Are infants with torticollis at risk of a delay in early motor milestones compared with a control group of healthy infants?

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PUBLICATION DATA
Accepted for publication 21st July 2008.
Published online 26th January 2009.

LIST OF ABBREVIATIONS

AIMS	Alberta Infant Motor Scale
CMT	Congenital muscular torticollis
SIDS	Sudden infant death syndrome

ACKNOWLEDGEMENTS
We are grateful to all of the infants and their families who participated and made this study possible. Financial support was received from the Petter Silfverskiöld memorial foundation and the FoU Södra Buhuslän and Göteborg foundation.

Recently it has been claimed that infants with congenital muscular torticollis (CMT) are at risk of a delay in early motor milestones. The aim of the present study was to investigate whether infants with CMT are indeed at risk in comparison with a control group of healthy infants. A second aim was to investigate whether the time spent in a prone position and plagiocephaly had any influence on motor development. Eighty-two infants with CMT (35 females and 47 males) were compared with 40 healthy infants (18 females and 22 males). Motor development was assessed with the Alberta Infant Motor scale (AIMS). Multiple regression showed that infants in the CMT group had a significantly lower AIMS score than the control group at 2 months (p=0.03) and 6 months of age (p=0.05). Infants who spent at least three occasions daily in a prone position when awake had significantly higher AIMS scores than infants who spent less time prone at 2 months (p=0.001), 6 months (p<0.001), and 10 months of age (p<0.001). The CMT group achieved early motor milestones significantly later than the control group until the age of 10 months, but the risk of delay seems to be more strongly associated with little or no time prone when awake than with CMT.

Some authors have recently claimed that infants with congenital muscular torticollis (CMT) are at risk of a delay in achieving motor milestones[1–3] but only one study supports these statements for infants below 1 year of age.[2] CMT is the third most common congenital musculoskeletal anomaly in infants after congenital hip dysplasia and talipes equinovarus. The reported incidence of CMT is 0.4 to 1.9%.[4] CMT is a result of shortening or excessive contraction of the sternocleidomastoid muscle; the infant holds the head tilted to one side and rotated to the opposite side. Often there is limited range of motion in both rotation and lateral flexion, imbalance in muscle function, weakness in the non-affected side, and increased strength in the affected side.[5] Schertz et al. found that infants with torticollis are at increased risk of early gross motor delay.[2]

When interpreting the infants' motor developmental progress, the influence of positioning when awake needs to be considered.[6] Since the launch of sleep position recommendations ('back to sleep'), many infants spend little or no time in a prone position when awake,[7–10] because parents are fearful of the possibility of sudden infant death syndrome (SIDS).[7,8] Nevertheless, a prone position encourages upper-body strength used in acquisition of many infant motor milestones, stimulates motor development,[6,7,9–14] and reduces the risk of plagiocephaly.[15,16] The traditional age ranges for the development of motor milestones were established when infant sleeping in the prone position was the norm.[7,9] Several studies have shown that children who spend little or no time in a prone position are at risk of a delay in early motor milestones compared with infants who spend time prone.[6,7,9–14] It

may be that supine-positioned infants are late in developing their milestones in the first year because the upper body contributes less to daily movement than for predominantly prone-positioned infants.[7]

The purpose of this study was to investigate whether infants with CMT are at risk of a delay in early motor milestones in comparison with healthy infants. A second aim was to investigate whether the time spent in the prone position or the presence of plagiocephaly had any influence on motor development.

METHOD

Participants

All infants treated for CMT at the Department of Physiotherapy, Queen Silvia Children's Hospital, Gothenburg, Sweden, who were aged 2 to 10 months at the time of the study were invited to participate. They had to be 2, 6 or 10 months old at the first assessment. Infants were excluded if they had any disorder other than CMT or were born before gestational week 37.

A control group consisting of healthy infants in the same age range was recruited from the Child Health Centre (in Sweden all infants attend a health centre in the first year to have their weight, length, etc. checked). They could be either 2 or 6 months old at the first assessment. Inclusion criteria were birth at a gestational week of at least 37 weeks and no suspected syndromes or medical conditions.

The local ethical committee approved this study and the parents gave informed consent to participate.

Procedure

Parents in the CMT group performed daily treatment for CMT, with instructions from the physiotherapist. Treatment including stretching (if the child had limited range of motion in rotation below 90° and in lateral flexion if a side difference in range of motion existed), muscle function exercises, and positioning. No additional physiotherapy treatment was given for late motor development. All parents in both groups were verbally encouraged to give their infants time in the prone position when awake. For ethical reasons, the parents of all infants with plagiocephaly were advised to avoid letting the infant spend time on the flat spot.

Parents were asked about their infant's gestational week, weight, and length at birth. The infants were assessed at the 'key' ages of 2, 6, and 10 months for motor development and deformational plagiocephaly. At each occasion the parents answered questions about sleeping position and were asked to estimate time spent in a prone position (i.e. occasions of at least several minutes prone when awake) during the previous weeks. Motor development was also assessed at 18 months of age to ensure that no child had remaining delay.

Measurements

To assess motor development, the Alberta Infant Motor scale (AIMS) was used. This is a norm-referenced measure and consists of a 58-item observational assessment for infants. It assesses the infants' sequential development of motor milestones from birth to independent walking.[17] The AIMS measures spontaneous movements which reflect the quality of weight bearing, posture, and antigravity skills in the prone, supine, sitting, and standing positions. The scale is reliable and valid in discriminating the motor performance of infants developing normally from infants at risk and motor-delayed infants, and for evaluating small changes in motor skills due to maturation.[18–20]

The infants were assessed through observation with minimal handling and no arbitrary stimuli or facilitation. The opportunity was given for the infants to demonstrate their entire movement repertoire and there was no minimum or maximum number of trials for an infant to perform an item.[17] No item was credited on the basis of development assumptions or parental reporting.

Deformational plagiocephaly was assessed according to a 4-point severity assessment scale for plagiocephaly (0=none, 1=minor, 2=moderate, 3=severe).[21]

The same physiotherapist (AÖ) evaluated all of the infants on all occasions.

Statistical analysis

Information about sex, gestational week at birth, birthweight, birth length, sleep position, prone time when awake, and plagiocephaly was summarized descriptively. The centile rank for the groups was compared using the Mann–Whitney test; the centile ranks that are used are from the AIMS manual. Suspected late motor development was tested using Fisher's exact test. Differences in AIMS raw scores between CMT and controls were assessed by analysis of covariance (ANCOVA), using prone time awake, plagiocephaly, sleep position, gestational week at birth, sex, birth length, and birthweight as covariates. The Wilcoxon signed-rank test was used to compare plagiocephaly at 2 and 10 months of age. The SPSS statistical programme (version 12.01) was used and p values of 0.05 or less were considered evidence of statistically significant findings.

RESULTS

A total of 122 infants aged 2 to 18 months participated in this study. Eighty-two infants with CMT (35 females and 47 males) and 40 healthy infants (18 females and 22 males) were assessed on at least one occasion (maximum four occasions at 2, 6, 10, and 18mo; Table I). Thirty-two infants in the CMT group were included at 6 months and 25 at 10 months of age. By the end of the study three

infants with CMT had only participated at 2 months of age. Some infants in the CMT group moved out of the area and could not continue to participate. Only one infant in the control group did not complete the study. Gestational week, weight, and length at birth were similar in both groups. All infants aged 10 months and most of those aged 6 months had been referred to physiotherapy at an earlier age before the study started (range 0.5–7.5mo).

At 2 months of age, the majority of infants slept in a supine position or in both supine and side positions, (72% in the CMT group [18 infants], 86% in the control group [31 infants]) and no infant in either group slept solely prone, but 17% of the CMT group and 22% of the control group spent at least three times a day in a prone position when awake at 2 months; the time spent prone increased with age (Table II).

The centile rank of motor development was significantly lower for the CMT group at 2 months (p=0.02) and 6 months (p<0.004) than for the control group (Table III). At 2 months (p=0.02) and 6 months (p=0.03), significantly more infants from the CMT group than from the control group were at or below the 10th centile according to the AIMS (Table IV). At 2 months (p=0.03) and 6 months (p=0.05), multiple regression on raw scores showed that infants in the CMT group had scored significantly lower on the AIMS than in the control group (Table V). None of the four assessed positions was significantly more impaired than the other positions. Infants who spent at least three times daily prone when awake had significantly higher scores on the AIMS than infants who spent less time prone at 2 months (p=0.001), 6 months (p<0.001), and 10 months (p<0.001) of age (Table V). Multiple regression showed that both CMT and prone time had an impact on the motor development at 2 and 6 months of age. Prone time had a higher impact and also had an impact at 10 months of age (Table VI). Gestational week at birth, sex, birthweight, birth length, and plagiocephaly had no significant impact on AIMS scores. Sleep position and AIMS scores were not analyzed as no infant slept solely in a supine position at 2 months of age, although two infants slept in all positions (i.e. supine, side, and prone) at that age. The infants in the control group spent significantly more time in a prone position when awake than the CMT group did at 6 months of age. In both groups the prone position became more common with increased age.

Plagiocephaly occurred at 2 months of age in 61% (15 infants) of the CMT group (95% confidence intervals [CI]

Table I: Numbers of infants participating

Assessments	Torticollis	Control	Total
2mo	25	35	60
6mo	54	39	93
10mo	57	39	96
1–4 assessments	82	40	122
3–4 assessments	57	38	95
All 4 assessments	22	35	57

Data are numbers of infants who had at least one assessment at the age of 2, 6, or 10 months; where possible, infants were also followed up at 18 months of age. All infants who participated at 6 months were also assessed at 10 and 18 months of age; for three of the infants who participated at 2 months, data are available at 10 months but missing at 6 months of age.

Table II: Infants spending ≥3 times/day prone when awake

Assessment age	Torticollis	Control	*p* value
2mo	5 (17%)	8 (22%)	0.13
6mo	37 (65%)	33 (85%)	0.02
10mo	63 (90%)	38 (97%)	0.21

Table III: Alberta Infant Motor Scale centiles

Assessment age	Torticollis	Control	*p* value
2mo			
Mean (SD)	20.4 (18.4)	31.8 (21.5)	0.02
Median (range)	12 (1–69)	23 (1–69)	
6mo			
Mean (SD)	34.4 (23.2)	46.9 (21.0)	0.004
Median (range)	27 (1–85)	41 (3–99)	
10mo			
Mean (SD)	49.8 (28.5)	53.8 (24.6)	0.59
Median (range)	67 (1–100)	67 (1–100)	

Table IV: Infants scoring ≤5th and ≤10th centiles on the Alberta Infant Motor Scale

Assessment age	Torticollis	Control	*p* value
2mo			
≤5th centile	3 (10%)	1 (3%)	
≤10th centile	11 (38%)	4 (11%)	0.017
6mo			
≤5th centile	6 (9%)	1 (3%)	
≤10th centile	11 (19%)	1 (3%)	0.025
10mo			
≤5th centile	11 (19%)	1 (3%)	
≤10th centile	13 (23%)	4 (10%)	0.286

44–77%) and 42% (14 infants) of the control group (CI 25–59%). Plagiocephaly decreased significantly between 2 and 10 months of age in both the CMT group (p=0.01) and the control group (p<0.001). At 10 months of age minor plagiocephaly continued in 39% (22 infants) of the CMT group but in none in the control group. Twenty-seven infants had minor signs of CMT at the age of 10 months, and six infants had still visible head tilt at 18 months' follow-up. At 18 months of age all of the infants in both groups achieved the total score of the AIMS. Two infants in the control group had noticeable idiopathic toe walking at this age.

Table V: Impact of congenital muscular torticollis and time spent prone while awake on Alberta Infant Motor Scale scores (AIMS; multiple regression analysis), by number of visits

Assessment age	All infants regardless of number of visits (n=122)	Infants missing one visit (n=96)	Infants attending all 4 visits (n=57)
2mo			
Torticollis vs control p value	0.03	0.02	0.02
Time spent prone p value	0.001	0.003	0.004
6mo			
Torticollis vs control p value	0.05	0.06	0.04
Time spent prone p value	<0.001	<0.001	0.007
10mo			
Torticollis vs control p value	0.92	0.73	0.52
Time spent prone p value	<0.001	<0.001	0.03

Multiple regression on raw scores showed that infants in the torticollis group scored significantly lower on the AIMS than the control group. Infants who spent at least three times daily prone when awake had significantly higher scores on the AIMS than infants who spent less time prone at 2, 6, and 10 months of age.

Table VI: Impact of congenital muscular torticollis and time spent prone on Alberta Infant Motor Scale scores (AIMS; multiple regression analysis), for all infants seen at least once

Assessment age	Standard errors	Beta[a]	p value
2mo			
Torticollis vs control	0.397	–0.252	0.03
Time spent prone	0.495	0.389	0.001
6mo			
Torticollis vs control	0.775	–0.177	0.05
Time spent prone	0.848	0.472	<0.001
10mo			
Torticollis vs control	1.157	–0.009	0.92
Time spent prone	2.254	0.521	<0.001

Number of times spent prone when awake is dichotomized as <3 vs ≥3. [a]Estimated differences in AIMS raw score between torticollis and control and between <3 and ≥3 times spent prone when awake.

DISCUSSION

The CMT group scored significantly lower than the control group on the AIMS at 2 and 6 months of age. Therefore infants with CMT seem to be at risk of a delay in early motor milestones. However the mean centile for both groups was below the 50th centile at 2 months of age. This might be because only about one-fifth of the infants spent at least three times a day in the prone position when awake at this age. Fleuren et al. found that 75% of Dutch infants scored below the 50th centile on the AIMS and they propose that this may be due to less time in the prone position after the 'back to sleep' campaign.[22] Little or no time prone when awake correlated more strongly with lower scores on the AIMS at 2 and 6 months of age than CMT did. The present study indicates that little or no time prone when awake implies a higher risk of having a delay in achieving early motor milestones than CMT; however, CMT seems to contribute to the risk of delay. A systematic review of 19 articles shows that no or limited time in the prone position for infants when awake affects motor development during the first 6 months of life.[23] In our study, all parents were encouraged to give their infants a lot of time in the prone position when awake; nevertheless the CMT group spent less time prone when awake than the control group. This was an unexpected finding as infants treated for CMT at the Department of Physiotherapy are encouraged to spend a lot of time prone to stimulate head posture and neck strength and to reduce the risk of plagiocephaly. We had expected the parents of the CMT group to be at least as motivated as the parents in the control group to put their infants in a prone position. Both our study and that of Schertz et al.[2] indicate that infants with CMT are at risk of a delay in early motor milestones, so it might be of increased importance for infants with CMT to spend time prone when awake to eliminate at least one risk factor. We need to improve the information to make the parents realize the benefits and make the effort to give their infant more prone time. Today we often see in clinic that infants have somewhat low tolerance of the prone position, especially if it is not introduced early in life. It is possible that an infant with CMT has a lower tolerance than healthy infants because of the muscular imbalance in the neck.

Schertz et al. had no control group of healthy infants and time spent in prone when awake was not investigated.[2] In their study, it could be questioned whether it was only the torticollis that affected the motor skills. Also in their study, infants with a delay in early motor milestones were given weekly physical therapy to promote motor development.[2] It is likely that prone positioning was a part of the physical therapy, but this does not necessarily mean that parents practised this at home. That study was performed after the 'back to sleep' campaign in Israel.[24] It is likely that the infants in their study spent little time in a prone position compared with infants born before this campaign. Davis et al. found that prone-sleeping infants spent significantly more prone time awake than supine sleepers (more than twice the time during their first 3mo of life).[7] Mildred et al. found that 26% of parents never placed their infants in a prone position for play.[8] Majnemer and Barr found that normally developing infants who were positioned to sleep supine had a delay in early motor milestones by the age of 6 months and this was significantly associated with limited exposure to a prone position when awake.[13] Monson et al. found higher scores on the AIMS for infants who had been placed in a prone position when awake than for infants with limited or no experience in the prone position (all slept supine).[6] If the sleep position is the same as the position awake, then the delay in achieving milestones could be explained by less opportunity for the infant to learn the motor skills.[12] The prone position seems likely to be of particular importance in promoting the development of antigravity extension and gaining head control.[8] It is generally accepted among physiotherapists and paediatricians that experience in a variety of positions is necessary for optimal motor development.[8] There seems to be an obvious connection between infant motor development and time spent prone when awake. However, the fact that the parents estimated instead of giving exact times that their infants spent in the prone position when awake is a limitation in our study.

At 18 months of age all infants in this study were considered to have normal motor development (i.e. all acquired total scores on the AIMS). It can be questioned whether the AIMS can detect minor or moderate differences between the groups at 18 months, as there are few items to be achieved after the age of 15 months, and the centile ranks are rather tight after this age. Liao and Campbell found the ceiling effect for AIMS to be 12 months of age and they do not believe that the AIMS is suitable for use once infants can lower themselves in a controlled manner from a standing position.[25]

In the present study there was a rather large prevalence of plagiocephaly in the control group (40%, CI 25–59%) and for most of the infants a mild asymmetry. It is eye-opening to discover that even healthy infants without neck problems can be at higher risk of developing deformational plagiocephaly than previously expected. From 1990 to the end of the last century the prevalence of plagiocephaly increased from 0.3 to 1.7%.[26] Recently van Vlimmeren et al. found the prevalence of deformational plagiocephaly to be 22.1% for infants at the age of 7 weeks.[16] Because of the risk of SIDS, infants need to sleep supine but they also need prone time awake to stimulate the gross motor development and to decrease the risk for plagiocephaly. The fact that parents avoid the prone position when their infant is awake can be due to misinterpretation of information about the SIDS risk factors leading to parents exercising unnecessary caution.[8] Parents also avoid putting their infants in a prone position when awake if the infant has low tolerance of this position.[6,7] Our clinical experience has shown the prone position to be accepted more easily if it is introduced very early in life. Community educators may need to clarify that a prone position for play is not a risk factor for SIDS and that it is desirable for infants to spend supervised time when awake in the prone position.[8]

CONCLUSION

The CMT group in our study had significant risk of a delay in early motor milestones compared with the control group until the age of 10 months. Torticollis seems to contribute to the risk of delay especially for infants below the age of 10 months. However, time spent in the prone position when awake seems to be of greater importance in reducing the risk. Gestational week at birth, sex, birthweight, birth length, and plagiocephaly had no influence on motor development in this study. The result of this study indicates that prone time awake is an advantage for motor development. Information about prone time awake ought to be improved in general and specifically for infants with CMT.

REFERENCES

1. Van Vlimmeren LA, Helders PJM, Van Adrichem LNA, Engelbert RHH. Torticollis and plagiocephaly in infancy: therapeutic strategies. *Pediatr Rehabil* 2006; **9:** 40–46.
2. Schertz M, Zuk L, Zin S, Nadam L, Schwarts D, Bienkowski RS. Motor and cognitive development at one-year follow-up in infants with torticollis. *Early Hum Dev* 2008; **84:** 9–14.
3. Children's Hospital and Clinics. Torticollis. http://xpedio02.childrensmn.org/stellent/groups/public/@manuals/@pfs/@childdev/documents/policyreferenceprocedure/031726.pdf (accessed 1 August 2008).

4. Cheng JCY, Wong MWN, Tang SP, Chen TMK, Shum SLF, Wong EMC. Clinical determinants of the outcome of manual stretching in the treatment of congenital muscular torticollis in infants. *J Bone Joint Surg Am* 2001; **83-A:** 679–87.
5. Emery C. The determinants of treatment duration for congenital muscular torticollis. *Phys Ther* 1994; **74:** 921–29.
6. Monson RM, Deitz J, Kartin D. The relationship between awake positioning and motor performance among infants who slept supine. *Pediatr Phys Ther* 2003; **15:** 196–203.
7. Davis BE, Moon RY, Sachs HC, Ottolini MC. Effects of sleep position on infant motor development. *Pediatrics* 1998; **102:** 1135–40.
8. Mildred J, Beard K, Dallwitz Z, Unwin J. Play position is influenced by knowledge of SIDS sleep position recommendations. *J Paediatr Child Health* 1995; **31:** 499–502.
9. Salls JS, Silverman LN, Gatty CM. The relationship of infant sleep and play positioning to motor milestone achievement. *Am J Occup Ther* 2002; **56:** 577–80.
10. Dudek-Shriber L, Zelazny S. The effects of prone positioning on the quality and acquisition of developmental milestones in four-month-old infants. *Pediatr Phys Ther* 2007; **19:** 48–55.
11. Dewey C, Fleming P, Golding J. Does the supine sleeping position have any adverse effects on the child? II. Development in the first 18 months. *Pediatrics* 1998; **101:** e5–e9.
12. Jantz JW, Blosser CD, Fruechting LA. A motor milestone change noted with a change in sleep position. *Arch Pediatr Adolesc Med* 1997; **151:** 565–68.
13. Majnemer A, Barr RG. Influence of supine sleep positioning on early motor milestones acquisition. *Dev Med Child Neurol* 2005; **47:** 370–76.
14. Jennings JT, Sarbaugh BG, Payne NS. Conveying the message about optimal infant positions. *Phys Occup Ther Pediatr* 2005; **25:** 3–18.
15. Hutchison BL, Thompson JMD, Mitchell EA. Determinants of nonsynostotic plagiocephaly: a case-control study. *Pediatrics* 2003; **112:** e316–22.
16. Van Vlimmeren LA, van der Graaf Y, Boere-Boonekamp MM, L'Hoir MP, Helders PJM, Engelbert RHH. Risk factors for deformational plagiocephaly at birth and at 7 weeks of age: a prospective cohort study. *Pediatrics* 2007; **119:** e408–18.
17. Piper MC, Darrah J. Motor assessment of the developing infant. Philadelphia: WB Saunders, 1994.
18. Piper MC, Pinnell LE, Darrah J, Maguire T, Byrne PJ. Construction and validation of the Alberta Infant Motor Scale (AIMS). *Can J Public Health* 1992; **83:**(Suppl 2) S46–50.
19. Jeng S-F, Yau K-IT, Chen L-C, Hsiao S-F. Alberta Infant Motor Scale: reliability and validity when used on preterm infants in Taiwan. *Phys Ther* 2000; **80:** 168–78.
20. Blanchard Y, Neilan E, Busanich J, Garavuso L, Klimas D. Interrater reliability of early intervention providers scoring the Alberta Infant Motor Scale. *Pediatr Phys Ther* 2004; **16:** 13–18.
21. Cranial Technologies, Inc. http://www.cranialtech.com (accessed 1 August 2008).
22. Fleuren KMW, Smit LS, Stijnen T, Hartman A. New reference values for the Alberta Infant Motor Scale need to be established. *Acta Pediatr* 2007; **96:** 424–27.
23. Pin T, Eldridge B, Galea MP. A review of the effects of sleep position, play position, and equipment use on motor development in infants. *Dev Med Child Neurol* 2007; **49:** 858–67.
24. Siegel-Itzkovich J. Israel: Government endorses back to sleep policy. *BMJ* 1995; **310:** 10.
25. Liao P-JM, Campbell SK. Examination of the item structure of the Alberta Infant Motor Scale. *Pediatr Phys Ther* 2004; **16:** 31–38.
26. Graham JM Jr, Gomez M, Halberg A, et al. Management of deformational plagiocephaly: repositioning versus orthotic therapy. *J Pediatr* 2005; **146:** 258–62.