THE INFLUENCE OF POSITIONING AND TORTICOLLIS
ON THE DEVELOPMENT AND TREATMENT OF
ABNORMAL INFANT HEAD SHAPES

A DISSERTATION

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BY

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DEDICATION

For my husband Curt who never wavered in his support of my further education. He is my partner in all of my efforts, my cheerleader, my caregiver and my biggest supporter. His love and support are the greatest gifts I have ever received.

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ABSTRACT

JULIE SHILL

THE INFLUENCE OF POSITIONING AND TORTICOLLLIS ON THE DEVELOPMENT AND TREATMENT OF ABNORMAL INFANT HEAD SHAPES

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The purpose of this project was to investigate how positioning and torticollis influence the development and treatment of abnormal head shapes. Study One is a regression analysis to determine if time in inclined positioning equipment, birth (singleton or multiple), and sex are predictors of brachycephaly and/or severe plagiocephaly. Charts of 4-month-old infants were reviewed. Two binomial logistic regression analyses were done. The two dependent variables were presence of brachycephaly (cephalic index ≥ 92) or not, and presence of severe plagiocephaly (cranial vault asymmetry ≥ 12 mm) or not. The model for presence or absence of cephalic index ≥ 92 was significant. Only hours spent in incline was significant. The model for presence or absence of cranial vault asymmetry (CVA) was not significant.

Study Two determined reliability and validity of using Image J software to measure torticollis from digital images. Using Image J software, two physical therapy students measured the head tilt of 24 infants from digital images of the infants. The images were printed and manually measured by the principal investigator. All measurers were blinded to the measurements of each other and to previous measurements from an earlier session. Intraclass Correlation Coefficient (ICC) analyses were done to determine inter and intra-rater reliability of the Image J software measurement method. A paired samples *t*-test was done comparing Image J measurements to the manual measurements to determine concurrent validity. All ICCs were above .75. Inter and intra-rater reliability was established. There was no significant difference comparing Image J measurements and manual measurements. Concurrent validity was established.

Study Three is a retrospective comparative analysis of the effect of age and head tilt on the amount of change in CVA in infants with plagiocephaly who receive dynamic orthotic cranioplasty (DOC) band treatment. Charts of infants with CVA ≥ 12mm who received DOC band treatment were reviewed. A two-way ANOVA was performed comparing age (< 5 months, ≥ 5 months) and head tilt (< 5 degrees ≥ 5 degrees) at the beginning of treatment. The dependent variable was change in CVA. The interaction effect between age and head tilt was not significant. Analysis of the main effect of age showed that infants < 5 months of age demonstrated significantly greater change in CVA than older infants. There was no significant difference in change in CVA measurements for infants with < 5 degrees of head tilt compared to ≥ 5 degrees.

This project provides new information on how inclined equipment contributes to the development of brachycephaly. It introduces a new software method for measuring torticollis. It informs that infants with torticollis achieve similar change in CVA from DOC band treatment as infants without torticollis, while emphasizing the need for physical therapy during DOC band treatment.

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CHAPTER I

INTRODUCTION

Brachycephaly and plagiocephaly are abnormal head shapes that are developed in a normal infant head from pressure on the cranium as it grows. It should be noted that the brachycephaly and plagiocephaly referred to in this project are deformational in nature as opposed to abnormal head shapes that are structural and caused by genetic syndromes, and/or synostosis of cranial sutures at birth. Brachycephaly is characterized by flattening of the occipital area resulting in a tall, wide head shape. The cephalic index (CI) is used to determine the severity of brachycephaly and is calculated using the length and width measurements (cranial width/length x 100). Plagiocephaly presents as asymmetry of the cranium with flatness on one side of the parietal occipital area and contralateral flatness on the frontal area. Facial asymmetry and a shift in the ear and jaw alignment are characteristics of severe plagiocephaly. Cranial vault asymmetry (CVA) is determined by measuring the length of the right and left diagonals of the cranial vault and recording the difference of one diagonal from the other in millimeters. A cranial vault asymmetry index is also commonly used to classify plagiocephaly [diagonal 1 – diagonal 2 /diagonal 1] x 100, where diagonal 1 < diagonal 2 (Wildbrand et al., 2012).

See Figure 1 for an illustration of the measurement of CI and CVA on brachycephalic and plagiocephalic head shapes.

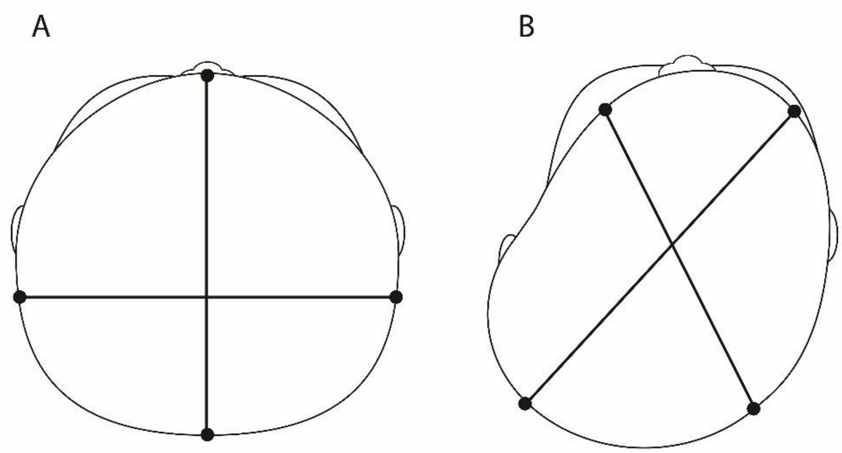

Figure 1. An illustration of the measurement of CI and CVA on a brachycephalic and plagiocephalic head shape. A. A brachycephalic head shape. CI = cranial width/length x 100. B. A plagiocephalic head shape. CVA= the difference of one diagonal from the other diagonal in millimeters.

The incidence of brachycephaly and plagiocephaly has increased since the American Academy of Pediatrics (AAP) recommended supine sleeping for infants in 1992 in order to reduce the risk of sudden infant death syndrome (SIDS; AAP Task Force on Infant Positioning and SIDS, 1992). The literature attributes the increased incidence of brachycephaly and plagiocephaly to supine sleeping (AAP Task Force on Sudden Infant Death Syndrome, 2011; Joganic, Lynch, Littlefield, & Verrelli, 2009; Kane, Mitchell, Craven, & Marsh, 1996; McKinney, Cunningham, Holt, Leroux, & Starr, 2008). Incidence of at least mild plagiocephaly in infants has been reported at 46% with moderate and severe plagiocephaly at 10% to 12% (Mawji, et al., 2014). Since 1992, the types and use of infant positioning equipment has become more plentiful (Bartlett & Kneal Fanning, 2003). Joganic, et al. (2009) identified premature birth, multiple birth, and gender as predictors of deformational plagiocephaly. Anecdotally, pediatric physical and occupational therapists have observed an increase in brachycephaly or plagiocephaly in infants who spend large amounts of time in positioning equipment. Further, in published studies, time spent in positioning equipment combined with time spent in supine has been established as a predictor of brachycephaly or plagiocephaly (Jorganic, et al., 2009; Mawji, et al., 2014). No studies have examined time spent in positioning equipment as a separate predictor of either condition.

The standard of care for infants with severe plagiocephaly and brachycephaly is referral to a plagiocephaly clinic for cranial band treatment. Since head circumference growth rate during the first 6 months of life (one-half inch per month) is twice as fast as the growth rate in the second 6 months (one-fourth inch per month), pediatricians refer infants for cranial band treatment early (typically by 4 months of age) to ensure better outcomes of treatment (John Hopkins Medicine). The DOC band, developed by Cranial Technologies in 1986, is a custom manufactured cranial band designed to provide a gentle holding pressure on the bulged areas of the infant's skull while leaving space in the band for the flattened areas of the head to grow into. The infant wears the DOC band 23 hours per day. The band is adjusted weekly or every two weeks depending on the growth rate of the infant. Adjustments allow for growth in the flatter areas of the head. The band is removed when it is outgrown, usually in 2 to 4 months. The desired outcome is a more symmetrical and round head shape than before treatment. Clinicians measure CVA both before and after treatment, and it is one outcome measurement of cranial band treatment for plagiocephaly. Similarly, CI is measured before and after treatment and is an outcome measurement of cranial band treatment of brachycephaly.

Infants with plagiocephaly also often present with torticollis. Torticollis is a contracture of one side of the neck muscles causing the head to be tilted to one side and rotated to the opposite side. Torticollis and plagiocephaly both contribute to facial asymmetry. Because the face hangs from the skull, the bony structural asymmetry of the plagiocephalic skull causes facial asymmetry. With torticollis, the tight neck muscles of one side pull on the soft tissues of the face also contributing to the facial asymmetry. Anthropometric measurements are done to measure facial asymmetry, but these measurements only reflect bony structural asymmetries. Shortening of the sternocleidomastoid muscle in infants with torticollis pulls on the soft tissues of the face including facial musculature, skin, eyes, and lips. In spite of anecdotal clinical observations that the presence of torticollis hinders improvement of cranial vault asymmetry during cranial band treatment, research has not been conducted to determine the validity of these observations. Since cranial band treatment alone does not address torticollis, it is not sufficient to correct facial asymmetry in infants with torticollis. Best outcomes are achieved if the infant is receiving physical therapy to treat the torticollis along with cranial band treatment to treat the asymmetry of the cranium (Aarnivala, et al., 2016; Kao, Tritasavit, & Graham, 2014).

It is important for pediatric physical therapists to understand how infant positioning and torticollis influence the development and treatment of both brachycephaly and plagiocephaly. The results of this project provide better understanding of the influence of infant positioning on the development of brachycephaly and plagiocephaly as well as the influence of torticollis on CVA improvement during cranial band treatment. This improved understanding supports the importance of physical therapy to address torticollis in infants receiving cranial band treatment to achieve best outcomes. The project used retrospective examination of patient charts. Image J software was used (freely available at http://imagej.net/). A new plug-in program for the software was designed (freely available at https://github.com/rcshill/angle_from_points.git) that enables clinicians to measure head tilt (necessary to determine the degree of torticollis) from digital images. Measurements determined using the software proved to be reliable; therefore, therapists are able to use the software to accurately measure head tilt from digital images (Shill, Beck, & Trudelle-Jackson, 2017).

Purpose

The overarching purpose of this three-study research project was to investigate how positioning and torticollis influence the development and treatment of abnormal infant head shapes. This purpose is accomplished with three distinct studies. Study One is a retrospective regression analysis to determine if time in inclined positioning equipment, birth, and sex are significant predictors of brachycephaly and/or severe plagiocephaly. Study Two is a study to determine inter- and intra-rater reliability of a software method of measuring head tilt from digital images. Concurrent validity of the software method of measuring head tilt from digital images compared with manual measurement of printed images is determined in this second study. It was necessary to determine reliability and validity of the software measurement method in order to use this method confidently to assess head tilt from images of infants in the third study. Study Three is a retrospective comparative analysis of the effect of age and head tilt on the amount of change in CVA in infants with plagiocephaly who receive cranial band treatment.

Study One had two hypotheses: (a) a model including time in inclined equipment, birth (singleton or multiple), and sex would significantly predict CI ≥ 92 at 4 months of age, and (b) a model including time in inclined equipment, birth (singleton or multiple), and sex would significantly predict CVA ≥ 12mm. at 4 months of age. Study Two had three hypotheses: (a) Image J software measurement of digital photographs would demonstrate good intra-rater and inter-rater reliability (ICC values of at least .75) with infants in supine and with infants in supported sitting, (b) Image J software measurement of 3D Digital Surface Images with infants in a supported sitting position would demonstrate good intra-rater and inter-rater reliability (ICC values of at least .75), and (c) there would be no significant difference of head tilt measurements comparing Image J software measurement of digital images and manual measurement of the printed digital images.

Hypotheses for Study Three were: (a) Infants with less than 5° of head tilt at the beginning of treatment would demonstrate greater change in CVA than infants with ≥ 5° of head tilt at the beginning of treatment, and (b) infants younger than 5 months of age at the beginning of DOC band treatment would demonstrate greater change in CVA than infants 5 months and older.

This project has clinical significance. It provides better understanding of the influence of infant positioning on the development of brachycephaly and plagiocephaly as well as the influence of torticollis on CVA improvement during cranial band treatment. It also supports the importance of physical therapy to address torticollis in infants receiving cranial band treatment to achieve best outcomes. Finally, a new software measurement method of measuring torticollis from digital images is presented.

CHAPTER II

STUDY ONE

Inclined Positioning and Development of Brachycephaly and Severe Plagiocephaly in 4-Month-old Infants

Inclined equipment is defined as equipment that positions infants on their back in an inclined position. The baby is securely strapped in the equipment with safety harnesses and belts. Examples of inclined equipment are swings, bouncy seats, inclined sleepers, and car seats. Car seats conveniently snap in and out of stroller bases allowing the baby to stay in the same static position during travel and after removal from the car. Swings rock and play music while the baby sleeps securely strapped in an inclined position.

Pediatric physical therapists who specialize in cranial band treatment report that parents often think their babies are more comfortable and sleep more soundly when they are positioned in inclined equipment. When an infant is positioned in inclined equipment, their chin is tucked with the neck in a flexed position and the head is often tilted to one side. The pressure of the equipment against the back of the head is primarily on the lower occiput, influencing the head to grow taller and wider with a flattened occiput. Unrestrained in the supine position (no incline), the pressure of the surface on the infant's head is on the upper posterior cranium. Since the baby is not strapped into a flexed position, there is freedom to extend the neck and turn the head from side to side reducing the sustained pressure on the lower occiput. Although the AAP (2011) infant sleep recommendations specify supine sleeping in a crib without any additional inserts or positioning devices, parents often think they are following the AAP's back to sleep recommendations by having their baby sleep in inclined positioning equipment because they are on their backs in the equipment. If an infant spends prolonged periods in inclined equipment, the pressure on the back of the infant's head could become deformational (Collett, Breiger, King, Cunningham, & Speltz, 2005). No studies have determined quantitatively how much time in inclined equipment affects the development of brachycephaly or plagiocephaly.

Brachycephaly is characterized by flattening of the occipital area resulting in a tall, wide head shape. The CI is used to determine the severity of brachycephaly and calculated using the length and width measurements (cranial width/length x 100). Plagiocephaly presents as asymmetry of the cranium with flatness on one side of the parietal occipital area and contralateral flatness on the frontal area. Facial asymmetry and a shift in the ear and jaw alignment are characteristics of severe plagiocephaly. CVA is determined by measuring the length of the right and left diagonals of the cranial vault and recording the difference of one diagonal from the other in millimeters (Wildbrand et al., 2012). Different classification systems have been developed to determine the severity of brachycephaly (Branch et al., 2015; Hutchison, Hutchison, Thompson & Mitchell, 2005; Pindrik, Molenda, Uribe-Carenas, Dorafshar & Ahn 2016; Wildbrand, et al., 2012). For the current study, a CI $\geq$ 92 was used as the threshold for brachycephaly. The threshold for severe plagiocephaly was CVA $\geq$ 12 mm.

Jorganic et al. (2009) found that premature birth, multiple birth, and sex were predictors of deformational plagiocephaly but that sleep position was the best predictor of plagiocephaly. The sleep positions examined in Jorganic et al.'s (2009) study included supine with head turned right or left, side-lying right or left, and prone. In this study, head position while sleeping was linked to deformational plagiocephaly, specifically in infants who preferred to position their head turned to one side. However, sleeping in inclined equipment was not examined. The amount of time an infant spends in inclined equipment has not been identified as a predictor of brachycephaly or plagiocephaly. Pediatric physical therapists specializing in cranial band treatment observe that infants who spend more time in inclined positioning equipment typically present with more severe brachycephaly than infants who spend less time in incline. Infants presenting with brachycephaly often have plagiocephaly as well, but infants who do not sleep in inclined equipment seem to be more likely to present with plagiocephaly without brachycephaly.

Therefore, the purpose of this first study was to determine if the amount of time infants spend positioned in incline, birth (multiple or singleton), and sex, are significant predictors of brachycephaly (CI > 92) and/or severe plagiocephaly (CVA ≥ 12 mm) at 4 months of age. To achieve this purpose, the following hypotheses were proposed: (a) a model including time in inclined equipment, birth (singleton or multiple), and sex would significantly predict CI ≥ 92 at 4 months of age, and (b) a model including time in inclined equipment, birth (singleton or multiple), and sex would significantly predict CVA ≥ 12mm. at 4 months of age.

Methods

Following approval for the study by the Texas Woman's University Institutional Review Board, the primary investigator reviewed charts of 4-month-old infants who came for an evaluation at Cranial Technologies in Dallas, Texas from April 2016 through October 2016, retrospectively. Charts were included if the infant was 4 months old at the time of evaluation. Charts were excluded if the infant had a diagnosis of craniosynostosis, genetic syndrome, or skeletal abnormality such as cleft lip, facial deformity, or cervical spine abnormality. Additionally, if the infant was born prematurely (≤ 36 weeks gestation), or if the parents reported the infant was positioned in prone for sleeping, the charts were excluded.

The charts included three-dimensional (3D) digital images that were taken with the Digital Surface Imaging (DSI) system at Cranial Technologies in Dallas, Texas (Littlefield, Kelly, Cherney, Beals, & Pomato, 2004). VAM software version 5.4.2 (Canfield Scientific Inc, Parsippany, New Jersey) was used to take CI and CVA measurements from the digital images and recorded in the patient's chart. Reliability and validity of using a digital 3D imaging system for cranial vault measurements has been established (Skolnick, Naidoo, Nguyen, Patel, & Woo, 2015). The amount of time the infants spent in inclined positioning equipment during the first 2 months of life was reported by the parents and recorded in the charts as the number of hours per day in incline. Parents had also reported whether or not their baby slept in inclined positioning equipment at night and whether or not they transitioned from sleeping in inclined positioning equipment to sleeping supine (no incline) at 2 months of age.

Statistical analysis. Data from 78 charts were analyzed using IBM SPSS Statistics for Windows, Version 24 (IBM Corp, Armonk, New York) and two binomial logistic regression analyses were done. The predictor variables in each of the logistic regression models were hours per day in inclined equipment, birth (singleton or multiple), and sex (male or female). The two continuous dependent variables were reduced to dichotomous variables as follows: presence of brachycephaly (CI $\geq$ 92) or not, and presence of severe plagiocephaly (CVA $\geq$ 12 mm) or not.

From the charts included in the study, infants whose parents reported they slept in inclined positioning equipment at night for at least 2 months were identified. Parents had also reported whether or not they repositioned their baby from inclined equipment for sleeping to supine sleeping (no incline) at 2 months of age. An independent samples *t*-test was conducted to compare the mean CI of infants who slept in inclined equipment for at least 2 months to the mean CI of infants who slept supine. An independent samples *t*-test was also conducted to compare the mean CI of infants who slept in inclined equipment from birth to 2 months and then transitioned to supine sleeping to the mean CI of infants who never transitioned out of inclined equipment for sleeping (slept in inclined equipment for 4 months).

Results

Table 1 shows descriptive data from the 78 charts of infants who were 4 months old upon evaluation.

Table 1

Descriptive data of infants who were 4 months old upon evaluation.

Variable	Total
Sex	
Male	51 (65%)
Female	27 (35%)
Birth	
Singleton	57 (73%)
Multiple	21 (27%)
Cephalic Index (CI)	
Minimum	77.40
Maximum	101.60
Mean	91.078
Standard Deviation	4.858
CI ≥ 92	
No	35 (45%)
Yes	43 (55%)
Cranial Vault Asymmetry (CVA) in mm	
Minimum	1
Maximum	20
Mean	8.77
Standard Deviation	4.615
CVA ≥ 12 mm	
No	57 (73%)
Yes	21 (27%)
Hours per Day in Inclined Equipment	
Minimum	0.50
Maximum	22.00
Mean	9.494
Standard Deviation	6.135
Sleep Position	
Supine (No Incline)	32 (41%)
Inclined Equipment all 4 months of life	22 (28%)
Transitioned to Supine from Incline at 2 months of age	24 (31%)

Presence or absence of cephalic index ≥ 92. Linearity of the continuous variable, Incline Hours, with respect to the logit of the dependent variable Presence of CI ≥ 92 was assessed with the Box-Tidwell procedure. The continuous variable, Hours in Incline, was found to be linearly related to the logit of the dependent variable Presence of CI ≥ 92 (p = .06). No outliers with studentized residuals ≥ 3 standard deviations were found in the data.

A binomial logistic regression was performed to determine the effects of hours in incline equipment per day, birth (singleton or multiple), and sex on the likelihood that 4-month-old infants will have brachycephaly (CI ≥ 92). The logistic regression model was statistically significant $\chi^2(3) = 16.40$, $p = .001$. The model explained 25.0% (Nagelkerke R^2) of the variance in brachycephaly and correctly classified 70.5% of cases. Sensitivity was 74.4%, specificity was 65.7%, positive predictive value was 72.7%, and negative predictive value was 67.6%. Of the three predictor variables, only the hours spent in incline was statistically significant ($p < .001$). Sex and birth (singleton or multiple) were not significant predictors, ($p = .510$ and $p = .559$, respectively). That is, as the number of hours in incline increased, the probability of babies having brachycephaly increased (OR = 1.183, 95% CI [1.079,1.297]). For each hour per day the infants spent in inclined equipment, their likelihood of exhibiting brachycephaly (CI ≥ 92) at 4 months of age increased by 1.183 times (see Table 2).

Table 2

Logistic Regression Predicting Likelihood of Cephalic Index ≥ 92 based on Sex, Birth (Singleton or Multiple) and Hours in Incline

	B	SE	Wald	*P*	Odds Ratio	95% Confidence Interval	
						Lower	Upper
Sex	-.371	.563	.434	.510	.690	.229	2.079
Birth (singleton or multiple)	-.357	.610	.342	.559	.700	.211	2.315
Hours in Incline	.168	.047	12.861	.000	1.183	1.079	1.297
Constant	-.464	.714	.421	.516	.629		

Presence or absence of cranial vault asymmetry ≥ 12 mm. Linearity of the continuous variable, Hours in Incline, with respect to the logit of the dependent variable Presence of CVA ≥ 12 mm was assessed with the Box-Tidwell procedure. The continuous variable, Hours in Incline, was found to be linearly related to the logit of the dependent variable Presence of CVA ≥12 mm (p = .01). No outliers with studentized residuals ≥ 3 standard deviations were found in the data.

A binomial logistic regression was performed to determine the effects of hours in inclined equipment per day, birth (singleton or multiple), and sex, on the likelihood that 4-month-old infants will have severe plagiocephaly (CVA ≥ 12 mm). The logistic regression model was not statistically significant, $\chi^2(3) = 7.39$, $p = .061$.

Mean CI comparisons and infant sleep positions. Of the 78 infants' charts reviewed, 46 were identified with parent report that the infants slept in inclined equipment at night for at least 2 months, and 32 charts were identified with parent report that the infants slept supine (no incline). Infants who slept in inclined equipment for at least 2 months had a higher CI (*M* = 92.33, *SE* = 0.58) than infants who slept supine (*M* = 89.27, *SE* = 0.97). An independent samples *t*-test was performed to determine if the mean difference between the CI values for the two groups was statistically significant. The mean difference in CI comparing infants who slept in inclined equipment for at least 2 months, and infants who slept supine (no inclined sleeping) was found to be statistically significant, $t(52.79) = -2.706$, $p = .009$, and $d = 0.56$ which represents a medium effect size.

Of the 46 infants whose parents reported they slept in inclined equipment for at least 2 months, 24 transitioned from sleeping in inclined equipment to sleeping supine (no incline) at 2 months of age, and 22 infants slept in inclined equipment for the entire 4 months of their lives. Infants who transitioned from sleeping in inclined equipment to supine at 2 months of age had a similar CI (*M* = 92.63, *SE* = 0.82) as infants who never transitioned out of the inclined equipment for sleeping (*M* = 92.02, *SE* = 0.95). An independent sample *t*-test was performed to determine if the difference in mean CI comparing the two groups was statistically significant. The difference in mean CI scores comparing infants who transitioned from sleeping in inclined equipment to supine sleeping at 2 months of age and infants who never transitioned out of inclined equipment for sleeping was not significant, $t(44.00) = .514$, $p = .610$; the effect size was small, $d = 0.15$.

Discussion and Conclusion

This study found that the amount of time an infant spent in inclined equipment the first two months of their lives was a significant predictor of brachycephaly (CI ≥ 92) at 4 months of age. Comparing infants who slept in inclined equipment to infants who slept supine (no incline), the mean CI of the infants who slept in inclined equipment was significantly higher than the mean CI of the infants who did not. In fact, the mean CI of infants who slept in inclined equipment met the threshold for brachycephaly, while infants who slept supine (no incline) did not. Repositioning at 2 months of age by transitioning from inclined sleeping to supine sleeping with no incline was not effective in reducing the cephalic index. Based on this information, it appears that the occipital flattening occurred in the first 2 months of life. Birth (singleton or multiple) and sex were not significant predictors of brachycephaly. The hypothesis that time in inclined equipment, birth (singleton or multiple), and sex would be significant predictors of brachycephaly (CI ≥ 92) was partially correct. The regression model was a significant predictor of brachycephaly but only time in inclined equipment was significant.

The hypothesis that time in inclined equipment, birth (singleton or multiple), and sex would be significant predictors of severe plagiocephaly was not correct. Based on the results of Jorganic et al. (2009) that found multiple birth and males were more likely to develop plagiocephaly, it was surprising that in the current study, birth (singleton or multiple) and sex did not predict severe plagiocephaly. The current study's dependent variable of presence or absence of severe plagiocephaly (CVA ≥ 12 mm) was different than the dependent variable in Jorganic et al.'s (2009) study. In their study of charts of infants referred to Cranial Technology clinics across the country, Jorganic et al. (2009) reported that charts were excluded if they had brachycephaly without plagiocephaly (flatness to the back of the head without asymmetry). Other exclusion factors reported were if the infants had craniosynostosis, hydrocephalus, achondroplasia, or Down syndrome. All other charts were included as infants with plagiocephaly. Jorganic et al. (2009) explained that this was done because the patients were referred to Cranial Technologies after they were given a diagnosis by their physician. No data on the severity of the plagiocephaly or mean CVA of the infants was included in their study (Jorganic et al., 2009). Therefore, charts of infants with mild, moderate, and severe plagiocephaly were likely included. Perhaps the different dependent variable definition of plagiocephaly accounts for the current study's different results. Another difference is that the current study included 78 charts compared with 20,691 charts in Jorganic et al.'s (2009) study. With a very large study, there can be greater sensitivity for positive predictive values. Jorganic et al. (2009) did not report sensitivity and specificity results. Kaplan, Chambers, Phil, & Glasgow, (2014), caution that with large sample sizes (like data obtained from large electronic data bases), it is necessary to exercise greater caution to be sure that a large sample size does not lead to inferential errors. Kaplan et al. (2014) found that large sample size can magnify the bias associated with error resulting from sampling or study design.

A potential reason multiple birth was not a significant predictor in the current study is that we excluded premature infants, while Jorganic et al.'s (2009) study did not. Pre-term births are more common in multiple births. The cranial vault is more malleable in pre-term babies making them more susceptible to deformational plagiocephaly. Including only multiple birth babies that were born full-term in the current study could have decreased the effect of multiple birth on the predictor of CVA $\geq$ 12 mm. It would be more informative to include premature infants in future research.

The findings of our study support the importance of education at the earliest interaction with parents, preferably before the baby's birth, or during the first week of the baby's life. If brachycephaly is identified in a 2-month-old baby, repositioning alone is not likely to reduce the cephalic index. The AAP's positioning recommendations should be reviewed, including back to sleep in a crib and increased tummy time when the baby is awake. Care should be taken to ensure parents understand that sleeping in inclined positioning equipment does not comply with the back to sleep recommendations and can lead to the development of brachycephaly. If parents use inclined positioning equipment for convenience or other reasons when the baby is awake or for naps during the day, healthcare providers should be aware of the risk of brachycephaly with prolonged time in the positioning equipment and educate parents of the risk.

A weakness in the current study is that the retrospective chart review relied on parent reports of the number of hours per day the infants spent in inclined equipment. Some parents may have underestimated the actual number of hours spent in inclined equipment or may have been reluctant to truthfully report the number of hours. Future research including an objective method of measuring actual hours per day in inclined equipment would provide a more accurate analysis of how the amount of time in inclined equipment affects infant head shape. Finally, a healthcare provider referred the infants in our study to Cranial Technologies, or the infants were self-referred by parents who noticed flattening of their baby's head. The mean CI of 91.07 and mean CVA of 8.77

indicate the infants had at least moderate plagiocephaly or brachycephaly. It would be informative to study infants at their well-baby pediatric visits in order to include infants not referred to a plagiocephaly clinic in future research.

Our study adds to the knowledge of how infant positioning and sleep position contributes to brachycephaly. While therapists advocate limiting the amount of time infants are positioned in inclined equipment, our study determined that each hour an infant is positioned in inclined equipment increases the likelihood of developing severe brachycephaly by 1.183 times. Our study also determined that infants who sleep in inclined equipment have a higher cephalic index than infants who sleep supine, and that repositioning an infant from sleeping in inclined equipment to supine at 2 months of age is not effective at reducing the cephalic index. The practical application of our findings for the primary healthcare provider is that early education (before the baby is 2 months old) to parents on limiting time in inclined equipment is important in order to prevent the development of brachycephaly. Education should include making sure parents understand that using inclined equipment for sleeping is not in accordance with the AAP back to sleep recommendations, and increases the risk of developing brachycephaly.

CHAPTER III

STUDY TWO

Reliability and Validity of Using Digital Images of Infants in Supported Sitting and Supine Positions for Measurement of Torticollis

Infant torticollis is the third most common musculoskeletal diagnosis treated by pediatric physical therapists. An infant with torticollis presents with a lateral head tilt towards the tightened sternocleidomastoid and cervical rotation towards the unaffected side (Ohman, Mardbrink, Stensby, & Beckung, 2010). The pediatric section of the American Physical Therapy Association developed clinical practice guidelines for evaluation and treatment of torticollis and identified evidence-based evaluation methods (Kaplan, Coulter, & Fetters, 2013). Most of the methods listed in the clinical practice guidelines involve measuring the head tilt with the infant in a supine position. However, the supine position can be complicated by an additional diagnosis of plagiocephaly, characterized by flattening of the cranial vault in an asymmetric manner (Chang, Tang, Chen, Wong, & Wong, 1999; Chang et al., 2000; Chang et al., 2001; Klackenberg, Elfving, Haglund-Akerlind, & Carlberg 2005; Ohman & Beckung, 2008; Rahlin & Sarmiento, 2010). When an infant with plagiocephaly is positioned in supine, the influence of the asymmetric head shape against the surface contributes to increased rotation and lateral head tilt. The influence of plagiocephaly on the amount of head tilt measured during supine measurement compared to the amount of head tilt in a sitting position has not been studied.

In a previous study, Rahlin and Sarmiento (2010) determined intra-rater and inter-rater reliability of measuring lateral head tilt using printed digital photographs of infants in the supine position. Digital photographs were printed, and one line was drawn through irises of the eyes and extended laterally toward the side of the head tilt. Another line was drawn through the acromion processes of the shoulders and extended laterally until it intersected the first line. The angle of the intersected lines was measured with a protractor (see Figure 2).

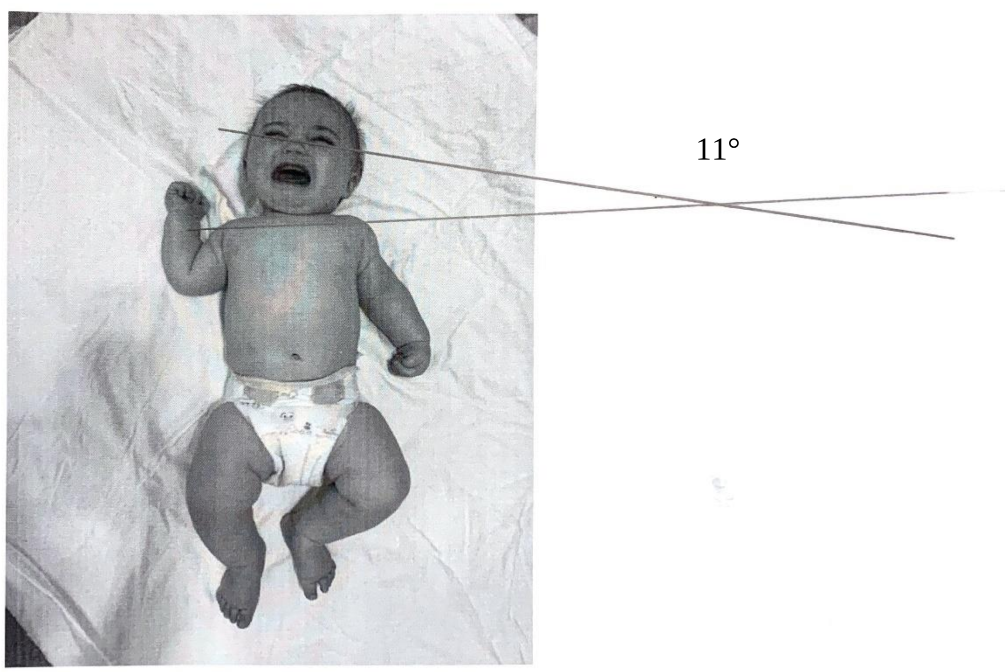

Figure 2. A Photograph of the Head Tilt Measurement Technique Described by Rahlin and Sarmiento (2010) that was Compared to the Computer Software Method in this Study.

The subjects in Rahlin and Sarmiento's (2010) study were being treated for plagiocephaly at a Cranial Technologies clinic. The authors did not report if they considered the effect of an asymmetric head while the baby was lying in supine, or if they attempted to position the head in neutral as much as possible.

Using digital photography of infants positioned in a supported sitting position eliminates the effect of the asymmetric head shape on head tilt. Without the influence of the cranial asymmetry on head positioning, better information is gained about the influence of the neck on the patient's torticollis. The physical therapist may want to consider using digital photography of infants in both the supine and seated positions for a more comprehensive evaluation.

The reliability and validity of using 3D imaging systems for cranial vault anthropometric measurements has been previously established (Metzler et. al., 2014; Skolnick, Naidoo, Nguyen, Patel, & Woo, 2015). Currently, 3D imaging is not used to measure lateral head tilt. However, since successful treatment of plagiocephaly often includes physical therapy for torticollis (Aarnivala, et. al., 2016; Kuo, Tritasavit, & Graham, 2014), a reliable method of torticollis measurement is needed to assess progress and treatment effectiveness. We wanted to establish reliability of measuring head tilt using a 2D digital image taken from a 3D image in a patient's electronic medical record. The head tilt measurement could then be included with the head shape anthropometric measurements, and the influence of the torticollis on the plagiocephaly included in the evaluation. It would be convenient to the clinician and to the family because the same 3D image could be used for the head tilt measurement without the need for an additional photograph.

The use of Image J software (Laboratory for Optical and Computational Instrumentation, Madison, Wisconsin) and digital images for measurement of lateral head tilt provides an updated method of measuring torticollis that is convenient for use with electronic medical records. Software measurement of digital photographs is more efficient for the pediatric therapist in any setting because it eliminates the need to access a printer to print out the images, manually measure the images, and then shred the printed patient identifiable image. The primary purpose of this second study was to determine inter-rater and intra-rater reliability of three new methods of measuring lateral head tilt seen in infant patients with torticollis. Two new methods used Image J software measurement of digital photographs of infants in supine, and supported sitting positions. The third new method used Image J software measurement of a 2D digital image obtained from a 3D digital image of infants in a supported sitting position. The last purpose of the study was to determine concurrent validity of using Image J software to measure the head tilt of digital images compared to manual measurement of printed digital images.

To achieve these purposes, the study had three hypotheses: (a) Image J software measurement of digital photographs would demonstrate good intra-rater and inter-rater reliability (ICC values of at least .75) with infants in supine and with infants in supported sitting, (b) Image J software measurement of 3D Digital Surface Images with infants in a supported sitting position would demonstrate good intra-rater and inter-rater reliability (ICC values of at least .75), and (c) there would be no significant difference of head tilt measurements comparing Image J software measurement of digital images and manual measurement of the printed digital images.

Methods

Participants. Twenty-four male and female infants aged 4-10 months who were diagnosed with plagiocephaly were included. This was a convenience sample of infants who were receiving cranial band treatment at Cranial Technologies in Dallas, TX. Infants were excluded if they had a syndrome that presents with atypical musculoskeletal facial or shoulder features, a skeletal defect of the trunk or face, or if they could not hold their head up in a supported sitting position.

Instrumentation. The Digital Surface Image (DSI) system (Littlefield et. al., 2004) used at the Dallas, Texas, Cranial Technologies clinic was used to take the 3D images (Cranial Technologies Inc., Tempe, AZ). The DSI system includes 15 digital cameras positioned around and above an adjustable stool that the infant is positioned on in a supported seated position. The system uses VAM software version 5.4.2 (Canfield Scientific Inc., Parsippany, NJ) to processes the 3D image.

Image J, a software program developed by the National Institutes of Health (Laboratory for Optical and Computational Instrumentation, Madison, Wisconsin) was used to measure the head tilt of the digital photographs as well as the “snipped” 2D images taken from the 3D digital images (Schneider, Rasband, & Eliceiri, 2012). Image J is freely available at http://imagej.net/. A plug-in that was developed by a computer programmer was used with the Image J software. The plug-in program is freely available at https://github.com/rcshill/angle_from_points.git. The plug-in allowed the program to automatically calculate the angle after using the computer mouse to click on the two eye landmarks (lateral commissures of the eyes) and the two shoulder landmarks (acromion processes). In Rahlin and Sarmiento's (2010) conclusion, the authors recommended using the lateral commissures of the eyes for the eye landmarks (the lateral corner of the upper and lower eyelids where they meet) instead of the irises of the eyes that they used in their study. The recommend landmarks were used in the current study.

Procedures

After obtaining IRB approval from the Texas Woman's University IRB committee (see Appendix), parents who gave permission for their infant to participate read and signed a consent form explaining the purpose of the study. The principal investigator (PI) asked the parents about the medical history of the infant to determine eligibility for the study.

3D imaging session. The parents were asked to undress the participant down to his/her diaper and bring the infant into the DSI room. The PI placed a thin nylon stockinet, with an opening cut out for the infant's face, on the participant's head. The PI then placed the infant on a stool in a supported sitting position and sat on a stool behind the infant to provide support to the infant's trunk while holding both arms at the infant's side. The infant was positioned facing one of the 15 digital cameras in the DSI system. Each participant was faced toward the same designated camera. Using a stimulating toy, a physical therapy student investigator provided a visual stimulus to the infant in order to facilitate as neutral a head position as possible. Head rotation to either side was minimized, and any leaning of the infant's body to one side or the other was avoided. Another physical therapy student investigator took three digital images before the infant was given back to his/her parent with the stockinet removed. Finally, the PI selected the image with the best posture (most neutral position of the head and neck) to be used for the study. The 3D image was saved to a digital folder.

Supported sitting photograph session. The infant was again positioned on the stool and held in the same supported sitting position by the PI. This time the infant was faced toward a physical therapy student who took a digital photograph of the infant, holding a Nikon D5000 digital camera directly in front of the infant at the infant's eye level. While the photograph was being taken, a second physical therapy student provided a visual stimulus with a toy to facilitate a neutral head position in midline. Three photographs of the infant were taken. Finally, the PI chose the photograph with the best posture and saved it in a digital folder.

Supine photograph session. The parent was directed to bring the infant to a designated area with a mat on the floor. A clean disposable sheet was placed on the mat, and the parent was asked to place the infant supine on the mat. While one physical therapy student provided a visual stimulus directly above the infant's head to facilitate a midline position, another physical therapy student took three pictures with the digital camera held directly over the infant. Finally, the PI chose the photograph with the best posture and saved it to a digital file.

Preparing the images for data collection. For the DSI measurement method, although VAM software is able to measure an angle from three identified points on the 3D image, there is not a method to measure the angle of the intersecting extended lines from the four landmarks (two eye landmarks and two shoulder landmarks). In addition, coding a plug-in for the 3D environment was difficult because the lines from the landmarks often were not on the same plane and therefore did not intersect. Therefore, for this study, a 2D image was created from the 3D image using the snipping tool from Microsoft Windows 8.1 Pro software. Following the collection of the digital images, the PI used the snipping tool to "snip" a 2D digital image from each 3D digital surface image. Nine digital copies of the 2D images were made for each participant and a designated participant code was labeled on each copy with an added 1A, 2A, 3A respectively on three of the copies, 1B, 2B, 3B respectively on three of the copies, and 1C, 2C, 3C respectively on three of the copies. The three "A" copies were placed in a digital folder labeled "DSI A." The three "B" copies were placed in a folder labeled "DSI B." The three "C" copies were placed in a folder labeled "DSI C."

Repeating this procedure, the principal investigator made nine digital copies of the supported sitting photographs for each participant. The designated participant code was labeled on each copy with an added 1D, 2D, 3D respectively on three of the copies, 1E, 2E, 3E respectively on three of the copies, and 1F, 2F, 3F respectively on three of the copies. The three "D" copies were placed in a digital folder labeled "Sitting D." The three "E" copies were placed in a folder labeled "Sitting E." The three "F" copies were placed in a folder labeled "Sitting F."

Once more, the principal investigator made nine digital copies of the supine photographs for each participant. The designated participant code was labeled on each copy with an added 1G, 2G, 3G respectively on three of the copies, 1H, 2H, 3H respectively on three of the copies, and 1I, 2I, 3I respectively on three of the copies. The three “G” copies were placed in a digital folder labeled “Supine G.” The three “H” copies were placed in a folder labeled “Supine H.” The three “I” copies were placed in a folder labeled “Supine I.”

Head tilt measurements. To measure the digital images, the two physical therapy student investigators (rater 1 and rater 2) used Image J software with the plug-in program. For each image, they identified the lateral commissure of each eye and the acromion process of each shoulder (the top lateral third of the shoulder) and used the computer mouse to click on the landmarks. After the landmarks were clicked the angle of the head tilt was displayed on the computer screen and the measurement was recorded and kept in the same digital folder that the image was in. For the digital surface image measurements, rater 1 measured the head tilt on the images in the digital folder labeled “DSI A.” Rater 2 measured the head tilt on the images in the digital folder labeled “DSI B.” The raters measured the images during different sessions in order to keep them blinded to the measurements of each other. They were only able to access the folders from which they were working. At least one week later, rater 1 repeated the process measuring the images in the digital folder labeled “DSI C.” This measuring procedure was repeated for the sitting photographs and the supine photographs. Rater 1 measured the photographs in folders Sitting D, Sitting F, Supine G, and Supine I while rater 2 measured the photographs in folders Sitting E and Supine H.

The DSI images and the sitting and supine photographs of each participant were printed. The PI manually measured each printed image using the method described in Rahlin and Sarmiento's (2010) study. Using a pencil and a ruler, a line was drawn through the lateral commissures of the eyes and extended laterally toward the side of the head tilt. Another line was drawn through the acromion processes and extended laterally until it intersected with the first line. If needed, additional paper was attached to the printed image in order for the lines to extend until they crossed. A protractor was used to measure the angle of the intersecting lines. The PI was blinded to the Image J measurements of rater 1 and rater 2.

Data analysis. Intraclass Correlation Coefficient ($ICC_{3,1}$) analysis was used to determine intra-rater reliability of measurements taken by rater 1 at each of the two measurement sessions for each image position. Intraclass Correlation Coefficient ($ICC_{2,1}$) analysis was used to determine inter-rater reliability of measurements taken by rater 1 and rater 2 at the first measuring session of each image position.

Paired sample *t*-tests were used to determine concurrent validity of Image J measurements of the digital images of rater 2 with the manual measurement of the printed images taken by the principal investigator.

Results

Twenty-four infants participated in the study. All of the infants were receiving cranial band treatment at Cranial Technologies. Their ages ranged between 6-12 months. There were 10 female and 14 male infants. Descriptive statistics are included in Table 3

Table 3

Descriptive Statistics

Image/Position/Rater	Mean Head Tilt (degrees)	Min Head Tilt (degrees)	Max Head Tilt (degrees)	Standard Deviation	Standard Error Mean
DSI (N=24)					
Rater 1	4.925	0.49	16.94	4.695	0.958
Rater 2	5.198	1.07	15.33	4.322	0.882
Photo Sitting (N=24)					
Rater 1	5.177	0.61	20.14	4.422	0.903
Rater 2	5.330	0.83	20.70	4.494	0.917
Photo Supine (N=24)					
Rater 1	5.419	0.59	15.94	4.088	0.835
Rater 2	6.134	0.31	18.84	4.704	0.960
Manual Measurement of Printed Images (N=24)					
DSI	5.979	1.00	15.00	4.898	1.00
Photo Sitting	5.208	0.00	17.00	4.657	0.951
Photo Supine	6.29	0.00	18.00	4.912	1.00

Note. DSI = 2D picture of a 3D Digital Surface Image taken of each infant in a supported sitting position.

Intraclass Correlation Coefficient Analysis. Intra- and inter-rater reliability results of Image J measurement of head tilt on all digital image positions are included in Tables 4 and 5. All ICCs were above .95.

Table 4

Intra-rater Reliability of measurements taken by rater 1 at each of the two measurement sessions for each image position

Image/Position	($ICC_{3,1}$)	95% CI	Standard Error of Measurement
DSI	.990	.977 to 0.996	0.870
Photograph in Sitting	.979	.951 to 0.991	1.321
Photograph in Supine	.995	.988 to 0.998	0.658

Note. DSI = 2D picture of a 3D Digital Surface Image taken of each infant in a supported sitting position. ICC = Intraclass Correlation Coefficient

Table 5

Inter-rater Reliability of measurements taken by rater 1 and rater 2 at the first measuring session of each image position

Image/Position	($ICC_{2,1}$)	95% CI	Standard Error of Measurement
DSI	.973	.939 to 0.988	1.473
Photograph in Sitting	.977	.947 to 0.990	1.344
Photograph in Supine	.970	.933 to 0.987	1.515

Note. DSI = 2D picture of a 3D Digital Surface Image taken of each infant in a supported sitting position. ICC = Intraclass Correlation Coefficient

Paired *t*-test Analysis. Results of the *t*-test analysis to determine concurrent validity of the Image J measurement method of digital images compared with manual measurement of printed images are included in Table 6. No significant differences were found when comparing the manual measurements of the printed images with Image J measurement of the digital images for DSI, sitting photos, and supine photos ($p > 0.05$). Regarding the DSI measurements, two outliers were detected that were more than 1.5 box-lengths from the edge of the box in a boxplot. Since inspection of these values did not reveal them to be extreme, they were kept in the analysis. The difference scores for the Image J DSI measurements and the manual measurements were normally distributed as assessed by Shapiro-Wilk's test ($p = 0.277$). Regarding the sitting photos, one outlier was detected that was more than 1.5 box-lengths from the edge of the box in a boxplot. Again, inspection of this value did not reveal it to be extreme, and it was also kept in the analysis. The difference scores for the Image J sitting photo measurements and the manual measurements were normally distributed as assessed by Shapiro-Wilk's test ($p = 0.364$). Finally, regarding the supine photos, there were no outliers in the data as assessed by inspection of a boxplot for values greater than 1.5 box-lengths from the edge of the box. The difference scores for the Image J supine photo measurements and manual measurements were normally distributed as assessed by Shapiro-Wilk's test ($p = 0.336$).

Table 6

Concurrent Validity of Image J measurements of the digital images of rater 2 with the manual measurement of the printed images taken by the principal investigator

Image/Position	$t(23)$	p	95% CI
DSI	1.052	0.304	-0.475 to 0.155
Photograph in Sitting	0.008	0.994	-0.449 to 0.453
Photograph in Supine	0.388	0.701	-0.558 to 0.382

Note. DSI = 2D picture of a 3D Digital Surface Image taken of each infant in a supported sitting position.

Discussion and Conclusion

All four hypotheses were confirmed. Image J software measure of: infant head tilt from 3D images of infants in a supported sitting position, digital photographs of infants in a supported sitting position, and digital photographs of infants in supine were all found to be reliable (ICC > .75). Concurrent validity of Image J software measurement was also established. The angle of head tilt calculated using Image J software from digital images was consistent with manual measurement of printed images.

A weakness of the study was that the mean head tilt value was low. Some of the infants did not have torticollis despite being diagnosed with plagiocephaly. Although there were some infants with a high degree of head tilt, it would be informative to include more infants with a greater degree of torticollis. In order for the measurers to have a greater range of head tilt to measure, a greater range may affect the reliability of the measurement method.

The computer software head tilt measurement method is an updated version of Rahlin and Sarmiento's (2010) manual measurement method described in their study. The new method used in the current study will be easier to incorporate in electronic medical records now used by most pediatric therapists. With reliability and validity established with the infant in both sitting and supine positions, the pediatric physical therapist can easily include head tilt measurements of an infant patient in both positions for a more comprehensive depiction of an infant's torticollis. The use of Image J software to measure the head tilt of DSI images can be done with the same image that is routinely taken in the plagiocephaly clinic for head shape analysis. This allows clinicians to obtain a more comprehensive evaluation of the plagiocephaly patient by including the head tilt measurement with the anthropometric head shape measurements.

CHAPTER IV

STUDY THREE

Results of Cranial Band Treatment for Plagiocephaly in Infants with Torticollis Compared with Infants without Torticollis

The literature is divided on whether or not plagiocephaly causes future functional and psychological issues (Collett, Breiger, King, Cunningham & Speltz, 2005; Feijen, Franssen, Vincken &Van der Hulst, 2015; Shamji, Fric-Shamji, Merchant & Vassilyada, 2012). However, when infant plagiocephaly is severe, it causes facial asymmetry and a shift in ear and jaw alignment. With moderate or severe plagiocephaly reported at 10-12%, more parents are choosing cranial band treatment to improve their baby's headshape (Mawji, et al., 2014).

Cranial band treatment has been found to be effective in improving facial asymmetry and jaw alignment, as well as cranial vault asymmetry (Kreutz, et al., 2018; Kunz et al., 2019). Cranial band treatment is typically provided to infants with moderate and severe plagiocephaly. Optimal results are obtained if cranial band treatment begins before the infant is 6 months of age (Aarnivala et al., 2016). This treatment involves the creation of a custom-made cranial helmet that provides gentle corrective pressure to the bulged parietal occipital area and contralateral bulged frontal temporal area while providing growing space in the contralateral areas. The PI is a physical therapist who has experience providing cranial band treatment using the DOC band (Cranial Technologies, Tempe, AZ). This study examined results of infants treated with the DOC band at Cranial

Technologies, a plagiocephaly clinic in Dallas, Texas.

Infants with severe plagiocephaly often also present with torticollis. While severe plagiocephaly causes facial asymmetry, torticollis also contributes to the facial asymmetry as the tight sternocleidomastoid muscle pulls on one side of the head and face (Stellwagen, Hubbard, Chambers, & Lyons Jones, 2008; Yu, Wong, Lo, & Chen, 2004). It can be difficult to determine if the plagiocephaly caused the torticollis or if the torticollis caused the plagiocephaly (Pan & Tong, 2017). With plagiocephaly, the infant's head rolls to the flatter posterior surface in the supine position and the contralateral sternocleidomastoid muscle shortens. The AAP's emphasis on the importance of supine sleep positioning, along with the increased availability of more supine positioning equipment, may contribute to torticollis. Torticollis can be difficult to resolve without aggressive physical therapy treatment that includes a home exercise program where parents do exercises with the infant multiple times a day.

CVA is determined by measuring the length of the right and left diagonals of the cranial vault and recording the difference of one diagonal from the other in millimeters. While providing cranial band treatment to infants with moderate and severe plagiocephaly, the PI observed that although all infants demonstrated improvements in CVA with DOC band treatment, infants with torticollis seemed to show less improvement than infants without torticollis. This was particularly evident in infants who began treatment with severe plagiocephaly. In order to determine if this clinical hypothesis had merit, DOC band treatment outcomes of infants with severe plagiocephaly with and without torticollis were examined in this study.

The outcome examined in this study was change in CVA. It should be noted that at Cranial Technologies, DOC band treatment is provided by physical and occupational therapists who evaluate the head and neck and provide a home exercise program for parents to address torticollis. Infants with torticollis are also referred to outside physical therapy. Therefore, while two groups of infants with severe plagiocephaly were compared (infants with torticollis and infants without torticollis), the infants in the torticollis group did receive intervention from a physical and/or occupational therapist that addressed the torticollis throughout DOC band treatment. Because there are few studies on the effect of torticollis on cranial band treatment outcomes, this study was conducted to add to the evidence available. Therefore, the purpose of this study was to determine if torticollis affects the DOC band treatment outcome of change in CVA. We hypothesized that infants with less than 5° of head tilt at the beginning of treatment would demonstrate greater change in CVA than infants with ≥ 5° of head tilt at the beginning of treatment. In addition, we expected that infants younger than 5 months of age at the beginning of DOC band treatment would demonstrate greater change in CVA than infants aged 5 months and older.

Methods

Participants. Following approval for the study by the Texas Woman's University Institutional Review Board, the PI reviewed charts of infants 3 to 15 months of age who received DOC band treatment at Cranial Technologies in Dallas, Texas from August 2017 through December 2017, retrospectively. Charts were included in the study if the infant began treatment with severe plagiocephaly (≥ 12 mm of CVA). Charts were excluded if the infant had a diagnosis of craniosynostosis, genetic syndrome, or skeletal abnormality such as cleft lip, facial deformity, or cervical spine abnormality. Additionally, if the infant was born prematurely (≤ 36 weeks gestation), the charts were excluded.

Procedures. The age at the beginning of DOC band treatment and CVA before and after treatment was extracted from the data. CVA measurements in the charts were obtained from the DSI system at Cranial Technologies. The DSI system takes 3D images of the infant's head and identifies the arthrometric landmarks on the head used to measure CVA. The measurements were then digitally obtained from VAM software version 5.4.2 (Canfield Scientific Inc, Parsippany, New Jersey). The reliability and validity of using a 3D imaging system for cranial vault anthropometric measurements has been established (Metzler, et al., 2014; Skolnick, et al., 2015). Change in CVA was obtained by subtracting the CVA measurement before DOC band treatment from the CVA measurement taken after the DOC band treatment. Head tilt in degrees was used as the measure of torticollis. Head tilt measurements were not part of the infant's charts. The charts included 3D digital images of the infants in a supported sitting position that were taken with the DSI system. The primary investigator obtained head tilt measurements of the infants from the supported sitting 3D images taken at the beginning of treatment using the computer software head tilt measurement method described in Chapter Three of this dissertation (Study Two). This method used Image J software (Laboratory for Optical and Computational Instrumentation, Madison, Wisconsin), a software program developed by the National Institutes of Health, to determine the angle of two intersecting lines. One line going through the eyes and extending laterally toward the head tilt, and a second line going through the acromion processes of the shoulders and extending laterally until it intersected with the first line. Reliability and validity of using Image J software with a designed plug-in program to obtain head tilt measurements from DSI images of infants in a supported sitting position was demonstrated in Chapter Three (Study 2). Head tilt measurements were used to group participants as being $< 5°$ or $\geq 5°$.

Statistical analysis. Data from 67 charts were analyzed using IBM SPSS Statistics for Windows, Version 24 (IBM Corp, Armonk, New York). A two-way ANOVA was performed comparing the independent **v**ariable age at beginning of treatment (< 5 months, ≥ 5 months) and head tilt ($< 5°$ of head tilt, $\geq 5°$ of head tilt). The dependent variable was change in CVA. The assumptions necessary for performing a two-way ANOVA were examined. Outliers were assessed by inspection of a boxplot, normality was assessed using Shapiro-Wilk’s normality test for each cell of the design and homogeneity of variances was assessed by Levene’s test.

Results

Table 7 shows descriptive statistics from the 67 charts of infants who had a CVA ≥ 12 mm according to the independent variables, age group and head tilt group. The infants were between 3-14 months of age.

Table 7

Descriptive Statistics of Infants 3-14 Months of Age with a Cranial Vault Asymmetry ≥ 12mm.

Variable	Group	Number (Total = 67)	Mean	Standard Deviation	Standard Error Mean
Head Tilt					
	< 5°	45	2.40	1.37	.20
	≥ 5°	22	10.70	5.81	1.20
Age					
	< 5 months	27	4.00	.67	.130
	≥ 5 months	40	7.51	1.57	.25

Table 8 shows the descriptive statistics for the dependent variable, change in CVA, by head tilt group. The parametric assumptions for ANOVA analysis were met. There were no outliers, residuals were normally distributed (p < .05) and there was homogeneity of variances (P = .875).

Table 8

Descriptive Statistics for Change in CVA by Head Tilt Group

Head Tilt Group	Age Group	Mean Change in CVA mm	Standard Deviation	Number
< 5°	< 5 months	8.00	2.30	18
	≥ 5 months	6.78	2.06	27
	Total	7.27	2.22	45
≥ 5°	< 5 months	7.89	2.26	9
	≥ 5 months	5.39	2.14	13
	Total	6.41	2.48	22
Total sample	< 5 months	7.96	2.24	27
	≥ 5 months	6.33	2.17	40
	Total	6.99	2.33	67

Note. All of the patients experienced improvement in CVA (CVA before DOC band treatment was greater than CVA after treatment).

The two-way ANOVA revealed that the interaction effect between age and head tilt on change in CVA was not statistically significant $F(1,63) = 1.244$, $p = .269$, partial $\eta^2 =.019$. Therefore, analysis of the main effects for age and head tilt were performed. Figure 3 is a clustered bar chart showing mean change in CVA by age group and head tilt group.

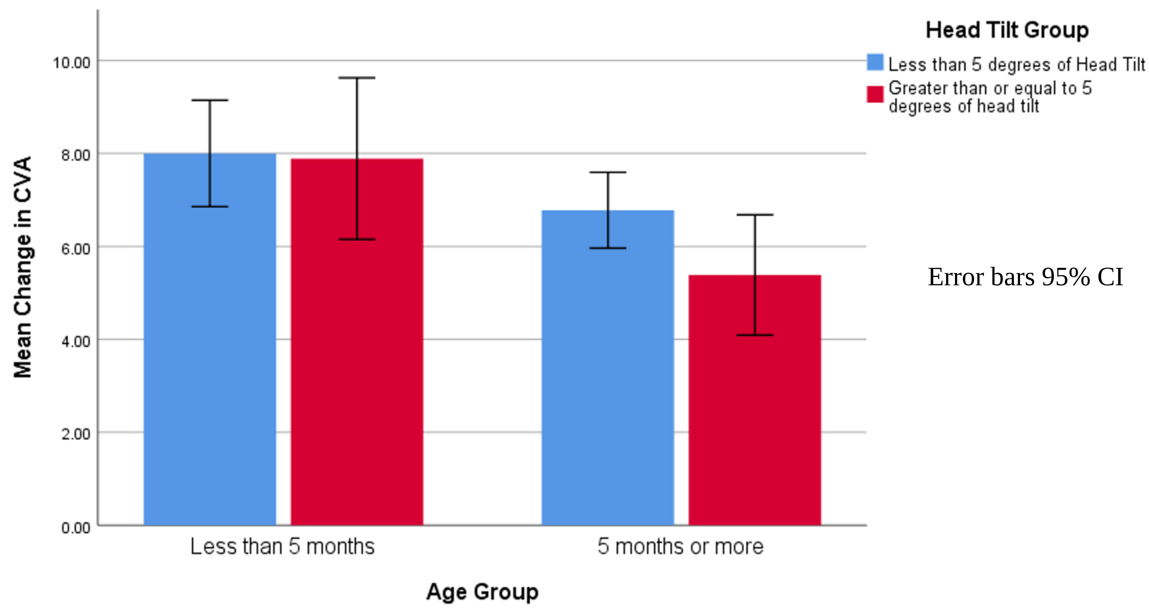

Figure 3 Clustered Bar Chart Showing Mean Change in CVA by Age Group and Head Tilt Group. Note: CVA = cranial vault asymmetry, CI = confidence Interval

When head tilt groups were pooled, there was a statistically significant difference in change in CVA measurements between infants < 5 months of age and infants ≥ 5 months of age, $F(1,63) = 10.510$, $p = .002$, partial $\eta^2 = .143$. Age accounted for 14.3% of the variance in change in CVA measurements. An age of < 5 months was associated with a mean change in CVA measurement of 1.86 mm [95% CI, .715 to 3.01] greater than an age ≥ 5 months.

When the age groups were pooled, there was no statistically significant difference in change in CVA measurements between infants with < 5° of head tilt, and those with infants with ≥ 5° of head tilt $F(1,63) = 1.713$, $p = .195$, partial $\eta^2 = .026$. Head tilt only accounted for 2.6% of the variance.

Discussion and Conclusion

The finding that torticollis as assessed by head tilt $\geq 5°$ in a supported sitting position did not have a significant effect on change in CVA was surprising. However, it was consistent with the findings of a previous study (Graham, Gilbert, Witthoff, Gregory & Walsh, 2019). Graham et. al. (2019) found that while torticollis contributed to a longer cranial band treatment time, it did not contribute to less improvement from the treatment. In the same study, age and severity were found to contribute not only to a longer treatment time, but also to less head shape improvement from the treatment (Graham, et al., 2019).

One explanation for the result of similar change in CVA among infants with and without torticollis is that the charts reviewed were from infants who received DOC band treatment at Cranial Technologies. At Cranial Technologies, DOC band treatment is provided by physical and occupational therapists in addition to orthotists. Babies who present with torticollis are provided with an assessment of the torticollis and a home exercise program is implemented in order to address the torticollis. Throughout treatment, the neck is monitored and exercises are reviewed with the parent or caregiver. In cases of severe torticollis, babies are referred to outpatient physical therapy services so that the torticollis-focused treatment can be carried out in parallel with DOC band treatment. The home program torticollis intervention from the Cranial Technologies physical and occupational therapists could have contributed to the result that infants with torticollis achieved similar improvements compared to infants without torticollis. Further study with infants who received cranial band treatment without physical or occupational therapy treatment for torticollis may provide more information. If there are infants who did not have therapy treatment for torticollis during cranial band treatment, the main effect may not be affected by the therapy treatment; however, it would not be ethical to design a study that denied infants access to physical therapy needed to treat the torticollis.

From anecdotal observations in the clinic, it seemed to the PI that the presence of torticollis resulted in a less optimal overall outcome from cranial band treatment, especially in infants who begin treatment with severe plagiocephaly. One possible explanation for this could be that the pull of the sternocleidomastoid muscle contributes to facial asymmetry. After treatment, if torticollis persists, the facial asymmetry is typically still present despite the improved head shape. Infants without torticollis typically do not have as severe facial asymmetry, and therefore the overall outcome from cranial band treatment appears better. The PI could have mistakenly assumed babies with torticollis and residual facial asymmetry had less change in CVA, when in fact the change was similar to the change experienced by babies without torticollis.

The result that age significantly affected the change in CVA was expected. From the PI’s clinical experience, and from evidence from previous studies, infants who begin cranial band treatment at younger ages experience a shorter treatment time (Kluba, Kraut, & Krimmel, 2011; Seruya, Oh, Taylor, Sauerhamma & Rogers, 2013). Furthermore, earlier treatment is typically associated with a more optimal outcome, especially for infants with more severe asymmetry (Kluba, et al., 2011; Seruya, et al., 2013). When an infant begins treatment with severe plagiocephaly, cranial band treatment at earlier ages typically results in better outcomes. This is because there is a limited amount of time during younger ages when the head growth is fast enough for a cranial band to be effective for the longer duration of treatment needed to correct the severe deformity. Pediatricians and other healthcare providers typically refer patients with severe plagiocephaly to plagiocephaly clinics for cranial band treatment prior to 5 months of age. Kluba et al. (2011), recommend that infants with plagiocephaly receive cranial band treatment when the infant is 5-6 months of age (preferably not older than 6 months). These researchers recommend infants receive physical therapy concurrently with cranial band treatment instead of doing physical therapy before cranial band treatment (Kluba et al., 2011). We agree with Kluba et al. (2011) that if a baby has severe plagiocephaly, cranial band treatment should be done before the baby is 6 months old. In our opinion, with severe plagiocephaly, optimal outcomes from cranial band treatment are achieved if treatment begins at 3-4 months of age.

In conclusion, all infants in our study with severe plagiocephaly experienced improved CVA with DOC band treatment. The amount of improvement achieved from the treatment was not influenced by torticollis. In addition, the amount of change in CVA was significantly greater among infants younger than 5 months, compared with infants 5 to 14 months. Parents of infants with severe plagiocephaly and torticollis should expect similar improvements in CVA from DOC band treatment as infants with severe plagiocephaly without torticollis. However, it is important for infants with torticollis to receive physical therapy treatment to treat the torticollis in addition to the DOC band treatment.

CHAPTER V

CONCLUSIONS

The overarching purpose of this project to investigate how positioning and torticollis influence the development and treatment of abnormal infant head shapes was achieved. The findings of all three studies provide new evidence of how positioning and torticollis influence the development and treatment of abnormal infant head shapes. The first study gives physical therapists and other pediatric practitioners quantitative information on the influence of inclined positioning equipment on the development of brachycephaly. With this information, pediatric clinicians will be able to educate parents of newborn infants that for every hour per day the infant spends in inclined equipment, they are 1.18 times more likely to develop brachycephaly. For example, if an infant sleeps in an incline sleeper during naps and at night and spends 15 hours a day in this equipment, the infant is 17.7 times more likely to develop brachycephaly than an infant who sleeps supine with no incline (15 x 1.18 = 17.7). The information from the first study provides quantitative evidence to the pediatric clinician's anecdotal professional opinion that parents should limit the amount of time they keep their baby in inclined positioning equipment. The pediatric clinician will also know that it is extremely important that parents have this information as soon as possible after the baby is born. The evidence from the first study showed that if brachycephaly is noticed when the baby is 2 months old, repositioning does not provide improvement when assessed at 4 months of age.

The third study provides information about how torticollis affects cranial band treatment in infants with severe plagiocephaly. Evidence from the third study shows that infants with torticollis, will demonstrate similar improvement of their head shape from DOC band treatment and physical therapy, as infants without torticollis. In addition, from Study Three, clinicians who provide DOC band treatment will have more evidence that better outcomes are achieved if an infant with severe plagiocephaly begins DOC band treatment before they are 5 months of age.

Information from Study Two provides pediatric physical therapists an updated tool for measuring torticollis. The software measurement method of measuring head tilt is more convenient, especially if the clinician uses electronic medical records that most practicing therapists now use. The method does not require a printer to print photographs of the infant, or the extra time it takes to mark the printed image and measure the head tilt with a protractor. Instead, after a photograph is uploaded to the computer, with four clicks of the mouse on the two eye landmarks and the two shoulder landmarks, the head tilt measurement is achieved.

Finally, in the plagiocephaly clinic, 3D images are a regular part of a head shape evaluation. Because the software head tilt measurement method for use on 3D images was found to be reliable and valid, head tilt measurements could be obtained from the 3D images taken for the head shape analysis, and included in the plagiocephaly evaluation. At Cranial Technologies, where DOC band treatment is provided by physical and occupational therapists, the additional head tilt measurement can help the therapist evaluate the neck before, during and after DOC band treatment. This quantitative evaluation of torticollis will help the clinician determine if it is improving, or if more intervention is needed.

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APPENDIX A

Texas Woman's University Institutional Review Board Approval Letter for the Research Study: Reliability and Validity of Using Digital Images of Infants in Supported Sitting and Supine Positions for Measurement of Torticollis

Institutional Review Board
Office of Research and Sponsored Programs
P.O. Box 425619, Denton, TX 76204-5619
940-898-3378
email: IRB@twu.edu
http://www.twu.edu/irb.html

DATE: November 30, 2016

TO: Ms. Julie Shill
Physical Therapy - Dallas

FROM: Institutional Review Board (IRB) - Dallas

Re: Approval for Reliability of Using Digital Images of Infants in Supine and Supported Sitting Positions for Measurement of Torticollis (Protocol #: 19309)

The above referenced study was reviewed at a fully convened meeting of the Dallas IRB (operating under FWA00000178). The study was approved on 11/22/2016. This approval is valid for one year and expires on 11/22/2017. The IRB will send an email notification 45 days prior to the expiration date with instructions to extend or close the study. It is your responsibility to request an extension for the study if it is not yet complete, to close the protocol file when the study is complete, and to make certain that the study is not conducted beyond the expiration date.

If applicable, agency approval letters must be submitted to the IRB upon receipt prior to any data collection at that agency. A copy of the approved consent form with the IRB approval stamp is enclosed. Please use the consent form with the most recent approval date stamp when obtaining consent from your participants. A copy of the signed consent forms must be submitted with the request to close the study file at the completion of the study.

Any modifications to this study must be submitted for review to the IRB using the Modification Request Form. Additionally, the IRB must be notified immediately of any adverse events or unanticipated problems. All forms are located on the IRB website. If you have any questions, please contact the TWU IRB.

cc. Dr. Mary Thompson, Physical Therapy - Dallas
Dr. Elaine Trudelle-Jackson, Physical Therapy - Dallas
Graduate School