Prevention of Deformational Plagiocephaly in Hospitalized Infants Using a New Orthotic Device

Michele DeGrazia, Deborah Giambanco, Gretchen Hamn, Amy Ditzel, Lindsay Tucker, and Kimberlee Gauvreau

Correspondence
Michele DeGrazia PhD, RN, NNP-BC, Boston Children's Hospital, 7 North NICU, 300 Longwood Avenue, Boston, MA, 02115. Michele.DeGrazia@childrens. harvard.edu

ABSTRACT

Objective: To measure the feasibility, safety, and efficacy of the cranial cup device in a sample of hospitalized infants at risk for deformational plagiocephaly (DP).

Design: A multisite, stratified, and randomized single-blinded study.

Setting: Neonatal intensive care units (NICU) from three urban and one suburban hospital participated.

Participants: Subjects included 62 infants with lengths of stay $\geq$ 14 days.

Methods: Nurses caring for infants in study group 1 used the moldable positioner. In study group 2, nurses rotated the moldable positioner and cranial cup devices using the cranial cup for a target goal of 12 hours/day. Both study groups received routine position changes. Outcome measures included hours of device use (feasibility), cardiorespiratory and emesis events (safety), and cranial measurements obtained at discharge (efficacy) by one of four, licensed orthotists who were blinded to the study.

Results: A total of 35 infants were randomized to study group 1 (moldable positioner) and 27 infants to study group 2 (moldable positioner and cranial cup). The median hours per day on the cranial cup was 10.7 (range 4.5 – 15.3). Emesis and cardiorespiratory events were equally distributed for the moldable positioner and cranial cup devices in study group 2. At discharge, more infants in study group 1 (46%, $n = 16$) exhibited abnormal cranial measurements than those in study group 2 (19%, $n = 5$) ($p = .03$).

Conclusion: Rotating the cranial cup with the moldable positioner provides a feasible, safe, and potentially efficacious therapy for prevention of DP.

Michele DeGrazia, PhD, RN, NNP-BC, is the Director of Nursing Research, Neonatal ICU and a neonatal nurse practitioner at Boston Children's Hospital, Boston MA; instructor in the Department of Pediatrics, Harvard Medical School, Boston, MA; and adjunct associate professor, Northeastern University, Boston, MA.

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Sick or premature infants spend more time in bed than healthy term newborns. The prolonged exposure of the hospitalized infant's head to the flat and often firm bed mattress can lead to deformational plagiocephaly (DP). This condition is characterized by a flattening of the head that results in an asymmetrical, elongated, narrowed, or widened head shape and may be accompanied by ear misalignment, frontal bossing, and facial asymmetry (Hummel & Fortado, 2005; Littlefield, 2003). Numerous researchers have described problems of parent/infant attachment and social isolation for infants with positional head shape deformities or DP (Alley, 1981; Badr Zahr & Abdallah, 2001; Hemingway & Oliver, 2000a; Ritter, Casey, & Langlois, 1991). Also concerning are reports linking DP to deficits in neurodevelopment (Constantin, Waters, Morielli, & Brouillette, 1999; Hunter & Malloy, 2002; Hutchison,

2009; Hutchison, Stewart, de Chalain, & Mitchell, 2010; Miller, Johnson, Duggan, & Behm, 2011; Robertson, 2011; Speltz et al., 2010).

Neonatal health care providers have implemented various interventions, including waterbed therapy, water-filled pillows, gel pillows, air-filled mattresses, foam mattresses, and repositioning procedures aimed at prevention of DP. However investigators have shown these interventions are not consistently successful in preventing DP (Cartlidge & Rutter, 1988; Chan, Kelley, & Khan, 1993; Hemingway & Oliver, 1991, 2000b; Hutchison et al., 2010; Marsden, 1980; Schultz et al., 2008; Schwirian, Eesley, & Cuellar, 1986).

The cranial cup is a sleep surface specifically designed to prevent or correct DP by supporting the infant's head and entire body in the supine and

semiside lying positions. In 2008, researchers in a nonrandomized prospective clinical trial found that an older and customized version of the cranial cup successfully corrected early positional head shape deformities in infants younger than age 4 months (Rogers, Miller, & Mulliken, 2008). Subsequently, the custom cranial cup was revised into a standardized device that can accommodate infants of various weights and lengths. In addition, a special scaled-down version has been developed specifically for use in the NICU. This newest model accommodates infants weighing between 1 and 2.5 kg.

Previous success in correcting mild positional head shape deformities in healthy infants with the cranial cup and the newly revised standardized cranial cup models supported testing in the NICU population (Seruya, Oh, Sauerhammer, Taylor, & Rogers, 2013). In this multisite, stratified, randomized, single-blinded study, we measured the feasibility, safety, and efficacy of the cranial cup device in a sample of hospitalized infants at risk for DP. The specific aims of this study were as follow:

1. Describe the feasibility of using the cranial cup for 12 hours per day on a sample of NICU infants.
2. Determine the safety of the cranial cup by comparing the incidence of cardiorespiratory (apnea, bradycardia, and oxygen desaturation) and emesis events for the subset of NICU infants rotating between the cranial cup and a traditional moldable positioner.
3. Compare the efficacy of rotating the cranial cup and moldable positioner device versus the moldable positioner alone on head shapes as measured by cranial index and cranial symmetry on a sample of NICU infants at hospital discharge.

Literature Review

Commonly observed forms of DP include brachycephaly, plagiocephaly, scaphocephaly, and dolichocephaly. Head shape deformities are further characterized as being unilateral or bilateral and are sometimes accompanied by ear misalignment, frontal bossing, and asymmetrical or distorted facies (Littlefield, 2003; Pollack, Losken, & Fasick, 1997). Risk factors for DP include restricted uterine environment, birth trauma, prematurity, lack of full bone mineralization, neurological deficits, torticollis or preferential head position when lying down, sedation, paralysis, multiple-birth infants, limited prone positioning, and the

> **Sick infants are more vulnerable to developing deformational plagiocephaly from spending more time in their beds and illness-associated positioning restrictions.**

application of continuous positive airway pressure devices (Hemingway & Oliver, 1991; Hutchison, Thompson, & Mitchell, 2003; Ifflaender, Rudiger, Konstantelos, Wahls, & Burkhardt, 2013; Littlefield, Kelly, Pomatto, & Beals, 1999; van Vlimmeren et al., 2007). Hospitalized infants spend more time in their beds and have positioning restrictions due to their medical needs; therefore they are more vulnerable to developing DP.

Deformational plagiocephaly has social and neurodevelopmental implications (Bialoskurski, Cox, & Hayes, 1999; Collett, Breiger, King, Cunningham, & Speltz, 2005; Miller & Clarren, 2000) and may interfere with parent/infant attachment due to the atypical appearance of the infant with DP. Most full-term healthy infants have round symmetrical head shapes. In contrast, many premature and sick term infants develop DP. Infants with DP exhibit asymmetrical, narrowed, or widened head shapes, sometimes accompanied by ear misalignment and distorted facial features. Due to these features, premature and sick infants do not look like full-term healthy infants and thus do not conform to parental expectations (Bialoskurski et al., 1999). Kelly, Vannostrand, Shiftlett, and Chan (1996) demonstrated this by asking mothers of preterm and term infants to rate pictures of premature infants with DP, premature infants without DP, and full-term infants. These investigators found that premature infants with DP were perceived to be less attractive than premature infants without DP and full-term infants (Kelley et al., 1996).

Even more concerning are the proposed neurodevelopmental implications of DP. In one retrospective study of 287 infants with DP, 36% of parents reported that their children had one or more developmental delays (Hutchison, 2009). However that study had significant limitations due to unknown birth data and medical histories of participants. In another study, researchers compared toddlers with and without positional head shape deformities, and toddlers with deformities scored lower than similar but unaffected peers using the Bayley Scales of Infant Development III (Collett et al., 2011). In 2002, Balan et al. demonstrated that depressed cortical sound processing was indicative of auditory processing dysfunction in

Deborah Giambanco MS, RN, NNP-BC, is a neonatal nurse practitioner and maternal-child health educator, Boston Children's Hospital, St. Elizabeth's Medical Center, Boston, MA.

Gretchen Hamn MS, RN, NNP-BC, is a neonatal nurse practitioner, Boston Children's Hospital, Shore Hospital, Weymouth, MA.

Amy Ditzel MS, RN, NNP-BC, is a neonatal nurse practitioner, Boston Children's Hospital, Shore Hospital, Weymouth, MA.

Lindsay Tucker MSN, APRN, is a neonatal nurse practitioner, Connecticut Children's Medical Center, Hartford, CT.

Kimberlee Gauvreau, ScD, is a senior staff biostatistician at Boston Children's Hospital and an associate professor in the Department of Biostatistics, Harvard School of Public Health, Boston, MA.

a sample of infants with posterior plagiocephaly. While studying the visual fields of 40 infants with DP, Siatkowski et al. (2005) found that 35% had constriction of one or both hemifields by at least 20 degrees, suggesting that positional head shape deformity may affect visual field development. Nevertheless, these researchers reported that the constriction and head flattening were not necessarily located on the same side of the infants' heads, raising uncertainty about a cause and effect relationship (Siatkowski et al., 2005).

Despite the numerous problems associated with DP, little attention is paid to its prevention. Typically DP does not become an issue until the deformity is visibly noticeable by a parent or an astute health care provider. Once it is recognized, parents and health care providers use conservative measures to address the deformity. Conservative measures consist of active repositioning techniques such as rotating the direction of the head during feedings and sleep and supervised time spent prone. Providing very young infants with time spent in the prone position and altering head positions was reported to reduce positional head shape deformities in the home setting (Hutchison et al., 2003). However, these conservative measures of prevention are not always practical in the hospital, and conservative measures are not always successful at preventing or correcting the deformity (Graham et al., 2005; Hemingway & Oliver, 2000b; Hutchison et al., 2010; Laughlin, Luerssen, Dias, & Committee on Practice and Ambulatory Medicine Section on Neurological Surgery, 2011; Lipira et al., 2010; Patry, 2008).

The inability to employ conservative measures in the hospital has led health care providers to examine alternative interventions for DP. These include a variety of positioning devices and bedding. Schwirian et al. (1986) demonstrated improved head shapes in a small sample of premature infants by using water pillows made from partially filled ice packs placed beneath the infant's head and by propping the infants in a supine position for several hours a day. In another study, soft air-filled apnea mattresses versus standard foam mattresses significantly reduced but did not eliminate head flattening in a sample of 34 premature infants (Cartlidge & Rutter, 1988). Waterbed mattresses ($N = 84$) and pressure relief mattresses ($N = 144$) had no effect on the development of DP in two separate randomized clinical trials of premature infants (Chan et al., 1993; Hemingway & Oliver, 1991). Additionally, in a single case report of twins, some improvement

in head shape was shown as measured by cranial index when a gel pillow was used beneath the head of one twin (Marsden, 1980), but this finding could not be replicated in a larger sample of premature infants (Schultz et al., 2008). Similarly, when 126 infants younger than age 3 months with DP were randomized to positioning techniques versus positioning techniques plus a Safe T Sleep wrap, neither group demonstrated improvement in head shapes as measured by cranial index or oblique cranial length ratio (Hutchison et al., 2010). Thus, results of multiple studies suggested that a more effective device was needed.

Methods

Study Design
This was a multisite, stratified, randomized, and single-blinded study.

Sample
Infants born at ≥ 22 weeks gestation who were < 14 days of age, had expected lengths of stay ≥ 14 days, and received medical clearance from their health care teams were targeted for enrollment. It was our experience that these infants were at greatest risk for developing DP. Infants were excluded from this study if they had any of the following: continuous ventricular drain, subgaleal shunt, craniofacial anomaly, cervical anomaly, critical airway or airway anomaly requiring prone positioning, cutis aplasia, craniosynostosis, significant skin breakdown to the scalp, or prolonged scalp IV access needs.

Infants from four participating NICUs were enrolled between April 2010 and July 2012. Of more than 800 infants screened, many were excluded for an anticipated length of stay of < 14 days or for medical reasons, and a few parents declined. Parents agreeing to have their infants participate signed informed consent and HIPPA forms. Because the cranial cup device was designed for infants weighing ≥ 1000 grams, randomization was stratified by study site and weight (< 1000 grams vs. ≥ 1000 grams).

In total, 88 infants were enrolled and 62 were included in the final analysis. Of the 26 not included, 14 were discharged or transferred before completing study requirements, five were withdrawn per parental request as they perceived their infants were uncomfortable on the cranial cup, three died as a result of illness, and four were assigned to the
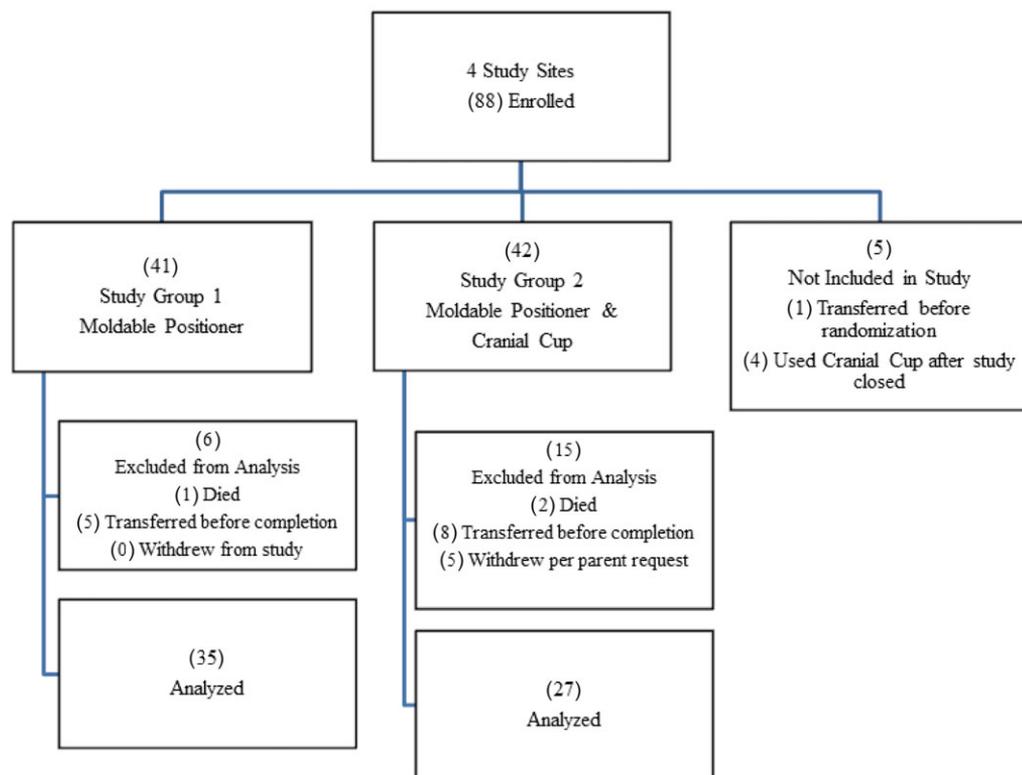
Figure 1. Study design.

cranial cup after the study was closed (Figure 1). Infants who did not complete the study did not differ from those who did with respect to primary clinical diagnosis ($p = .72$), gender ($p = 1.0$), age at NICU admission ($p = .74$), birth weight ($p = .76$), or gestational age ($p = .74$).

Sample Size Calculation

A sample size of 160 infants was calculated based on an estimated incidence of DP from prior studies at 20% with a reduction to 5% in the experimental group, assuming 80% power for a 2-sided chi-squared test with alpha .05; 76 infants were required per study group with an additional eight participants included for missing data and attrition. The assumed incidence of DP was based on infants presenting to outpatient clinics. To prevent enrolling more infants than would be necessary if our participants experienced a higher incidence of DP in the NICU, a series of sequential evaluations of the data with more stringent levels of significance were planned to occur after complete information was available for 40, 80, and 120 participants (Whitehead, 2000).

Setting

This study was conducted in three urban and one suburban NICUs. Collectively these NICUs cared for an average 5000 preterm and term infants per year. The main study site was a 24-bed Level IV NICU (Committee on Fetus and Newborn, 2012) located in an urban tertiary care center that received approximately 680 local, national, and international transfers for admission per year and provided care to infants with diverse medical and surgical needs. The secondary study sites consisted of three Level III NICUs caring for infants with critical care and intermediate care needs and included the following: an 18-bed unit that admitted approximately 300 infants per year, a 46-bed unit admitting approximately 3600 infants per year, and a suburban 30-bed unit admitting approximately 1100 infants per year. Infant admissions to these three units often occurred on the day of birth or shortly thereafter.

Devices

Cranial Cup. The cranial cup, a child safe device, is made from cross-linked polyethylene foam. It is

Figure 2. Infant positioned on cranial cup.

designed to support the infant's head and entire body in the supine and semiside lying positions (Figure 2). Preterm and full- term cranial cup devices are made to fit traditional NICU bedding; both models have smooth edges to prevent dislodgement of tubes and monitoring equipment. Both models are finished with soft, washable, jersey covers. The foam layers of the cranial cup are removable to accommodate the growing infant. To provide containment for the infant, straps and a tubular bean bag–like device provided with the cranial cup were used to form a nest around the infant, and when appropriate infants were swaddled using a blanket.

Moldable Positioner. The latex and di (2-ethylhexyl) phthalate (DEHP) free standard fluidized neonatal positioner was approved for use with infants placed in the supine, side-lying and prone positions. This 9'' x 15'', pillow-shaped, moldable device was used to support the infant's head, neck, and shoulders. As recommended by the manufacturer, to help facilitate normalized head shape development, nurses were instructed to make a round indentation in the device where the infant's head would lay (Figure 3). To provide containment on the moldable positioner, nurses placed the positioner in a premanufactured or nurse-made nest-like bed or swaddled the infants in a blanket while on the device (Figure 3). The moldable positioner fit comfortably inside the flattened area of the nest-like bed, so function of the moldable positioner was not altered.

Procedures

Ethical review committees at each of the participating institutions approved this study. Written informed consent and HIPPA authorization was obtained from the parents of study participants prior to the initiation of study procedures.

Education of Bedside Nurses. Prior to enrollment and throughout the study, the investigators and study nurses provided scheduled and impromptu (group and one-on-one) in-services for nurses caring for study participants. The nurses were educated about the study protocol, including blinding of orthotists, proper use the moldable positioner and cranial cup devices, and documentation on the study logs. Study personnel were available by page operator 24 hours per day, 7 days per week for questions or issues.

Education of Orthotists. Cranial measurements were obtained by one of four skilled, licensed orthotists. All orthotists completed an orientation that included training on study procedures, obtaining the cranial measurements (including infection control), and completion of the cranial measurement form. Calipers accurate to 1 mm and a plastic-coated measuring tape were used to assess head circumference. Manual and laser scan cranial measurements and head circumference were obtained and recorded on the cranial measurement form. A standardized method of measurement using anatomic landmarks was employed to ensure accuracy of measures. Intraclass

correlation coefficients for pairs of the four orthotists across all head shape measures for 17 randomly selected infants were between .97 and .99.

Blinding of Orthotists. Blinding of orthotists was achieved by having the bedside nurses remove the study devices from the infants' bedsides prior to the orthotists' arrival on the unit.

Stratification and Randomization. Computer-generated and sequentially numbered randomization cards were prepared by the clinical research program affiliated with the main study site by staff not connected with the study. To stratify by study site, two unique sets of the randomizations cards (one set for infants born weighing < 1000 grams and one set for infants born weighing ≥ 1000 grams) were prepared and delivered to each of the four study sites. To stratify by weight, research personnel at each site selected a card that corresponded with the infant's birth weight.

Infants weighing < 1000 grams at enrollment were positioned using the moldable positioner and received routine position changes every 3 to 4 hours minimum until they reached 1000 grams. Once these infants reached 1000 grams they were reassessed to confirm study eligibility and verify they

Numerous scientific reports link deformational plagiocephaly to social and neurodevelopmental problems.

were not being transferred to a lower level of care. These infants then underwent head shape measurements and were randomized, utilizing cards from the < 1000 grams strata (Figure 4). In contrast, after obtaining consent for infant participants weighing ≥ 1000 grams, head shape measurements were obtained and the infants were randomized without delay utilizing cards from the ≥ 1000 grams strata.

Infants randomized to study group 1 received the moldable positioner, and regular position changes every 3 to 4 hours. Infants in study group 2 received the experimental treatment. They were routinely positioned every 3 to 4 hours minimum and rotated between the moldable positioners and cranial cup devices. When on the cranial cup, infants were positioned supine or semiside lying; when not using the cranial cup nurses were instructed to use the moldable positioner. Nurses caring for infants in study group 2 were instructed to use the cranial cup for ≥ 12 hours out of 24 hours.

Figure 3. On the left the positioner is prepared, and on the right the infant is placed on the moldable positioner.
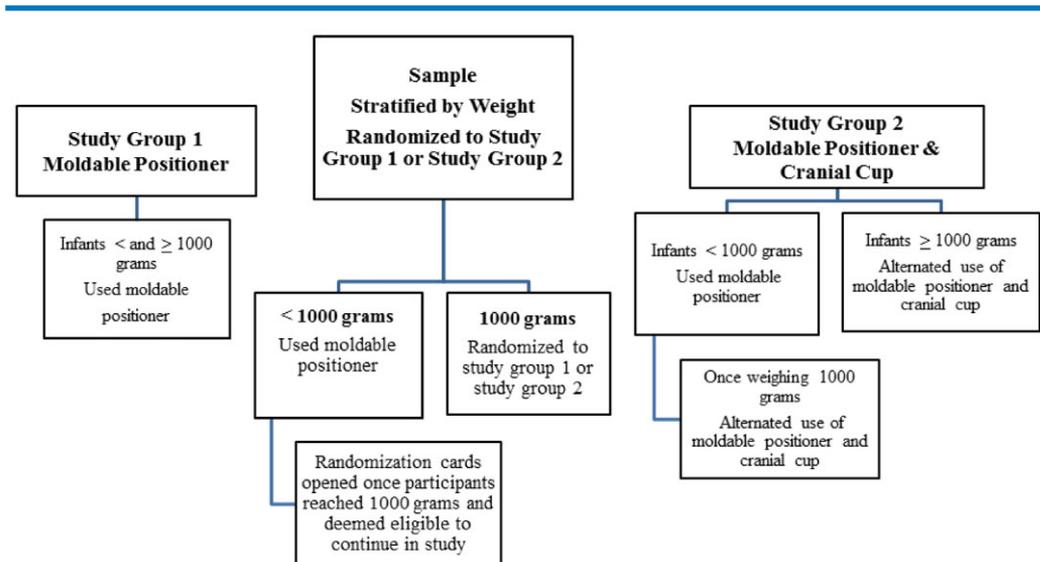
Figure 4. Intervention by study group.

Intermittent Digressions. The protocol allowed for intermittent digressions (5 days or fewer) from using the cranial cup. Intermittent digressions were defined as feeding problems, perioperative care, special procedures, or medical management; these digressions were not protocol deviations. We believed that 5 days or fewer would have little effect on infant's head shapes and would provide a more realistic evaluation of the cranial cup. During these digressions, infants were positioned on the moldable positioner. No infants were removed from this study due to prolonged digressions extending beyond 5 days. Because we wanted a realistic evaluation of the cranial cup despite the need for intermittent digressions, we did not make any adjustments to account for these digressions during data analysis.

Outcome Measures
Feasibility. Our first aim was to determine if nurses could use the cranial cup for 12 hours per day with sick, fragile NICU infants. To measure feasibility, bedside nurses maintained hourly recordings of study device use with the infants. Device use was measured as the total hours spent on each device per day. A study nurse and the investigators monitored device use and completion of study logs, ensuring that the devices were being used and logs were filled out properly.

Safety. Cranial cup use requires supine or semi-side lying positioning of infants. Published studies on premature infants have demonstrated small increases in oxygen saturation, increased incidence of apnea, and more frequent regurgitation of feedings in the supine position (Balaguer, Escribano, & Roque, 2003; Bredemeyer & Foster, 2012; Jenni et al., 1997; Orenstein & Whitington, 1983). Therefore, our second aim was to ensure that the cranial cup was safe to use with NICU patients, particularly with those born prematurely.

To measure safety, bedside nurses recorded cardiorespiratory and emesis events on the study logs. Because infants often experience clustering of apnea, bradycardia, and oxygen desaturation, each of these events was measured as a singular event and referred to as a cardiorespiratory event. A *cardiorespiratory event* was defined as the clinically relevant constellation of symptoms including one or more of the following: apnea: cessation of breathing for > 20 seconds; bradycardia: heart rate < 80 for a full- term infant and < 100 for a premature infant; or desaturation: oxygen saturation of < 90% for full- term infant or < 87% for a premature infant for > 10 seconds (DeGrazia, 2007). Emesis events were defined as each singular episode of regurgitation of breast milk or formula recorded by the nurses. To ensure the accuracy of the cardiorespiratory events and emesis data, our research staff carefully verified the documentation of these events on the study logs against each of the infants' medical records.

Efficacy. Our third aim was to measure effectiveness of the study devices; therefore cranial

measurements (cranial index and cranial symmetry) of study participants were obtained at hospital discharge by the orthotists. Because standardized cranial index and cranial symmetry measurements were not available, we adopted those used in the plagiocephaly clinic at the main study site. We found these measures were comparable with those reported in the literature (Hummel & Fortado, 2005; Looman & Kack Flannery, 2012). Cranial index (normal measurement between 73%–85%) was obtained by dividing the medial-lateral (M-L) by anterior-posterior (A-P) dimensions of the cranium, then multiplying it by 100%. This measure was used to assess for brachycephaly, dolichocephaly, or scaphocephaly. Cranial symmetry (normal measurement of < 8 mm) was obtained to assess for an asymmetrical head shape, simply termed "plagiocephaly," by calculating the difference in the right and left anterior-posterior measures. Abnormalities in either cranial index or cranial symmetry measures were indicative of an abnormal head shape.

Data Analysis

Tests for normality were conducted to determine if continuous data followed a normal distribution; if not, nonparametric methods were used. Baseline patient characteristics were compared between the two study groups using either the two-sample t test or Wilcoxon rank-sum test for continuous variables and Fisher's Exact Test for categorical variables. To determine if the cranial cup could be used for 12 hours per day, the median hours of cranial cup use was calculated for those infants assigned to study group 2.

To assess safety, the number of cardiorespiratory and emesis events was analyzed for the subset of infants randomized to the moldable positioner and cranial cup (study group 2; $n = 27$). As prespecified, analyses included only the timeframe when infants were using both devices. This circumscribed timeframe allowed participants to serve as their own controls and prevented confounding factors such as intermittent digressions, age, illness, and changes in care management from influencing the findings. Comparisons were performed using Wilcoxon signed-rank test for medians and the Poisson distribution for incidence rates.

Our third aim was to gain preliminary information on preventing all types of DP using the cranial cup; therefore cranial index and cranial symmetry measures were combined for the analyses.

Analyses for cranial measurements were performed on participants with endpoint measurements that had been obtained ≥ 14 days following randomization. Categorical variables were compared for the two study groups using Fisher's Exact Test; continuous variables were compared using either the two-sample t test or Wilcoxon rank-sum test. Complicating factors such as head molding, nonfixed mobile sutures, trauma from vacuum-assisted deliveries, caput succedaneum, cephalohematoma, and edema present in the newborn period, and delayed measurement of unstable infants, precluded making comparisons between initial and discharge head shape measures.

Results

Interim Analysis

Following the first interim analysis of 40 infants, we observed a greater difference between study groups than anticipated, and the sample size calculation was subsequently revised. The new sample size of 62 infants (31 per treatment arm) was based on an observed reduction from 41% in the moldable positioner group to 11% in the experimental group, with a significance level of 0.01.

At the time of the second interim analysis, a data safety monitoring board meeting (DSMB) was convened. A report on the study progress to the DSMB revealed that nurses and doctors were observing and openly discussing their observations of improved head shapes for infants in study group 2 (cranial cup and moldable positioner) compared to study group 1 (moldable positioner). The DSMB was informed that staff members began requesting the cranial cup device for patients not enrolled in the study and for their own children at home with DP. The DSMB recommended early closure of the study at that time due to loss of study equipoise. They also recommended that the experimental treatment be offered to parents of all remaining infants enrolled in the study.

Patient Characteristics

At the time of study closure, 35 infants had randomized to study group 1 (moldable positioner alone) and 27 infants had randomized to study group 2 (moldable positioner and cranial cup). There were no statistically significant differences in patient characteristics (Table 1). Initial head shape measures revealed equally distributed abnormalities as measured by cranial index ($p = .22$) and no abnormalities of cranial symmetry when comparing the two study groups.

Table 1: Participant Characteristics by Study Group

Characteristic	Study Group 1: Moldable Positioner (n = 35)		Study Group 2: Moldable Positioner & Cranial Cup (n = 27)		p Value
	Median	*Range*	*Median*	*Range*	
Chronological Age at Admission (days)	0	0–7	0	0–12	0.51
Gestational Age at Birth[a] (weeks)	31	24–39	30	23–39	0.79
Birth Weight (grams)	1575	455–3710	1495	510–3370	0.84
	n	*%*	*n*	*%*	
Gender					0.79
Male	22	63%	18	67%	
Female	13	37%	9	33%	
Birth Mode					1.0
Cesarean	29	83%	22	81%	
Vaginal	6	17%	5	19%	
Primary Diagnosis					0.48
Prematurity	24	69%	16	59%	
Esophageal Atresia	3	9%	2	7%	
Gastroschisis	2	6%	3	11%	
Dysplastic Kidney	2	6%	0	0%	
Congenital Heart Disease	1	3%	0	0%	
Other	3	9%	6	22%	

Note. [a]For the characteristic Gestational Age at Birth, data from 26 participants in Study Group 2 were available for analysis.

Feasibility

The median hours per day on the moldable positioner for study group 1 was 18.1 (range 9.7 – 21.8). The median hours per day on the cranial cup was 10.7 (range 4.5 – 15.3) versus 9.3 (range 0.8 – 11.7) on the moldable positioner for study group 2.

Safety

There were 5.4 cardiorespiratory events on the cranial cup versus 5.9 on the moldable positioner per 100 hours on the devices. Likewise there were 1.1 emesis events on the cranial cup versus 1.0 on the moldable positioner per 100 hours on the devices. There were no statistically significant differences in the number of cardiorespiratory events or emesis when comparing the moldable positioner versus cranial cup. Also there were no significant adverse events associated with use of the cranial cup device.

Effectiveness

At discharge, more infants in study group 1 (n = 16, 46%) the moldable positioner group had abnormal cranial measures when compared to study group 2, the moldable positioner and cranial cup group (n = 5, 19%) (p = .03) (Table 2). Of the 16 infants in study group 1 with abnormal cranial measures, 15 infants had abnormal cranial indexes. Twelve of these infants had narrow dolichocephalic or scaphocephalic head shapes (median 71.4, range 66.4 – 72.8); and three had wide, brachycephalic head shapes (median 88.2, range 85.5 – 88.9). Only one infant had an abnormal cranial symmetry measure (consistent with plagiocephaly) and another exhibited abnormalities in both cranial measures. In comparison, of the five infants in study group 2 that exhibited

Table 2: Cranial Measurement Outcomes by Treatment Group at Discharge

Cranial Measurement	Study Group 1: Moldable Positioner ($n = 35$)		Study Group 2: Moldable Positioner & Cranial Cup ($n = 27$)		*p* Value
	n	%	*n*	%	
Cranial Index Abnormality					0.12
Low	12	34%	4	15%	
High	3	9%	1	4%	
Cranial Symmetry Abnormality	2	6%	1	4%	1.0
Cranial Index or Cranial Symmetry Abnormality	16	46%[a]	5	19%[a]	0.03

Note. [a]Each group had one infant with cranial index and cranial symmetry abnormalities; these infants were only counted once.

abnormal cranial measures, four had abnormal cranial indexes (median 71.95, range 71.4 – 72.5) demonstrating narrowed head shapes and one infant exhibited abnormalities in both cranial measures.

We compared the two study groups to see if potential confounding variables influenced our findings. There was no statistically significant difference detected between the two study groups when controlling for the number of days on CPAP. When examining weight as a confounder, we found twice as many infants weighing < 1 kg in study group 1 that had abnormal head shapes at discharge as compared to study group 2; however this difference was not statistically significant. Also we looked at lengths of study participation. There were more infants with abnormal cranial measurements in study group 1 as compared to study group 2, when the lengths of study participation was < 40 days ($p = .04$). However, there was no statistically significant differences between the groups ($p = .49$) when the length of study participation was ≥ 40 days (Table 3).

Discussion

We examined the use of the standardized cranial cup with a sample of sick, hospitalized infants. The 62 premature and term gestation infant participants had medical and surgical needs and were acutely ill upon enrollment. These infants were at very high risk for developing DP because of prematurity, prolonged illness and hospitalization, inactivity and positioning restrictions related to medical treatments, sedation, intubation, and the need for continuous positive airway pressure (CPAP). In this study, our goal was to evaluate the cranial cup for feasibility, safety, and efficacy.

Feasibility

At the start of the study, we anticipated a number of potential obstacles that could have influenced the feasibility of the cranial cup in the NICU setting. First the cranial cup is a firmer sleep surface than traditional NICU bedding, thus we thought the device might be perceived as uncomfortable. Next, our literature review suggested that supine positioning of NICU patients, a requirement for using the cranial cup, could lead to increased cardiorespiratory instability and increased regurgitation of feedings (Balaguer et al., 2003; Bredemeyer & Foster, 2012; Jenni et al., 1997; Orenstein & Whitington, 1983). Furthermore, the cranial cup had not been deemed compatible for use during commonly performed procedures in the NICU, such as X-rays; therefore nurses caring for participants on the cranial cup would have to creatively integrate the device into the patients' care regimen.

During the course of this study, concerns about comfort, cardiorespiratory instability, gastrointestinal events, and integration of the device waned. In total five parents decided to withdraw their infants from the study due to concerns over comfort, and a few health care providers were reluctant to use the cranial cup. However, most parents and providers embraced the cranial cup and many nurses were able to incorporate it seamlessly into the infants' care regimens using the cranial cup a median of 10.7 hours per day, slightly under the targeted goal of 12 hours per day.

Safety

With concerns about cardiorespiratory instability and gastrointestinal disturbances, we wanted to be sure that our study participants were safe

Table 3: Potential Confounders by Study Group

Cranial Index or Cranial Symmetry Abnormality	Study Group 1: Moldable Positioner $n = 35$		Study Group 2: Moldable Positioner & Cranial Cup $n = 27$		p Value
	Median	*Range*	*Median*	*Range*	
Number of days on CPAP	1	0–76	0	0-45	0.21
	n	*%*	*n*	*%*	
Birth weight <1 kg $n = 14$[a] $n = 7$[b]	6	43%	3	43%	1.0
Length of study participation <40 days $n = 15$[a] $n = 14$[b]	7	47%	1	7%	0.04
Length of study participation ≥ 40 days $n = 20$[a] $n = 13$[b]	9	45%	4	31%	0.49

Note. [a]indicates the sample size available for study group 1.
[b]indicates the sample size available for study group 2.

when using the cranial cup. As a measure of cardiorespiratory stability we considered recording continuous oxygen saturation. However, because variables such as wandering atelectasis of the pulmonary bed, variations in airway secretions, locations of endotracheal tubes in intubated infants, and pulmonary edema were inherent to our study population, we felt we would not be able to directly attribute fluctuations in oxygen saturations to use of the study devices.

For our analyses we conducted subgroup comparisons of cardiorespiratory and emesis events, with the infants in study group 2 (cranial cup and moldable positioner) serving as their own controls. By infants serving as their own controls, we were able to compare equivalent groups, preventing extraneous stressors such as sepsis and ophthalmologic exams that are known to increase cardiorespiratory and emesis events in this patient population, from affecting our results. Because there was no difference in the number of cardiorespiratory or emesis events while using the cranial cup versus moldable positioner, and no serious adverse events observed during our study, we believe our findings indicate that the cranial cup can be safely used in the NICU with trained neonatal nurses.

Efficacy

Our results showed that more infants in study group 1 ($n = 16$, 46%) the moldable positioner group, had abnormal cranial measures when compared to study group 2, the moldable positioner and cranial cup group ($n = 5$, 19%) ($p = .03$) at hospital discharge. To better understand this difference in outcomes for the two study groups, we examined birth weight, length of study participation, and the use of CPAP as possible confounders. Findings from these analyses showed no statistically significant differences in outcomes between the two study groups based on birth weight or the use of CPAP. However, our findings did show that more infants in study group 1 had head flattening at discharge, when length of study participation was < 40 days compared to ≥ 40 days. These findings may support the gradual improvement in head shapes for some infants over time, as the infant become more active and have less positioning restrictions (Cavalier et al., 2011; Wilbrand et al., 2012). However, overall there were fewer infants in study group 2 (cranial cup and moldable positioner) with abnormal head shapes in the < 40 days and the ≥ 40 days subsets, suggesting that the noninvasive cranial cup helps to prevent the development of DP.

Implications for Future Study

To achieve the best results with the cranial cup, its inventors had recommended that the device be used anytime the infant is lying in bed. However, our study team determined that round-the-clock cranial cup use was not a realistic goal. NICU patients require frequent repositioning, including prone, to facilitate good skin integrity, skeletal development, biomechanical alignment, and proprioceptive/tactile stimuli (Sweeney & Gutierrez, 2002); they need to undergo frequent tests and procedures; and they sometimes require surgical interventions. Accordingly, we determined a more practical goal for cranial cup use in the NICU setting would be 12 hours/day.

In this study, nurses used the cranial cup for a median of 10.7 hours/day, slightly less than our set goal. Future studies are needed to determine if the cranial cup can be used more than 12 hours/day or longer in different subsets of the NICU patient population, and to examine relationships between device exposure and head shape outcomes, as well as patient characteristics and device use. Furthermore, studies are also needed to determine if the cranial cup device is compatible for use with X-rays and other frequently performed procedures such as ophthalmological exams. Confirming that the cranial cup is compatible for use during these frequently performed tests and procedures will enhance the utility of this device in the NICU.

Limitations

There were several limitations to this study. First, we overestimated our ability to predict the length of stay and the viability of potential study participants which resulted in an attrition rate for randomized participants equaling 30%; 16% of our study participants were discharged to convalescent care units, 6% were withdrawn by parents, 3% died before they completed all of the study requirements, and another 5% were not included in analysis due to study closure. It is not known if inclusion of these infants would have influenced our study results.

Second, participating NICUs employed between 30 and 100+ nurses; therefore, differences in nursing management and the repositioning of the infants may have occurred. These differences may have influenced the study results. To help ensure that all nurses received the necessary education and adhered to the study protocol, we enlisted research nurses and study nurse liaisons to perform surveillance and round-the-clock education.

> **Rotating the cranial cup and moldable positioner provides a feasible, safe, and promising therapy for prevention of deformational plagiocephaly in the NICU.**

Finally, because the weight requirement for using the cranial cup was ≥ 1000 grams, extremely low-birth-weight study participants were managed differently in this study. We enrolled these infants at ≤ 14 days of age, however initial cranial measurements and the use of the cranial cup was delayed until these infants reached 1000 grams. To address this issue we stratified this study sample by weight to achieve equal numbers of extremely low-birth-weight infants in both study groups. We analyzed these two strata together and separately. Our findings suggest that weight was not an important determinant of normal versus abnormal head shape at hospital discharge in this study.

Conclusion

DP is a preventable condition that has important social and neurodevelopmental implications for NICU patients and their families. The cranial cup is unique in that it provides the necessary support to the cranium that cannot be achieved through routine repositioning or the use of other positioning aids. It is our experience that the noninvasive cranial cup is feasible, safe, and a potentially efficacious therapy for DP prevention in the NICU.

Still, more studies with the cranial cup are needed to determine if it is feasible, safe, and efficacious for use in specific patient populations and during commonly performed tests and procedures. A more recent study using the cranial cup to correct DP in a small sample of convalescent premature infants is nearly complete; findings from this study suggest that stable infants are good candidates for cranial cup use. Additionally, our research team is currently considering yet another study of the cranial cup, this time using a repeated-measures design. From this study we hope to gain a better understanding about device exposure and lengths of treatment as they relate to achieving and sustaining normal head shapes with the cranial cup.

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