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Impact of Parent Practices of Infant Positioning on Head Orientation Profile and Development of Positional Plagiocephaly in Healthy Term Infants

Amy Leung [a,b], Allison Mandrusiak[a], Pauline Watter[a], John Gavranich[c], and Leanne M. Johnston[a]

[a]School of Health and Rehabilitation Sciences, The University of Queensland, St Lucia, Queensland, Australia; [b]Child Development Program, Brisbane, Queensland, Australia; [c]Ipswich Hospital, Ipswich, Queensland, Australia

ABSTRACT

Aims: The influence of infant positioning on the development of head orientation and plagiocephaly is not clear. This study explored the relationship between infant body and head positioning, with the development of asymmetrical head orientation and/or positional plagiocephaly. *Methods:* Clinician measurement of head orientation profile and parent-reported infant positioning data were collected for 94 healthy term infants at 3, 6, and 9 weeks of age. Plagiocephaly was measured at 9 weeks with the modified Cranial Vault Asymmetry Index. *Results:* More severe plagiocephaly was associated with longer *supine-sleep-maximum* ($p = 0.001$) and longer *supine-lying-total* ($p = 0.014$) at 6 weeks. Prone positioning was not associated with plagiocephaly. Parent-reported head asymmetry during awake and sleep time at 3 weeks identified infants with clinician-measured head asymmetry at 9 weeks. Better symmetry in head turning was associated with more *side-lying-total* time by 9 weeks ($p = 0.013$). *Conclusions:* Our results showed that infant positioning is associated with early head orientation and plagiocephaly development. Early parent-reported asymmetry during awake and sleep time is an important indicator for the need for professional assessment and advice. A *Plagiocephaly Prevention Strategy* and *Plagiocephaly Screening Pathway* are provided for clinicians and parents.

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Community; head orientation; infant positioning; plagiocephaly; parent practice

Parenting choices around infant positioning are now largely guided by international best practice recommendations associated with preventing Sudden Infant Death Syndrome—which recommend that infants should sleep in supine at all times (Adams, Good, & Defranco, 2009; American Academy of Pediatrics Task Force on Infant Positioning and SIDS, 1992, 2000). This recommendation has been followed by a significant increase in the incidence of positional plagiocephaly in infants (Argenta, David, Wilson, & Bell, 1996; Kane, Mitchell, Craven, & Marsh, 1996; Loveday & de Chalain, 2001; McKinney, Cunningham, Holt, Leroux, & Starr, 2008; Turk, McCarthy, Thorne, & Wisoff, 1996). Positional plagiocephaly is the most common atypical asymmetrical head shape. It occurs in up to 33% of infants within the first year of life, with a peak around 2–3 months (Aarnivala et al., 2015; Boere-Boonekamp & van der Linden-Kuiper, 2001; Hutchison, Hutchison, Thompson, & Mitchell, 2004; Leung,

CONTACT Amy Leung yuen.leung@uqconnect.edu.au Division of Physiotherapy, School of Health and Rehabilitation Sciences, The University of Queensland, St. Lucia, 4072, Australia.
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Mandrusiak, Watter, Gavranich, & Johnston, 2016; Peitsch, Keefer, LaBrie, & Mulliken, 2002; van Vlimmeren et al., 2007). It occurs twice as often on the right side compared to the left (Hutchison, Stewart, & Mitchell, 2009; Hutchison, Thompson, & Mitchell, 2003; Leung, Watter, & Gavranich, 2014; van Vlimmeren et al., 2007, 2008); and up to twice as often in boys compared to girls (Bialocerkowski, Vladusic, & Ng, 2008; Boere-Boonekamp & van der Linden-Kuiper, 2001; Hutchison et al., 2003; Kelly, Littlefield, Pomatto, Manwaring, & Beals, 1999; Leung et al., 2014; Oh, Hoy, & Rogers, 2009). Long-term unresolved significant plagiocephaly can contribute to considerable and persistent craniofacial deformity and/or musculoskeletal or developmental implications (de Chalain & Park, 2005; Rekate, 1998; Roby, Finkelstein, Tibesar, & Sidman, 2012; Shamji, Fric-Shamji, Merchant, & Vassilyadi, 2012; Steinbok, Lam, Singh, Mortenson, & Singhal, 2007; van Wijk et al., 2014).

Two mechanisms have been proposed to explain deformation of the neonatal and/or infant cranium. Initially, authors proposed the "Water balloon theory," which suggests that cranial deformity is caused by external force exerted on a certain area of the soft and malleable skull bones (Littlefield et al., 1998; Persing et al., 2003). More recently however, Rogers (2011a) has proposed the "Pumpkin Analogy" which suggests that the development of a flat spot on the skull is similar to the asymmetrical growth in pumpkins which occurs when they are growing against a hard surface. In infants, this reflects the occipital flattening that occurs when cranial expansion and growth are consistently restricted in the area of contact with the supporting surface. Thus, brain growth is re-directed over time to the area where there is no resistance, causing corresponding skull expansion in that area. This is particularly significant in young infants, who are experiencing rapid growth while spending prolonged periods of time in supine with their head in one position, usually turned to one side (Hutchison et al., 2004). Therefore, infant body positioning and parent practices of head positioning are important factors to consider in prevention and intervention protocols for plagiocephaly.

Many parents receive information about plagiocephaly prevention and head orientation preference intervention strategies during inpatient maternity and/or outpatient child health visits (Aarnivala et al., 2015; Cavalier et al., 2011; Hutchison, Stewart, & Mitchell, 2007; Lennartsson, 2011; Persing et al., 2003; Rogers, 2011b). Studies investigating the relationship between infant body positioning and the development of plagiocephaly have produced mixed results. Some studies have found the *prone-play* position (sometimes termed tummy play) to be a protective factor against plagiocephaly development when this is encouraged for more than five minutes per day, and a risk factor for plagiocephaly development if the duration was less than 5 minutes per day, or the frequency was less than three occasions per day (Hutchison et al., 2003; van Vlimmeren et al., 2007). However, in other studies neither duration nor frequency of *prone-play* were associated with plagiocephaly (Hutchison et al., 2004; Mawji et al., 2014).

A second risk factor, the cumulative time spent in supine when awake and asleep *(supine-lying-total*) has been associated with presence of plagiocephaly at six weeks when the duration is more than 19 hours per day (Hutchison et al., 2004; Hutchison et al., 2003). On the contrary, others have found no difference in supine sleeping time in infants with and without plagiocephaly aged between two to seven weeks (van Vlimmeren et al., 2007) or four months (Cavalier et al., 2011). Further prospective research is needed to investigate the influence of *prone-play* and *supine-lying-total* on plagiocephaly development. In this context, it would also be relevant to investigate other potentially important positioning factors such as the *supine-sleep-maximum,* which would reveal whether there is any differential impact of prolonged versus intermittent periods of force on the cranial surface during long sleeps versus short naps.

The influence of asymmetrical head orientation (AHO) on the development of plagiocephaly has also received some attention (Boere-Boonekamp & van der Linden-Kuiper, 2001). Up to 18% of all infants demonstrated AHO on clinical examination within the first few months of life (Boere-Boonekamp & van der Linden-Kuiper, 2001; Gesell, 1938; Hopkins, Lems, Janssen, & Butterworth, 1987; Michel, 1981; Rönnqvist & Hopkins, 1998; van Vlimmeren et al., 2007, 2008), which is identified when an infant turns their head to one side in supine >75% of the time. However, findings from clinical examination may underestimate AHO, with more parent-reported AHO prevalent, up to 43.4% (van Vlimmeren et al., 2007) or 58% (Mawji et al., 2014). The relative contribution of AHO when an infant is asleep versus awake has not been determined. Further, the relationship between parent-reported AHO and clinically measured AHO, and plagiocephaly development are unknown. Our recent research has shown that infants with AHO on awake clinical examination at 3-weeks and 6-weeks were more likely to demonstrate plagiocephaly at 9-weeks of age (Leung et al., 2016). Further research is needed to explore the associations between infant positioning and its impact on AHO and positional plagiocephaly.

The aim of this study was to explore the relationship between infant body positioning and parents' use of proactive or corrective infant head positioning practices, and the presence of AHO, and/or positional plagiocephaly in healthy young infants. The first and second aims were to determine whether parent positioning practices (across 24 hours) are related to: (1) AHO—as measured clinically at 3, 6, and 9 weeks by Head Orientation Duration (*HOD*), Head Orientation Strength (*HOS*), and Latency To Turn (*LTT*) from each side and/or (2) positional plagiocephaly—as measured at 9 weeks by the modified Cranial Vault Index (*mCVAI*). The third and fourth aims were to examine the relationships between parent-reported AHO at 3, 6, and 9 weeks and (1) clinical measurement of AHO measured at 3, 6, and 9 weeks and (2) plagiocephaly measured at 9 weeks.

Method

This study was part of a larger prospective study of healthy term infants conducted between June 2011 and July 2013 (Leung et al., 2016). The study received approval from Human Research Ethics Committees of The University of Queensland (EC00179) and West Moreton Hospital and Health Service (EC00184). Parents provided written informed consent.

Participants

Full term infants and their parents were recruited through community child health clinics, antenatal clinics and as community volunteers. Infants were excluded if they presented with: APGAR scores of <7 at 1 minute or 5 minutes, with an identified neurological insult; low birth weight (less than 2500g at term); or a diagnosed medical or orthopedic condition.

Measures

Head Orientation Profile (HOP)

Head orientation duration (*HOD*), head orientation strength (*HOS*) and latency to turn (*LTT*) were used to determine each child's HOP according to our previously published protocol (Leung et al., 2016). The protocol is based on that used in Goodwin and Michel (1981) study which was reported to have high inter-rater reliability. *HOD* was calculated as the time that the infant's head was in each position (*HOD-midline, HOD-left, HOD-right*) after commencing in four start positions (midline-right-left-midline or midline-left-right-midline) with each start position held for 60 s by the examiner, then examined for 60 s (total = 240 s). In this study, we were interested in the absolute value of the difference between *HOD-right* and *HOD-left*, which yields the *HOD-diff*. When there is more preference to one side, there is an increase in *HOD-diff*.

HOS was calculated by extracting head position at each 10s interval from the observational data for the four conditions combined. Counts for each head position were summed, then *HOS* was calculated according to the formula: $HOS(\text{z-score}) = (R-L)/(R+L)^{1/2}$ (Goodwin & Michel, 1981).

LTT was defined as the duration taken for the infant to rotate their head from the initial position (in seconds) (Cornwell, Barnes, Fitzgerald, & Harris, 1985). In this study, the difference in *LTT* from the preferred side compared to the non-preferred side was used to determine the *LTT-diff*, which increases when there is more preference to one side. In all measures, raw scores were positive if there was a right preference and negative if there was a left preference. Absolute values were used to enable data to be pooled for right and left cases.

Plagiocephaly

Anthropometric measurement of plagiocephaly was performed using the modified Cranial Vault Asymmetry Index (*mCVAI*) (Leung, Watter, & Gavranich, 2013). As for AHO measures, the absolute value of *mCVAI* was used in this paper in order to pool right and left cases. The cut-off point indicating significant plagiocephaly was set at 3.5% (Leung et al., 2013; Loveday & de Chalain, 2001). Infants with a *mCVAI* $< 3.5\%$ were categorized as having non-significant plagiocephaly and those with a *mCVAI* $\geq 3.5\%$ were categorized as having significant plagiocephaly.

Infant Positioning Survey

We developed a structured parent completed survey to record parents' recall of their infants' positioning over the most recent 3-day period. Parents reported their infants': (1) body positions in 24-hour period (*supine-lying-total*, *prone-lying-total*, *prone-play* [≤ 5 mins or > 5 mins], *side-lying-total* and *upright-total*); (2) *supine-sleep-maximum* (in one sleep); (3) main sleeping positions at night or during the day (*supine-main-sleep-position*); (4) AHO; (5) proactive practice of alternating their infant's head when placing them down to sleep (*alternate-head-positioning*); (6) corrective practice of turning their infant's head to the non-preferred side when placing them down to sleep (*passive-head-repositioning*), and (7) corrective active stimulation of their infant's head to turn to the non-preferred side (*active-head-stimulation*). Parent-reported AHO was divided into four categories: (1) during asleep and awake time (*AHO-Both*); (2) asleep time only (*AHO-Sleep*); (3) awake time only (*AHO-Awake*); (4) no asymmetry during asleep or awake time (*AHO-None*).

Procedures

Testing occurred at the infant's home or a community health center at 3 weeks, 6 weeks, and 9 weeks of age +/− 6 days. At the time of assessment, infants were in a quiet alert state. If crying occurred, a pause was taken and parents were encouraged to settle their infant before testing recommenced. During testing infants were positioned in supine and away from bright lights and colorful visual signs within their visual field. Infants were assessed on head orientation profile and *mCVAI*. Parents also completed the survey to record their practices of infant positioning at home during the previous three days.

Data Analysis

Data were analyzed using STATA version 14.1 (StataCorp, 2015). Descriptive statistics including frequencies (percentages), means and standard deviations (SD) were computed for the dependent variables (*HOD-diff, HOS, LTT-diff, mCVAI*); independent variables were parent-reported infant asymmetrical head orientation, infant body positioning time, parent practices of infant head positioning and main sleep positions. Paired *t*-test was used to examine change in infant body positioning across all age periods. Head orientation profile variables (*HOD, HOS and LTT-diff*) with mixed effect modeling were used to test the association between infant body positioning and their parent's practices of infant head positioning.

Uni-variate linear regression was used to investigate the plagiocephaly severity (*mCVAI*). Logistic regression was used to investigate plagiocephaly categories, with infant body positioning variables and parent practices of infant head positioning as co-variants. In both linear and logistic regression modeling, those independent variables with $p < 0.10$ (Bursac, Gauss, Williams, & Hosmer, 2008) were tested for their collinearity and then included for further analysis in multiple regressions with stepwise backward elimination. The assumptions of homoscedasticity, outliers, and normality were evaluated for the final models.

The association of parent-reported infant asymmetrical head orientation with clinically measured HOP was analyzed using pair-wise *t*-test. Odd ratios (OR), standardized coefficients (β), percentage of variance (r^2), and rho (intra-class correlation) were reported. The confidence level (CI) was set at 95% and p-value < 0.05 was established as the level of significance in all analyses.

Results

Ninety-four parents consented for their infants to participate. The mean (±SD) age of the infants at 3 weeks was 21.4 (± 2.29) days; at 6 weeks was 42.1 (± 2.19) days and at 9 weeks was 63.2 (± 2.53) days. Demographic data were presented in an earlier study (Leung et al., 2016). Seventy-two (77%) infants completed all three assessments, 13 (14%) completed two assessments and nine (10%) completed only one assessment. Eighty-two infants (87%) had mCVAI measured at 9 weeks.

Descriptive statistics for infant positioning and position tolerance, parent-reported AHO, and parent practices of infant head positioning across all ages are presented in Table 1. The time spent in supine was consistent across all ages. Infants spent more time in prone ($p = 0.009$) and upright ($p = 0.025$) but less time in side-lying ($p = 0.004$) at 3 weeks compared to 6 weeks and then these times stayed consistent at 9 weeks. Infants increased time for their *supine-sleep-maximum* from 3 weeks to 9 weeks ($p < 0.001$). There was an increase in the number of infants who were able to tolerate prone play for >5 mins between 3 weeks and 6 weeks ($p = 0.041$) and between 6 weeks and 9 weeks ($p = 0.004$).

The number of infants without AHO (*AHO-None*) increased between 3 weeks and 6 weeks ($p = 0.004$) and between 6 weeks and 9 weeks ($p = 0.03$). The number of infants who presented with *AHO-Sleep* and *AHO-Awake* did not differ across all ages. There was a reduction in infants with *AHO-Both* from 6 weeks to 9 weeks ($p < 0.001$). *Alternate-head-positioning* was performed by an average of 57% of parents at 3, 6, and 9 weeks. Parents who practiced *alternate-head-positioning* were either aware of the strategy from antenatal or postnatal advice, or had noticed their infant was starting to develop some degree of AHO. There was

Table 1. Parent-reported (A) infant positioning time, (B) infant position tolerance, (C) infant asymmetrical head orientation, and (D) their practice of infant head positioning.

	3 weeks ($n = 91$)		6 weeks ($n = 75$)		9 weeks ($n = 83$)	
	Mean (SD)	(Min, max)	Mean (SD)	(Min, max)	Mean (SD)	(Min, max)
(A) Infant positioning (hours/day)						
Upright-total	2.7 (1.4)	(0.5, 8)	3.3 (2.2)	(0.5, 14)	3.7 (2)	(1, 12.5)
Side-lying-total	5.8 (3.7)	(0.5, 18)	4.8 (2.5)	(0.5, 12)	4.5 (2.5)	(0, 14)
Prone-lying-total	1.9 (1.5)	(0, 7)	2.8 (3.3)	(0.2, 18)	2.8 (3.4)	(0.1, 18)
Supine-lying-total	14.0 (4.6)	(1, 21.5)	13.3 (4.6)	(1, 21.5)	13.5 (4.5)	(1, 21)
Supine-sleep-maximum	4.9 (1.5)	(1.5, 10)	5.5 (1.5)	(3, 10)	6.5 (1.9)	(2, 12)
	Frequency	% Yes	Frequency	% Yes	Frequency	% Yes
(B) Infant position tolerance						
Supine-main-sleep-position	87	95	69	92	76	92
Prone-play position						
Tolerates $\leq$ 5 mins	38	58	27	39	34	43
Tolerates $>$ 5 mins	28	42	42	61	45	57
(C) Infant head preference (yes/no)						
Neither awake or asleep (AHO-None)	39	42	37	49	44	53
When asleep only (AHO-Sleep)	30	33	14	19	25	30
When awake only (AHO-Awake)	10	11	4	5	1	1
Both awake and asleep (AHO-Both)	13	14	20	27	13	16
(D) Parent positioning practices						
Alternate-head-positioning	55	60	45	60	43	52
Passive-head-repositioning	33	36	28	38	28	33
Active-head-stimulation	17	19	24	32	13	16

Note. SD = standard deviation; Min = minimum; max = maximum.

a trend for *alternate-head-positioning* to decrease if the sole reason for use was awareness of the recommendation (64% at 3 weeks, 53% at 6 weeks, 47% at 9 weeks). Likewise, there was a reduction in the *passive-head-repositioning* ($p < 0.001$) and *active-head-stimulation* ($p = 0.003$) from 6 weeks to 9 weeks.

Relationships Between Clinical Measures of HOP and Infant Positioning

Uni-variate regression showed a greater *HOD-right* was associated with parents commencing *active-head-stimulation* at 3 weeks and 9 weeks, however, only *active-head-stimulation* at 9 weeks was significant in the multiple regressions (Table 2, Model 1). For infants with greater *HOD-midline*, parents were more likely to have implemented proactive *alternate-head-positioning* by 6 weeks and were marginally less likely to perform *active-head-stimulation* at 9 weeks (Table 2, Model 2). There was no association between *HOD-left* with infant body or head positioning practices. Stronger *HOS* asymmetry was associated with parents commencing *active-head-stimulation* at 9 weeks and marginally associated with the infant's *supine-main-sleep-position* at 6 weeks (Table 2, Model 3). More symmetry in head turning (smaller *LTT-diff*) was associated with more time spent in s*ide-lying-total* position by 9 weeks (Table 2, Model 4).

Relationships Between Clinical Measures of HOP and Parent-Reported AHO

Infants whose parents identified them as having *AHO-Both* at 3 weeks, were observed clinically to have more *HOD-diff* and stronger *HOS* asymmetry than other groups combined (Table 3). When parents did not identify their infants as having AHO at 6 weeks and 9 weeks,

Table 2. Significant results of (A) mixed effect modeling analysis of clinical measurement of head orientation profile variables (HOD, HOS, LTT-diff), and (B) positional plagiocephaly, with infant body and head positioning.

	Mixed effect modeling			
Clinical measure of infant head orientation	β	95% CI	*p*-value	rho
(A) Head orientation profile (3, 6, 9 weeks)				
Model 1: HOD-right				
Active-head-stimulation at 9 weeks	9.59	0.12–19.06	0.047	0.22
Model 2: HOD-midline				
Alternate-head-positioning at 6 weeks	8.73	0.59–16.87	0.036	0.18
Active-head-stimulation at 9 weeks	− 10.47	− 21.11–0.16	0.054*	
Model 3: HOS				
Active-head-stimulation at 9 weeks	0.99	0.05–1.93	0.040	0.21
Supine-main-sleep-position at 6 weeks	1.17	− 0.17–2.51	0.086*	
Model 4: LTT-diff				
Side-lying-total at 9 weeks	− 1.97	− 3.53–0.42	0.013	0.13
	Multiple regression			
(B) Positional plagiocephaly at 9 weeks				
Severity of plagiocephaly	β	95% CI	*p*-value	r^2
Model 5: mCVAI (absolute value)				
Supine-sleep-maximum at 6 weeks	0.47	0.21–0.72	0.001	0.24
Supine-lying-total at 6 weeks	0.12	0.02–0.21	0.014	
	Logistic regression			
Presence of plagiocephaly	OR	95% CI	*p*-value	r^2
Model 6: Significant vs. non-significant**				
Supine-sleep-maximum at 6 weeks	2.23	1.40–3.57	0.001	0.25
Passive-head-repositioning at 9 weeks	3.97	1.15–13.75	0.029	

Note. HOD = head orientation duration; HOS = head orientation strength; LTT-diff = difference in latency to turn; mCVAI = modified Cranial Vault Asymmetry Index; β = coefficient; OR = odd ratio; CI = confidence interval; rho = intra-class correlation (% of variance); r^2 = percentage of variance.
*Marginally significant variables which were included in the model.
**Significant plagiocephaly group with absolute value of mCVAI ≥3.5%.

both *HOD-diff* and *HOS* asymmetry were less than other groups combined. The *LTT-diff* was not significant for any group.

Relationships Between Positional Plagiocephaly and Infant Positioning

Multiple regression showed that infants with a greater *mCVAI* (more severe plagiocephaly) at 9 weeks had a longer *supine-sleep-maximum* at 6 weeks and spent more time in *supine-lying-total* at 6 weeks – these variables accounted for 24% of the variance in plagiocephaly (Table 2, Model 5). Using the regression equation for model 5 [$mCVAI = -1.53 + 0.47(supine\text{-}sleep\text{-}maximum) + 0.12\,(supine\text{-}lying\text{-}total)$] and the assumption that there are 24 hours in a day, the risk of developing significant plagiocephaly ($mCVAI \geq 3.5\%$) at 9 weeks is predicted at 6 weeks when the time relationship between *supine-sleep-maximum* and *supine-lying-total* are: 5:22 hours; 6:18.5 hours; 7:14.5 hours; or 8:10.5 hours. That is, a longer supine sleeping time requires shorter total supine time to reach the threshold.

Prone positioning was not associated with plagiocephaly. Logistic regression showed that presence of positional plagiocephaly ($n = 24$) at 9 weeks was also associated with *supine-sleep-maximum* at 6 weeks as well as the parent performing corrective *passive-head-repositioning* at 9 weeks—these variables accounting for 25% of the variance in presence of plagiocephaly (Table 2, Model 6). There was no association in this cohort between positional plagiocephaly with parents proactively alternating their infant's head when placing them down to sleep at any age.

Table 3. Significant results of subgroup comparisons (A) parent-reported infant AHO-Both vs. other groups (AHO-None, AHO-Sleep, AHO-Awake)[a,b], (B) parent-reported infant AHO-None vs. other groups (AHO-Both, AHO-Sleep, AHO-Awake) with difference in head orientation duration between (HOD-diff) and direction of head orientation strength (HOS) and latency to turn difference (LTT-diff[c]).

	Mean (SD)	95% CI	t(df)	*p*-value
(A) Parent-reported infant AHO-Both vs. others				
3 *weeks*				
HOD-diff (AHO-Both)	47.69 (25.50)	30.56–64.82		
HOD-diff (Others)	27.88 (21.90)	22.88–32.89		
Mean difference	−19.81	−34.14–5.47	−2.75 (85)	0.007
HOS (AHO-Both)	2.47 (1.38)	1.60–3.35		
HOS (Others)	1.64 (1.20)	1.37–1.92		
Mean difference	−0.83	−1.59–0.07	−2.17 (86)	0.03
(B) Parent-reported infant AHO-None vs. others				
6 *weeks*				
HOD-diff (AHO-None)	21.88 (20.85)	14.82–28.93		
HOD-diff (Others)	31.92 (22.36)	24.57–39.27		
Mean difference	10.04	0.01–20.08	2.00 (72)	0.05
9 *weeks*				
HOD-diff (AHO-None)	23.79 (20.08)	17.45–30.13		
HOD-diff (Others)	33.56 (22.75)	25.74–41.38		
Mean difference	9.77	−0.02–19.56	1.99 (74)	0.05
HOS (AHO-None)	1.59 (1.13)	1.23–1.95		
HOS (Others)	2.22 (1.22)	1.80–2.64		
Mean difference	0.63	0.09–1.17	2.33 (74)	0.02

[a]AHO-Both = asymmetrical head orientation during awake and asleep; AHO-None = no asymmetrical head orientation during awake or asleep; SD = standard deviation; CI = confidence interval; df = degree of freedom.
[b]Between groups analysis for awake only (AHO-Awake) at 9 weeks was not possible because there was only one infant in the group.
[c]No significant difference in LTT-diff from preferred and non-preferred side across all age groups. HOD-diff = (HOD-right% - HOD-left%).

Relationships Between Positional Plagiocephaly and Parent-Reported AHO

In infants who did not have parent-reported AHO when awake and asleep (*AHO-None*) at 3 weeks there was a lower rate (16%) of positional plagiocephaly at 9 weeks compared with 41% in other infants ($p = 0.015$). There was no association between presence of positional plagiocephaly at 9 weeks with parent-reported AHO (*AHO-Both, AHO-Awake or AHO-Asleep*) at 3 weeks or 6 weeks.

Discussion

Our study has prospectively monitored the parent practices of infant body positioning and infant head positioning in healthy term infants from 3 weeks to 9 weeks of age. In our cohort, approximately half of the parents were implementing proactive head alternating strategies during this age range and this practice was associated with improved midline head orientation on clinical assessment (Model 2). Parent practice of active head stimulation was associated with clinical identification of increased head orientation asymmetry (HOD and HOS) (Model 1 & 3). Parents showed a good ability to identify head orientation asymmetry, with parent report of asymmetry consistent with clinical identification of asymmetry (HOD and HOS) at 3 weeks. Similarly, parent report of absence of head orientation asymmetry was consistent with clinical identification of better symmetry (HOD and HOS) at 6 weeks and 9 weeks. Proactive use of side-lying was identified as promoting more head orientation symmetry. Plagiocephaly at 9 weeks was associated with more time in supine and longer sleep durations in supine, but not prone-play time.

This study supported the hypothesis that positioning is related to the development of symmetrical head orientation and/or head orientation asymmetry in healthy young infants. Approximately 60% of parents practiced proactively alternating their infant's head at 3, 6, and 9 weeks and this was associated with improved midline orientation on clinical assessment. Studies have showed that targeted parent education programs focused on plagiocephaly and asymmetrical head orientation prevention are able to reduce the prevalence of these conditions (Aarnivala et al., 2015; Cavalier et al., 2011; Lennartsson, 2011). These programs have included an educational session in the maternity ward with a pediatrician, or ongoing infant check-ups by child health nurses. Furthermore, we confirmed that prolonged supine-lying together with long supine sleeping time at 6 weeks of age are risk factors for plagiocephaly development at 9 weeks. The longer of these times related to more severe plagiocephaly. We are not suggesting infant sleeping time should be reduced, instead, prolong supine positioning during awake hours should be avoided. Literature shows that parents frequently place infants on their backs and can lack confidence, skills or intent to carry out tummy time advice (Koren et al., 2010; Shweikeh, Nuno, Danielpour, Krieger, & Drazin, 2013). In our study, prone-play position was not associated with plagiocephaly, whereas side-lying positioning was related to head orientation symmetry. In line with research evidence, we recommend a 3-part *A-B-C Plagiocephaly Prevention Strategy* (Appendix I): (A) "*Active Baby*"—which encourage frequent change of play positions and symmetrical free movement of head and body; (B) "*Balanced Handling*"—which encourages symmetrical parent positioning and handling practices; and (C) "*Corrective Strategies*"—which encourages active and passive correction of infant's head to the non-preferred side if any asymmetry is identified.

This study highlights the importance of parents being able to identify asymmetry in their infants in order to instigate reactive management strategies. Parent-reported infant asymmetrical head orientation was associated with clinical identification of head orientation asymmetry, particularly at 3 weeks of age. Other studies have shown that physiotherapy is effective in reducing progression to severe plagiocephaly when asymmetrical head orientation was identified and timely treatment commenced (van Vlimmeren et al., 2008). Since asymmetrical head orientation is a possible precursor for plagiocephaly, we suggest that young infants who are identified by their parents as having asymmetrical head orientation during both asleep and awake times should be reviewed by their primary care professional and/or referred on for pediatric physiotherapy intervention. Similarly, when parents do not report their infants as having any head orientation asymmetry, clinicians can be confident that the majority of these infants will have a normal head shape at 9 weeks, and follow up is only needed if observations change. In light of these findings, we propose a *Plagiocephaly Screening Pathway* (Appendix II) to enhance primary health care professionals' identification and management of infants at risk of asymmetrical head orientation and positional plagiocephaly.

It is recognized that cervical dysfunction, principally sternocleidomastoid (SCM) muscle imbalance, may be commonly found in infants with positional plagiocephaly and may be expressed as asymmetry in head orientation (Golden, Beals, Littlefield, & Pomatto, 1999; Hutchison et al., 2004; Hutchison et al., 2009; Rogers, 2011a). In the current study, infants with congenital muscular torticollis were excluded since this muscular condition has a different pathological pathway for plagiocephaly development. We recommended investigation of secondary SCM muscle imbalance as a potential contributor to the development of plagiocephaly.

Limitations and Recommendations for Further Research

All HOP measurements were conducted by the principal researcher who had extensive experience in measurement of head orientation in infants. Further evaluation of the psychometric properties of these measures is warranted to support clinical implementation. The parent survey relied on recall of infant positions over a 3-day period, which may pose some errors. Future research may involve a positioning diary (paper-based or electronic) to record a child's daily activities by their parents (Thomas, Hunt, Hurley, Robertson, & Carter, 2010). In our study, parents indicated whether they performed the proactive and corrective strategies, but not how often they performed each strategy, which may have an impact on head asymmetry and plagiocephaly prevalence. We recommend further research to investigate such parameters, which could be incorporated into the positioning diary. Finally, further research is needed regarding the optimal dosage and intensity of the *A-B-C Plagiocephaly Prevention Strategy,* and the effectiveness of the *Plagiocephaly Screening Pathway* to prevent positional plagiocephaly.

Conclusion

Infant positioning appears to impact early head orientation development and asymmetrical head shape at 9 weeks of age. The length of supine sleeping time (in one sleep) and total supine time (across 24 hours) were found to be associated with plagiocephaly development. This supports advice to avoid prolonged supine time during awake hours. Parents who notice their infants have asymmetrical head orientation during awake and asleep time, should be encouraged to seek review by a primary healthcare professional for timely intervention. Using our data, we have now proposed two new clinical tools, the *A-B-C Plagiocephaly Prevention Strategy* and the *Plagiocephaly Screening Pathway* to help parents and primary healthcare professionals to prevent and/or identify infants at risk of asymmetrical head orientation or positional plagiocephaly.

Declaration of Interest

The authors reported no conflicts of interest. The authors alone are responsible for the content and writing of the article.

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About the Authors

Amy Leung, MPhty(Paed), PDPhty, is an experienced paediatric physiotherapist working in child development and a Doctoral Student at The University of Queensland, Australia. She completed this research as part of her PhD program. **Allison Mandrusiak**, PhD, GCHEd, BPhty(HonsI), is a Senior Lecturer in Physiotherapy at The University of Queensland, Australia. She completed her PhD in paediatric cardiorespiratory physiotherapy, and her teaching and research interests cover a range of areas across the lifespan. **Pauline Watter**, PhD, BPhty, is an Honorary Senior Lecturer in Physiotherapy at The University of Queensland, Australia. Her research has involved children with DCD, CF, ELBW outcomes, plagiocephaly, and implementation of ICF in clinical settings and research. **John Gavranich**, MBBS, FRACP, is Director of Paediatrics, Ipswich Hospital, Queensland, Australia. He has actively encouraged and supported staff to participate in research, particularly in evidence based practice and knowledge translation. **Leanne M. Johnston**, PhD, BPhty(HonsI), is a Senior Lecturer in Physiotherapy at The University of Queensland, Australia and a Director on the Childrenâ..s Health Queensland Hospital and Health Service Board. She is the Chair of the Childrenâ..s Motor Control Research Collaboration for research with children and youth with motor disorders.

ORCID

Amy Leung http://orcid.org/0000-0001-6050-2019

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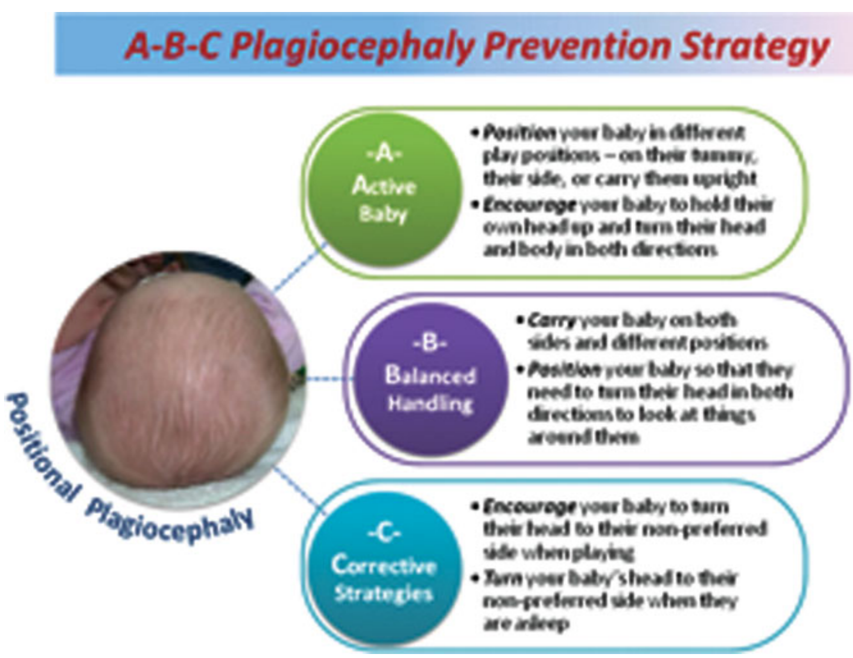
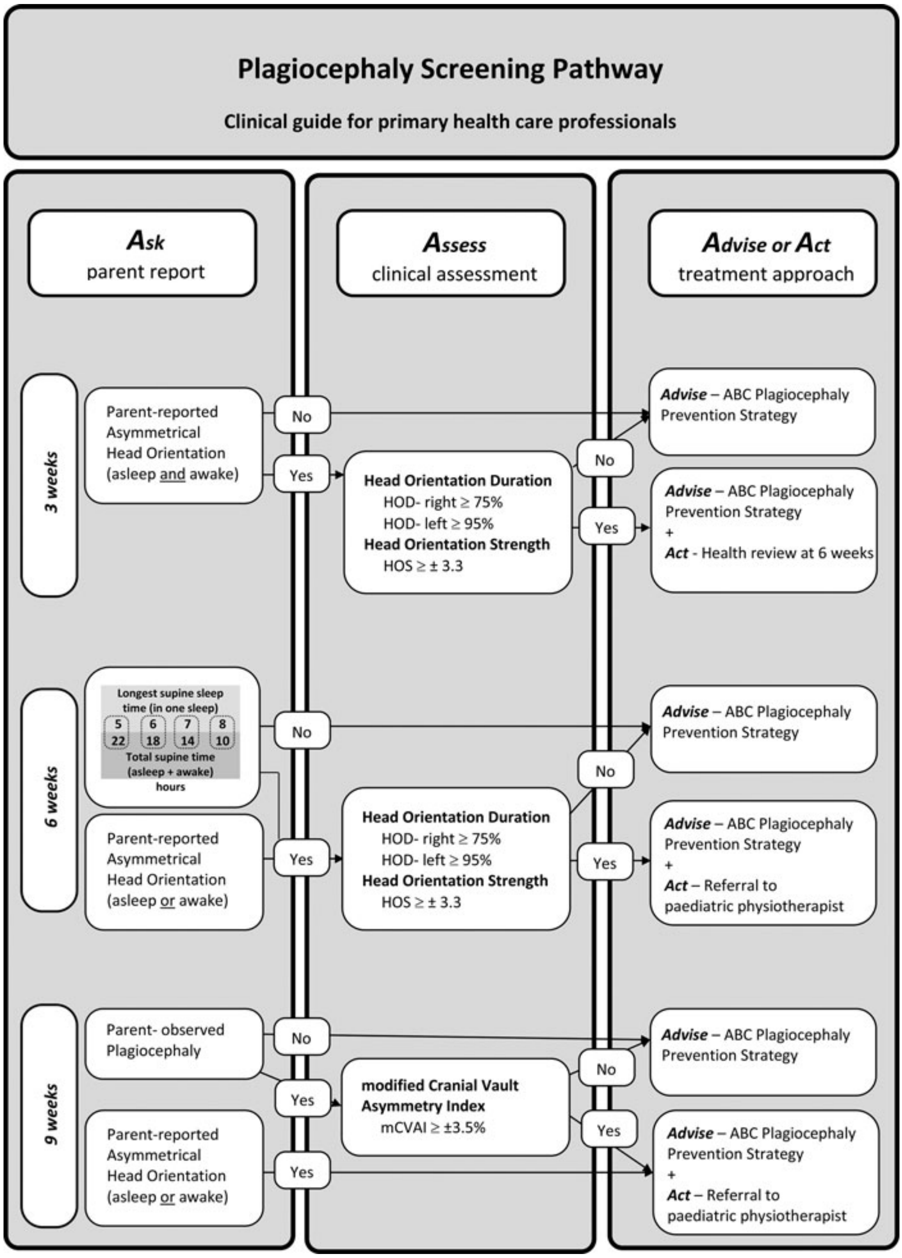
Appendix I



Appendix II


Plagiocephaly Screening Pathway
Clinical guide for primary health care professionals
Ask
parent report
Assess
clinical assessment
Advise or Act
treatment approach
3 weeks
Parent-reported Asymmetrical Head Orientation (asleep and awake)
No
Yes
Head Orientation Duration
HOD- right ≥ 75%
HOD- left ≥ 95%
Head Orientation Strength
HOS ≥ ± 3.3
No
Yes
Advise – ABC Plagiocephaly Prevention Strategy
Advise – ABC Plagiocephaly Prevention Strategy
+
Act - Health review at 6 weeks
6 weeks
Longest supine sleep time (in one sleep)
5 6 7 8
22 18 14 10
Total supine time (asleep + awake) hours
Parent-reported Asymmetrical Head Orientation (asleep or awake)
No
Yes
Head Orientation Duration
HOD- right ≥ 75%
HOD- left ≥ 95%
Head Orientation Strength
HOS ≥ ± 3.3
No
Yes
Advise – ABC Plagiocephaly Prevention Strategy
Advise – ABC Plagiocephaly Prevention Strategy
+
Act – Referral to paediatric physiotherapist
9 weeks
Parent- observed Plagiocephaly
No
Yes
Parent-reported Asymmetrical Head Orientation (asleep or awake)
Yes
modified Cranial Vault Asymmetry Index
mCVAI ≥ ±3.5%
No
Yes
Advise – ABC Plagiocephaly Prevention Strategy
Advise – ABC Plagiocephaly Prevention Strategy
+
Act – Referral to paediatric physiotherapist