Deformational plagiocephaly in normal infants: a systematic review of causes and hypotheses

Freia De Bock,[1,2] Volker Braun,[3] Herbert Renz-Polster[1]

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[1]Mannheim Institute of Public Health, Social and Preventive Medicine, University Medicine Mannheim, Heidelberg University, Mannheim, Germany
[2]Centre for Child Neurology, Frankfurt am., Germany
[3]University Library, University Medicine Mannheim, Medical faculty Mannheim, Heidelberg University, Mannheim, Germany

Correspondence to
Priv Doz Dr Freia De Bock,Mannheim Institute of Public Health, Social and Preventive Medicine, Medical Faculty Mannheim, Heidelberg University, Ludolf-Krehl-Strasse 7-11, Mannheim 68167, Germany;
freia.debock@medma.uni-heidelberg.de

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ABSTRACT

Background Deformational plagiocephaly (DP) is one of the most prevalent abnormal findings in infants and a frequent reason for parents to seek paediatric advice.

Objective To systematically review the literature and identify evidence and hypotheses on the aetiology and determinants of DP in otherwise healthy infants.

Design Systematic keyword search in all major biomedical databases to identify peer-reviewed publications reporting (a) empirical research or (b) hypotheses on the aetiology of DP in healthy, term infants. 3150 studies published between 1985 and 2016 and containing relevant keywords were screened. In a two-pronged approach, results were summarised separately for the body of empirical work (22 studies) and the body of hypotheses (110 articles).

Review findings Only a few empirical studies have examined risk factors in non-selected patient populations on a higher grade methodological level. The most commonly reported risk factors were: male gender, supine sleep position, limited neck rotation or preference in head position, first-born child, lower level of activity and lack of tummy time. Agreement between empirical studies was poor for most exposures, including supine sleep position, tummy time and use of car seats. The articles reporting hypotheses on the aetiology of DP cover a wide field of environmental and biological factors, but include little suggestions as to the potential influence of the everyday care environment of the baby.

Conclusions and relevance The evidence on the aetiology of DP is fragmentary and heterogeneous. In addition, factors possibly relevant to the development of DP have not been appreciated in the scientific discussion.

What is already known on this topic?

- Postnatally acquired posterior deformational plagiocephaly (DP) is one of the most prevalent abnormal paediatric findings and a frequent reason for seeking paediatric advice.
- However, little is known about the factors that lead to DP in the normally developed, healthy child.

What this study adds?

- Influences on head shape development are complex and not fully understood.
- It is unclear if the supine sleep position is an independent risk factor for the development of DP.
- Larger prospective cohort studies in primary care settings could clarify if there may be other risk factors for DP not detected as of yet.

INTRODUCTION

Flattening of the skull by external forces has been termed positional or deformational plagiocephaly (DP). Depending on the severity, DP presents as an abnormal head shape, facial asymmetry, frontal bossing, ear misalignment and asymmetrical orbits.[1] The moulding can occur in utero, during birth, or it may develop postnatally. The latter form of DP is one of the most prevalent abnormal findings in otherwise healthy infants and a frequent reason for seeking paediatric advice.

While there is ample knowledge about risk factors for the development of DP in infants with diseases or developmental delay, much less is known about why DP should develop in a normal child. Risk factors proposed include supine sleep position, bottle propping, lack of tummy time, or frequent use of car seats, swings or bouncy seats.[2–6] However, research has been ambiguous about their significance and relevance. It has been proposed that this ambiguity may reflect an incomplete understanding of how DP develops altogether.[2–6]

To gain a fuller understanding, we systematically reviewed the literature for empirical studies that have explicitly examined determinants of DP (key question 1) and for articles in which novel, as of yet untested, hypotheses on potentially contributing factors of DP have been proposed (key question 2).

METHODS/LITERATURE SEARCH

In November/December 2013 and again in February 2016, we performed two identical systematic keyword searches in all major biomedical databases and study registries, including PubMed, Embase, Web of Science Core Collection, Cochrane Library, LILACS and CINAHL (for details on our search strategy, see online supplement). Peer-reviewed articles in English, Spanish and French published between 1985 and 2016 were eligible to be included.

We defined case series as the minimum quality category for key question 1 and did not restrict study designs for key question 2. After elimination of duplicates, one person (JB) screened all articles by title with regard to the following inclusion criteria: (1) outcome— DP, (2) sample—healthy children and (3) exposure—lifestyle or environmental

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risk factors for DP. Articles with samples of only newborns, children with specific conditions (like cerebral palsy), preterm children only or hospitalised children were excluded, as our interest was on the development of postnatally acquired DP in the normal, otherwise healthy child. Screening of abstracts and, if needed, full texts was performed in duplicate (HR-P and JB). Articles were categorised as containing either original, empirically tested data or hypotheses on the aetiology of DP.

Data extraction and quality rating in the set of n=22 empirical studies were performed in duplicate by two independent investigators (HR-P and JK). For aetiological factors with three or more comparable risk estimates from case–control or cohort studies, I^2 was calculated to assess heterogeneity (see online supplementary table).

The quality rating of the identified 22 empirical studies included both study-level (eg, study design, recruitment) and outcome-level assessment (table 1, column 6) of potential bias. As many well-known quality ascertainment instruments for the assessment of study-level bias (eg, Cochrane,[7] Critical Appraisal Skills Programme (CASP))[8] could not be used for this sample of relatively low-grade evidence studies, we applied the Effective Public Health Practice Project Quality Assessment Tool (table 1, footnote and second row)[9] and a score developed by the Oxford Centre for Evidence-based Medicine (table 1, footnote and last column). In the case of substantial inter-rater differences, discussion between the original duplicate raters (HR-P and JK) and a third experienced researcher (FDB) was carried out until agreement or consensus was reached. The review protocol according to PRISMA statement[10] can be accessed via http://miph.umm.uni-heidelberg.de/miph/cms/upload/pdf/Plagiocepahly_Review_protocol_De_Bock_et_al..pdf.

RESULTS

Of 3150 studies retrieved by the above-mentioned search strategy (see flow chart, figure 1), 2951 were removed as they were duplicates or were identified as irrelevant by title, abstract and full-text screening, leaving 199 articles for detailed full-text screening. In the second step (review of the full texts), 71 more articles had to be excluded based on predefined exclusion criteria, such as being not peer-reviewed articles in the general press, not meeting language criteria or fulfilling content-related exclusion criteria (see figure 1 and Methods section). Another four articles (including one from 1981) were included post hoc by screening references of all relevant articles, leading to 132 relevant peer-reviewed articles containing information on the aetiology of this condition (figure 1). Of these, only 22 studies reported original, empirically tested data. Table 1 summarises these studies in terms of formal characteristics and potential bias (columns 8–14); table 2 reports the risk factors identified using a roster of exposure catergories. An online supplementary table informs about the heterogeneity of these studies.

The remaining 110 publications covered hypothetical discussions and debates on potential aetiological factors of DP. This body of non-empirical work was analysed and summarised in table 3, which reports all potential risk factors discussed *over and above* those found in the empirical studies.

Of the n=22 empirical studies on risk factors of DP (total study population of 27 782 children), 19 studies (with a total study population of 26 860 children) were conducted with highly selected populations. Only three studies were performed using a primary care patient base without significant selection bias. Study designs were all weak but for four studies (see table 1). Most studies (13 of 22; 59%) were retrospective chart reviews. Only six (27%) studies used a case–control or prospective cohort design. No controlled experiment was identified.

The methods of outcome measurement differed widely among the 22 studies: 11 (50%) defined the outcome 'diagnosis of DP' based upon the criteria that were either not defined or not reported in the majority of cases, 3 (14%) used anthropometric criteria and 8 (36%) applied imaging methods (eg, photography or neuroimaging) (table 1). The sample sizes of the studies range from n=23 children to n=19 685 children, with the largest studies showing the most serious selection bias, as data were taken from tertiary care registries. None of the 22 articles reported a statistical substantiation of sample sizes.

The concordance among empirical studies as to the 60 risk factors identified was poor (table 2). Not one single risk factor was reported in all studies. Most risk factors (63%) were only reported in one single study. The direction of effect was not homogeneous among the 22 studies for most exposures (eg, tummy time: significant protective effect in three studies, no significant effect in other three studies; see table 2 for more examples). Agreement for the direction of effect was only noted for two exposures (smoking and APGAR scores, both without significant effect). Supine sleep position was reported in only 14 out of the 22 studies, and was found to be significant in only 6 of them (table 2). Other exposures commonly reported were male gender, preferred head orientation or limited head rotation, developmental delay, level of activity, birth order and tummy time. All these factors were found to be significant in only about half of the higher quality empirical studies (prospective cohort and case–control studies; see table 1 and the online supplementary table).

In the 110 articles on putative or hypothetical aetiological factors of DP, the scope of risk factors mentioned was wide, ranging from metabolic influences (eg, folic acid supplementation during pregnancy) to influences of care routines (eg, placement of the crib in relation to the parents' bed). Eleven (64%) of the seventeen aetiological factors mentioned *over and above* the risk factors identified in the original, empirical studies were related to the concept of uniform positioning and stimulation by the caregivers (table 3). No publication was identified with reference to the social sleep arrangement (co-sleeping vs not co-sleeping) or modality of baby transport (use of pram or stroller vs babywearing).

DISCUSSION

Our systematic review shows that empirically verified evidence on the aetiology of DP is rare and heterogeneous, with only a few studies examining risk factors in non-selected patient populations on a high-grade methodological level. The overwhelming majority of empirical data on determinants of DP has been generated through retrospective chart reviews and uncontrolled case series in tertiary referral centres. Most of the studies were also characterised by poor reporting of methods, population characteristics and outcome measurement. Thus, DP as a common concern both for parents and paediatricians is currently not well described in terms of its contributing factors.

The poor methodological quality of the empirical studies may explain why the evidence on the aetiology of DP is conflicting —another prominent finding in our review. Agreement between studies was poor for most exposures, including supine sleep position, tummy time and use of car seats (table 2). As a matter of fact, in our sample of 22 studies, not one single risk factor was uniformly identified as such if investigated by more than two research teams. For only two determinants, the concordance

Table 1 Characteristics and final quality rating of the 22 empirical studies on risk factors of DP included in this review

No.	Author	Year	Country	Study design	Outcome measurement*	N	Selection bias	Recruitment		Confounder list†		Follow-up (in longitudinal studies)		Quality rating scheme‡
EHPP rating component				B	E		A			C		F		
								Representative? §		*Recruitment rate*		*Dropouts reported?*	*Participants completing study*	
1	Chadduck[19]	1997	USA	Retrospective case series	Clinical criteria	121	Yes	3	Children's hospital	NA	Weak	NA	NA	4
2	Clarren[20]	1981	USA	Baseline data of a non-randomised trial	Degree of deviation (skull radiographs)	43	Yes	3	Specialty clinic	NA	Weak	NA	NA	4
3	Glasgow[21]	2007	USA	Retrospective case series	Transcranial diameter difference >0.6 cm	192	Yes	2	Primary care	NA	Weak	NA	NA	4
4	Habal[22]	2004	USA	Retrospective case series	Clinical criteria	37	Yes	3	Specialty clinic	NA	Weak	NA	NA	4
5	Hutchison[23]	2003	NZ	Case–control study	Clinical diagnosis of 'relatively severe' DP +'visual and anthropometric examinations'	100	Yes	NA	Specialty clinic	80%–100%	Strong	NA	NA	3
6	Hutchison[4]	2004	NZ	Prospective cohort study	Photography+calculation of cranial length ratio and cephalic index	200	No	1	Primary care	80%–100%	Strong	Yes	80%–100%	3
7	Hutchison[5]	2009	NZ	Retrospective case series	Photography+calculation of cranial length ratio and cephalic index	287	Yes	3	Specialty clinic	NA	Weak	NA	NA	4
8	Joganic[24]	2009	USA	Retrospective case series, comparison with 'natural population statistics'	Register of children receiving orthostatic headbands with (clinical) diagnosis of DP	19 685	Yes	3	Patient register	NA	Weak	NA	NA	4
9	Kane[14]	1996	USA	Retrospective case series	Clinical diagnosis +radiographic exclusion of fused sutures	269	Yes	3	Specialty clinic	NA	Weak	NA	NA	4
10	Littlefield[25]	2003	USA	Retrospective case series	Register of children with (clinical) diagnosis of DP	636	Yes	3	Specialty clinic	NA	Weak	NA	NA	4
11	Littlefield[26]	2004	USA	Retrospective case series, comparison with 'natural population statistics'	Clinical criteria	342	No	2	Primary care	80%–100%	Weak	NA	NA	4
12	Losee[27]	2007	USA	Retrospective case series	Clinical examination	128	Yes	3	Neurosurgery	80%–100%	Weak	NA	NA	4
13	Martinez-Lage[28]	2006	ES	Retrospective case series+follow-up	Clinical criteria +neuroimaging (skull radiographs, CT, 3D CT, MRI)	23	Yes	3	Neurosurgery	NA	Weak	NA	NA	4
14	Martinez-Lage[29]	2012	ES	Retrospective case series+follow-up	Clinical criteria+partly (66%) neuroimaging (skull radiographs, CT, 3D CT, MRI)	158	Yes	3	Neurosurgery	NA	Weak	NA	NA	4

Continued

Table 1 Continued

No.	Author	Year	Country	Study design	Outcome measurement*	N	Selection bias	Recruitment		Confounder list†		Follow-up (in longitudinal studies)		Quality rating scheme‡
EHPP rating component				B	E		A			C		F		
15	McKinney[30]	2008	USA	Retrospective case series	Register of children with (clinical) diagnosis of DP	2733	Yes	3	Specialty clinic	NA	Moderate	NA	NA	4
16	Oh[31]	2009	USA	Prospective case series +retrospective risk assessment	Transcranial diameter difference	434	Yes	3	Children's hospital	NA	Weak	NA	NA	4
17	Seoane[32]	2006	AR	Case–control study with follow-up	Neuroimaging (radiographs, CT)	41 (34 with positional plagiocephaly)	Yes	3	Neurosurgery	NA	Weak	NA	NA	3
18	Sergueef[33]	2006	FR	Retrospective case series	Clinical criteria (palpatory)	649	Yes	3	Specialty clinic	NA	Weak	NA	NA	4
19	Stefani[34]	2005	IT	Prospective clinical case series +retrospective risk assessment	Not specified	64	Yes	3	Children's hospital	<60%	Weak	NA	NA	4
20	Van Vlimmeren[6]	2007	NL	Prospective cohort study	Plagiocephalometry (skull anthropometry)+diameter difference index > 104%	380	No	2	Primary care	NA	Strong	Yes	80%–100%	3
21	Mawji[35]	2014	CA	Retrospective cohort study	Clinical criteria (Argenta's 5-point scale)	435	Yes	3	Primary care	<60%	Strong	Yes	80%–100%	3
22	Weernink[36]	2014	NL	Case–control study	Plagiocephalometry +transcranial diameter difference index >104%	832	Yes	3	Primary care and specialty clinic	NA	Strong	NA	NA	3
Mean values				3× Prospective cohort 3× Case–control 2× Prospective clinical case series Rest: retrospective case series		27.782		2.6	6× Primary care setting Rest: special care setting					

For further explanation, see also Methods and Results sections.
*Different methods were used across studies to measure DP. 'Clinical criteria' stands for diagnosis by a physician on the grounds of clinical presentation (eg, unilateral occipital flattening).
†Rating criteria for control of confounders:
1. Strong: studies that controlled for at least 80% of relevant confounders.
2. Moderate: studies that controlled for 60%–79% of relevant confounders.
3. Weak: studies that controlled for <60% of relevant confounders.
‡Quality rating scheme for studies and other evidence (*modified from the Oxford Centre for Evidence-based Medicine):* 1: Properly powered and conducted randomised clinical trial; systematic review with meta-analysis. 2: Well-designed controlled trial without randomisation; prospective comparative cohort trial. 3: Case–control studies; retrospective cohort study. 4: Case series with or without intervention; cross-sectional study. 5: Opinion of respected authorities; case reports.
The *second row of this table* indicates to which of the *EHPP components* the extracted information belongs. **A**: Selection bias in terms of quality of recruitment strategies was measured by the question Q1—'Are the individuals selected to participate in the study likely to be representative of the target population?' (with responses: 1=very likely, 2=somewhat likely, 3=not likely, 4=cannot tell) and Q2—'What percentage of selected individuals agreed to participate?' (with responses: 1=80%–100%, 2=60%–79%, 3=<60% agreement, 4=not applicable, 5=cannot tell). B*: Study design: strong=RCT* **or** *CCT, moderate=cohort, case–control, weak=any other design method*. **C**: Bias through confounding was assessed by checking how many exposures of the list of potential exposures were investigated or adjusted for. E: Tools for primary outcome measures were described. However, reliability assessment was not possible due non-standardised, clinical nature of measurements and lacking details. For all outcomes, 'face' validity could be assumed. (Q2; rating: strong=> 80% of the potential confounder list, moderate=60%–79%, weak=<60%). **F**: Withdrawals and dropouts in longitudinal studies were assessed by two questions: (Q1) 'Were withdrawals and dropouts reported in terms of numbers and/or reasons per group?' (1=yes, 2=no, 3=cannot tell, 4=not applicable) and (Q2) 'Indicate the percentage of participants completing the study' (for grading, see table 1).
In addition to the Effective Public Health Practice Project questions, we added a global assessment for selection bias ('Was the sample highly selective?') and limitations to generalisability ('Was the sample representative for the 'normal child' without prenatal, perinatal or early postnatal developmental problems?').
Exposures were identified as relevant risk factors for DP if (1) supportive evidence from at least two of the six cohort and case–control studies and (2) at least one more significant result from cross-sectional or case series study was present.
§Rating criteria for representativeness of recruitment: 1—very likely; 2—somewhat likely; 3—not likely.
CCT, controlled clinical trial; DP, deformational plagiocephaly; EHPP: Effective Public Health Practice Project; RCT, randomised-controlled trial.

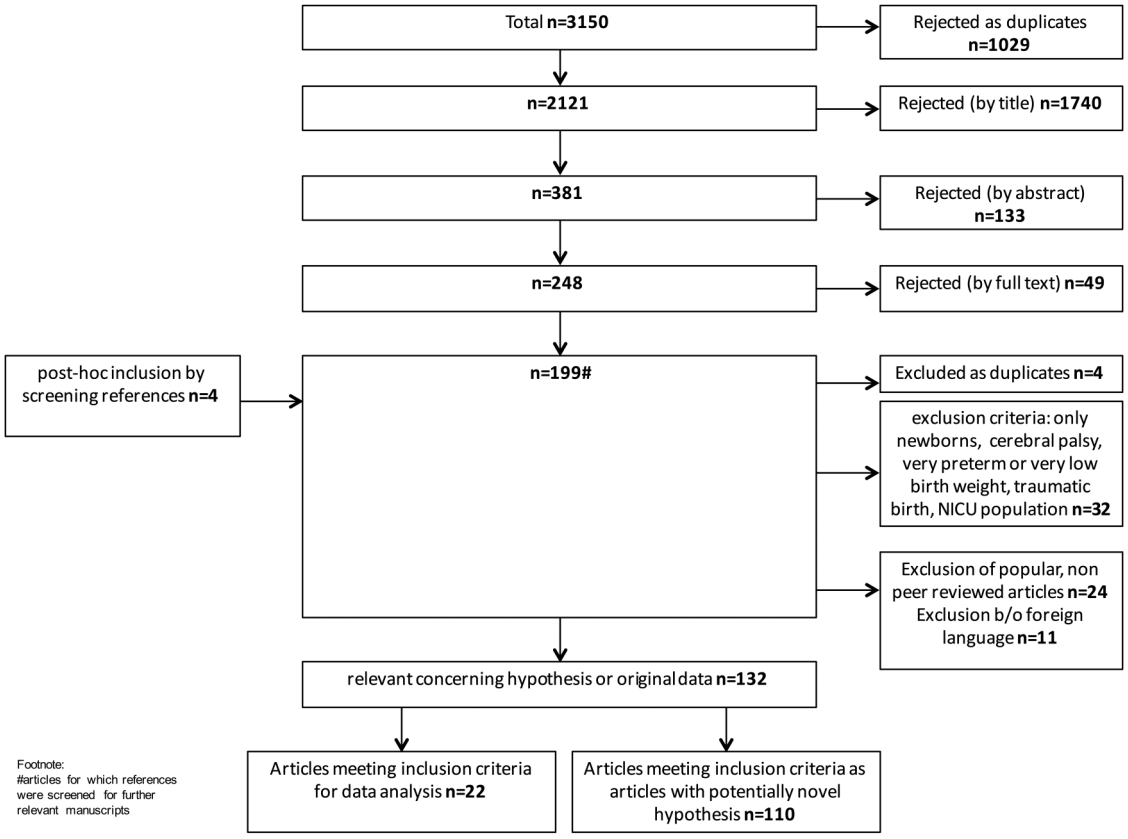

Figure 1 Flow of information in the systematic review: number of records identified, included and excluded, with reasons for exclusion.

rate for significance of the exposure was more than 50% between studies (male gender and supine sleeping position).

Most determinants reported in the empirical studies had been identified as possible risk factors in an earlier review on the aetiology of DP by Bialocerkowski.[2] The fact that obstetric factors seem less important in our review may be explained by our search strategy, which excluded studies of newborns only.

Most of the risk factors reported in our sample of studies fall into the clinical or biological category (eg, gender or birth order). Determinants related to lifestyle have been reported to a lesser extent (eg, feeding mode, different sleep or transport environments). Indeed, within the evidence base summarised in this review, it is currently not possible to identify a single environmental or lifestyle risk factor that could be advocated for preventive measures with any degree of scientific confidence.

Controversies, limitations of research and new hypotheses

While the risk profile of DP thus remains controversial, there seems to be agreement within the expert community on a basic biophysical framework of how DP develops.[3] According to this pathogenetic framework, DP is the result of gravitational forces that act on the same spot of the skull for too long a period of time. Many of the risk factors summarised in our review, like the use of car seats, or supine sleep position, seem to fit well into this model. The pathogenetic influence of the supine sleeping position seems to be additionally supported by epidemiological data, according to which the incidence of DP has increased significantly since the 'back to sleep campaign,' launched in the 1990s to prevent sudden infant death syndrome.[12–14]

Yet, the evidence from this systematic review cautions this interpretation: the supine sleep position did not emerge as a consistent risk factor. The same holds true for the use of bouncers, rockers and car seats, which are not consistently associated with a higher likelihood of DP.

By separately presenting a review of hypotheses, we were able to identify potential risk factors currently not tested through empirical research which may fertilise future prospective studies on DP aetiology. We have reviewed this body of hypotheses for possible explanations on why such major everyday exposures such as infant sleep position should be so inconsistently related to the development of DP. However, in the 110 papers reviewed, we have not identified suggestions to advance the debate.

Notably, none of the hypotheses refers to the infant care practices where they differ most in contemporary Western societies, that is, in regard to the social sleep arrangement and the modality of baby transport. More than half of American mothers of infants from birth to 12 months of age report occasional bed-sharing,[15] and the rate of American parents who routinely use co-bedding, co-sleeping or baby bays ('side cars)' as preferred sleep environments for infants has doubled to about 13.5% within 17 years.[16] Also, a significant proportion uses slings, wraps or baby carriers to transport their infants instead of (or in addition to) prams, strollers or buggys. In our review, there is neither empirical evidence nor any hypothetical suggestion on which role these influences may play in the development of DP.

This may be a serious void as some of the exposures mentioned may have a significant bearing on head shape development and may therefore act as risk modifiers for DP. For example, there is good evidence that different sleep and transport environments are associated with vastly different moulding effects on the developing cranium. Experimental research in the sleep lab has shown that babies sleeping close to their nursing mother experience more body and head repositioning during sleep. In addition, they spend more time in active sleep phases

Table 2 Exposures that have been identified as potential risk factors of positional plagiocephaly in any of the 22 empirical studies reviewed (listed in table 1)

Category	Exposure	Examined in article nr.*†	Frequency and proportion‡
Biological infant factors	Male gender	1†,5†,6,7,8†,9†,11,12,15,16†,17,18,19,20†,21†, 22†	16/22 (72%)
	Torticollis, limited head rotation, head rotational asymmetry	1†,2, 5†, 9,12†,13,14,16†,17†,19†	10/22 (45%)
	Preferred head position/orientation	2,3†,5†,6,7,16†,20†, 21†	8/22 (37%)
	Higher birth weight	2,8†,11,13,14,15,17†, 20,22	8/22 (37%)
	Developmental delay	1,5†,6†,7,20	5/22 (23%)
	Head circumference, macrocephaly	1,3,13†, 17†,20	5/22 (23%)
	Lower level of activity	5†,6†,7†	3/22 (14%)
	APGAR score	17,19	2/22 (9%)
	Others§: malformations (2), prenatal bone mineral density (4), head shape at birth (5), temperament (6†), snoring (6†), limitation of head function (7†), zygosity (dizygotic)(8†), abnormal cerebrospinal fluid spaces (13), 'high rate of pericerebral fluid collections' (14), head tilt (16†), neurological problems (17†), 'lateral strain patterns of spheno-occipital synchondrosis' (18†), pattern of occipito-atlantal motion (18†), asymmetrical movements of trunk (20), brachycephaly (20), siblings with plagiocephaly (3)		
Obstetric factor	Birth order (parity)	4,5†,6,8†,11,15,18†,19,20†, 21	10/22 (45%)
	Mode of delivery: forceps, vacuum or assisted delivery	3,5,7,9†,11,15,18†, 20,21†, 22†	10/22 (45%)
	Prematurity	1,5,9,11,12,19, 22†	7/22 (32%)
	Intrauterine position/cranial immobility	1,2,5,8†	4/22 (18%)
	Lower gestational age	5†,15,16†,17	4/22 (18%)
	Multiple birth	5,15,16†	3/22 (14%)
	Multiple gestation pregnancies	12†, 21	2/22 (9%)
	Others§: birth injury (15), diagnosis of oligohydramnios during pregnancy (15), other obstetric factors (17), birth season (22)		
Infant care practices, lifestyle of mother/ parents	Supine sleep position	3†,4,5†,6†,7†,8,9,11,14,16,17†,19,20, 21†	14/22 (63%)
	Little time spent prone ('tummy time')	3,4,5†, 6†, 20†, 21	6/22 (27%)
	Feeding pattern (bottle feeding)/non-varying position during feeding	6, 7, 12†, 20†, 21, 22†	6/22 (27%)
	Use of car seats,¶ swings, carriers, bouncy seats, rockers	3, 6, 10(†)	3/22 (14%)
	Smoking	4, 15	2/22 (9%)
	Others§: mother's holding position (5), pacifier use (6), mattress type and softness (5, 6), pillow use (6), advice received about plagiocephaly (3), soft drinks (caffeine) (4), alcohol consumption (4), attended antenatal classes (4), late or early begin of prenatal care (4), chemicals (5†), medication (15), insufficient vitamin D intake (22†)		
Sociodemographic factors	Lower parental age	4,5,6,11†, 15, 21, 22†	7/22 (32%)
	Lower educational level	5†, 6, 19, 20, 21, 22†	6/22 (27%)
	Others§: SES (6), marital status of mother (15), maternal number of years lived in Canada (21), maternal language barriers (21), ethnicity (22)		

*Numbers and relevant articles are listed in table 1.
†Exposure found to be significant in the cited study.
‡Proportion and percentage of empirical studies in which the respective exposure was reported.
§Exposures examined in only one single study.
¶Significant for 'heavy use of car seats' according to author (without presentation of data).

during which the gravitational effects on the skull are minimised due to more arousals and a higher muscle tone.[17 18] Thus, the social sleep arrangement seems to be able to modify or even nullify the effect of the supine sleep position exposure, making the latter a necessary rather than a sufficient exposure for DP to develop. This could explain why in this review supine sleep position appears as such an inconsistent determinant for DP.

Similar risk-modifying effects may come from alternative modes of baby transport: The deforming impact on the back of the skull is clearly different in quality and duration for a baby carried in a sling or wearing device than for one transported in a classic pram, stroller or car seat. Therefore, a baby routinely transported in a sling may show a different head shape development, even if subject to other risk factors, such as supine sleep position.

Strengths and limitations of the systematic review

To our knowledge, this study is the second[2] systematic review on the aetiology of positional plagiocephaly and the first to use a two-pronged approach to summarise results, combining an in-detail summary of empirical studies on the determinants or risk factors of DP and a narrative summary of articles on novel hypotheses about potentially contributing factors to DP. The search strategy used was systematic and based on primary and secondary searches to identify published and unpublished evidence. Through a quality rating system (table 1), we were able to assess risk of selection, detection and assertion bias in all empirical studies. Raters of quality were not blinded to authors, which theoretically could lead to biased ratings. However, the independent raters' assessments were consistent in >95% of ratings. Although the PRISMA statement guided the methods in

Table 3 Exposures within predefined exposure categories that have been mentioned as hypothetical risk factors for the development of positional plagiocephaly in the sample of 110 articles on novel hypotheses[1–110]* over and above the exposures identified as correlates in any of the 22 empirical studies reviewed (see table 2)

Category	Exposures (risk factors)
Uniform positioning and stimulation	▶ One-sided positioning during sleep[102]* ▶ Side preferences in infant handling while awake: addressing baby from the same side,[102] unilateral stimulation,[15] non-varying nursing habits,[104] side preference while holding and playing,[68] routinely feeding the infant from the same side[57] ▶ Positioning of crib, so that infant turns its head to the parents while sleeping[85] ▶ Lack of awareness by the caregiver of the need to vary the infant's head position when placed supine[38] ▶ No nightly alternating the supine head position[78] ▶ No periodically changing the orientation of the infant to outside activity during sleep[78] ▶ No alternate directions of the face during sleep[58]
Others	▶ Swaddling[42] ▶ Lack of breast feeding ('sucking on the breast exercises the facial muscles of the baby, which in turn prevents cranial moulding')[31] ▶ Result of greater awareness of cranial dysmorphisms by primary care physicians[95] ▶ Reduced intake of folic acid[65] ▶ No holding and hugging of the infant[57]

*Reference numbers in table 3 refer to reference list for the data set of 110 articles mentioning hypothetical risk factors for the development of positional plagiocephaly. This reference list can be found in the online supplemental material.

our systematic review, we could not consider publication bias as we only included completed studies.

Conclusion and implications

The lack of sound evidence for many of the measures advocated for the prevention of DP poses a challenge for the anticipatory guidance of parents. Yet, it has a benign face: many of the recommendations currently given to parents, such as providing adequate tummy time or positioning babies supine for sleep, are important, regardless of their effectiveness for the prevention of DP. On the other hand, parents do ask specific questions around their specific lifestyles: if I carry my baby in a sling instead of transporting her in a pram—will that prevent her from developing a flat head? To find more specific answers, we need more specific research. For the case of DP, this translates into larger prospective cohort studies in paediatric primary care settings with both rigorous assessment of potential confounders, outcome measures and risk modifiers—including the diverse lifestyle factors associated with today's parenting practices.

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Contributors FDB designed the study and conceptualised the methods, drafted the Methods section, reviewed in detail all other sections of the manuscript and approved the final manuscript as submitted. FDB had full access to all the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis. VB designed the search strategy, performed the searches in all databases, reviewed the manuscript and approved the final manuscript as submitted. HR-P conceptualised the study, extracted the data (supported by Juliane Klein and Janina Beckmann; see acknowledgements), drafted the main manuscript, reviewed the Methods section and approved the final manuscript as submitted.

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Freia De Bock, Volker Braun and Herbert Renz-Polster

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