Review

Prevalence, risk factors, and natural history of positional plagiocephaly: a systematic review

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This review synthesized current research evidence on the prevalence, risk factors, and natural history of positional plagiocephaly. Research published between 1985 and 2007 was sourced from 13 databases. Evidence was categorized according to a hierarchy and rated on a standardized critical appraisal tool. These evaluations were incorporated into a narrative synthesis of the main results. Eighteen studies met inclusion criteria (prevalence: $n=3$, risk factors: $n=17$, natural history: $n=1$). The methodological quality of studies was fair. The point prevalence of positional plagiocephaly appears to be age-dependent and may be as high as 22.1% at 7 weeks of age. Point prevalence tends to decrease with age and may be as low as 3.3% at 2 years. When compared with historical data, the prevalence of positional plagiocephaly appears to have remained stable over the last four decades. Assisted delivery, first born child, male sex, cumulative exposure to the supine position, and neck problems may increase the risk of positional plagiocephaly. To reduce the risk of positional placiocephaly, infants should experience a variety of positions, other than supine, while they are awake and supervised, and early treatment may be warranted for infants with neck problems and/or strong head preference.

Positional plagiocephaly (also referred to as deformational plagiocephaly or non-synostotic plagiocephaly)[1] is a paediatric condition that has featured prominently in lay publications, such as the Wall Street Journal,[2] and seems to have increased in prevalence since the introduction of the 'Back to Sleep' campaign which recommended placing healthy infants on their backs to sleep. It is characterized by changes in skull shape,[3,4] resulting from mechanical factors applied in utero, at birth, or postnatally.[5,6] Infants may have altered skull shape at birth[7,8] and this is thought to revert to normal in the early postnatal period.[5,9] Therefore, positional plagiocephaly refers to infants with altered skull shape, who are older than 6 weeks of age.[10] The cranial sutures are open and normal, and no craniosynostosis is present.[11]

Positional plagiocephaly was once considered a purely cosmetic disorder. Consequently, the management approach has traditionally been based on 'normalizing' skull shape.[2] However, recent research suggests that infants with the condition may also experience developmental difficulties (psychomotor and cognitive),[1,8,12–16] which may require intervention during primary-school years.[16] Therefore, it is important to determine the prevalence and natural history of the condition so that appropriate resources can be allocated for the management of these infants. From an epidemiological perspective, it is equally important to identify and address modifiable factors that place an infant at risk of positional plagiocephaly. This would potentially reduce its prevalence and the cost of its management.

This is a systematic review of the evidence on the prevalence, risk factors, and natural history of positional plagiocephaly. The authors aimed to determine whether there was epidemiological evidence to support an increase in its

prevalence during the 1990s, following the introduction of the 'Back to Sleep' campaign,[17–20] compared with historical studies.[21,22] This review also sought evidence regarding the natural history of positional plagiocephaly, as it is assumed that changes in skull shape will correct with time during infancy.[5,9] As positional plagiocephaly may be the consequence of a combination of sociodemographic and socioeconomic variables, obstetric, infant, and infant care factors,[1,23] this review aimed to identify evidence-based modifiable risk factors which could be used in preventative programmes to reduce the likelihood of the condition.

Method

Published and unpublished epidemiological studies on positional plagiocephaly were sourced from 13 library and internet databases (MEDLINE, CINAHL, Proquest 5000, ISI Current Contents, ISI Web of Science, Expanded Academic ASAP, ScienceDirect, Journals@Ovid, PubMed, Strathclyde, OAIster, Proquest Digital Dissertations, and Australian Digital Theses Program). A well-established search strategy was used,[24,25] including positional plagiocephaly synonyms in a keyword search of all databases except MEDLINE and CINAHL. For these databases, the synonyms were mapped to subject headings, and both synonyms and subject headings were used in the final search strategy. The terms 'prevalence', 'risk factor' and 'natural history' were not used in the search strategy as it was found on preliminary searches that they limit the number of 'hits', and omitted relevant studies. All searches were limited to English-language papers published over the last two decades (January 1985 to October 2007), as during this time there have been reports of increased treatment referral rates of infants with positional plagiocephaly and an explosion of research in this area.[17–20] Secondary searching (reviewing reference lists of included studies) was also conducted.

The studies identified from the searching process were independently assessed by two reviewers against the following inclusion criteria: (1) used a quantitative study design with an epidemiological focus (case–control, cohort, cross-sectional survey, case series); (2) investigated the prevalence, risk factors, or the natural history of positional plagiocephaly in infants. Studies were excluded if their participants were diagnosed with genetic disorders or syndromes, such as Down syndrome, as these comorbidities may confound the development of the condition.[5] Where there was disagreement regarding study eligibility, decisions were made by an independent, experienced researcher.

All included studies were appraised in terms of quality of study design and potential biases using the University of Sheffield's Hierarchy of Evidence.[26] This hierarchy ranges from level 1 evidence (systematic reviews) which, potentially contains the least amount of bias, to level 8 evidence (anecdotal information), which potentially contains the most amount of bias. To address the study aims, evidence was sourced from cohort, case–control, cross-sectional surveys, and case series, which correspond to levels 3, 4, 5, and 6 respectively. However, cohort (level 3 evidence) and case–control (level 4 evidence) study designs were considered the most appropriate to answer the research aims. They can identify causation, can provide evidence regarding prevalence, and natural history, and have fewer potential sources of bias compared with other quantitative methodologies.[27] In addition, the Critical Review Form – Quantitative studies[28,29] was used to evaluate the internal and external validity of the studies and their findings. To provide a summary of study quality, the closed-ended questions were scored as either 1 (completely fulfills the criterion) or 0 (does not fulfill the criterion). Scores for these questions were tallied to produce a total score.[30] The maximum score of 15 indicated excellent quality.

A description of all the included studies was undertaken, including sample characteristics, method of diagnosis of positional plagiocephaly, and main results regarding prevalence, risk factors, and natural history of the condition. Two reviewers extracted this information independently, with decisions made by an independent, experienced researcher if disagreements occurred. Data were synthesized in a narrative format and evidence regarding prevalence, risk factors, and natural history of positional plagiocephaly were interpreted with consideration given to hierarchy of evidence, methodological quality, sample characteristics, and method of diagnosing the condition. Potential risk factors were identified based on significant results from cohort or case–control studies, and supportive evidence from at least one cross-sectional study or case series.

Results

The search strategy yielded 1385 studies. After removal of studies that were sourced from more than one database, 18 were included in this review.[1,6,8–10,19,20,23,31–40]

PREVALENCE AND NATURAL HISTORY OF POSITIONAL PLAGIOCEPHALY

Positional plagiocephaly prevalence was investigated by three research teams,[1,8,31] all of which used different definitions of the condition (Table I). The most robust evidence was produced by Hutchinson et al.[1] and van Vlimmeren et al.[8] who conducted high-quality (quality score: 12/15) prospective cohort studies (Table II). Its 2-year prevalence may be as high as 29.5%;[1] however, prevalence appears to be age-dependent with most cases manifesting in the first months of life (point prevalence at 6–7 weeks = 16–22.1%;[8] at 4 months = 19.7%[1]). By 2 years of age, the point prevalence of positional plagiocephaly may be as low as 3.3%.[1] Although not explicitly stated, it was inferred that infants in the Hutchinson et al. study[1] did not receive treatment for positional plagiocephaly. Thus, this study provides evidence regarding its natural history, i.e. that skull shape naturally changes within the first 2 years of life.

RISK FACTORS

Seventeen of the 18 studies investigated 64 potential risk factors for positional plagiocephaly (Table III). The methodological quality of the studies was only fair, with the average quality score being 7 from a maximum of 15 (SD 2.9, range 2–12[1,8]; Tables I and II). Methodological quality was significantly higher, on average, in studies that used cohort and case–control designs[1,8,23,36,38] compared with those that were cross-sectional studies and case series[6,9,10,19,20,32-35,37,39,40] ($z=-2.82$, $p=0.005$). Very few or no study fulfilled the following criteria: no biases present ($n=0$), used valid outcome measure ($n=0$), used reliable outcome measures ($n=0$), acknowledged limitations of the study ($n=3$), or gained informed consent ($n=5$). In most studies, data were extracted from medical records.

Table I: Study design, quality score, study population, definition of positional plagiocephaly (PP), and method of diagnosis

Author	*Study design (hierarchy)*	*Quality score*	*Study population*	*Definition of postitional plagiocephaly and method of diagnosis*
Bruneteau and Mulliken[6]	Case series (6)	6	Reviewed medical records of 60 patients with frontal plagiocephaly (synostotic and positional) from the Craniofacial Centre, Harvard Medical School, US	Open sutures and gradual improvement of head shape on follow-up, evaluated by photography
Golden et al.[32]	Case series (6)	7	Reviewed medical records of 100 PP patients referred for orthotic management in 1 month (August 1996) from the South West Craniofacial Centre, Phoenix, US	Asymmetry of occipital skull and forehead evaluated by physical examination
Habal et al.[33]	Cross-sectional survey (5)	5	Random selection of 37 patients whose child was diagnosed of developmental problems related to PP, from Tampa Bay Craniofacial Centre, Tempa, US	Not stated
Hutchinson et al.[23]	Case–control (4)	11	*Cases*: 100 consecutive infants diagnosed with PP from Middlemore Hospital Craniofacial Clinic and Auckland Pediatric Physiotherapy Clinics, New Zealand, between January and August 2001. *Controls:* every sixth infant from a community health nursing database in the Auckland region (New Zealand)	Visual and anthropometric measurements of skull shape
Hutchinson et al.[1]	Cohort study (3)	12	Every fourth child born between September 2001 and February 2002 at North Shore Hospital Auckland, New Zealand	Cephalic index ≥93% and or oblique cranial length ratio ≥106% evaluated by digital photography
Kane et al.[19]	Case series (6)	7	Reviewed medical records of patients referred to Cleft Palate and Craniofacial Deformities Institute, St Louis, US, between January 1979 and December 1994	Unilateral occipital flattening and contralateral occipital prominence, evaluated by physical examination; normal cranial sutures on radiographs
Keusch et al.[34]	Case series (6)	5	Reviewed medical records of 35 twin pairs where one child or both exhibited a craniofacial anomaly, who presented to the Craniofacial Clinic at Harvard Medical School, US	Severity rated on the posterior occipital deformational score (scoring criteria were not defined)
Littlefield et al.[35]	Case series (6)	7	Reviewed medical records of 140 sets of twins, where at least one child was treated for PP. Data from a larger twin study were used	Occipital flattening, ear misalignment, facial asymmetry
Losee et al.[40]	Case series (6)	11	Reviewed medial records of 105 children with occipital plagiocephaly who were treated between January 2001 and January 2003 at the Cleft–Craniofacial Centre, Children's Hospital of Pittsburg, US	Not stated
Martinez-Lage et al.[38]	Case–control (4)	9	*Cases:* 23 children with head deformities excluding true craniosynostosis from medical records of 110 consecutive children at University Hospital, Murcia, Spain. *Controls:* 9 children assessed for macrocephaly, diagnosed with benign extracerebral collections of cerebrospinal fluid	Skull radiograph, 3D computerized tomography or magnetic resonance imaging to identify head structure and patency of cranial sutures
Pomatto et al.[9]	Case series (6)	5	1 set of 8-month-old triplets who presented for treatment at Cranial Technologies, Phoenix, US	Not stated
Sawin et al.[36]	Case–control (4)	9	*Cases:* 31 infants treated for PP, between 1990 and 1995 at the University of Iowa Hospital, Iowa City, US. *Controls:* 20 age- and sex-matched controls without intracranial and calvarial pathology	Posterior calvarial flattening
Sergueef et al.[37]	Case series (6)	9	Reviewed 649 children observed and treated at an osteopathic practice in Lyon, France between 24 January 1999 and 30 May 2000	Shape of skull using osteopathic clinical (palpatory) diagnostic methods
Slate et al.[10]	Case series (6)	6	Reviewed medical records of 26 patients referred with craniofacial asymmetry without craniosynostosis and congenital muscular torticollis to the Hospital for Sick Children, Toronto, Canada, between 1987 and 1990	Craniofacial asymmetry
Stefani et al.[39]	Cross-sectional survey[a] (5)	4	64 (56 with PP) of 122 children with occipital plagiocephaly between 1998 and 2003 at the Paediatric Department of Padua Hospital, Italy	Flattening of posterior cranium with contralateral occipital and ipsilateral frontal bossing with or without facial asymmetry

[a]Case series and cross-sectional survey. Quality score ranges from 0 to 15, with 15 indicating excellent quality. Hierarchy based on the University of Sheffield Hierarchy of Evidence, which ranges from 1 (systematic reviews, potentially containing the least amount of bias) to 8 (anecdotal information, potentially containing the most amount of bias).

Positional plagiocephaly was often defined in general terms and visually evaluated (Table I). Moreover, it is not known whether the variables of interest were measured or recorded in a standardized or reliable manner and whether there was consistency in participant selection for each study.[6,9,10,19,20,32,34,35,37,39,40]

As psychometric properties of the outcome measures used were not reported, the magnitude of measurement error is not known. Although sample sizes were justified in pragmatic terms, sample size or power calculations were not provided by any of the authors. Thus small, underpowered cohort or case–control studies may have only detected strong associations.[41] None of the authors conducted sensitivity analyses to explore the heterogeneity of their groups, or controlled for or discussed the implications of confounding.[42] Thus all results of the studies in this review should be interpreted with caution.

Socio-demographic and socioeconomic variables

The effect of socio-demographic (family structure, parents' age, ethnicity) and socioeconomic variables (parents' educational qualifications, occupation, household income) on positional plagiocephaly were studied by five research teams.[8,23,35,39,40] Although lower maternal education levels were associated with the condition[23] (Table IV), this result was not supported by the work of others. Moreover, maternal education levels could potentially be a confounding factor, as it may influence obstetric, infant, and infant care variables.

Obstetric factors

Based on the investigations of 13 research teams,[1,6,8,9,23,32-35,37,39,40] obstetric risk factors for positional plagiocephaly are being first-born[1,8,23,33,37] or delivered with assistance (e.g. forceps or vacuum extractor;[19,23,37] Table IV). These factors could potentially be confounders; however, these associations were not investigated. There was some evidence to suggest that antenatal education was associated with positional plagiocephaly,[23] and conflicting evidence regarding the role of preterm birth in the development of positional plagiocephaly.[23,39] There was little evidence to suggest that multiple births,[9,40] being the lower utero twin,[35] vertex delivery,[35] low birthweight,[35] and maternal exposure to caffeine and tobacco[33] were risk factors.

Infant factors

Fourteen research teams investigated the role of infant factors in positional plagiocephaly.[1,6,8,10,19,20,23,32,35-40] Neck problems were consistently associated with the condition[1,6,23,32,35,37] (Table IV). However, neck problems were variably defined (Tables III and IV) and evaluated, either by parental report[8,23] or by clinical assessment.[1,6,10,19,20,23,32,35,37,40] A standardized definition and method of assessing the neck is required to confirm the relationship between positional plagiocephaly and neck problems.[43] Male sex consistently appears to be associated with positional plagiocephaly.[1,8,23,32,39] There was some evidence to suggest that larger cerebrospinal fluid spaces,[36,38] slower motor development,[1,8] infant inactivity,[23] difficult infant temperament,[1] and snoring[1] may be associated with the condition; however, further investigation is warranted.

Infant care factors

Based on the work conducted by eight research teams,[1,6,8,23,32,35,39,40] it appears that cumulative exposure to the supine position may be associated with positional plagiocephaly [1,8,23,32] (Table IV). Feeding position,[8] not varying the position of the infant's head when they are put to sleep,[1,23] varying the infant's head position but it not being maintained,[1] sleeping on firmer mattresses,[23] and being bottle fed[40] may contribute to the effect of supine positioning (Table IV). These associations have been rarely investigated.

Discussion

This is the first known systematic review that has collated evidence regarding the epidemiology of positional plagiocephaly. The search strategy used was systematic and thorough, using primary and secondary searches to locate published and unpublished evidence.[44,45] Hand searching and contacting experts in the field was not undertaken, as these searching strategies are difficult to perform in a systematic manner and are difficult to replicate. Moreover, it was highly likely that most research evidence would be indexed

Table I: Continued

Author	*Study design (hierarchy)*	*Quality score*	*Study population*	*Definition of postitional plagiocephaly and method of diagnosis)*
Turk et al.[20]	Case series (6)	2	Reviewed medical records of 52 consecutive patients over a 2-year period presenting with PP to the Variety Club Centre for Craniofacial Rehabilitation, New York, US, between January 1992 and December 1994	Occipital flattening, contralateral forehead flattening, contralateral lowering of brow, contralateral ear shearing or asymmetry in the posterior-inferior direction by clinical examination, patent cranial sutures
van Vlimmeren et al.[8]	Cohort study (3)	12	Healthy, consecutively-born neonates born between December 2004 and September 2005 at General District Hosspital Bernhoven in Veghel, The Netherlands (>36 weeks gestation, with no dimorphisms or syndromes)	Oblique cranial length ratio ≥104%, using plagiocephalometry

Table II: Methodological quality of studies in systematic review

Quality criteria	*Boere-Boonekamp and van der Linder-Kuiper*[31]	*Bruneteau and Mulliken*[6]	*Golden et al.*[32]	*Habal et al.*[33]	*Hutchinson et al.*[23]	*Kane et al.*[19]	*Keusch et al.*[34]	*Littlefield et al.*[35]	*Losee et al.*[40]	*Martinez Lage et al.*[38]	*Pomatto et al.*[9]	*Sawin et al.*[36]	*Sergueef et al.*[37]	*Slate et al.*[10]	*Stefani et al.*[39]	*Turk et al.*[20]	*Total*
Purpose clearly stated	1	1	1	1	1	1	1	1	1	1	0	1	1	1	0	1	14
Literature review relevant	1	1	1	0	1	0	0	0	1	1	1	1	1	1	0	0	10
Study design appropriate to study aims	1	1	1	1	1	1	1	1	1	1	0	1	1	0	1	0	13
No bias present	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
Sample described in detail	0	0	1	1	1	1	0	1	1	1	1	1	1	1	1	0	12
Sample size justified	1	0	1	0	0	1	1	1	1	0	1	1	1	1	1	1	12
Informed consent gained	0	0	0	0	1	0	0	1	1	0	0	0	0	0	1	0	4
Valid outcome measures used	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
Reliable outcome measures used	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
Results reported in terms of statistical significance	0	1	0	0	1	1	0	1	1	1	0	1	1	0	0	0	8
Analyses appropriate	0	1	0	1	1	1	1	1	1	1	1	1	0	0	0	0	10
Clinical importance of results reported	1	1	1	0	1	1	0	0	1	1	0	1	1	0	0	0	9
Appropriate conclusions	1	0	0	1	1	0	1	1	1	1	0	1	1	1	0	0	10
Clinical implications reported	1	0	1	0	1	0	0	0	1	1	1	0	1	1	0	0	8
Limitations of study acknowledged	0	0	0	0	1	0	0	0	0	0	0	0	0	0	0	0	1
Total	7	6	7	5	11	7	5	8	11	9	5	9	9	6	4	2	–

1, criterion fulfilled; 0, criterion not fulfilled.

in the 13 databases searched. Although only English language studies were included in this review, no evidence was found in other languages. Reviewers were not blinded to the author or source of the paper, which may theoretically affect methodological quality ratings. However, consistent results were independently gained from both reviewers.

PREVALENCE AND NATURAL HISTORY OF POSITIONAL PLAGIOCEPHALY

The prevalence of positional plagiocephaly appears to be age-dependent, with the prevalence peaking within the first 6 months of life.[1,8] The variation in prevalence rates reported between studies can be attributed to different methods of assessment, definitions, and assessment times. Errors associated with measurement processes were not stated in either of these articles.

The point prevalence of positional plagiocephaly may be as high as 22.1% (in the first months of life).[8] This prevalence estimate is similar to the prevalence of altered skull shape in healthy neonates (25% of singleton neonates had skull flattening or unusual shaped heads).[7] However, historical data suggest that the point prevalence of the condition at 1 year of age may range between 5%[21] and 48%,[22] with the prevalence rate gained by Hutchinson et al.[1] (6.8%) being similar to that gained by Watson[22] in the early 1970s. Therefore, in comparison with historical data, it is difficult to draw conclusions regarding the effect of the 'Back to Sleep' campaign on the prevalence of positional plagiocephaly, and on the relationship between its prevalence and treatment referral rates. An increase in referral rates may be the result of artifact[36] and increased awareness of early referral for evaluation of infants with skull deformities,[20,36] rather than an increase in its prevalence over the past two decades. Moreover, reported referrals rates do not take into account those infants who have been diagnosed with positional plagiocephaly but have not been referred for treatment and those infants who have not been diagnosed with the condition but have changes in skull shape.

Hutchinson et al.[1] also documented a decrease in the prevalence of positional plagiocephaly up to 2 years of age. This appears to be the first high-quality evidence that suggests that head shape continues to change substantially beyond the early neonatal period when treatment is usually advocated. Anecdotal clinical opinion suggests that skull-shape remodeling does not tend to occur beyond 1 year of age, although Hutchinson et al.[1] found that the point prevalence of positional plagiocephaly halved between 1 and 2 years of age (point prevalence = 6.8% and 33% respectively). This suggests that without treatment head shape may normalize during early childhood. The long-term follow-up of infants diagnosed with positional plagiocephaly is required to determine if skull shape changes occur later in childhood and adolescence,[10] as historical data suggest that a large percentage of infants diagnosed with positional plagiocephaly at 1 year of age continued to have altered skull shape at 10 years of age.[21]

RISK FACTORS ASSOCIATED WITH POSITIONAL PLAGIOCEPHALY

There is little high-quality evidence regarding the factors that increase the likelihood of positional plagiocephaly. There is little epidemiological research on this topic, which

Table III: Investigated risk factors for positional plagiocephaly

Socio-demographic and socioeconomic factors	*Obstetric factors*	*Infant factors*	*Infant care factors*
Parents' ages[23]	Multiple birth[1,6,8,9,19,23,34,35,40]	Sex[1,6,8,19,23,32,39]	Sleep position[1,6,8,23,32,35,39]
Mother's age at birth[23]	Primagravidity[1,6,8,23,35,39,40]	Race[35]	Head position when sleeping[8]
Parents' ethnicity[23]	Duration of pregnancy[1,6,8,19,23,35,39]	Apgar scores[1,8]	Head position when awake[8]
Parents' occupation[23]	Prenatal history [32]	Head circumference[1,8]	Head positioning practices[1,23]
Parents'/mother's education[35,39]	Exposure to environmental agents which influence bone mineral density [33]	Weight[1,8]	Type of bed[23]
Family structure[8]	Labour and delivery[1,8,32]	Height[1,8]	Type and firmness of mattress[1,23]
Principal carer[8]	Delivery complication[39]	Morphometric factors[23,35]	Under bedding[1]
Household income[40]	Presentation[1,8,19,23,35]	Presence of hair loss on the back of the head[1,23]	Pillow use[1]
Race[40]	Mode of delivery[1,8,19,23,35,37]	Temperament[1,23]	Position and orientation of cot[23]
Number of parents in household[40]	Abnormal uterine or pelvic structure[35]	Activity level[1,23]	Infant position on change table[8]
		Developmental delay[1,8,23,35]	Use of infant chairs[8]
		Posture[1,8]	Use of mobiles[23]
		Preferential head orientation[1,8,23]	Other resting places such as car seats[1,23]
		Torticollis[6,19,35,40]	Upright time[1]
		Cervical active and passive movement[1,8,23,32,35]	Daily duration in supine[23]
		Joints and synchondroses mechanics in the cranial base region[37]	Daily duration of tummy time[1,23]
		Poor head control[19]	Infant's age when first put into prone and side-lying[8]
		Neck examination[10,20]	Frequency and duration of prone and side-lying positions when awake[8]
		Health problems[1]	Hand dominance of mother[23]
		Ear infections[35]	Parental holding positions and duration[1,23]
		Cerebrospinal fluid spaces[36,38]	Method of feeding[1,8,40]
			Position of infant when fed[8]
			Pacifier use[1]

Table IV: Potential risk factors for positional plagiocephaly (PP)

Risk factors		*Evidence from level 3 and 4 studies*	*Supportive evidence from level 5 and 6 studies*
Socio-demographic and socioeconomic factors			
Mother's education	Completed high school	OR=2.5 (1.1–2.7) [23]	
Obstetric factors			
Parity	First born	OR=2.9 (1.6–5.5)[23] OR=1.8 (1.1–3.0) at birth and 7 weeks of age[8]	Left and right sided PP correlated to primiparity (p=0.024)[37] 51%[39] and 76%[35] sample with PP were first born
Delivery method	Assisted delivery	OR=2.5 (1.1–5.7)[23]	Correlation between PP and the use of forceps (p=0.055) and vacuum extractor suction (p=0.055)[37] Proportion of infants with PP delivered with forceps was significantly greater than expected (χ^2=3.84, p=0.05)[19]
Period of gestation	<37 weeks	OR=3.3 (1.02–10.5)[23]	17% of PP sample were preterm (mean gestational age 38.6 weeks)[39]
Antenatal education	Attended antenatal education	OR=2.1 (1.4–3.2)[23]	
Infant factors			
Neck problems	Infant difficulty turning head	OR=14.0 (6.6–29.7)[23]	64% of infants with PP had torticollis[6]
	Decreased cervical rotation	OR=22.0 (7.5–64.6)[23]	20% of infants with PP had torticollis, a significantly higher percentage compared with population norms (p<0.05)[40]
	Limited passive cervical rotation	OR=6.2 (2.0–18.8) at 6 weeks of age[1]	
	Limited active cervical rotation	OR=7.8 (2.6–23.7) at 4 months of age[1]	76% of infants with PP had neck involvement[32]
		OR=2.7 (1.2–5.8) at 4 months of age[1]	PP was significantly more likely in twins with neck involvement (p<0.001)[35]
			50% of infants with PP had C1–2 subluxation[37]
Potential neck problems	Early established head preference	OR=29.7 (8.7–101.0)[23] OR=9.5 (5.3–17.0)[8]	
	Consistently sleeps with head turned to one side	OR=7.1 (3.9–12.8) at 7 weeks of age[8]	
Sex	Male	OR=2.2 (1.2–3.9)[23] OR_adjusted=5.4 (1.9–15.3) at birth[8]	64%[39] and 71%[32] of infants with PP were male
		OR=1.87 (1.1–30.0) at 7 weeks of age[8]	Sex (male:female) ratio in infants with PP=1.58:1[1] 76% of infants with PP were female[6]
Cerebrospinal fluid spaces	Larger cerebrospinal fluid spaces	PP infants had larger Sylvian fissures (p<0.0001), frontal subarachnoid spaces (p<0.001), perimesencephalic cistern (p<0.001), and suprasellar cistern (p=0.007) than infants without PP[36] PP infants had enlarged Sylvian fissures with anterior fluid collections (p=0.02)[38]	
Activity	Inactive infant	OR=2.8 (1.4–5.6)[23] OR=3.3 (1.4–7.8) at 4 months[1]	
Temperament	Average to difficult	OR=2.6 (1.1–6.3) at 4 months[1]	
Snoring	Snoring reported	OR=5.6 (1.6–19.5) at 4 months[1]	
Development	Abnormal PDQII test	OR=18.1 (2.0–166.5) at 4 months[1]	
	Increased motor development	OR=0.6 (0.4–0.9)[8]	
Infant care factors			
Position of infant	>20 hours per day in supine position	OR=6.4 (3.3–14.74)[23] OR=3.2 (1.5–7.0) at 6 weeks[1]	89% of infants with PP were recommended to sleep in supine[32]
	<1 hour per day upright	OR=2.4 (1.5–5.2) at 4 months[1] OR=2.5 (1.1–5.6) at 6 weeks[1]	41% of infants with PP lay in an abnormal position[6]

is generally only fair in methodological quality, and has mainly used study designs that cannot attribute causation. Its variable definitions and different methods of measurement were used across the studies. Confounding variables were not adjusted for or discussed and thus the results of these studies must be interpreted with caution. The studies sourced for this systematic review investigated a wide variety of possible risk factors for the condition. There is some evidence to suggest that possible risk factors include assisted delivery,[19,23,37] first-born child,[1,8,23,33,37] male sex,[1,8,23,32,39] cumulative exposure to the supine position,[1,8,23,32] and infant neck problems.[1,6,23,32,35,37,40] Positional plagiocephaly may also be the consequence of a variety of inter-related factors, with some of these factors being confounders.

Most of the research has focused on obstetric and infant factors. Obstetric factors, such as first-time mothers and assisted delivery, cannot be modified by preventative programmes. In contrast, infant factors, such as neck problems, may be amenable to treatment, thus preventing the development of positional plagiocephaly.[37] However, inconsistent definitions of what constitutes a neck problem and the variable criteria used to diagnose these problems limits the interpretation of the literature.[43] It appears that early detection and management of neck problems should be advocated to prevent positional plagiocephaly, despite the possibility that neck problems could develop subsequently.[46] Neck problems could occur as a consequence of intrauterine[6,35] or postnatal head position,[6] birth trauma,[6] or a congenital musculoskeletal abnormality.[10] It is hypothesized that spasm or injury to the sternocleidomastoid muscle leads to its shortening and the development of a head preference.[6] When in supine, preference to turn the head to only one side results in uneven distribution of pressure over the occiput, which may lead to positional plagiocephaly.[10] If sternocleidomastoid spasm or injury is recognized early, treatment can be commenced, which could reduce the likelihood of the condition.[32]

It is plausible that males tend to be at greater risk of positional plagiocephaly compared with females,[1,8,23,32,39] as male fetuses have larger heads and are less flexible in their body compared with females.[7,47] Thus their skulls may be more susceptible to deformation during delivery.[47] Male infants grow more quickly compared with females, especially within the first 3 months of life.[48] Given that the sleep position exerts the most molding effects on the skull at this time,[48] the strength of the skull may be insufficient to withstand the pressure imposed on it by gravity.[14]

Little research has been focused on socio-demographic and socioeconomic variables. Although these variables are not modifiable by preventative programmes, this information identifies families at risk to whom preventative programs should be delivered. It appears that infants born to women with lower levels of education may be at increased risk of developing positional plagiocephaly.[23] These mothers may be anxious regarding sudden infant death syndrome, and may use the recommendation to put their infant to sleep in supine during other times of the day, e.g. while their baby is awake and supervised.[33] Cumulative exposure to the supine position was evaluated in various ways in the studies reviewed, including time per day spent in the supine position, time spent in the sideline and prone positions, sleeping and feeding positions. It has been hypothesized that lack of experience of different positions, while the infant is awake and supervised, may contribute to developmental delay, especially in upper body motor strength and coordination.[16,49-51] Moreover, slower to develop infants tend to spend more time in supine which may increase their risk of developing positional plagiocephaly.[13,16] Therefore, it seems that strategies for the prevention of positional plagiocephaly may not only reduce the risk of abnormal skull shape, but also potentially reduce developmental difficulties.

Table IV: Continued

Risk factors		*Evidence from level 3 and 4 studies*	*Supportive evidence from level 5 and 6 studies*
Position of infant	Placed in prone <3 times per day	OR=2.7 (1.1–6.6) at 7 weeks[8]	
	Infants frequently held in supine	OR=1.9 (1.1–3.4)[23]	
	Supine sleeping	OR=11.5 (2.7–49.8) as newborn and at 6 weeks[8] OR=4.7 (1.8–11.9) at 6 weeks[8] 2.3 (1.1–4.9) at 4 months[1]	
	Position of head not varied when infant put to sleep	OR=6.3 (3.1–12.9)[23] OR=2.8 (1.1–6.8) at 4 months[1] OR=1.8 (1.2–2.9) at 7 weeks [8] OR=5.2 (1.9–14.0) at 6 weeks[1]	The side of occipital flattening was strongly correlated to the side of supine head sleeping position (χ^2 test, p=2.256 e–09)[39]
	Position of head varied but not maintained	OR=4.2 (1.7–10.3) at 4 months[1]	
	Only bottle fed	OR=1.9 (1.0–2.6) at 7 weeks[8]	Breastfed babies had a lower rate of PP compared with population norms (p<0.05)[40]
	Bottle fed on same arm of carer	OR=1.9 (1.2–3.1) at 7 weeks[1]	
Mattress	Firmer mattress	OR=2.0 (1.01–3.9)[23]	

PDQII, Revised Denver II Prescreening Developmental Questionnaire. Potential risk factors were identified based on significant results from level 3 and 4 studies (cohort and case–control studies) and supportive evidence from at least one level 5 or 6 study (cross-sectional study or case series).

IMPLICATIONS FOR RESEARCH AND CLINICAL PRACTICE

Large-scale cohort studies are required to investigate the prevalence of positional plagiocephaly, the effect of treatment on its prevalence, and associated risk factors. Future research should use standardized criteria to define the condition and psychometrically sound measures to quantify skull shape. Detailed investigation of the effects of confounding variables should be undertaken. However, based on the current evidence, the present authors recommend that infants should experience a variety of positions other than supine, while they are awake and supervised. They should have regular periods of supervised prone play.[7,19,23] Early identification of infants with strong preference for turning the head to one side and/or with decreased active cervical rotation.[1,6,8,23,32,37,40] is warranted. Head preference may be discouraged by alternating the infant's head position while settling them to sleep[7,19,23] or regularly changing the position of their cot in the room.[5] Early treatment may be warranted for infants with neck problems and/or strong head preference.[1,6-8,10,23,32,37,40]

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