Plagiocephaly and Developmental Delay: A Systematic Review

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ABSTRACT: ***Objective:*** **Deformational plagiocephaly (includes plagiocephaly and brachycephaly) is a common pediatric condition. Infants who present with altered head shape often experience developmental delay. It is uncertain how common developmental delay is in infants with plagiocephaly and how sustained this is, when present. This review explores the association between plagiocephaly and developmental delay to guide clinical practice.** ***Study Design:*** **A systematic review was conducted. MEDLINE, EMBASE, CINAHL, and PEDro databases were searched. Data from relevant studies were extracted regarding study: sample, follow-up, design, and findings. Methodological quality of each study was rated using a critical appraisal tool.** ***Results:*** **The search recovered 1315 articles of which 19 met the inclusion criteria. In the included studies, the children's ages ranged from 3 months to 10 years. Study limitations included selection bias, nonblinding of assessors, and reuse of the same study population for multiple papers. Most papers (11/19) rated "moderate" on methodological quality. A positive association between plagiocephaly and developmental delay was reported in 13 of 19 studies, including 4 of 5 studies with "strong" methodological quality. Delay was more frequently in studies with children ≤24 months of age (9/12 studies) compared with >24 months of age (3/7 studies). Motor delay was the most commonly affected domain reported in high-quality papers (5/5 studies).** ***Conclusion:*** **This review suggests plagiocephaly is a marker of elevated risk of developmental delays. Clinicians should closely monitor infants with plagiocephaly for this. Prompt referral to early intervention services such as physiotherapy may ameliorate motor delays and identify infants with longer term developmental needs.**

*(J Dev Behav Pediatr 38:*67–78, 2017) **Index terms:** plagiocephaly, brachycephaly, developmental delay, infants, motor skill disorders.

Positional or deformational plagiocephaly describes the common head shape disorder that is characterized by occipital flattening and asymmetric cranial vaults that has increased among children born after 1992 when the "Back to Sleep" Campaign began.[1–3] It occurs in infants due to external or mechanical forces acting on the skull prenatally or postnatally.[2,4,5] Benign positional skull deformities are common in infants with a head preference to one side, limited bilateral neck movements, sternocleidomastoid muscle dysfunction such as torticollis, and often accompany prematurity and many conditions associated with developmental delay.[5] Although skull deformity due to uterine constraint generally improves within the first few weeks of life, for most infants the flattening of the skull as a result of supine postnatal positioning tends to worsen

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A. L. C. Martiniuk conceptualized and designed the study, provided analysis and interpretation of the data, provided critical revision and intellectual content to subsequent versions of the manuscript and approved the final manuscript submitted. C. Vujovich-Dunn contributed to the study design, ran one of 2 independent searches for the acquisition of the data and reviewed each article, provided analysis and interpretation of the data, wrote the first draft of the manuscript and prepared the final manuscript. M. Park provided critical revision and intellectual content to the initial and subsequent versions of the manuscript. W. Yu contributed critical interpretation of study findings, contributed in particular to the results section and provided critical revision and intellectual content to subsequent versions of the manuscript. B. R. Lucas interpreted study findings and their implications, assisted with interpreting study methodological quality, and provided critical revision and intellectual content to subsequent versions of the manuscript. All authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

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over time without intervention.[6] Estimates of prevalence suggest rates as high as 22.1% at 7 weeks and as low as 3.3% at 2 years[7] indicating manifestation early in infancy. To date, treatment for plagiocephaly includes increasing the usual developmental recommendations for "tummy time" or upright positioning, physiotherapy exercises, and cranial orthoses such as helmet therapy for more severe cases.[8,9] However, treatment with cranial orthotics is now under debate following a randomized trial published in 2014, which found equal effectiveness of helmet therapy with the control group (natural course).[10] Delayed development in areas such as gross motor, fine motor, problem solving, and personal social skills is associated with positional plagiocephaly,[11] suggesting developmental assessment should be considered in the examination of infants with plagiocephaly.

Referrals for the treatment of plagiocephaly have increased significantly since the "Back to Sleep" campaign was launched in 1992 by American Academy of Pediatrics.[1] The key strategy of this campaign is to place infants to sleep in a supine position to reduce the risk of sudden infant death syndrome (SIDS).[1] Although this has led to a successful reduction in SIDS-related infant mortality, it has contributed to an increase in positional plagiocephaly.[12] This increase in skull deformation has been attributed to reduced variability in postnatal positioning, producing constant pressure on specific areas of the cranial vault during periods of rest and play in supine positions.[12–14] Before the campaign, the incidence of plagiocephaly was considered rare at 1 in 300 infants (0.3%).[15] Current prevalence rates vary widely between 8.2% and 48% of infants affected[10,16–20] with contributing factors for the differences between these estimates likely related to the age when plagiocephaly is detected and the criteria used to define plagiocephaly.

Positional skull deformities include deformational scaphocephaly, plagiocephaly, and brachiocephaly.[21] This review focuses on positional plagiocephaly and brachycephaly, which are the most common presentations.[22] For ease of reading and writing, we have used plagiocephaly as a term throughout this review, but taken to include both plagiocephaly and brachycephaly as their management principles are similar.

Several studies suggest that infants with plagiocephaly are more likely to be developmentally delayed in comparison with infants without this condition.[14,23–37] The delay may be acquired due to limited head movement,[23] which then causes skull deformity. Conversely, a preexisting developmental pathology can also cause reduced head movement leading to skull deformity. In addition, a preexisting skull deformity affecting brain shape and therefore producing brain changes has also been suggested to cause developmental delay.[28] Other studies have not found that plagiocephaly is a predictor of developmental delay alone[38] or long-term poor developmental trajectories.[2,39] To our knowledge, no systematic review has been undertaken of published literature examining the potential association between plagiocephaly and short-term or long-term developmental delay. We, therefore, systematically reviewed existing published literature to investigate, and characterize, the association between plagiocephaly and developmental delay. Given the high level of concern by parents of infants with plagiocephaly and the number of referrals for management of this condition, the information from this systematic review is timely for parents and clinicians to guide the need for screening, early intervention, and to ascertain the likely prognostic outcomes for infants with plagiocephaly.

METHODS

Design

A systematic review of published literature was conducted and reported according to the PRISMA (Preferred Reporting Items for Systematic reviews and Meta-Analyses) statement.[40]

Data Sources and Searches

Electronic data sources were systematically searched using a highly sensitive search strategy and by approaching experts in the field. Searches were restricted by language to English publications. MEDLINE, EMBASE, and CINAHL databases were searched from 1946 to present (February 2016) and PEDro databases from 1982 to present (February 2016) (Supplemental Digital Content 1 and 2, Appendix 1 and 2, http://links.lww.com/JDBP/A117, http://links.lww.com/JDBP/A118). Search terms used were plagiocephaly, nonsynostotic, craniosynostoses, brachycephaly, and developmental disabilities, intellectual disability, motor skill disorders, cognition, comprehension, psychomotor performance, motor skills, child development, intelligence, social behavior, communication disorders, learning disorders, cognition disorders, vision, ocular, and auditory perception. Craniosynostoses was included in the MEDLINE database search as plagiocephaly did not become a MESH term until 2011 and was mapped to craniosynostoses before this date. In addition, reference lists of all included articles were hand searched.

Study Selection

Two reviewers (A.L.C.M. and C.V.-D.) screened all relevant titles and abstracts of the retrieved publications to exclude irrelevant titles, then independently assessed the full reports for eligibility against the study inclusion and exclusion criteria (Table 1) using standardized Forms (Supplemental Digital Content 3, Appendix 3, http://links.lww.com/JDBP/A119).

Data Extraction and Quality

The same reviewers (A.L.C.M. and C.V.-D.) independently extracted data using standardized forms (Supplemental Digital Content 4, Appendix 4, http://links.lww.com/JDBP/A120). Disagreements were resolved by discussion with other authors (W.Y. and B.R.L.). From

Table 1. Inclusion and Exclusion Criteria

Inclusion Criteria
Design
All study types including randomized-controlled trials, cohort studies, case-control studies, cross-sectional, and case series studies of infants and children
Participants
Aged between 0 to ≤18 yr
Conditions
Deformational (positional) plagiocephaly or brachycephaly according to the authors' definition based on clinical diagnosis *or* skull measurement *or* imaging
Outcomes
Any article reporting developmental outcomes such as motor, language, cognition, adaptive behavior.
Exclusion criteria
Studies summarizing what is known about positional plagiocephaly
Studies reporting synostotic plagiocephaly including craniosynostoses
Letters, editorials without new data
Not published in English

studies meeting eligibility criteria, information was extracted on condition, age of participants, study design, sample size, intervention (if any), comparison group/controls, and outcomes/findings reported quantitatively with a corresponding measure of variability/error (SE or SD or confidence interval).

Methodological Quality

The methodological quality of studies was assessed using a critical appraisal tool that we developed from items recommended from a systematic review of quality assessment tools for observational studies.[41] Included studies were independently assessed by A.L.C. Martiniuk (an epidemiologist) against a 7-point rating scale (Supplemental Digital Content 5, Appendix 5, http://links.lww.com/JDBP/A121). Items assessed included (1) population source, (2) exposure, (3) outcome, (4) methods to deal with bias, (5) methods to deal with confounding, (6) use of statistics including adequate power, and (7) declaration of conflicts of interest. Studies were classified as having low (0–2), moderate (3–5), or strong (6–7) methodological quality.

Data Synthesis and Analysis

A meta-analysis was not conducted because of the heterogeneity of plagiocephaly measures and developmental outcomes.

RESULTS

Literature Search

The database search identified 1327 titles following the removal of duplicates. After screening titles and abstracts, 72 full articles were assessed for inclusion, 19 of these met the inclusion criteria and were included in the systematic review[2,14,19,23–37,39] (Fig. 1).

Study Characteristics

No population-based cohort studies were identified by the literature search. Of the 19 studies, most were case series (n = 8) or case-control studies (n = 7). The remaining 4 studies were prospective cohort studies and two[23,29] of these reported outcomes at different time points from the same cohort (Supplemental Digital Content 6, Table 1, http://links.lww.com/JDBP/A124). Studies assessed children at mean ages up to 12 months (n = 10),[2,24–26,28,30–32,37,39] up to 24 months (n = 2),[19,27] and up to 36 months (n = 3).[23,29,35] Other studies examined developmental outcomes for children who had previously been diagnosed with plagiocephaly, with follow-up data presented on 6 and 9-year-old children, respectively (n = 2),[33,36] or at "school-age" (n = 1)[14] or provided data on children aged 3 months to 13 years (n = 1).[34] Study populations were from the United States (n = 10 studies), New Zealand (n = 4 studies), Canada (n = 3 studies), and Finland (n = 2 studies).

Risk of Bias and Quality Assessment

The methodological quality of included studies is presented in Table 2. Most of the studies were rated as having moderate (11/19 studies) or strong (5/19 studies) methodological quality. The most common methodological problems were failing to avoid or minimize bias (18/19), measure or control for confounding (11/19), use appropriate statistical methods (10/19), or declare conflicts of interest (9/19). Selection bias was common among included studies (including referral bias because most of the study groups were enrolled from tertiary care hospitals), "length sampling bias," whereby cases with plagiocephaly with long duration would be more easily included in the studies and often also healthy volunteer bias for the controls.[42] Ten studies used self-report tools completed by parents that were likely affected by recall bias. The direction of effect of these biases is to increase the likelihood of finding an association between plagiocephaly and developmental delay. Several of the studies compared data with developmental test norms. However, some of these norms were set decades ago, before the Back to Sleep Campaign of the 1990s, and thus developmental norms for infants may differ for infants today. The inability to control for potential confounders, including early life factors such as prematurity, hospitalization, time spent in the prone position, torticollis, or the "cohort effect" when comparing with test norms—to name a few—may have biased the estimates of risk toward or away from the null. In about half of the studies, no power calculation was shown and statistical methods rarely used multivariable or other appropriate methods to take into account confounding or test for interactions. Most

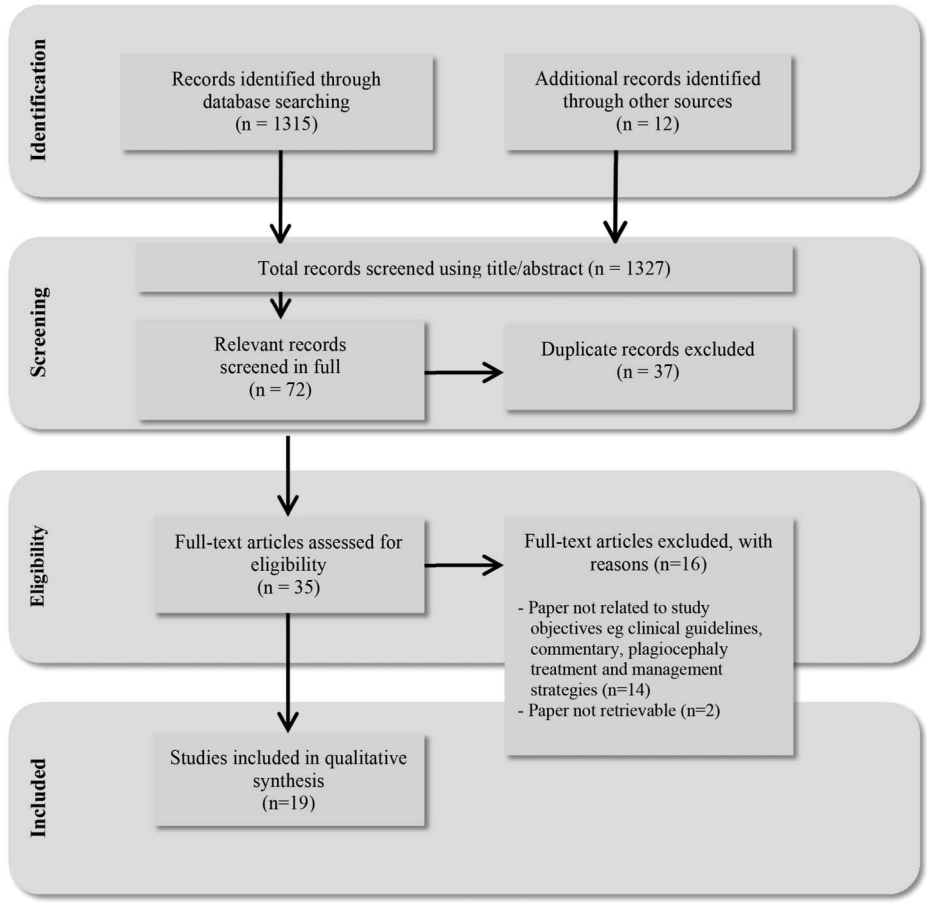

Figure 1. Identification and selection of studies for the review (N = 19).

studies used appropriate measures of exposure (17/19) and outcome (16/19).

Overview of Measurement of Developmental Delay

Studies were categorized into 3 groups: (1) normative referenced developmental screening tools, (2) self-report or interview questionnaire, and (3) physiological measurement (Table 3). For all studies, the highest level of evidence for primary outcome measures provided by each study's authors was reported. Where possible, a size effect and *p*-value or confidence level was reported and where statistical analysis was not performed, provided proportions or percentages were reported instead.

Normative Developmental Screening Tools (n = 11)

Bayley Scales of Infant Development

Six studies[24–29] (33%) used the Bayley Scales of Infant Development (BSID), a validated screening tool that is administered by trained health care professionals to assess young children (1–42 months) across 5 major domains: cognitive, motor, language, social-emotional, and adaptive behavior. Higher scores represent better performance. The participants mean age was similar for 4 studies[24–26,28] (range, 7.2–8.9 months) and 2 studies provided outcomes at longitudinal time points at 18.6[27] and 36.5 months.[29] All these studies used objective measures to define DP such as radiographical[24,25] or 3D imaging[26–29] evidence. The methodological quality of these studies ranged from strong to low (range: 6/7 to 3/7; Table 2). Two studies used the Second Edition (BSID-II), which provides 2 development outcomes measures relevant to this study; the Mental Development Index (MDI) and Psychomotor Developmental Index (PDI). The first of these studies[25] (scored 3/7) examined 46 children with plagiocephaly with a mean age of 8.4 months. Compared with the standardized distributions, significant differences were found in the MDI group where more infants with plagiocephaly were in the "Normal" scoring group (82.6% against an expected 68.7%, $p = .002$). Significant differences were also observed in the PDI group as there was a dramatic increase in the "Severely Delayed" scoring group (13.0% against an expected 1.6%, $p < .001$). No child with plagiocephaly scored within the accelerated groups against an expected rate of 16.5% and 14.8% for the MDI and PDI, respectively. A discrepancy in the tabulated number of children with plagiocephaly against the reported methods is noted (n = 46 against n = 42). The findings of the second study[24] (scored 5/7) of infants whose mean age was 9 months (n = 110) were similar, with no children

Table 2. Methodological Quality of the Included Studies (N = 19)

First Author	Appropriate Source Population	Appropriate Measurement for Exposure (s)	Appropriate Measurement for Outcome (s)	Appropriate Methods to Deal with Bias	Appropriate Methods to Deal with Confounding (Design or Analysis)	Appropriate Use of Statistics, Including Adequate Power	Declaration of Conflicts of Interest (Including by Funding Source)	Score/of 7
Balan et al.[37]	O	—	O	—	O	O	—	4
Collett et al.[27]	O	O	O	—	O	O	O	6
Collett et al.[28]	O	O	O	—	O	O	O	6
Collett et al.[29]	O	O	O	—	O	O	O	6
Fowler et al.[32]	O	O	O	—	—	O	—	4
Habal et al.[34]	O	O	O	—	—	—	—	3
Hashim et al.[2]	O	O	O	—	—	—	—	3
Hutchinson et al.[19]	O	O	O	O	O	O	O	7
Hutchinson et al.[30]	O	O	O	—	—	—	O	4
Hutchinson et al.[23]	O	O	O	—	—	O	O	5
Hutchinson et al.[31]	O	O	O	—	—	—	O	4
Kennedy et al.[39]	O	O	O	—	O	O	O	6
Kordestani et al.[24]	O	O	O	—	O	O	—	5
Korpilahti et al.[35]	O	—	O	—	—	—	O	3
Miller et al.[14]	O	O	—	—	—	—	—	2
Panchal et al.[25]	O	O	O	—	—	—	—	3
Shamji et al.[36]	O	O	—	—	—	—	—	2
Speltz et al.[26]	O	O	O	—	O	O	O	6
Steinbok et al.[33]	O	O	—	—	—	—	—	2

A checklist was developed based on recommendations for scoring study quality.[41] O = Yes denotes criterion was satisfied; — No denotes criterion was not satisfied or where it was unclear if the criterion was satisfied. Methodological quality was defined as low (0–2), moderate (3–5), or strong (6–7).

Table 3. Summary of Association Between Plagiocephaly and Developmental Delay

		Outcome	Notes						
Author	**Instrument**	**Indicates Plagiocephaly Is Associated with Developmental Delay**	**Studies Reporting Educational Outcomes**	**Studies Reporting Outcomes at 2 or More Time Points**	**Subjects Mean/ Median Age: <12 mo**	**Subjects Mean/ Median Age: 13 to <24 mo**	**Subjects Mean/ Median Age: >25 mo**	**Subjects Age Range: 6 wk to 2 yr**	**Subjects Age Range: 3 mo to 10 yr**
Normative Developmental Screening Tools	—	—	—	—	—	—	—	—	—
Kordestani et al.[24]	BSID-II	O	—	—	O	—	—	—	—
Panchal et al.[25]	BSID-II	O	—	—	O	—	—	—	—
Collett et al.[27]	BSID-III	O	—	—	O	—	—	—	—
Collett et al.[28]	BSID-III	O	—	—	—	O	—	—	—
Collett et al.[29]	BSID-III	O	—	—	—	—	O	—	—
Speltz et al.[26]	BSID-III	O	—	—	O	—	—	—	—
Korpilathi et al.[35]	RDLS-III	O	—	—	—	—	O	—	—
Kennedy et al.[39]	AIMS	—	—	—	O	—	—	—	—
Kennedy et al.[39]	PDMS	—	—	—	O	—	—	—	—
Fowler et al.[32]	HINE	O	—	—	O	—	—	—	—
Habal et al.[34]	NCST	O	—	—	—	—	O	—	—
Total studies showing an association	—	9/11	—	—	—	—	—	—	—
Self- Report Tools	—	—	—	—	—	—	—	—	—
Hutchinson et al.[19]	RDQ-II	O	—	O	—	—	—	O	—
Hutchinson et al.[30]	ASQ-3	—	—	—	—	O	—	—	—
Hutchinson et al.[23]	ASQ-3	—	—	O	—	—	O	—	—
Hutchinson et al.[31]	ASQ-3	—	—	O	O	—	—	—	—
Fowler et al.[32]	ASQ-3	—	—	—	O	—	—	—	—
Kennedy et al.[39]	AIMS	?	—	—	O	—	—	—	—
Steinbok et al.[33]	SRQ	?	O	—	—	—	O	—	—
Miller et al.[14]	PI	O	O	—	—	—	O	—	—
Shamji et al.[36]	SRQ	?	O	—	—	—	O	—	—
Habal et al.[34]	NCST	O	O	—	—	—	O	—	—
Total studies showing an association	—	3/10	—	—	—	—	—	—	—
Physiological Measures	—	—	—	—	—	—	—	—	—

(*Table continues*)

Table 3. Continued

Author	Outcome: Instrument	Outcome: Indicates Plagiocephaly Is Associated with Developmental Delay	Notes: Studies Reporting Educational Outcomes	Notes: Studies Reporting Outcomes at 2 or More Time Points	Notes: Subjects Mean/Median Age: <12 mo	Notes: Subjects Mean/Median Age: 13 to <24 mo	Notes: Subjects Mean/Median Age: >25 mo	Notes: Subjects Age Range: 6 wk to 2 yr	Notes: Subjects Age Range: 3 mo to 10 yr
Balan et al.[37]	ERP	O	—	—	O	—	—	—	—
Hashim et al.[2]	ERP	—	—	—	O	—	—	—	—
Collett et al.[28]	MRI and BSID-III	O	—	—	—	O	—	—	—
Balan et al.[37]	ERP	O	—	—	O	—	—	—	—
Total studies showing an association	—	2/3	—	—	—	—	—	—	—

AIMS, Alberta Infant Motor Scales; ASQ-3, Ages and Stages Questionnaire, Third Edition; BSID II, Bayley Scales of Infant Development, Second Edition; BSID III, Bayley Scales of Infant Development, Third Edition; ERP, event-related potentials; HINE, Hammersmith Infant Neurological Assessment; MRI, magnetic resonant imaging; NCST, Nurse Child Satellite Training; PDMS, Peabody Developmental Motor Scales; PDQ-II, Revised Denver Prescreening Questionnaire, Second Edition; PI, phone interviews; RDLS-III, Reynell Developmental Language Scales, Third Edition; SRQ, Self-report Questionnaires, O = Yes denotes criterion was satisfied; — No denotes criterion was not satisfied or where it was unclear if the criterion was satisfied.

in the accelerated groups, a markedly expanded "Normal" group in the MDI series (90% against an expected 68.7%, $p = .0001$) and significantly reduced scores in the PDI series (7% "Severely Delayed" against an expected 1.6%, and 19% "Mild Delay" against an expected 11%, $p = .0001$). The other 4 studies[26–29] used the newer Third Edition (BSID-III) and were conducted by the same cohort of authors, with 3 studies based on the same cohort[27–29] and the fourth unclear.[26] One case-control study[26] (scored 6/7) of infants whose mean age was 7.2 months (n = 233 cases) demonstrated consistently reduced test scores across cognitive (mean difference: −4.68, $p < .001$), language (mean difference: −4.96, $p < .001$), and motor domains (mean difference: −9.96, $p < .001$). In the motor domain, 19.7% of children with plagiocephaly were classified as "Delayed" compared with 9.0% in the control group ($p = .008$). However, the reported "Delayed" proportion for children with plagiocephaly (19.7%) was close to the expected rate in normative data (16.0%), whereas controls were slightly lower (9.0%). This difference may have influenced the studies' significant findings. A longitudinal follow-up study[27] (scored 6/7) of infants whose mean age was 7.9 months (n = 227) with plagiocephaly compared developmental outcomes against controls (n = 232) at 7 (Time 1) and 18.6 months (Time 2), respectively. Excepting gross motor scoring, all Time 2 score differences were larger. Although the authors did not provide the absolute difference for Time 1 for comparison, Time 1 scores were reportedly taken into account in adjusted analyses. At age 18 months, the relative risk (RR) of "Delayed" scoring across adaptive, motor, language, and cognitive scales was elevated in children with plagiocephaly (RR = 1.8, 95% confidence interval [CI], 0.9–3.8; RR = 3.2, 95% CI, 1.1–13.1; 3.9, 95% CI, 1.6–6.2; and RR = 13.8, 95% CI, 1.8–10.5, respectively). This cohort[29] (scored 6/7) was assessed again at mean age of 36 months (Time 3); and the RR of "Delayed" scoring across adaptive, motor, and language scales was further elevated for children with plagiocephaly (RR = 1.7; 95% CI, 0.7–4.4; RR = 4.3, 95% CI, 1.0–17.9; and RR = 7.9, 95% CI, 1.8–35.1, respectively). The RR for cognitive delay was not calculated in this report because no controls were considered "Delayed"; however, 3.1% of children with plagiocephaly were considered "Delayed" in the cognitive domain. Only developmental outcomes were reviewed at Time 2[27] and Time 3[29] to compare against Time 1.[26] Deformational plagiocephaly was not remeasured at Time points 2[27] and 3[29] to determine if the plagiocephaly criteria were still met. The final study[28] (scored 6/7) attempted to associate BSID-III scoring with brain volume and shape in toddlers aged 18.6 months. It found inverse associations of BSID-III motor scores with brain shapes most notably the angle of the corpus callosum. All these studies (6/6) reported a positive association between plagiocephaly and developmental delay[24–29] and had strong to moderate methodological scores (Table 2).

Reynell Developmental Language Scales, Third Edition

The Reynell Developmental Language Scales, Third Edition (RDLS-III) measures receptive and language impairment. Only one study (scored 3/7) used a language outcome to measure developmental delay.[35] This study (n = 61) included children with various skull deformities. There were 29 children whose mean age of 3.3 years identified with plagiocephaly, and 17 of these required surgical correction. Using the Reynell Receptive Scale, children with nonoperated plagiocephaly (n = 12) were 10.31 times more likely to have deficiencies than children with operated plagiocephaly (n = 49); however, this was not statistically significant (p = .14). Specific language impairment was found in 25% of children with nonoperated plagiocephaly (n = 3) compared with an expected 7% in the Finnish population. In the broader context of plagiocephaly, whether surgically corrected or not, 24% (n = 7) were classified as having specific language impairment. Based on the comparison of nonsurgical plagiocephaly and the Finnish population, this study reported a positive association between plagiocephaly and language measures of developmental delay, and had moderate methodological quality (Table 2).

Alberta Infant Motor Scale

The Alberta Infant Motor Scale (AIMS) is an observational scale to measure gross motor maturation from birth to walking in domains of prone, supine, sitting, and standing. Higher scores indicate better performance. In a cohort (scored 6/7) of infants (n = 27), mean age 5.0 months, 18.5% of infants with plagiocephaly scored below the 10th percentile on the AIMS compared with 3.7% in the control group (p = .08).[39] When the authors corrected for other factors, time in prone position was the only factor influencing AIMS score (r^2 = 0.40, p = .01). No association was therefore reported between plagiocephaly and developmental delay. This study had strong methodological quality (Table 2).

Peabody Developmental Motor Scales

The Peabody Developmental Motor Scale (PDMS) was also scored in the same cohort of children (n = 27).[39] This clinical measure of motor skill performance is also norm-referenced. It includes scales of both gross and fine motor acquisition. Although the findings of this study found that infants with plagiocephaly had reduced motor scores compared with controls (22.2% compared with 14.8% with mean developmental quotient <1.5 SD, respectively), this was not statistically significant. No association was therefore reported between plagiocephaly and developmental delay.

Hammersmith Infant Neurological Examination

The Hammersmith Infant Neurological Examination (HINE) is an objective scorecard system for physiologically predictable neuromotor development in infants between 2 and 24 months. Other domains of development such as language and cognition are not included in the HINE. One study[32] (scored 4/7) examined 49 infants, mean age 8.1 months, with plagiocephaly using the HINE and including a muscle tone scoring system. Compared with controls (n = 50), children with plagiocephaly had lower muscle tone scores (p = .03) and greater variability in muscle tone scoring indicating that both hypertonia and hypotonia may play a role in plagiocephaly pathogenesis. This study suggests a positive association between plagiocephaly and gross motor developmental delay. This study had moderate methodological quality (Table 2).

Nurse Child Assessment Satellite Training

One further study[34] (scored 3/7) examined 2 cohorts of children with plagiocephaly (n = 25 and n = 14, respectively; total n = 39), mean age 22 months. Using a composite of validated screening tools that included the Teach subscale of the Nurse Child Assessment Satellite Training (NCST) (scored observation of interaction between parent and child), the Denver Developmental Screening Test (screening for developmental problems including a behavioral checklist in preschool children, but does not screen specifically for cognitive problems), and the Boston Naming Test (testing for word retrieval/aphasia), children with plagiocephaly (n = 35) met the criteria for disorders of psychosocial, psychomotor, interactive, and cognition/language domains (29%, 34%, 40% and 51%, respectively). This study suggests a positive association between plagiocephaly and developmental delay. Moderate methodological quality was found for this study (Table 2).

Parent Report Tools (n = 10)

Ages and Stages Questionnaire, Third Edition

The Ages and Stages Questionnaire, Third Edition (ASQ-3) is a popular and validated parent-completed screening tool that involves both parental recall and task-oriented testing at the time of assessment. It covers five domains (communication, gross motor skills, fine motor skills, problem-solving, and personal-social skills) for children aged 2 to 60 months. Scores below the cutoff correspond with higher risk of developmental delay in that specific domain. Four of the 19 studies (22%) in this review used the ASQ-3.[23,30–32] The methodological quality of these studies ranged from strong to moderate (range: 7/7–4/7; Table 2). In a case series study[30] (scored 4/7), 287 children referred for plagiocephaly were classified as noncases (62/287; 21.8%), brachycephalic (47/287; 16.5%), plagiocephalic (107/287; 37.5%), and both (69/287; 24.2%). In infants defined as cases, the greatest number of "high risk of delay" categories was seen in the gross motor domain (18% of infants), followed by problem solving (17%), personal-social (15%), fine motor (14%), and communication (7%), with 36% of infants identified as having one or more delay risk category. A second case series[23] (scored 5/7) followed up a cohort of 129 infants with deformational plagiocephaly at 3 and 4 years of age (80% follow-up), and demonstrated significant amelioration of ASQ-3 risk categories over the average follow-up period

of 47 months. Initially, 41% and 22% of the cohort had one or two risk factors of delay, respectively, on the ASQ-3. This reduced to 11% and 2%, respectively, at the end of follow-up, which was comparable with expected (15%) rates in the ASQ-3 normative data ($p = .26$). A third study with a prospective cohort design[31] (scored 4/7), assessed developmental outcomes at 3, 6, and 12 months of age in 126 children with plagiocephaly. At 12-month follow-up, the risk categories measured by the ASQ-3 reduced to 23% from an initial 30%, and were predominantly found in gross motor function, with 10% of infants experiencing more than 4 delays in total over all 4 assessments. These delays approached expected levels of development by 17 months. The final study[32] (scored 3/7), was a case-control design and demonstrated a difference in tone scoring and tone variability in infants (mean age: 8.1 months; range: 4–13 months) with plagiocephaly compared with infants without plagiocephaly ($p = .003$). The abnormality observed was variable tone, not simply decreased tone. This case-control study did not demonstrate a difference in gross or fine motor scores on the ASQ-3. Overall, none of these studies report a long-term association between plagiocephaly and developmental delay.

The Revised Denver II Prescreening Questionnaires

The Revised Denver Prescreening Questionnaire, Second Edition (PDQ-II) is a parent self-report developmental screening tool assessing several developmental domains including motor skills. A study[19] with strong methodological quality (7/7; Table 2) used a prospective cohort design to follow-up 200 infants at 5 time points (6 weeks, 4 months, 8 months, 12 months, and 2 years). The PDQ-II was used to ascertain delays, and activity level was evaluated using a crude activity scale (0–10). At 4 months, cases with plagiocephaly were more likely to have low activity level (adjusted odds ratio [OR]: 3.28; 95% CI, 1.16–9.29) and an abnormal result on the 6-week PDQ-II (crude OR: 18.06; 95% CI, 1.96–166.54). No adjusted OR was reported by study authors for the 6-week PDQ-II result. Results at other time points using these measures were not reported. A positive association between plagiocephaly and developmental delay is suggested at the 4-month time point in this study.

Other Self-Report Tools

There were five[14,33,34,36,39] studies in this category. The methodological quality of these studies ranged from strong to low (range: 6/7–2/7; Table 2) with more than half scoring low. One study[39] with strong methodological quality (score 6/7; Supplemental Digital Content 6, Table 1, http://links.lww.com/JDBP/A124) used a parental recording diary of prone time to establish an association of prone time to the AIMS score as previously described in infants whose mean age was 5.1 months. A case series[33] (score 2/7) using self-reported questionnaires for children whose mean age was 8.9 years found 34% of cases (n = 21) had received learning assistance, whereas 14% were in a special class. This study had a low response rate of 23% and no objective standardized assessment for development and scored low on methodological quality (Table 2). A case study[14] (scored 2/7) using phone interviews to determine whether there was an increased rate of developmental delay in "school-aged" children who had presented as infants with plagiocephaly also had low methodological quality (Table 2). The families self-reported that 39.6% of cases (n = 63) showed evidence of developmental delays as they received special help in primary school including; special education assistance, physical therapy, occupational therapy, and speech therapy. They determined that children with plagiocephaly were more likely to require special education services in school compared with 7% of the controls (n = 91) needing similar services. It is unclear how each child was referred for special assistance, which is a limitation of this study. The cases were identified as having plagiocephaly between 1980 and 1991, before the Back to Sleep campaign, which may result in cohort differences between this group of identified cases, and more recently identified cases. We note inconsistencies in reporting of sizes of the group without delay: in places, 36 cases with no delay are reported, whereas in other places, 38 children with no delay are reported. Another case series[36] (scored 2/7) of children whose median age was 6 years collected parental responses to a questionnaire that tested cosmetic and cognitive impressions and encompassed a spectrum of both objective and subjective questions. Parental impression of developmental delay (n = 80) was higher than the expected population percentage: 21% with language difficulties, 28% with motor difficulties, and 15% requiring special education against the expected 5% to 6%. Left-sided plagiocephaly was strongly related to requiring special education (27 vs 10%, $p < .05$), evidence of fine motor delay (41 vs 22%, $p < .05$), and speech delay (36 vs 16%, $p < .05$). This study had low methodological quality (Supplemental Digital Content 6, Table 1, http://links.lww.com/JDBP/A124). The final study[34] (scored 3/7) in this category assessed 39 infants and children with plagiocephaly ranging from 3 months to 10 years age using composite scores from differing metrics described earlier in this review. In this study, the self-reported outcome measures, which included parental and school reporting, were collated into one categorical scale, which found children displayed high rates of delays/disorders in psychosocial (29%), psychomotor (34%), interactive skills (40%), and cognitive language (51%) areas. This study had moderate methodological quality (Supplemental Digital Content 6, Table 1, http://links.lww.com/JDBP/A124). Only 2 of these studies (2/5) reported a positive association between plagiocephaly and developmental delay[14,34] (Table 3).

Physiological Measurement (n = 3)

Auditory Event-Related Potentials

The 2 studies in this category had moderate methodological quality (Table 2) and mixed findings. A case-control study[37] (scored 4/6) published findings which

determined that auditory event-related potentials (ERPs) revealed brain dysfunction in infants whose mean age was 11.1 months with plagiocephaly. Children with plagiocephaly (n = 15) differed from the controls (n = 15) with respect to reduced P150 ($p < .002$) and N250 amplitudes ($p < .002$). The authors concluded that the children diagnosed with plagiocephaly were at a greater risk of auditory processing disorders. In contrast, a recent case-control study[2] (scored 3/7) found infants, mean age 7.3 months, with plagiocephaly (n = 16) did not differ from controls (n = 18) with respect to the maximum P150 amplitude and latency. No significant P150 abnormalities were observed over the frontal or central scalp, the responses indicated normal auditory processing in the infants with plagiocephaly. Only 1 of the 2 studies therefore found a positive association between plagiocephaly and reduced ERP amplitudes.

Physiological Measurement: Magnetic Resonance Imaging

This study[28] (scored 6/7) compared magnetic resonance imaging measurements with BSID-III outcomes. It examined 20 children, whose mean age was 7.9 months, with a history of plagiocephaly and compared them with a control group of 21 children. The MRI measurements were taken from the participants and only brain asymmetry, rather than total volume, was significantly associated with plagiocephaly ($p < .001$). Of note, motor development was positively correlated with posterior brain length (Beta = .44, $p = .015$). Brain width and width-length ratio however was inversely related to BSID-III motor scores (Beta = −.55 and −.42, respectively; $p = .002$ and .014, respectively) and the angle of the corpus callosum was inversely related to BSID-III motor scores (Beta = −.61, $p < .001$). The strength of these associations and the magnitude of the findings strongly suggest that the metrics used by the BSID-III are sensitive enough to screen for subtle motor deficits associated with underlying measurable intracranial anatomical changes. This study has strong methodological quality (Table 2) and suggests a positive association between plagiocephaly and developmental delay.

Summary

Of included papers in the systematic review, the highest level of evidence comes from those with "strong" methodological score ratings indicated in 6 studies.[19,26–29,39] These included prospective cohorts (n = 3)[19,27,29] and case controlled studies (n = 2).[26,28,39] Six of these studies used normative referenced developmental tools (BSID III [3/5],[26–29] AIMS [1/5],[39] PDMS [1/5]),[39] one used a parent self-report measure, PDQ-II (1/5)[19] and one measured brain volumes through MRI imaging.[28] All (6/6) these studies[19,26–29,39] found plagiocephaly to be associated with developmental delay and this delay was detected from 6 months to 3 years (Table 3). Developmental outcomes used to indicate this included motor (5/6),[26–29,39] language (5/6),[19,26–29] cognition (4/6),[26–29] and self-report (1/6).[39] Regardless of methodological quality, 68% of the 19 studies (13/19) in this review suggest an association between plagiocephaly and developmental delay, whereas 16% (3/19) were inconclusive (Table 2). This relationship was shown more clearly in studies using normative developmental screening tools (9/11) and physiological measures (2/3) compared with self-report tools (3/10) (Table 3). Of those studies that suggest a positive association (13/19), 75% reported this in subjects whose mean/median age was ≤24 months (9/12)[19,24–28,32,34,37] compared with only 43% (3/7) of studies where the subjects were in the mean age of >24 months[14,29,35] (Table 3).

DISCUSSION

This review identified 19 studies that reported developmental outcomes in infants and children with plagiocephaly. The association between plagiocephaly and developmental outcomes was explored. This review indicates that a positive association exists, with delay most commonly reported in motor domains followed by language. Our conclusion is supported by the replication of this finding in 12 of 19 studies, including 4 of 5 studies with the highest methodological quality and the correlation of altered brain dimensions using MRI with BSID-III scores in one study.[28] Most studies reporting developmental delay were in infants less than 2 years old, with associations in older children not as strong. The normative developmental screening tools and physiological measures showed this relationship most consistently.

The finding that an association exists between plagiocephaly and developmental delay is not surprising given head control is a core component of early development.[43] Similarly, the finding that the motor domain was the most commonly affected aligns with early intervention motor-based strategies, which show that conservative interventions such physiotherapy are effective in reversing plagiocephaly and in many cases developmental delay.[7] This reviews shows the higher quality of evidence studies (both "strong"[19] and "moderate"[31] quality of evidence scores) demonstrate an association between plagiocephaly and early developmental delay. Most cases with developmental delay will have resolved by 2 years of age. "Moderate" quality of evidence scores[23] also exists of improvement by 4 years of age. These studies provide the basis of reassurance to parents by clinicians and guide the importance of correctly identifying infants at risk of not improving.[23,24] Factors identified in this review that may isolate infants at higher risk of long-term developmental sequelae of plagiocephaly include the presence of low or variable tone,[32] left-sided plagiocephaly[36] (given the majority of presentations are right sided)[13,44] and delay in multiple developmental domains.[29] Long-term implications of plagiocephaly are less clear mainly because of

the poorer methodological quality of these studies including self-report instruments, poor follow-up response rates, and classification of school aged delay. Previous systematic reviews have explored prevalence, risk factors, and natural history of plagiocephaly,[45] or the conservative,[7] nonsurgical,[46] and helmet treatment[47] of plagiocephaly, but none have explored the association between plagiocephaly and developmental delay.

Strengths and Limitations

The key limitation found in studies included in this systematic review is sources of bias. They include selection bias, where cases were frequently drawn from craniofacial centers at tertiary hospitals, and sample bias because of low response rates occasionally as low as 23% of the eligible follow-up sample in one study.[33] Many papers were from the same study population or same study group, i.e., the Seattle United States group had 5 publications,[14,26–29] the Auckland NZ group had 4 publications[19,23,30,31]; however, many of these were prospective cohorts. The most obvious source of bias was the difficulty of blinding assessors (whether clinicians or parents) to developmental delay outcomes as plagiocephaly is visually easy to detect. Report bias may have played a role in parent self-report measures, where only 22% of studies showed a positive association suggesting parents may not be aware or deny delay. Other limitations include the inability to control for confounders such as positioning and limited developmental input, which may have contributed to developmental delay or conversely early intervention, which may have ameliorated developmental delay.

The strengths of this study include the use of a sensitive and extensive search strategy including hand searching of included studies to locate the best available evidence to reduce bias. Two reviewers screened all titles and extracted data from the final 19 retrieved papers to ensure accuracy of data extraction. A critical appraisal tool was developed according to recognized methods to rate the quality of evidence and studies with least bias. Although many studies were linked to the one study population, included studies were derived from several countries including Canada, the United States, New Zealand, and Finland suggesting these findings are generalizable at least to Westernized countries. The review is reported according to PRISMA (Preferred Reporting Items for Systematic reviews and Meta-Analyses)[40] guidelines.

Future Directions

Further longitudinal studies may help delineate what is considered pathological from normal variation by means of clinically relevant deficits that do not self-correct in the intermediate term (4–5 years). This systematic review was originally developed to determine the causative relationship between plagiocephaly and developmental delay, i.e., whether preexisting developmental delay caused plagiocephaly, or whether preexisting plagiocephaly caused developmental delay, but no study design allowed for interrogation of this question.

Our most important recommendation for future research therefore is the development of a prospective cohort following all children born, ideally in a community or nontertiary care hospital and followed every 3 months up to primary school age to assess for plagiocephaly as well as any potential long-term developmental health and educational impacts. Multiple confounders (or potential explanatory factors) need to be measured in future research including: age, gender, ethnicity, socioeconomic status of family, education of mother, birth weight, prematurity, hospitalizations, time spent prone, time spent in supported sitting or standing positions, time spent on back/sides, muscle tone, presence of torticollis, activity levels, and behavioral characteristic of the child. Samples should exclude or measure other conditions known to predispose to developmental delay as well as any interventions applied for plagiocephaly (e.g., physiotherapy, helmet). Objective measures of plagiocephaly and standardized developmental outcomes need to be used by clinicians and ideally compared with control populations versus normative data as often the normative data are very old. Replication of prospective cohorts in differing cultural settings with contrasting infant care practices, e.g., transporting babies in prams (Westernized countries) versus baby carrying devices (developing countries) may show differences in severity of plagiocephaly and longevity of developmental delay.

CONCLUSION

This is the first systematic review to examine and then also provide evidence of an association between plagiocephaly and developmental delay. Although the longevity of this relationship beyond 2 years of age is unclear from the available evidence, our findings indicate that plagiocephaly is a marker of elevated risk of developmental delay. Clinicians should closely monitor infants with plagiocephaly for this. Prompt referral to early intervention services such as physiotherapy may ameliorate motor delays and identify infants with potentially longer term developmental needs.

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