,18-21] However, the head molding deformation has the potential for a negative physical and psychosocial effects.[17] Parents fear that unattractive facial characteristics will induce adverse effects, such as teasing, poor self-perception, and teacher bias.[14,18]

Epidemiological studies have shown that prone and side sleeping are major risks for sudden infant death syndrome.[22-24] Therefore, the American Academy of Pediatrics (AAP) stated in the 1992 Task Force on Infant Positioning and Sudden Infant Death Syndrome that healthy term infants should be positioned on their side or back to sleep.[25,26] This statement was followed by the start of the "Back to Sleep" campaign in the United States and in many countries over the world. In the decade that followed, a dramatic rise in the prevalence of positional preference was observed.

In the Netherlands in 1995 and 2004, positional preference prevalences of 8.2%[27] and 12.2%,[28] respectively, were reported in children <6 months. The boy/girl ratio of positional preference was 3:2, whereas first-born children, premature children, and children with breech presentation at birth proved to have a higher risk for developing positional preference.[27]

The supine sleeping position of the child and a strong preference in offering the feeding bottle always from the right or the left side were positively associated with positional preference.[27] Concurrent with the increase in supine sleeping, consistent with the AAP recommendations, a rise in the prevalence of DP has been observed. This strong association suggests a causal relationship between supine sleeping and the development of DP.[29]

Conservative strategies to prevent and to intervene in positional preference and DP are parental counseling, counterpositioning, physical therapy, and orthotic devices.[6,13,18,30] Studies on the effectiveness of these interventions are of moderate-to-poor methodologic quality and randomized, controlled trials were not found.[18,30] The purpose of the present study was to document the prevalence of positional preference and DP at birth, to study prevalence changes over time until the age of 7 weeks, and to identify risk factors that influence the occurrence and possible progression of DP.

PATIENTS AND METHODS
Patients

Between December 2004 and September 2005, 400 healthy consecutively born neonates were included in the study within 48 hours after birth at the nursery of General District Hospital Bernhoven in Veghel. To be included, children had to have been born after >36 weeks' gestation and

had to show no dimorphisms or syndromes. Children with congenital muscular torticollis were excluded from this study.

Baseline Assessment

The following anamnestic data were collected within 48 hours of birth: (1) general characteristics of the child including gender, birth rank (first born or later born), parents' age, parents' educational level, family structure, and principal carer of the infant; and (2) obstetric data including gestation, pregnancy rank, presentation at delivery, mode of delivery (vaginal, vacuum-assisted, or cesarean section), length of labor, multiple birth, Apgar scores at 5 and 10 minutes, birth weight, and birth head circumference.

Through physical examination by the principal investigator, the following aspects were assessed: (1) posture and active movements of the child, with special attention for possible asymmetries; (2) passive range of motion (ROM) of the cervical spine in the supine position; (3) head circumference (centimeters); and (4) transversal shape of the skull, measured by plagiocephalometry.[31]

Assessment at Follow-up

At 7 weeks of age, the following anamnestic data were collected in 380 infants: (1) specific characteristics of nursing habits: the method of feeding (breast, bottle, or a combination), the position of the infant when being bottle fed (alternate positions on left/right arm or the child symmetrical in front of the carer), positioning on a chest of drawers (with the head alternately at the right or the left side, always at the same side, or with the infant's feet pointing toward the carer), usage of an infant chair (minutes per day), sleeping position during the day and night (supine, prone, or side), position of the head while asleep (most of the time turned to the same side or spontaneously altered), positioning of the child while awake (mostly supine, prone, or laying on his or her side), the age (in weeks) of the child when put in the prone and in the side position for the first time, frequency (per day) and duration (in minutes) of prone and side positioning when awake; and (2) whether the parents had observed a positional preference of the head when the infant was awake (and, if so, to which direction) and their opinion about the shape of the head, face, and posture (symmetry).

A physical examination was performed by a member of a team of 12 well-trained pediatric physiotherapists. The environmental circumstances (temperature, light, and positioning) during the assessments were the same for all of the children. In addition to the aspects that were measured at the baseline, the following aspects were assessed: (1) the presence of positional preference[27]; (2) qualitative motor development using the Alberta Infant Motor Scale (AIMS)[32,33]; and (3) quantitative motor development using the Bayley Scales of Infant Development, Second Edition (BSID-II).[34]

Measurement Instruments

The ROM of the cervical spine was assessed in the supine position by promoting gentle passive movements. At birth, measuring of the maximum ROM was avoided because of the vulnerable cervical structures; at the age of 7 weeks we assured to measure the maximum ROM. The cervical ROM was defined as normal when bilateral rotation of 90°, lateral flexion of 30°, and flexion and

extension of 45° was possible.[35] At the start of the study, we measured ROM by 360° goniometry in 10 infants, and it was decided that when the chin was above the acromion (rotation), the ear touched the shoulder (lateral flexion), the chin touched the sternum (flexion), and the occiput touched the thoracic trunk (extension), the above-mentioned degrees were reached. For logistic reasons, goniometry was not used on every newborn.

Plagiocephalometry was described recently as a reproducible, valid, noninvasive, easily applicable method to measure the (a)symmetry of the skull. Plagiocephalometry is performed with a strip of thermoplastic material, which is positioned around the infant's head at the widest transverse circumference. Landmarks (both ears and nose) are marked perpendicular on the ring in a standardized manner. Plagiocephalometry measures the relations between transversal shape of the skull related to the exact position of the ears and nose and thereby the location and amount of flattening of the skull (Fig 1 A and B). Cutoff points, based on clinical and psychometrical characteristics, to differentiate between abnormal and normal shape of the skull have been defined.[31] In this study, the oblique diameter difference index (ODDI) parameter (the ratio between oblique diameter left [ODL] and oblique diameter right [ODR] calculated as longest/shortest oblique diameter × 100%) served as an indicator of the presence (ODDI ≥104%) and severity of DP and was used to follow-up asymmetry of the skull over time. Cranial proportional index (CPI) is another plagiocephalometry parameter indicating severity of brachycephaly and is calculated as the ratio sinistra dextra/anterior posterior × 100%. Plagiocephalometry was always performed by the same investigator (Dr van Vlimmeren; Fig 1).[31]
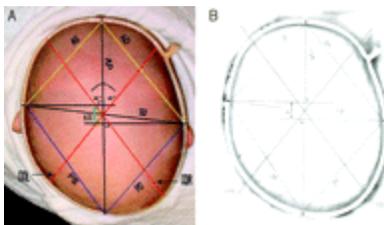
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FIGURE 1

Illustration of plagiocephalometry[31]: asymmetry DP left occipital of the skull (4-month-old boy). A, Photograph of a child with the thermoplastic measuring ring and landmarks. The digitally drawn lines are made to illustrate the agreement with the paper copy and to explain the names of the lines. B, Paper copy of the same ring with drawn and measured lines (ODDI: 109.6%; CPI: 88.1%; ODL: 125; ODR: 137; ED: 12 mm; PDPS: 8 mm). AP indicates anterior posterior; SD, sinistra dextra; ED, ear deviation; and PDPS, PD (posterior dextra) minus PS (posterior sinistra).

The presence of positional preference was assessed, indicating the condition in which the infant, in the supine position, revealed head rotation to either the right or the left side for ~75% of the time of

observation (minimal time of observation: 15 minutes), without active rotation of the head over the full range of 180°.[27]

The AIMS is a highly reliable and valid, norm-referenced, performance-based observational measure that examines the spontaneous qualitative gross motor movement repertoire of infants (until 18 months of age) in supine, prone, sitting, and standing positions.[33,36] The motor scale described in the BSID-II is a highly reliable, valid, and norm-referenced method of assessing the motor and mental abilities of children up to the age of 42 months.[34]

The medical ethics committees of the Wilhelmina Children's Hospital (University Medical Center Utrecht) and the Bernhoven Hospital approved this study. Informed consent was obtained from al of the parents.

Statistical Analysis

Means (SDs), medians (interquartile range), or proportions were calculated for the relevant variables. The relation between risk factors and deformity was analyzed by means of cross-tabulation, linear regression, and logistic regression. In the univariate analyses, putative risk factors with a $P < .15$ were selected[37] for inclusion in the multivariate models, and their effect for each other was estimated. In the multivariate linear regression analysis, the effect of risk factors on the dependent factor ODDI (continuous) was assessed. The regression coefficient β is interpreted as an increase of the outcome variable, when the determinant increases by 1 unit. In the logistic models, severe deformity was defined as ODDI ≥104% (yes or no), and odds ratios (ORs) and their 95% confidence intervals (CIs) were calculated. The AIMS raw score was transferred into a standardized z score (individual score minus the average score divided by the SD).[33] Scaled scores of the BSID-II were transformed into a psychomotor developmental index (PDI; mean: 100; SD: 16; range: 50-150).[34] Motor development and positional preference cannot be considered as risk factors for deformity but should be considered as intermediate factors in the development of severe deformity. The magnitude of their effect was estimated in a separate multivariate model. Statistical analyses were performed using SPSS for Windows 12.0.1 (SPSS Inc, Chicago, IL).

RESULTS
Clinical Characteristics

Twenty (5%) of the 400 children were lost to follow-up because of the child having physical problems (craniosynostosis: $n = 1$; congenital muscular torticollis: $n = 4$), decreased parental motivation ($n = 12$), moving out of the area ($n = 2$), or severe illness of the mother ($n = 1$). Therefore, data of 380 children (boys: 46.8%) could be analyzed at follow-up. The clinical characteristics of the participants are presented in Table 1. The participating children were healthy and were mostly born at term.

Baseline Assessment

The first assessment was performed at an average of 16.9 hours (SD: 8.7) after birth. Of all of the neonates, after 5 minutes, 99% had an Apgar score ≥7 and 100% after 10 minutes.

Passive ROM of the cervical spine was within reference range without asymmetries. The mean of deformity measure ODDI (%) was 101.7% (SD: 1.7%; range: 100.0%–110.9%); the mean of CPI was 78.9% (SD: 3.6%; range: 66.9%–89.2%). Boys' heads were significantly larger (mean head circumference for boys: 35.3; SD: 1.37 cm; girls: 34.3 cm; SD: 1.31 cm; $P < .0001$) and more asymmetrical (mean ODDI: boys: 101.9%; SD: 1.81%; girls: 101.5%; SD: 1.31%; $P = .016$) but less brachycephalic (mean CPI: boys: 78.3%; SD: 3.70%; girls: 79.4%; SD: 3.41%; $P = .003$). Boys' birth weight was significantly higher (mean boys: 3.47 kg; SD: 0.46 kg: girls: 3.27 kg; SD: 0.48 kg; $P < .0001$). DP (ODDI ≥104%) was present in 23 (18 boys and 5 girls) of 380 infants (6.1%). The flat occipital area was located more often on the right side than on the left side (11:9). The prevalence of DP was higher in boys (adjusted OR [aOR]: 5.4; 95% CI: 1.92–15.28), also after adjustment for birth rank (first born: aOR: 2.2; 95% CI: 0.89–5.26) and brachycephaly (CPI: aOR: 1.1; 95% CI: 1.00–1.26). Infant characteristics, sociodemographic factors, obstetric factors, and head circumference were not significantly associated with DP. Passive ROM of the cervical spine was within the reference range and without asymmetries.

Assessment at Follow-up

Positional Preference and DP

The second assessment was performed at a mean age of 7.4 (SD: 0.9) weeks after birth. In 68 of 380 infants, positional preference was observed by the physiotherapist (17.9%; 42 boys and 26 girls). In 41 (60.3%) of the 68 children with positional preference, DP was found (crude OR: 9.5; 95% CI: 5.30–17.01; Table 2). The flat occipital area was located twice as often at the right side as on the left side. Parents reported observing positional preference ~2.5 times as often as was measured by the physical therapist (in 165 children [43.4%]).

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TABLE 2

Determinants of Deformational Plagiocephaly (ODDI ≥104%) at 7 Weeks of Age

Variable	Severe Skull Deformity ($n = $ 84), n (%)	No Severe Skull Deformity ($n = $ 296), n (%)	Univariate OR (95% CI)	P	Multivariate OR (95% CI)
Gender					
Boy	49(58)	129(44)	1.8 (1.11–2.96)	.02	2.0 (1.12–3.41)

Variable	Severe Skull Deformity (n = 84), n (%)	No Severe Skull Deformity (n = 296), n (%)	Univariate OR (95% CI)	P	Multivariate OR (95% CI)
Girl	35(42)	167(56)	1.0		
Educational level of mother[a]					
Low	30(35)	76(26)	1.8 (0.93–3.43)	.08	
Medium	35(42)	131(45)	1.2 (0.65–2.25)	.6	
High	19(23)	86(29)	1.0		
Educational level of father[a]					
Low	32(39)	81(28)	1.7 (0.87–3.31)	.1	
Medium	34(41)	134(47)	1.1 (0.57–2.08)	.8	
High	17(20)	73(25)	1.0		
Pregnancy rank					
First	41(49)	101(34)	1.8 (1.13–3.01)	.01	
Second or more	43(51)	195(66)	1.0		
Birth rank					
First born	48(57)	126(43)	1.8 (1.10–2.94)	.02	2.4 (1.36–4.22)
Later born	36(43)	170(57)	1.0		
Presentation at birth					

Variable	Severe Skull Deformity (n = 84), n (%)	No Severe Skull Deformity (n = 296), n (%)	Univariate OR (95% CI)	P	Multivariate OR (95% CI)
Unusual	13(15)	37(13)	1.3 (0.65–2.54)	.5	
Breech (n = 29)					
Occipito–posterior (n = 14)					
Sinciput (n = 7)					
Usual	71(85)	259(87)	1.0		
Occipito–anterior (n = 330) Breech presentation					
Yes	4(5)	25(8)	0.5 (0.18–0.60)	.3	
No	80(95)	271(92)	1.0		
Delivery Cesarean section	18(21)	72(24)	0.9 (0.50–1.67)	.8	
Vacuum assisted	13(16)	30(10)	1.6 (0.77–3.25)	.2	
Normal vaginal	53(63)	194(66)	1.0		
ODDI at birth ≥104%					
Yes	9(11)	14(5)	2.4	.04	

Variable	Severe Skull Deformity (n = 84), n (%)	No Severe Skull Deformity (n = 296), n (%)	Univariate OR (95% CI)	P	Multivariate OR (95% CI)
			(1.01–5.80)		
No	75(89)	282(95)	1.0		
Sleeping supine after 2 wk of age					
Yes	68(81)	217(73)	1.6 (0.85–2.83)	.2	
No	16(19)	79(27)	1.0		
Head position when sleeping					
Turned to same side	34(41)	26(9)	7.1 (3.90–12.78)	<.0001	7.5 (3.94–14.37)
Alternated or symmetrical	50(59)	270(91)	1.0		
Position on chest of drawers					
Head to same side	38(45)	94(32)	1.8 (1.08–2.91)	.02	1.8 (1.00–3.09)
Alternated or symmetrical	46(55)	202(68)	1.0		
Kind of feeding					
Only bottle feeding	34(40)	89(30)	1.6 (0.99–2.61)	.07	1.8 (0.99–3.29)
Not only bottle feeding	50(60)	207(70)	1.0		
Positioning bottle feeding					
Same side	35(42)	81(27)	1.9	.01	1.8 (1.01–

Variable	Severe Skull Deformity (n = 84), n (%)	No Severe Skull Deformity (n = 296), n (%)	Univariate OR (95% CI)	P	Multivariate OR (95% CI)
			(1.15–3.14)		3.30)
Alternated or symmetrical	49(58)	215(73)	1.0		
First tummy time[b]					
≥3 wk of age	32(38)	120(41)	0.9 (0.55–1.49)	.7	
<3 wk of age	52(62)	176(59)	1.0		
Tummy time when awake					
<3 times per d	78(93)	245(83)	2.7 (1.12–6.55)	.02	2.4 (0.90–6.20)
≥3 times per d	6(7)	51(17)	1.0		
Tummy time					
≤5 min/d	51(61)	187(63)	0.9 (0.55–0.48)	.7	
>5 min/d	33(39)	109(37)	1.0		
Side laying position[c]					
<1 time per d	60(71)	221(75)	0.9 (0.50–0.46)	.6	
≥1 time per d	24(29)	75(25)	1.0		
Side laying position					
≤5 min/d	14 (17)	57 (19)	0.8 (0.44–	.6	

Variable	Severe Skull Deformity ($n = $ 84), n (%)	No Severe Skull Deformity ($n = $ 296), n (%)	Univariate OR (95% CI)	P	Multivariate OR (95% CI)
			1.60)		
>5 min/d	70 (83)	239 (81)	1.0		
Usage of a baby chair					
≥30 min/d	49 (58)	170 (57)	1.0 (0.64–1.70)	.9	
<30 min/d	35 (42)	126 (43)	1.0		
Principal carer in daytime					
Also others than mother	67 (80)	222 (75)	1.3 (0.77–2.38)	.4	
Mother	17 (20)	74 (25)	1.0		
Positional preference when awake					
Yes	41 (49)	27 (9)	9.5 (5.30–17.01)	<.0001	
No	43 (51)	269 (91)	1.0		
Motor development: AIMS z score					
Less than −1 SD	16 (19)	40 (14)	1.5 (0.80–0.85)	.2	
Between +2 SD and −1 SD	68 (81)	256 (86)	1.0		
Motor development: BSID–PDI					

Variable	Severe Skull Deformity (n = 84), n (%)	No Severe Skull Deformity (n = 296), n (%)	Univariate OR (95% CI)	P	Multivariate OR (95% CI)
Less than −1 SD	7 (8)	12 (4)	2.2 (0.82–0.66)	.1	
Between +2 SD and −1 SD	77 (92)	284 (96)	1.0		

↵a Low education level was defined as lower technical and vocational training and lower general secondary education. Medium education level was defined as intermediate vocational training and advanced secondary education. High educational level was defined as higher vocational education (college education) and university.

↵b Infant is placed prone when awake and under supervision.

↵c Infant is positioned on side, when awake and under supervision.

TABLE 2

Determinants of Deformational Plagiocephaly (ODDI ≥104%) at 7 Weeks of Age

Passive ROM, Alignment, and Head Circumference

The assessment at 7 weeks of age revealed asymmetrical active movements of the trunk in 13 children (3.4%). This was significantly associated with DP (crude OR: 6.1; 95% CI: 1.95–19.26). Passive ROM of the cervical spine illustrated normal outcomes without asymmetries. Mean ROM at 7 weeks of age were: bilateral rotation at 97° (SD: 5.1°); lateral flexion at 45° (SD: 3.1°); flexion at 44° (SD: 2.2°); and extension at 45° (SD: 2.5°). All of the children had a symmetrical alignment. Head circumference at 7 weeks of age (mean: 38.3 cm; SD: 1.4 cm) was not associated with DP.

Motor Development

The AIMS showed a mean z score of −0.26 (SD: 0.72; range: −2.81 to 2.91). The motor scale of the BSID-II showed a mean PDI score of 101.80 (SD: 10.51; range: 68–134). Having an AIMS z score of more than −1 SD was associated with positional preference (aOR: 2.1; 95% CI: 1.10–4.13), even after adjustment for tummy time less than once per day (aOR: 2.1; 95% CI: 1.20–3.83).

Severity of DP

The prevalence of DP (ODDI ≥104%), increased from 6.1% at birth to 22.1% at 7 weeks (49 boys and 35 girls). Only in 9 of 23 children who presented DP at birth was DP present at follow-up, whereas in 75 other children, DP developed after the first assessment.

The mean of the ODDI at 7 weeks of age was 102.6% (SD: 2.3%; range: 100.0%–113.0%). The mean of CPI was 79.7% (SD: 4.6%; range: 68.0%–94.4%).

Risk Factors at 7 Weeks of Age

Significantly more boys than girls had DP (crude OR: 1.8; 95% CI: 1.11–2.96), and more children born after a first pregnancy (crude OR: 1.8; 95% CI: 1.13–3.01) and a first delivery (crude OR: 1.8; 95% CI: 1.10–2.94). Children with DP were significantly more likely to always have their head turned to the same side when sleeping (crude OR: 7.1; 95% CI: 3.90–12.78), were more often positioned with their head to the same side of the chest of drawers (crude OR: 1.8; 95% CI: 1.08–2.91), were more often only bottle fed (crude OR: 1.6; 95% CI: 0.99–2.61), were more often always bottle fed on the same arm of the carer (crude OR: 1.9; 95% CI: 1.15–3.14), and were more frequently put in the prone position (tummy time) <3 times per day (crude OR: 2.7; 95% CI: 1.12–6.55; Table 2).

Associations were also found with univariate linear regression analysis, confirming the strong influence of the risk factors found by means of cross-tabulation. Univariate linear regression analysis revealed that the presence of DP was significantly associated with gender (boys; β = .5; 95% CI: 0.06–0.10), CPI (β = .1; 95% CI: 0.09–0.18), pregnancy rank (β = .5; 95% CI: 0.06–1.02), birth rank (β = .4; 95% CI: −0.03 to 0.91), motor development BSID–II PDI (β = −.03; 95% CI: −0.05 to −0.01), always being bottle fed on the same arm (β =.7; 95% CI: 0.16–1.17), sleeping in supine position from ≤2 weeks of age (β = .6; 95% CI: 0.05–1.13), head rotation preference in supine sleeping in the first 4 weeks (β = 2.4; 95% CI: 1.80–3.00), tummy time <3 times per day (β =.9; 95% CI: 1.53–0.22), unilateral positioning on chest of drawers (β = .47; 95% CI: −0.02 to 0.96), and positional preference when the child was awake (β = 2.8; 95% CI: 2.23–3.32).

The factors that were significant at the univariate level (except pregnancy rank, because of the strong correlation with birth rank) were all adjusted to identify environmental risk factors. Positional preference was not entered in the model, because it is the result of several of these significant variables. Gender and birth rank in the final multivariate model showed slightly increased ORs (aORs: 2.0 and 2.4, respectively). Positioning variables (head rotation preference in supine sleeping in the first 4 weeks of life, only bottle feeding, always being bottle fed on the same arm, always being positioned in the same way on the chest of drawers, and tummy time <3 times per day) all had almost the same (adjusted) ORs as in the univariate analyses.

DP at birth (ODDI ≥104%), when entered as variable in the model, lost significance, indicating that DP at follow-up was not associated with DP at birth. Otherwise, motor development (measured by the AIMS z scores) entered as a continuous variable in the model demonstrated an inverse, protective effect on DP (aOR: 0.6; 95% CI: 0.43–0.93), illustrating that an increase in the motor development repertoire was associated with a decrease in the prevalence of DP.

No significant differences were found between children with (n = 84) and without (n = 296) DP for obstetric factors (birth presentation, mode of delivery, and length of labor), multiple birth,

sociodemographic factors, head circumference, birth weight, age when put in the prone or side position for the first time, duration of prone or side positioning, usage of an infant chair, principal carer of the child, and BSID-II PDI scores.

Parental Educational Levels

DP was more prevalent when the mother was educated at the lowest level (crude OR: 1.8; 95% CI: 0.93–3.43). The parents of the 9 children with persistent DP had a lower educational level than the parents of the 14 children who recovered ($P = .08$).

Mothers with low educational levels are significantly positioning their infant on the same arm during bottle feeding (crude OR: 1.8; 95% CI: 1.09–2.81), gave more often only bottle feeding (crude OR: 2.0; 95% CI: 1.27–3.14), and gave their infant tummy time for the first time at ≥3 weeks of age (crude OR: 1.7; 95% CI: 1.09–2.68). After adjustment, only bottle-fed aOR was 2.0 (95% CI: 1.22–3.11) and aOR for tummy time for the first time at ≥3 weeks of age was 1.7 (95% CI: 1.06–2.66).

DISCUSSION

In this cohort study, it was demonstrated that DP in early life was primarily caused by postnatal, external factors (positioning and care) and inversely associated with achievement of motor milestones. DP at birth was positively associated with gender (boys), brachycephaly (high CPI), and birth rank (first-born children). DP at birth was not a predictor for DP at 7 weeks of age. Our results support the hypothesis[7,14] that positioning, motor development, positional preference of the child, and DP are associated with DP. The study contributes to the current literature because of its contemporary nature, the large sample size, the longitudinal design, the starting at birth, and the results concerning predictive risk factors.

The prevalence of DP increases dramatically in the first 7 weeks after birth. Male and right occipital preponderance are in accordance with the literature and are more prominent when the infant grows older.[5,6,11,38] We confirm that the right occipital spot of the skull is more likely to become flattened,[5,6,38] and the right/left ratio evolves in the same period from 11:9 to 2:1. It is suggested that the position of the infant in utero is responsible for the right occipital predominance; 85% of the vertex-presented children are laying on the left occipital anterior position.[8,39] Parents had preferences too; bottle feeding was given 4 times as often on the left arm than on the right arm, and parents placed their child also 4 times more often with the head on the left side of the chest of drawers than on the right side (both stimulating right rotation of the head).

DP at birth is significantly associated with gender (boys). The preponderance of boys with DP was explained by the suggestion of the larger male head circumference and more rapidly growing male head, together with less flexibility of the male fetuses.[8,13] This could not be stated in our study. First-born children are more likely to have a flattened head at birth, probably because of the more tightened uterus and vaginal structures.

At 7 weeks of age, gender (boys), birth rank (first-born children), specific nursing habits (bottle feeding, prone positioning, and positioning when bottle fed and on the chest of drawers), side

preference of the head when asleep, infrequent tummy time, and qualitative motor development (AIMS) remained independently associated with DP. Motor development was not significantly associated with DP when using cutoff points such as -1 SD or -2 SD for delay in motor development. Remarkably, an inverse association was found indicating that a higher AIMS z score was a protective factor for the development of DP. A higher z score indicates an earlier achievement of motor milestones, also in the prone position, thereby improving the lifting of the head in the prone position, influencing head balance positively with a possible preventive effect on skull flattening. Infants spent less time on the flat spot of the head, and the muscular imbalance, initiating positional preference,[11] might disappear by training cervical muscles.

No associations were found with obstetric factors (gestation, presentation at delivery, mode of delivery, length of labor, and multiple birth), child factors other than CPI (birth weight, head circumference, and active movements of the extremities), sociodemographic factors, feeding method, and infant chair positioning. The passive ROM of the cervical spine was normal in all of the children at both assessments and did not influence the severity of DP.

Although it is frequently suggested that DP at birth influences the development of DP at a later age,[6,5,9,13,20] we could not confirm this suggestion. Despite the small number of children, we found that the educational levels of the parents of the 9 children with persistent severe DP were significantly lower. This might indicate that handling and positioning of the child is not performed adequately in this subgroup.

Selection bias might be present because we recruited a hospital born population. Because in the Netherlands 30% of women deliver in their homes, our newborn study population shows a relatively high number of assisted deliveries and cesarean sections. However, adjustment of mode of delivery did not substantially influence the results of our study.

Although information about preventing positional preference and DP was not given to parents, participation in this study may have alerted them to be more cautious for developing DP, and they may have been aware of positioning advice to avoid head flattening in their children, even more so for those parents who were informed at birth that their infant's cranium was "deformed." This hypothesis would really drive home the importance of early diagnosis and intervention in prevention of DP.

The first measurements were performed within 48 hours after birth, and it could be argued that the deformity that we found was transient, caused by external obstetric molding forces. Because it is not exactly known how long deformity caused by delivery will persist, the time span termed as "at birth" will always be arbitrary.

We found that, for clinical and practical reasons, the best plagiocephaly indicator was the ODDI. By leaving out other DP indicators, we probably missed some children with specific head flattening: those with symmetrical oblique diameters (low ODDI) but with a severe occipital flattening spot or a remarkable ear deviation. The best choice of a cutoff point of the ODDI could be debatable. However, the analysis of the continuous variables showed the same tendency in risk factors. ODDI $\geq 104\%$ represents a distinct visible asymmetry of the skull. Using this cutoff point, the prevalence of DP

shows much resemblance with the prevalence determined with the slightly higher cutoff point in the heads-up method of Hutchison et al[7]

A classification of severity based on a clinical evaluation looking for all details of deformity that contribute to severity classification could alter the findings of this investigation. But taking all of these factors into account is far to complicated for clinical use and research in clinical situations. Choosing 1 clear indicator of plagiocephalometry as ODDI, which represents asymmetry of the skull in 2 dimensions, is most suitable for such a clinical study. In our experience, parents and observers are focused most on the asymmetry of the skull and less on facial asymmetry. Although the prevalence of DP is much higher than commonly reported in the literature, this incidence is similar to more recently reported findings and strongly suggests that the incidence of DP is far higher than originally reported.

Regarding development of skull flattening, the initial period of life has not been analyzed previously in detail. Assumptions about determinants and risk factors have now been explored in detail for the first time, using a recently developed and reliable measuring method, plagiocephalometry.[31]

Just as in previous DP studies, boys[5,6,11,38] and first-born children[3,6,7,40,41] were more frequently affected; the right occipital skull was always flattened more frequently,[6,13,38] and the important role of positional preference was confirmed.[7,11,27,42] A hypothesis for the higher incidence of DP in first-born children is that their parents have little child rearing experience and can be overwhelmed by the amount of information they get. Once their first-born infant has developed DP, the parents will be more likely with subsequent children to be cognizant of head shape and methods for preventing distortion. It could be argued that the lack of correlation between DP at birth and DP at 7 weeks was related to the fact that the plagiocephalometry measurement device is not sensitive enough to detect these deformities within the first 48 hours of life or that the temporary distortion of the cranium because of the birthing process influences the validity of the plagiocephalometry at this age. Conclusions regarding multiple birth infants should be interpreted carefully, because our selection criteria excluded preterm infants, thereby reducing the number of plural birth infants in this study.

Positioning factors were shown to be very significant, illustrated by our finding that placing the child in the prone position $\geq$3 times a day minimized the risk of developing DP. Also the difference between bottle feeding and breastfeeding is a postnatal positioning issue. In both bottle feeding and chest-of-drawer positioning, even if the child is just a few weeks of age, the child "instinctively" is visually and auditively directing to the parent/carer. Handling attitudes stimulate and facilitate active cervical rotation toward the parent/carer. Because these handling activities are performed frequently throughout the day, it supports our hypothesis that the positioning factors are highly responsible for the development of positional preference.

We did not find evidence for a number of assumptions and suggestions described in other studies on the determinants of DP. In our study, DP could not be associated with a prenatal start of deformity[5,6,9,13,20] or obstetric factors.[3,6] The larger head circumference of boys is frequently suggested to be the reason for the higher prevalence of DP in boys,[6,9,13] but this association was not found in our study.

In establishing the ROM of the cervical spine after birth, we avoided measuring the end ROM because of the vulnerable cervical structures. For logistic reasons, goniometry was not used on every newborn. Only at 7 weeks of age, passive cervical ROM was measured to find the maximum ROM. All of the children revealed normal passive ROM, indicating that problems with the cervical column caused by delivery[7,43] are unlikely to occur.

Although supine sleeping seems to be the primary cause of obtaining skull flattening,[4,9,12,15] in our study the association between supine sleeping and DP was not significant. So, supine sleeping is probably not the main cause of development of DP but only becomes a risk factor when combined with otdevelopment of DP but only becomes a risk factor when combined with other issues, such as delayed motor milestones, positional preference, and/or negative environmental factors.

A final interesting variable is motor development. Different etiologic pathways linking DP with neurodevelopment are hypothesized[14,15] but have not been studied in detail before. There is evidence that prone positioning while awake is positively correlated with AIMS scores.[44] We found an inverse association between motor development and DP, but the direction of the causal pathway is difficult to establish. Parents with low educational levels provide shorter and less frequent tummy time to their children, thereby probably inhibiting motor development and varying positions less, which might stimulate the development of positional preference.

Our study suggests possible mechanisms in causing DP. Positioning might influence developmental delay, resulting in positional preference and, eventually, DP. Varying positions (prone and side laying) of the child from birth onward, when awake and under supervision, in combination with varying head positions when sleeping in the supine position, seems to be the best way of avoiding DP. Parents should be aware of the increased rapidity of deformation of the skull in the first weeks of life. They have to become extensively motivated to alternate nursing positions and certainly change the position of their child when bottle feeding and when nursing on the chest of drawers as soon as the first signals of DP occur. Of course intrinsic factors, such as temperament and activity level of the child, also influence this process[7]; this requires creativity of the parents in stimulating their child. Implementation of practices based on this new evidence of preventing and diminishing DP in child health centers is of utmost importance.

The prevalence of positional preference that we found was 17.9%, whereas 43.4% of the parents thought that they observed a positional preference in their child. It seems that parents overinterpret the AAP recommendations and avoid prone positioning during the daytime.[14] Parents should be provided with adequate information about the mechanisms causing DP and of its likely consequences. We were especially interested in the development of DP in early childhood, because this has not been explored in detail before. Further analyses might provide additional information on determinants.

CONCLUSIONS

Three determinants were associated with an increased risk of DP at birth: male gender, first born, and brachycephaly. Eight factors were associated with an increased risk of DP at 7 weeks of age:

male gender, first born, positional preference when sleeping, head to the same side on chest of drawers, only bottle feeding, positioning to the same side during bottle feeding, tummy time when awake <3 times per day, and slow achievement of motor milestones. The study supports the hypothesis that specific nursing habits, as well as motor development and positional preference, are primarily associated with the development of DP. Earlier achievement of motor milestones probably protects the child from developing DP. Implementation of practices based on this new evidence of preventing and diminishing DP in child health care centers is very important.

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Footnotes

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DP—deformational plagiocephaly · AAP—American Academy of Pediatrics · ROM—range of motion · AIMS—Alberta Infant Motor Scale · BSID-II—Bayley Scales of Infant Development, Second Edition · ODDI—oblique diameter difference index · ODL—oblique diameter left · ODR—oblique diameter right · CPI—cranial proportional index · OR—odds ratio · CI—confidence interval · PDI—psychomotor developmental index · aOR—adjusted odds ratio

her issues, such as delayed motor milestones, positional preference, and/or