Nonsynostotic Plagiocephaly

Prevention strategies in child health care

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Cover illustration by Brenda Bergreen

Success is the sum of small efforts repeated day in and day out.

Robert Collier

To children

Abstract

The aim of this project was to assess whether it is possible to prevent non-synostotic plagiocephaly while still promoting safe infant sleeping practices. A continuing education including guidelines was developed for child health nurses, and a clinical intervention and three supporting studies were planned. Nurses were recruited and allocated to two groups. Intervention group nurses were educated about nonsynostotic plagiocephaly while control group nurses were not. Then nurses recruited infants to the clinical intervention.

Five individuals recruited externally were trained to assess infant cranial asymmetry and then reliability-tested. Results indicate substantial strength of intra- and inter-rater assessor agreement. A survey was conducted to compare what information intervention and control group parents received from their nurses during early infancy. A significantly higher proportion of intervention group parents compared to control group parents were aware of the three regular and five newly introduced recommendations. The continuing education for nurses was evaluated by asking intervention and control group nurses and parents two open-ended questions regarding what they did to prevent and to reverse cranial asymmetry. Intervention group nurses reported providing regular and newly introduced re-positioning strategies to parents, and intervention group parents who perceived severe cranial asymmetry at 3-4 months reported implementing regular and new re-positioning strategies in their reversal efforts. The effect of the intervention on infant cranial shape was evaluated by assessing cranial asymmetry in 176 intervention group and 92 control group infants at 2-, 4-, and 12-months of age. It was four times more common that cranial asymmetry at two months reversed by four months in the intervention group compared to the control group infants (OR = 4.07 [1.23; 13.44], p = 0.02) when adjusted for parental awareness of written recommendations from their nurse. An infant's risk of having asymmetry persist from two to 12 months was reduced nearly threefold in the intervention group (RR = 0.35, p = 0.03). However, preventing brachycephaly was difficult.

In conclusion, assessors were considered reliable and interchangeable; educating nurses increased parental awareness of recommendations and promoted integration of new re-positioning recommendations in practice; and the intervention was associated with early reversal, but prevention was difficult. The nurse education and prevention efforts need more research.

Keywords: Assessments, child health, education, infants, intervention, non-synostotic plagiocephaly, nurses' instruction, parents, prevention, reversal

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Sammanfattning

Bakgrund: Frekvensen av lägesbetingad skallasymmetri har ökat efter införandet av rekommendationen att lägga spädbarn på rygg när de sover med syftet att förebygga plötsligt spädbarnsdöd. Den bästa möjligheten att påverka barnets skallform är under de första levnadsmånaderna. Därefter ökar risken att barnet får en kvarstående asymmetri.

Syfte: Att bedöma om det är möjligt att förebygga lägesbetingad skallasymmetri samtidigt som råden att förebygga plötslig spädbarnsdöd efterlevs.

Frågeställning: Kan lägesbetingad skallasymmetri förebyggas genom att fortbilda BVC-sjuksköterskor som i sin tur informerar och vägleder spädbarnsföräldrar?

Metoder: Förebyggande riktlinjer och en fortbildning för BVC-sjuksköterskor utvecklades samtidigt som en interventionsstudie med tillhörande tre stödstudier planerades. Fem individer utanför barnhälsovården anlitades som bedömare för den kliniska interventionen. De utbildades i att bedöma skallasymmetri. Därefter fick de delta i ett reliabilitetstest där de bedömde eventuell skallasymmetri genom att studera 50 fotografier på småbarn samt huvudena på sex spädbarn. Intra- och inter-bedömaröverensstämmelse bedömdes med hjälp av en reliabilitetsanalys. Femtiosex BVC-sjuksköterskor, 274 nyfödda och deras föräldrar rekryterades för att delta i interventionsstudien. De delades in i en interventions- och kontrollgrupp. Allokering till respektive grupp baserades på huruvida BVC-sjuksköterskan hade exponerats för projektets pilotarbete tidigare eller ej. Om ingen BVC-sjuksköterska på arbetsplatsen hade exponerats för projektet tidigare placerades alla BVC-sjuksköterskor, nyfödda och deras föräldrar på den barnavårdscentralen i kontrollgruppen, annars interventionsgruppen.

I samband med barnens 4-månaders kontroll på BVC fyllde föräldrarna i interventions- och kontrollgruppen i en checklista med 22 punkter om nonsynostotisk plagiocefali. De redovisade kunskaperer i de två grupperna jämfördes statistiskt. Fortbildningen för BVC-sjuksköterskorna utvärderades när barnen var 12 månader. Två frågor med öppna svar ställdes till BVC-sjuksköterskorna och föräldrarna i interventions- och kontrollgruppen om vad de gjort för att förebygga skallasymmetri eller för att få en begynnande asymmetri att gå tillbaka. En kvalitativ innehållsanalys och en fall-till-fall analys

med process-orienterad ansats användes för att jämföra vilka kunskaper som integrerades i praktiken i de två sjuksköterske- och föräldragrupperna. I den kliniska interventionsstudien utbildades BVC-sjuksköterskor i interventionsgruppen om lägesbetingad skallasymmetri. De ombads att arbeta enligt specifika riktlinjer medan kontrollgruppens sjuksköterskor arbetade utan förändrade direktiv. Interventions- och kontrollgruppbarnens skallform analyserades statistiskt vid 2-, 4-och 12-månadersålder med hjälp av regressionsanalys och reliabilitetsanalys för att utvärdera hur interventionen påverkat skallformen.

Resultat: Bedömarnas skallasymmetri bedömningar påvisade en hög reliabilitet. En signifikant högre andel av interventionsgruppens föräldrar var medvetna om de sedvanliga rekommendationerna samt fem av sju nya rekommendationer jämfört med kontrollgruppens föräldrar. I interventionsgruppen konstaterades att BVC-sjuksköterskor gav både de sedvanliga och men också de nya rekommendationerna till föräldrarna. Det var också tydligt att de interventionsgruppsföräldrar som ansåg att barnen uppvisade svår asymmetri vid 3-4-månaders ålder använde de nya rekommendationer i högre grad för att få asymmetrin att gå tillbaka. Det var fyra gånger vanligare att barn med lägesbetingad skallasymmetri vid två månader hade reverserat vid fyra månader i interventionsgruppen jämfört med barnen i kontrollgruppen (OR = 4.07 [1.23; 13.44], p = 0.02) när man justerat för föräldramedvetenhet om skriftliga rekommendationer från sin BVC-sjuksköterska. Barnen i interventionsgruppen hade nästan tre gånger lägre risk att skallasymmetri vid två månader kvarstod vid 12-månadersålder än barnen i kontrollgruppen (RR = 0.35, p = 0.03). Lägesbetingad skallasymmetri visade sig dock på det hela taget vara mycket svårt att förebygga i båda grupperna, framför allt brakycefali.

Slutsatser: Bedömarna ansågs både vara tillräckligt tillförlitliga och tillräckligt konsistenta för att utgöra utbytbara bedömare i den kliniska interventionen. I en vidare klinisk kontext tyder resultaten på att BVC-sjuksköterskor kan lära sig att bedöma lägesbetingad skallasymmetri, vilket kan användas för tidigt upptäckt. Att utbilda BVC-sjuksköterskor om lägesbetingad skallasymmetri bidrog till att öka föräldrarnas medvetenhet om rekommendationer och bidrog till att nya rekommendationer integrerades i praktiken. Interventionen bidrog till tidig reversering och minskade spädbarnens risk att lägesbetingad skallasymmetri vid 2-månadersålder kvarstod vid 12-månadersålder. Dock har lägesbetingad skallasymmetri visat sig vara svårt att förebygga, framför allt brakycefali. Mer forskning behövs bland annat för att förbättra fortbildningen för BVC-sjuksköterskor och förebyggande insatser.

List of papers

This thesis is based on the following studies, referred to in the text by their Roman numerals.

I.   Lennartsson F, Wennergren G & Nordin P.
Reliable Assessors of Infant Cranial Asymmetry in Child Health Care
Open Nurs J 2015; 9: 33-41.

II.   Lennartsson F, Nordin P & Wennergren G.
Teaching Parents How to Prevent Acquired Cranial Asymmetry in Infants
J Pediatr Nurs 2016; 31(4): e252-61.

III.   Lennartsson F, Ahlberg B & Nordin P.
Integrating knowledge into practice: An evaluation study on a continuing education for Swedish child health nurses on non-synostotic plagiocephaly
Nurs Open 2018; 5(3): 329-40.

IV.   Lennartsson F & Nordin P.
Nonsynostotic Plagiocephaly: a child health care intervention in Skaraborg, Sweden
BMC Pediatr 2019; 19(48): 1-12.

Content

Abbreviations

AC2   Agreement coefficient 2

BVC   Child health clinic [Barnavårdscentral]

CI   Cranial index

CVAI   Cranial vault asymmetry index

NSP   Nonsynostotic plagiocephaly

OR   Odds ratio

RR   Relative risk

SIDS   Sudden Infant Death Syndrome

Introduction

Nonsynostotic Plagiocephaly

Definition

Nonsynostotic plagiocephaly (NSP) is acquired cranial asymmetry that develops pre- or postnatally from external pressure on the infant's cranium (Rekate, 1998). NSP needs to be distinguished from lamdoid craniosynostosis, a rare congenital condition with premature fusion of a cranial suture where surgery is nearly always indicated (Kabbani & Raghuveer, 2004). NSP falls into three main groups: plagiocephaly - skewed occipital flattening, brachycephaly - symmetric occipital flattening, and combined plagiocephaly/ brachycephaly (Wilbrand et al., 2012). Other terms used for acquired cranial asymmetry include: deformational plagiocephaly, occipital plagiocephaly, posterior plagiocephaly, and positional plagiocephaly. When terms including the word plagiocephaly are used, clarifying whether brachycephaly is included or not will help avoid confusion.

Prevalence

In the early 1990s there were reports on an increase of NSP referrals to craniofacial centers, coinciding with the "back to sleep" campaign where parents were recommended to place infants supine for sleep in order to prevent Sudden Infant Death Syndrome (SIDS) (Argenta, David, Wilson, & Bell, 1996; Biggs, 2004; Kane, Mitchell, Craven, & Marsh, 1996). Although there are few reports on prevalence prior to this campaign, in 1971, Watson reported a prevalence of 48% in 65 healthy infants less than one-year old (Watson, 1971). In a Canadian cohort study including 440 infants, an estimated 47% of infants had some degree of NSP at 7 to 12 weeks of age (Mawji, Vollman, Hatfield, McNeil, & Sauve, 2013). In an Italian cohort study including 283 infants, an estimated 38% of infants had plagiocephaly and 12% also had brachycephaly at 8 to 12 weeks of age (Ballardini et al., 2018).

Prevalence of NSP is difficult to calculate for a number of reasons. In the first weeks postpartum, pre-natal NSP is difficult to differentiate from cranial molding from the birth process. No study has established when cranial molding leaves off and NSP begins (Lima, 2004).

Prevalence is a measure which is calculated at one point in time but NSP is not static. There is an on-going process of asymmetry development and asymmetry reversal. In addition, the definition of NSP is not standardized, and there are different methods of taking measurements. Prevalence estimates vary, depending on the population studied and criteria for diagnosis (Glasgow, Siddiqi, Hoff, & Young, 2007). The prevalence of NSP in Texas increased from 3.0 cases per 10 000 live births in 1999 to 28.8 in 2007, an average increase of 21.2% per year. However, the researchers suspect that this so-called increase was due mainly to changes in available therapies and insurance reimbursement practices, because the trend was observed mainly in patients visiting certain health care facilities, among mild cases and among cases requiring minimally invasive procedures (Sheu, Ethen, Scheuerle, & Langlois, 2011).

Age differences are important when calculating prevalence. The prevalence seems to rise during the first four months and then gradually decreases (Hutchison, Hutchison, Thompson, & Mitchell, 2004). The point prevalence may be as high as 22.1% at seven weeks of age and as low as 3.3% at two years according to a systematic review (Bialocerkowski, Vladusic, & Wei Ng, 2008), but this can vary.

Pathophysiological mechanism

Nonsynostotic plagiocephaly occurs when an external force is applied consistently to a specific area of an infant's head over a period of time (Rekate, 1998). When a young infant is placed on a resting surface, a contact force is generated between the head and the surface, resisting cranial growth in the area of contact, and displacing growth to areas where there is no resistance. This compensatory growth of a stationary infant cranium leads to NSP, analogous to how a pumpkin flattens as it grows in a field – it cannot expand into the ground and must grow along it (Rogers, 2011a) (Photograph 1).

Photograph 1. Contact force from firm surface causes occipital flattening in supine lying infant. Signed parental consent to publish photograph has been obtained.

Potential risk factors

A systematic review with a total study population of 27 782 children found poor agreement among the 22 studies regarding 60 identified risk factors, including biological infant factors, obstetric factors, infant care practices/lifestyle of mother/parent factors, and sociodemographic factors. The most commonly reported risk factors in these studies were: male, supine sleep position, limited neck rotation or preference in head position, firstborn, lower infant activity level, and lack of tummy time (De Bock, Braun, & Renz-Polster, 2017) (Photograph 2). Three factors deterring recovery include: supine sleep position, limited head rotation, and lower infant activity level (Hutchison et al., 2004).

Reversal with time?

Capitalizing on the plasticity of the neonatal cranium is the essence of intervention (Morrison, 2006). Although NSP might disappear as a child grows older and increased mobility relieves pressure on the cranium, it

persists in some children. In a study of 235 infants diagnosed with NSP and seen at ages 18 and 36 months, head shape became more rounded especially between infancy and 18 months. However, 86% continued to show persistent NSP at 36 months (B. R. Collett et al., 2018). In a study of 129 children diagnosed with NSP in infancy and whose parents had been given information on counter-positioning strategies, 39% had not reverted to the normal range of symmetry at mean age of four years (Hutchison, Stewart, & Mitchell, 2011). Results of that study indicate that giving parents information on counter-positioning is not always sufficient. In a study evaluating the clinical course of NSP in 40 children treated with helmet therapy – median age seven months - matched at baseline to 41 untreated children selected with the greatest degree of NSP – median age 7 months, researchers found clear evidence of improvement in the group treated with helmet therapy, while in the untreated group, no clear evidence of improvement was found at the five year follow-up (Wilbrand et al., 2016). Results of that study indicate that without orthotic treatment of severe NSP, reversal is uncertain. The prevalence of NSP was 2% in a cross-sectional study of 1 045 teens born after the "back to sleep" campaign and before orthotic helmet treatment became widely available locally (Roby, Finkelstein, Tibesar, & Sidman, 2012). This indicates that most NSP, but not all, reverses with time.

Suspected consequences

Severe plagiocephaly effects facial asymmetry. A noticeable asymmetric face is often considered less attractive, and although difficult to measure, the psycho-social developmental consequences can be significant (Hummel & Fortado, 2005). Persistent severe NSP can lead to teasing, poor self-conception, and teacher bias (B. Collett, Breiger, King, Cunningham, & Speltz, 2005).

Possible consequences of NSP beyond the psycho-social concerns of an altered appearance are being researched. In a study evaluating the neurologic profiles of infants between four and 13 months, infants with NSP were found to have significantly more altered tone - deflecting abnormally high and low tone - than infants without NSP (Fowler et al., 2008). In a study examining language acquisition in three-year-old children with NSP, 25%of the children who had not been operated had severe problems in receptive language skills (Korpilahti, Saarinen, & Hukki, 2012). In a study examining

Photograph 2. Stationary infant activity center is fun for infants but lying in this position on the floor can cause occipital flattening and can deter recovery. Signed parental consent to publish photograph has been obtained.

development of 36-month-old children, children with NSP scored lower on all of the Bayley Scales of Infant and Toddler Development than children without NSP, and differences were largest in cognition, language, and parent-reported adaptive behavior. Even 36-month-old children not previously diagnosed with NSP, but who were later rated by pediatricians to have at least mild NSP, had lower developmental scores than unaffected children. The researchers are careful to point out that findings do not imply that NSP causes developmental problems but suggest that NSP could serve as a marker of developmental risk (B. R. Collett et al., 2013). In a follow-up study at age 3-4 years of 129 infants diagnosed in infancy with NSP, 11% had one or more delays on the parent-completed age-appropriate Ages and Stages Questionnaires, and 13 % of parents reported still being somewhat or very concerned (Hutchison et al., 2011). A positive association between NSP and developmental delay was found in 13 of the 19 studies in a systematic review, and the researchers of the systematic review suggest that NSP could be seen as a marker of elevated risk of developmental delay (A. L. Martiniuk, Vujovich-Dunn, Park, Yu, & Lucas, 2017). In a prospective, nonrandomized study of infants referred to a cranial facial clinic, researchers found that the severity of NSP cannot be used to predict presence or degree of developmental delay (Fontana et al., 2016). The relationship between NSP and early developmental delays remains poorly understood (Andrews & Fontana, 2017), if at all existent. However, this suspicion requires attention.

Swedish recommendations

From 2003 until late 2013, the Swedish Board of Health and Welfare's sleep position recommendations to prevent NSP while infants sleep safely were: place the infant supine, make sure the infant is comfortably warm and can move, alternate the direction of the infant's head, and use a pillow until the infant starts to turn over (Socialstyrelsen, 2006). However, the Swedish pillow recommendation was removed in late 2013 because there was scarce evidence that pillow-use helped to prevent NSP (Socialstyrelsen, 2013). In Sweden, the use of an infant-adapted pillow is considered safe until the infant starts turning over. The incidence of SIDS in Sweden did not rise when the pillow recommendation was introduced and did not decrease when the pillow recommendation was removed in late 2013. The incidence of SIDS in Sweden was 0.21 per 1 000 live births in 2002, 0.18 per 1 000 live

births in 2013, and 0.14 per 1 000 live births in 2014 (Socialstyrelsen, 2015), i.e., before, during, and after the pillow recommendation respectively (III).

Background

Sweden has a national Child Health Care Program which is voluntary, free of charge and an integrated part of primary health care. The attendance rate in 2005 was nearly 100% (Hallberg, Lindbladh, Petersson, Rastam, & Hakansson, 2005). Nurses at the child health clinics are the primary health care providers responsible for monitoring infants' growth and development. The nurses have the responsibility of informing parents of newborns about the Swedish Board of Health and Welfare's SIDS prevention and current infant sleep position recommendations (III). In the mid-1990s, child health nurses in Skaraborg County, Sweden began noticing that some infants coming for well-child visits had obvious NSP. In some cases, the asymmetry had become severe before anyone reacted. These nurses had never been educated about NSP, so reversal advice consisted of repeating prevention recommendations and encouraging parents to increase "tummy time", time infants spend lying prone when awake. But on subsequent visits, many parents explained that their infants refused tummy time, and the asymmetry had not decreased (III).

Since nearly all infants in Sweden attend the child health clinics, these clinics provide an ideal venue for both monitoring infant cranial shape and imparting knowledge to parents. A prevention project was therefore initiated in 2008. Following a literature search on prevention practices, guidelines for nurses were developed (F. Lennartsson, 2011a). The idea was to develop a working tool on NSP prevention for busy nurses. The guidelines were tested in a pilot study (F. Lennartsson, 2011b), and revised. A continuing education on NSP, which included the revised guidelines, was developed for the nurses and a clinical intervention was planned.

The continuing education

The continuing education for nurses included the following information: How NSP develops; which infants are especially vulnerable; how to assess cranial shape; the national recommendations; the Child Health Care Program's recommendations; further recommendations on positioning infants to prevent NSP and re-positioning infants to reverse incipient asymmetry;

infant safety aspects; and when to consult a physician (II, III).

Assumption

Most NSP can be prevented if child health nurses are educated about NSP and provide parents of infants with early and on-going tailored advice, and if parents in turn implement this knowledge in their infant care.

Research questions

Can NSP be prevented by educating child health nurses who in turn provide parents with tailored recommendations regarding infant positioning? Can the intervention help reverse NSP that develops in the early months of infancy?

Aim

The overall aim is to assess whether it is possible to prevent NSP while still promoting safe infant sleeping practices.

Specific aims

I.   To evaluate reliability of assessors judging infant cranial asymmetry in order to assess if they could be considered reliable interchangeable assessors in the planned intervention.

II.   To compare intervention and control group parents' responses regarding cranial asymmetry prevention recommendations they had received from their nurses during their infant's first four months.

III.   To assess what knowledge from a continuing education intervention and control group nurses imparted to parents and parents implemented in their infant care.

IV.   To evaluate the effect of the clinical intervention on prevention and reversal of acquired cranial asymmetry in infants.

Methods

Project plan

A longitudinal single-blinded clinical intervention study and three supporting studies were conducted to evaluate how well the NSP prevention strategies worked. An overview of the methods of the four studies is seen in Table 1. The framework of the project is seen in Figure 1. The intervention was two-armed. Firstly, 35 intervention group nurses were educated about NSP in one-to-one or small groups at their workplace and asked to work according to specific guidelines, while the 18 control group nurses did not participate in the education and were not given guidelines to follow. Secondly, the clinical intervention was conducted where parents brought their infants to well-child visits according to the national Child Health Care Program's schedule. This created a long chain of information flow from project leader providing knowledge to nurses→ nurses learning →nurses providing recommendations to parents → parents' infant care → infants' cranial shape. The study was designed keeping in mind that this long chain needs to flow without being biased or diluted. This necessitated setting up check-up points.

There were five evaluations at different points in time to check how well the strategies were working. 1) Five individuals who had been taught by the project leader to assess infant cranial asymmetry were reliability-tested pre-intervention in order to evaluate if they could be considered trustworthy and interchangeable assessors in the clinical intervention (I). They were blinded to group and assessed cranial asymmetry in 176 intervention group and 92 control group infants in conjunction with two-, four-, and 12-month well-child visits. 2) When infants were four months old, parent knowledge was evaluated during the "window of opportunity" for NSP prevention (II). 3) when infants were 12 months old, knowledge nurses provided to parents and parents integrated in their infant care was assessed to evaluate the educational intervention (III). 4) 12-month cranial

Table 1. Overview of methods of the studies

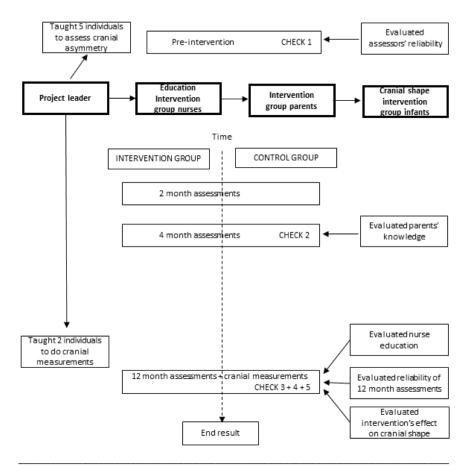| Study	Role	Design	Participants	Data collection and time point	Primary analysis
I	supportive study	reliability test	5 assessors	photograph test-retest and infant test, prior to clinical intervention	agreement analysis
II	supportive study	cross-sectional survey	272 parents	parent checklist in conjunction with 4-month well-child visits	statistical association
III	supportive study	evaluation study	50 nurses 270 parents	nurses and parents asked two open-ended questions when infants were 12 months old	qualitative content analysis and qualitative case-by-case analysis with process-oriented approach
IV	main study	longitudinal clinical intervention with 2 arms	274 infants 53 nurses 274 parents	cranial asymmetry assessments in conjunction with 2-, 4-, and 12-month well-child visits, cranial measurements at 12 months	longitudinal examination, statistical association, agreement analysis, regression analysis

Figure 1. Framework of the project

assessments were matched with 12-month cranial measurements in order to reliability test 12-month assessments using cranial measurements as the gold standard. 5) The effects of the intervention on infant head shape were evaluated (IV).

Recruitment and group allocation

Child health nurses were recruited to the project in December 2011. Nurses were allocated into groups because some nurses had previous exposure to the project, either through participation in the pilot study conducted in 2008 and/or by attending a group lecture on NSP prevention presented in 2010. Randomization into groups was excluded in order to avoid a so-called spill-over effect where nurses who were exposed to new knowledge might influence colleagues who had not. Therefore, if any nurse at a clinic had been exposed to the project by participating in the pilot study and/or attending the lecture held on NSP in December, 2010, all nurses at that clinic were placed in the intervention group. Nurses at clinics where no nurse had participated in the pilot study or attended the lecture held on NSP in December 2010 were placed in the control group. Infants and parents were placed in the same group as their nurse. Group allocation was retained in the supporting studies.

Nurses in both groups were asked to recruit infants born in February 2012 to the intervention study. Additional infants born other months were recruited until the sample size was deemed sufficient. The estimated sample size needed was 160 intervention group and 80 control group infants, when taking into account a 95% confidence interval, a 90% power, and an estimated effect size of 17% derived from the pilot study (IV).

Participants and setting

Participants included 35 intervention group and 18 control group nurses, 182 intervention group and 92 control group infants, and the infants' parents. The intervention was conducted at 26 child health centers in Skaraborg.

Evaluating assessors' reliability

Five individuals not working in the child health clinics were reliability tested after being trained to assess NSP using Severity Assessment for Plagiocephaly (Appendix A) and Severity Assessment for Brachycephaly (Appendix B). It was important to ascertain if these trained individuals could be considered reliable and interchangeable assessors for the clinical intervention. Reliable assessments were required for the trustworthiness of results. Assessors' interchangeability was important for logistic purposes since 816 cranial asymmetry assessments were conducted. The 274 infants were assessed on three different occasions in conjunction with appointments at 26 different child health clinics, and arranging the same assessor for each individual's three appointments was not feasible.

The five individuals, using the Severity Assessments as assessment tools, took a test-retest using 50 photographs of young children and a test involving six live infants. In the agreement analysis, Agreement Coefficient 2 (AC2) was chosen because it is still the only agreement coefficient that both weights disagreement and is appropriate when there are multiple raters. Furthermore, AC2 adjusts for random agreement in a way that is consistent and avoids inflating results by limiting random agreement to 50%, which other agreement coefficients do not (Gwet, 2002) Furthermore, the AC2 adjusts for number of raters, number of subjects, and number of response categories to avoid inflating results (Gwet, 2012). Quadratic weights were used to reflect our assumption on the non-linear severity across the four-picture series' scale in the Severity Assessments. Results were adjusted according to Gwet's model and interpreted (I).

Evaluating parents' prevention knowledge

A cross-sectional survey was conducted alongside the clinical intervention by distributing a checklist to 272 parents in conjunction with 4-month well-child visits. The checklist included 22 points pertaining to NSP recommendations. Parents' knowledge was investigated during the early months when the infant's cranium is most vulnerable to NSP development and when re-positioning can reverse NSP – the "window of opportunity" for

prevention and reversal. Parents' responses in the intervention and control group were compared using Fisher's Exact Test (II).

Evaluating the continuing education for nurses

An evaluation study of the continuing education for nurses on NSP was conducted when infants were 12 months old. The investigation concerned what knowledge on NSP prevention and reversal was implemented in practice, manifest content. Nurses were asked two open-ended questions regarding what recommendations they provided to parents to prevent NSP and to reverse cases that had begun to develop. Parents were asked two open-ended questions regarding what they did to prevent NSP from developing, and what they did to reverse NSP that had already begun to develop. Parents were also asked to rate their infant's cranial shape at 3-4 and 12 months.

Qualitative content analysis and qualitative case-by-case analysis were conducted with a process-oriented approach. Responses were coded. Then categories of knowledge were identified and each code was given a category of knowledge denotation. In the content analysis, coded responses were organized in a matrix. The codes in each nurse and parent column were arranged in descending order of importance and given bars where the height of each bar represents the relative importance of a code based on its' occurrence. This enabled group comparisons and examination of the process. In the qualitative case-by-case analysis, including cases parents perceived as severe at 3-4 months, the analysis followed the process from: nurse's sources of knowledge → parent's perceptions of severe asymmetry at 3-4 months → nurse's recommendations to parents → parent's infant care practices → parent's perceptions of asymmetry at 12 months (III).

Evaluating the reliability of 12-month assessments

The inter-rater agreement between 12-month plagiocephaly assessment scores and 12-month Cranial Vault Asymmetry Index (CVAI) measurement scores was analyzed with AC2. The inter-rater agreement between 12-month brachycephaly assessment scores and Cranial Index (CI) measurement scores was analyzed with AC2. Results were adjusted according to Gwet's model and interpreted (IV).

Evaluating effects of the clinical intervention on cranial shape

Statistical analysis included longitudinal examination, statistical association, agreement analysis, and regression analysis. Evaluating the true effect of the intervention on cranial shape is not straightforward. Given a symmetric cranium, asymmetry can be prevented between assessments or asymmetry can develop - prevention failure. Given an asymmetric cranium, asymmetry can reverse between assessments or asymmetry can persist - reversal failure (Figure 2). This approach examines what happens during the process, whereas net results sum up the process and miss what happens. One can see individuals' changes over time and the course of success and failure (IV).
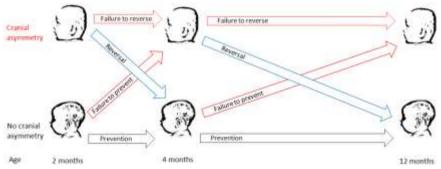

Figure 2. The ongoing processes of cranial asymmetry prevention and reversal in infants

Prevention and reversal are ongoing processes which can be influenced. The natural course, parents' habits, and national recommendations can also influence cranial shape of infants. However, it is difficult to separate these influences. Therefore, as a tool for evaluating the effect of the intervention, NSP prevention and reversal were analyzed separately. The *change* that occurred between assessments, i.e., prevention failure and reversal, was analyzed because the occurrence of change is something that can easily be detected (IV).

Results

Assessors: Intra-rater agreement was examined when five assessors and the project leader took the test the first and second time. Inter-rater agreement was analyzed when two different raters took the first test and when two different raters took the retest. Perfect agreement, the core of every agreement analysis, is seen on the diagonal of an agreement matrix. Perfect agreement is expressed as frequencies on the diagonal, as in the photograph retest (Table 2). Mean percentage of perfect intra-rater agreement was 73 (I). This is sometimes referred to as crude or unadjusted agreement.

Adjusted intra-rater AC2 was 0.69 [0.63; 0.76]. Adjusted inter-rater AC2s were 0.72 [0.64; 0.81] in the first test and 0.71 [0.63; 0.79] in the retest. In the infant test, the adjusted inter-rater AC2 was 0.74 [0.60; 0.87]. Each of these results indicate substantial strength of assessor agreement. Furthermore, assessors' detection of asymmetry in the infant test corresponded with the reference-raters' in 23 of 24 instances – which is nearly perfect. Thus, these five individuals were deemed to be reliable and interchangeable assessors of cranial asymmetry assessments in the clinical intervention (I).

Parents' awareness of recommendations from their nurse: In the 4-month survey, a significantly higher proportion of intervention group parents compared to control group parents were aware of regular recommendations - alternate direction of the infant's head when putting the child to bed (82%: 64%, p = 0.001), which pillow to use (92%: 80%, p = 0.01), and when to remove the pillow (48%: 31%, p = 0.006) - and five of seven newly introduced recommendations. The five new recommendations introduced to nurses include: begin to introduce awake infant to tummy time by two weeks of age (51%: 34%, p = 0.007), limit time in an infant car seat to car rides (50%: 26%, p = 0.001); limit time in the infant bouncer (54%: 26%, p = $\leq$ 0.001); avoid subjecting the infant's head to pressure from hard surfaces (67%: 42%, p = $\leq$ 0.001), and change arms when bottle feeding (54%: 17%, p = $\leq$ 0.001) (II).

Table 2. Frequencies of inter-rater agreement sorted by type of asymmetry in 50 photographs when 6 assessors judged cranial asymmetry in the retest

RETEST		ASSESSOR 2											
		Vertex view skew				Vertex view flat				Lateral view flat			
ASSESSOR 1		0	1	2	3	0	1	2	3	0	1	2	3
Vertex view skew	0	42*	16										
	1	7	13	12	6		2	1					
	2		10	28	9			4					
	3	2	10	14	62								
Vertex view flat	0		1			42*	2						
	1	1					7	1					
	2			3	2	2	9	4	1				
	3							4	10				
Lateral view flat	0									180	23		
	1									58	41	19	4
	2									9	8	39	25
	3										1	11	47

Degree of asymmetry: 0 = none, 1 = mild, 2 = moderate and 3 = severe.
Perfect agreement is expressed as frequencies on the diagonal. Disagreements regarding degree of asymmetry are expressed as frequencies in non-diagonal cells where increased distance from diagonal indicates stronger disagreement. Frequencies in areas outside four by four blocks indicate a photograph was judged as skewed by one assessor and flat by another.
*Counts included in 2 cells when a photograph was judged to have no asymmetry since that indicates both 0 skew *and* 0 flat agreement. These counts are only used once in calculations.

The proportion of intervention group compared to control group parents that reported receiving verbal recommendations from their nurse was 96% vs 85% (p = 0.002), and the proportion that reported receiving written recommendations was 68% vs 58%, although the observed 10% difference between the groups was not statistically significant. Five control group parents did not report having received either verbal or written recommendations from their nurse (II).

Integrating knowledge into practice: In the qualitative content analysis, intervention group nurses imparted both regular and newly-introduced

knowledge on positioning strategies to parents. The national pillow rec-ommendation at the time predominated in parents' responses and there were only sporadic parental reports of new knowledge. Nevertheless, an indication that intervention group parents had even incorporated new knowledge into infant care can be seen in the case-by-case analysis. Inter-vention group parents who perceived severe cranial asymmetry when in-fants were 3-4 months old reported implementing regular and new infant positioning recommendations, and most reported complete reversal of se-vere cranial asymmetry. No control group parent reported complete rever-sal. We compared responses in the two groups in order to understand why only the intervention group seemed successful in reversing severe cranial asymmetry. Intervention group nurses and parents reported re-positioning strategies from the official national recommendations, child health pro-gram recommendations, and nurse education. This infers that severe cra-nial asymmetry reversed when regular and new recommendations were implemented in joint nurse-parent efforts (III).

Infant safety: In the 4-month survey, most parents in both groups reported that their nurse provided information on why an infant should have tummy time under surveillance (85%: 82%; p = 0.601) and most reported having been informed about which infant pillow is appropriate to use (92%: 84%; p = 0.001). However, less than half of the parents in both groups reported having been informed *when* to remove the pillow (48%: 31%; p = 0.006), and less than half of the parents in both groups reported having received an explanation regarding *why* they should remove the pillow (45%: 29%; p = 0.009), safety aspects of pillow use (II).

In the 12-month qualitative content analysis, most nurses and parents that reported tummy time did not report the safety details of tummy time - that infants are awake and under surveillance when they are placed prone. Also, some parents in both groups reported placing their infant prone for sleep, a risk factor for SIDS. In addition, side sleep, also a risk factor for SIDS, is seen in both nurses' and parents' lists (III).

Reliability of assessments: The adjusted inter-rater agreement between plagiocephaly assessment scores and CVAI measurement scores at 12 months was 0.74 [0.68; 0.80] using AC2. The adjusted inter-rater agreement between brachycephaly assessment scores and CI measurement scores at

12 months was 0.81 [0.78; 0.84]. When interpreted, the size of both coefficients corresponds with substantial agreement. This indicates that assessments made by the assessors can be considered trustworthy when using the CVAI and CI measurements as the gold standard (IV).

The course of NSP development: In the clinical intervention, the proportion of infants with NSP was lower in the intervention group compared to the control at each of the three assessments (Figure 3) (IV). However, these results are not statistically significant. The proportion of infants with plagiocephaly and the proportion of infants with brachycephaly was lower in the intervention group compared to the control at each of the three assessments, and these results are not significant either. Yet in a longitudinal analysis, intervention group infants with combined plagiocephaly/brachycephaly showed a significantly different course of development than control group infants (p = 0.04) (Figure 4) (IV). The two groups took different paths. The proportion of infants with combined plagiocephaly/brachycephaly began to decrease in the intervention group between two and four months and then stayed at this level until twelve months, while the proportion of control group infants increased slightly between two and four months and then decreased between four and twelve months (IV).

Prevention: In the clinical intervention, the prevention effect was estimated by considering the opposite phenomenon to prevention, namely, the occurrence of prevention failures. Non-cases at the outset of each time were the starting point. In the subgroups of infants who were non-cases at two months, six of 138 (4%) intervention group and seven of 65 (11%) control group infants developed plagiocephaly, and 34 of 138 (25%) intervention group and 14 of 65 (22%) control group infants developed brachycephaly by 12 months. Thus, overall brachycephaly prevention failure (24%) was three times more common than overall plagiocephaly prevention failure (8%). Nine intervention group infants developed brachycephaly after four months. However, we are unable to explain this late development. It turned out that NSP prevention was difficult despite our efforts, and that brachycephaly was more difficult to prevent that plagiocephaly in both groups (IV).
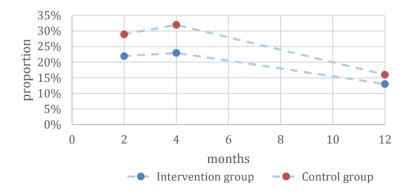
Figure 3. Proportions of intervention and control group infants at 2, 4, and 12 months with nonsynostotic plagiocephaly
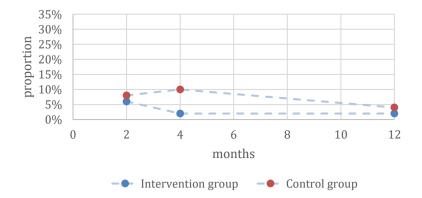

Figure 4. Proportions of intervention and control group infants at 2, 4, and 12 months with combined plagiocephaly/brachycephaly

Reversal: In the clinical intervention, NSP was assessed by trained assessors blinded to group assignment. There was a 24% difference between groups (65% intervention group; 41% control group) in NSP reduction from two to four months (p = 0.06), considered early reversal. There was a 50% difference between groups (50% intervention group; 0% control group) in combined plagiocephaly/brachycephaly reduction from two to four months (p = 0.03). Plagiocephaly and brachycephaly reduction from two to four months pointed in the same direction, but these subgroups are small and results are not statistically significant. There was a 10% difference between groups in NSP reduction from two to 12 months, indicating the intervention did not contribute much to late reversal. Six (3%) intervention group and nine (10%) control group infants had NSP at each assessment, i.e. persistent asymmetry, (RR = 0.35 [0.13; 0.94], p = 0.03), indicating intervention group infants seemed to have nearly a threefold lower risk for persistent NSP at 12 months, but the numbers are low (IV).

Factors that might help explain reversal were investigated. In the subgroups of infants with NSP at two months, it was four times more common that NSP reversed by four months in intervention group compared to control group infants (OR = 4.07 [1.23; 13.44], p = 0.02) when adjusted for parent awareness of written recommendations from their nurse. However, it was nine times more common that NSP at two months reversed by four months when parents were aware of written recommendations from their nurse (OR = 9.09 [0.02; 0.48], p = 0.004) when adjusted for group. It was 6½ times more common that NSP at two months reversed by four months when parents were aware of written recommendations from their nurse (OR = 6.5 [0.04; 0.61], p= 0.007). In addition, it was >5 times more common that NSP at two months reversed by 12 months when parents were aware of written recommendations from their nurse, (OR = 0.19 [0.04; 0.95], p = 0.04). Thus, it turned out that parents' awareness of written recommendations on NSP from their nurse had more effect on reversal than the intervention itself (IV).

Role of background factors: The two infant groups in the sample were, on the whole, similar regarding birth-related risk factors. At two months, a larger proportion of intervention group infants compared to control group infants had parent-reported side preference (44%; 36%, p = 0.18); a smaller proportion of intervention group infants compared to control group infants were solely bottle-fed (21%; 35%, p = 0.01); and there was a wide

range in parent-reported time infants in both groups spent in positional devices daily, most markedly the bouncer. The minimum - maximum time infants spent daily in a bouncer at two months was zero minutes - eight hours in the intervention group, and zero minutes - 9 hours 40 minutes in the control group (IV).

No single risk factor stood out, but 13 of the 15 infants who failed to reverse had two or more of the following risk factors: firstborn, side preference at two months, solely bottle-fed at two months, and spent $\geq$ 2 hours daily in positional devices at two months. Birth-related factors, side preference, and care factors of these 15 infants were compared with those of infants whose NSP did reverse between two and 12 months (Table 3) (IV). A larger proportion of infants whose NSP failed to reverse were firstborn (60% vs 44%), had a vacuum-assisted delivery (27% vs 11%), and were solely bottle-fed at two months (53% vs 33%). Moreover, their median daily bouncer time at two months was higher (90 minutes vs 30 minutes). However, no significant differences were detected, and no conclusions can be drawn regarding associations between risk factors during early infancy and failure to reverse by 12 months. Thirteen of the 15 infants who failed to reverse had brachycephaly at 12 months. Of the 12 late developers, 11 developed brachycephaly, and four spent $\geq$ 3 hours daily in positional devices at two months (IV).

Table 3. Comparison of parent-reported birth-related factors, side preference, and care factors of infants whose nonsynostotic plagiocephaly at 2 months failed to reverse and did reverse by 12 months

	Failed to reverse n = 15	Reversed n = 45
Birth-related factors		
male	7 (46 %)	22 (49 %)
birth weight (g)	3538 (3035 - 4015)	3540 (2875 - 4600)
gestational age (wks)	40 (38 - 42)	40 (36 - 42)
vacuum-assisted delivery	4 (27 %)	5 (11%)
first born	9 (60 %)	20 (44 %)
twin	2 (2 %)	6 (3 %)
born with a flat spot	2 (13 %)	4 (8 %)
Side preference at 2 months	8 (53 %)	23 (51 %)
Care factors at 2 months		
solely bottle-fed	8 (53 %)	15 (33%)
estimated time spent daily (min.)		
in infant car seat	24 (08 - 60)	20 (0 - 120)
in infant bouncer	90 (10 - 150)	30 (0 - 360)
in stationary infant activity center	33 (0 - 90)	1 (0 - 270)
total daily time in positional devices (min.)	139 (71 - 285)	98 (15 - 373)

n (%) or medians (min-max)

Discussion

The overall aim is to assess whether it is possible to prevent NSP while still promoting safe infant sleeping practices. The plan was to intervene by providing child health nurses with new knowledge in an attempt to improve practice. The assumption was that if child health nurses participated in a continuing education on NSP, were provided with guidelines to follow, and in turn provided tailored recommendations to parents of newborns, nearly all NSP would be prevented. The five phases of evidence-based nursing practice, i.e., ask the clinical question, search for the evidence, critically appraise the evidence, apply the evidence to practice, and evaluate the effectiveness of the evidence (Hockenberry, Wilson, & Barrera, 2006), were utilized from the conception of the project.

This thesis includes a longitudinal clinical intervention (IV) and three supporting studies (I, II, III), Each study contributed in a different way to the project. The reliability study assessed the agreement of the assessors who were specifically trained to judge infant cranial shape for the clinical intervention. This study was conducted pre-intervention. It was needed so that assessments in the clinical intervention could be considered trustworthy and so that the assessors could be considered interchangeable for logistic purposes (I).

The intervention had two arms - the continuing education on NSP for nurses and the clinical intervention. These created a long chain of information flow from project leader providing knowledge to nurses→ nurses learning → nurses providing recommendations to parents → parents' infant care → infants' cranial shape. We examined what was going on at different points in time in order to check how strategies were working. The cross-sectional study provided data on what parents learned from their nurse during the early months – the "window of opportunity" for cranial asymmetry prevention and reversal (II). The qualitative study at 12 months evaluated the continuing education for nurses by assessing what knowledge intervention and control group nurses and parents had integrated into

practice (III). In the clinical intervention study, the effectiveness of the strategies on infant cranial shape was evaluated at two months, four months and 12 months (IV).

Each study was conducted with a deductive approach including outcome-oriented research questions. Three studies (I, II, IV) were analyzed quantitatively and one study (III) was analyzed utilizing qualitative comparison. Each study explored different aspects of the intervention in an attempt to evaluate which components of the process worked or did not work, and to identify areas that need improvement.

Strategies that worked

Findings indicate that child health nurses and parents of young infants were motivated to improve practice, the assessors were reliable; the intervention had a significant effect in increasing knowledge nurses provided to parents; nurses and parents integrated new knowledge into practice; and nurses' and parents' joint efforts helped in reversal of NSP and in decreasing infants' risk of having persistent asymmetry at 12 months.

Motivation: Nurses in both groups were motivated to participate in the project. This can be seen in the high proportion of child health nurses employed at the time who agreed to participate in the project – 57 of 72 (79%) – and the high proportion of participating nurses who followed through – 53 of 57 (93%). This indicates they wanted to improve practice regarding NSP prevention. Parents in both groups were motivated to participate in the studies and to learn from their nurses. All parents followed through unless the family moved – 278 of 284 (98%) (IV). The Swedish child health care setting turned out to be an ideal venue for motivating both nurses and parents to participate in NSP prevention efforts.

Assessors' reliability: Findings indicate that when assessors were taught how to assess cranial asymmetry using Severity Assessments and then reliability tested, there was substantial strength of assessor agreement. Assessors showed excellent ability to detect NSP in the clinical setting, although numbers were low (I). Reliability was checked once more just to make sure. Assessments at 12 months showed substantial strength of agreement when using cranial measurements as the gold standard. We believe this trustworthiness can be extended to assessments conducted at

two months and four months as well, since these assessments were done in the same way (IV).

Imparting knowledge to parents: Findings indicate that the Swedish child health care program is a good starting point regarding imparting knowledge to parents. In the 4-month survey, most parents in both groups reported that their nurses had provided verbal recommendations on NSP prevention (96%: 85%; p = 0.002) and that their nurse had explained why infants should have tummy time under surveillance when awake (85%: 82%; p = 0.601). Findings also indicate that educating child health nurses about NSP worked to increase parents' awareness of recommendations. Intervention group parents reported having received significantly more recommendations from their nurse than control group nurses, both regular recommendations and newly introduced recommendations during the early months of infancy when parents' knowledge can influence infants' head shape (II).

Integration of new knowledge into practice: Intervention group nurses seemed to have integrated cranial asymmetry assessments in daily practice using the Severity Assessments because of the 184 intervention group infants, nurses provided assessments for 179 infants at 2-months and 180 infants at 4-months (IV). Also, according to parents' reports, intervention group nurses provided significantly more regular recommendations and five newly introduced recommendations during the first four months compared to control group nurses (II). Furthermore, intervention group nurses reported recommending new re-positioning details to parents regarding prevention and reversal, including how to accomplish occipital pressure relief when infants are awake, asleep, and being fed (III). Intervention group parents reported implementing both regular and new recommendations in their infant care, and intervention group parents who perceived severe cranial asymmetry at 3-4 months reported incorporating new re-positioning details in their infant care (III). A further indication that new knowledge was integrated into practice is the early reversal success in the intervention group (IV). This could be due to the intervention including specific reversal recommendations while the national recommendations do not. Another reason could be that intervention group nurses learned how to assess cranial asymmetry, while control group nurses did not. However, it is not known if intervention group nurses continued to integrate new

knowledge into practice after the clinical intervention ended. This would be interesting to explore.

Joint reversal efforts: Nurses and parents collaborated in efforts to help reverse incipient NSP. In the case-by-case analysis there were observations that intervention group nurses gave reversal advice to parents using both regular and newly introduced knowledge, and that parents who reported that their infants had severe asymmetry at 3-4 months implemented both regular and newly introduced knowledge in their infant care (III).

Decreased risk for persistent asymmetry: The risk for persistent asymmetry at 12 months was significantly lower for intervention than control group infants (RR = 0.35, [0.13; 0.94], p = 0.03) in the group of infants who had NSP at two months. Although the numbers of infants with persistent asymmetry were low, six intervention group and nine control group, this indicates that intervention group nurses and parents were effective in decreasing an infant's risk of having persistent asymmetry at 12 months (IV).

Strategies that did not work

Intervention group nurses were trained to assess cranial asymmetry in the same way as the assessors, yet their cranial asymmetry assessments did not always agree with assessors' assessments. In a sensitivity analysis of the data intervention group nurses provided from 2-month cranial asymmetry assessments, they showed a 65% sensitivity in detecting NSP when using the assessors' 2-month assessments as the gold standard. Intervention group nurses failed to detect 22 of 37 (59%) cases detected by the assessors (IV). Early detection is the key to prevention, so these findings suggest there is need for improvement. However, these results are based on our rating system. It is worth noting that intervention group nurses detected at least some asymmetry in 31 of the 37 (84%) 2-month assessor-detected cases. Yet whenever mild asymmetry was detected, it did not meet our rating system's criteria for NSP. We need to remember that the Severity Assessments are not precision tools. Another aspect is that comparing nurses' assessments with the assessors' is not entirely fair. Assessors had one duty – to do cranial asymmetry assessments - while assessing cranial asymmetry was only one small part of the child health nurses' job during a well-child visit, and these visits can be stressful.

Recommendations did not "get through" to all intervention group parents. Although all parents received "This is Your Child's Health Book" (Djäken, 2007) from their nurse which included the national prevention recommendations and the Child Health Care Program's prevention recommendations, and intervention group nurses were given a pamphlet to distribute to parents, only 68% of intervention group parents reported having received written recommendations on NSP from their nurse during their infants' first four months (II). Thus, they probably did not read the written recommendations that were provided. Furthermore, only 48% of intervention group parents reported having received information from their nurse regarding when to remove the recommended infant pillow and why to remove the pillow, safety aspects of infant pillow use (II). Some parents reported placing their infants prone or on the side for sleep, both considered unsafe sleeping positions. Furthermore, it is unclear how many intervention group parents were aware of the need for surveillance during tummy time, because few reported this safety aspect (III).

Recommendations on infant positioning devices did not get through to all intervention group parents either. Only 50% reported having received information on using infant car seats only during car rides, and only 54% reported having received information to limit time in infant bouncers (II). Parent-estimated minimum-maximum time intervention group infants spent daily in a bouncer was 0-480 minutes (IV). We do not know if the intervention group nurses forgot to inform some parents about some positioning details, if some parents chose not to comply, or if for some reason parents did not understand the provided information. Parents may have different abilities. Some may have language difficulties. Too much information at one time may make it difficult to remember details.

Prevention of NSP was difficult even in the intervention group (IV) despite the nurses' and parents' high motivation to participate and follow through in the study, and despite intervention group parents' increased knowledge about NSP at four months compared to control group parents (II). In the sensitivity analysis of intervention group nurses' 2-month assessments using assessors as the gold standard, there was a sensitivity of 65%. Thus, nurses failing to detect about three in five cases at two months is one possible explanation for the early prevention failure.

Brachycephaly was most difficult to prevent in both groups. In the sub-groups of infants who were non-cases at T1 and subsequently developed brachycephaly, overall brachycephaly prevention failure (24%) was three times more common than overall plagiocephaly prevention failure (8%). Of the nine "late developers" in the intervention group – infants that developed NSP after the 4-month peak -, all nine developed brachycephaly (IV).

What was missed

Early identification of head positional preference was not included in the continuing education for nurses or in the guidelines. Intervention group nurses learned to evaluate the cervical range-of-motion in infants who were old enough to support their heads, and were instructed to ask about side preference, not head positional preference. According to Rogers, 2011, the most important risk factor to find out about is whether an infant has a head positional preference. Rogers recommends asking parents about head positional preference at the first well-child visit and evaluating the cervical range-of-motion with the neonate lying supine (Rogers, 2011b). Whereas asking parents about side preference can help identify risk for developing plagiocephaly - a skewed head shape – and torticollis, asking parents about head positional preference could help in early identification of risk for developing brachycephaly – central occipital flattening - as well. And, as it turned out, brachycephaly was both difficult to prevent and to reverse in the intervention (IV).

Ethical considerations

Although the supine sleep position puts pressure on the occiput, SIDS prevention is the unquestionable priority in safe infant positioning. No infant should come to harm from NSP prevention and reversal efforts. Moreover, efforts to help children to have their normal head shape can conceivably protect them from possible teasing regarding an altered head shape, even later on in life. Furthermore, we want to help each child reach their full potential, and as long as there is suspicion regarding an association between NSP and developmental delays, prevention and reversal efforts are warranted.

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Clinical implications

The main principle of NSP prevention is simple **–** to relieve pressure on the infant's occiput. However, the situation is more complex, and the chain of information flow is long. The supine sleep position is recommended for infant safety and infant safety must always be the priority. Yet, infants sleep a lot. Sleeping supine during early infancy when infants lack muscle strength to change their own position, puts consistent pressure on the formable occiput. Therefore, it is important that parents, the caregivers, are provided with current and safe repositioning recommendations.

Tummy time under surveillance when an infant is awake is an important principle of safe NSP prevention. Tummy time provides total pressure relief on the occiput. However, there are several important aspects that parents need to be made aware of: tummy time should be initiated in the early neonatal period to get the infant accustomed to the prone position to avoid protests; even short periods several times daily are helpful; tummy time should be increased successively as the infant becomes awake for longer periods; and surveillance is necessary for infant safety whenever infants are placed prone. However, surveilling infants during increased tummy time is time consuming. Families' situations and life styles are not always conducive to frequent and increasingly lengthy periods of surveilling infants in the prone position. So, parents' understanding of the tummy time recommendation is important for both infant safety and parent compliance.

Parents have different capabilities, motivation, and backgrounds which can all effect compliance. Therefore, recommendations need to be presented clearly, and tailored to parents understanding and the situation at hand. It is important not to present too much information at once, otherwise some parents can become overwhelmed and/or forget. Repeating information can be useful in some instances. It is important that parents do not to mix up what to do during infant awake time with infant sleep time. Language difficulties need to be dealt with for the immigrant population in Sweden.

Although all parents in both groups had received written recommendations from their nurse at the first home visit, we found that more parents were aware of receiving verbal recommendations about NSP prevention from their nurse in the early months (II). Since parents seem to remember the spoken word, child health nurses should discuss recommendations with parents. Discussing with parents instead of merely relaying information can increase parents' understanding and motivation. This requires both good communication skills and up-to-date knowledge on NSP prevention (III). A finding from the regression analysis that can be useful in clinical practice is the importance of parent awareness of written recommendations from their nurse on reversal of NSP (IV). Parent awareness implies compliance. However, this needs to be investigated further.

Update

An infant-adapted pillow was recommended in the intervention for supine sleeping infants until they began to turn over, in accordance with Swedish national recommendations at the time. It is not known how pillows influenced results. However, in late 2013 after the intervention ended, the national pillow recommendation was removed. In order to comply with this change in national recommendations, a pillow is no longer recommended. Thus, intensified efforts are needed to reduce pressure on the occiput when infants are awake.

In "This is Your Child's Health Book" provided to parents at the time of the intervention, NSP prevention information is on page 38. In the new edition provided to parents since 2015, NSP prevention information is on page 14 (Central barnhälsovård Västra Götaland, 2015), much earlier and easier to find. Safe infant positioning and explanations on the importance of early tummy time and varying infants' head position are included.

Contributions to practice

The Severity Assessment for Plagiocephaly and the Severity Assessment for Brachycephaly, two non-intrusive assessment tools suitable for early detection in child health care clinics, were introduced to the nurses. These could be used in daily practice as a reminder to nurses on what aspects of asymmetry to include when doing cranial assessments. Nurses were handed to opportunity, but it is not known whether they continued to use

the Severity Assessments after the study. This would be interesting to explore in a future study.

A large part of the project was method development. The reliability test of assessors taught to judge cranial asymmetry using the Severity Assessments indicate that nurses can also learn to judge cranial asymmetry using the Severity Assessments. This can be useful for early detection. A continuing education was developed which provided child health nurses with knowledge on NSP development and re-positioning recommendations that went beyond the national recommendations. Guidelines for nurses were developed which encourage regular assessments to promote prevention, early detection, and reversal. They provide a step-by-step working tool for busy child health nurses which guides them in assessing NSP and when to consult a physician, and include prevention and reversal recommendations for parents. New knowledge gained from the four investigations together with the change in official national recommendations resulted in a new version of the guidelines (Appendix C). The focus of prevention efforts is now on informing parents to alternate sides of the neonate's head when placed for sleep and occipital pressure relief when infants are awake. Nurses are encouraged to ensure that safe infant positioning recommendations reach all parents and to screen for head positional preference in the neonatal period.

Further method development in the project includes introducing the concept of NSP prevention and reversal as on-going processes, and the approach of analyzing prevention and reversal separately. This enables an examination of what happened during the process, because net results only sum up what happened. This new analysis approach can be targeted at alleviating problems, whereas the model with only net results is more difficult to amend.

Conclusions

The assessors were considered reliable and interchangeable in the clinical intervention. Educating child health nurses to assess infant cranial asymmetry in the same way as the assessors can be used for early detection (I). Educating child health nurses about NSP and providing them with specific guidelines to follow increased parent's awareness of NSP prevention recommendations during the early months when infants are most vulnerable to NSP development (II), and contributed to new knowledge on infant positioning being integrated into practice (III). However, safety aspects did not get through to all parents (II, III).

Findings from the clinical intervention helped answer the two research questions. 1) Can NSP be prevented by educating child health nurses who in turn provide parents with tailored recommendations regarding infant positioning? No, the intervention did not succeed in preventing NSP. Prevention was difficult, most notably brachycephaly prevention. 2) Can the intervention help reverse NSP that develops in the early months of infancy? Yes, partly. The intervention was associated with early reversal and reducing infants' risk for persistent asymmetry (IV).

The intervention, starting with educating the nurses and resulting in head shape of infants, did work to some extent. The intervention seemed to boost the national recommendations and to promote new knowledge being integrated into practice. Providing nurses with an education on NSP who in turn provide parents with recommendations can work. However, nurses need to learn more about how to assess NSP, how to effectively communicate recommendations to parents, and to ensure that the parents of every infant understand safe infant positioning. Further research is needed to improve the education for nurses and to investigate why brachycephaly was so difficult to prevent.

Future Perspectives

Since the supine sleep position is the safest sleep position for infants, this sleep position will continue to be recommended. Thus, parents of infants will continue to need recommendations on how to concomitantly prevent NSP. It would be helpful if all child health nurses are provided with a continuing education on NSP.

Findings indicate that the strategies are on the right track, yet further research is needed. More research is needed to investigate effective ways for child health nurses to communicate knowledge to parents. Since parents' awareness of written recommendations from their nurse had a significant effect on reversal, it would be helpful to investigate why this was so. It would be interesting to investigate if the nurses continue to use the Severity Assessments in practice, and what other new knowledge they continue to integrate into practice. More research is needed to investigate why brachycephaly prevention was so difficult.

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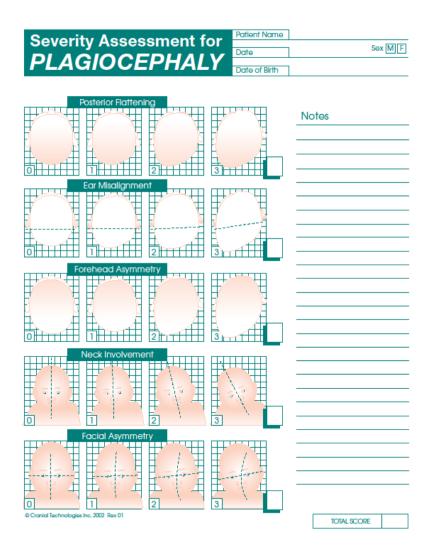
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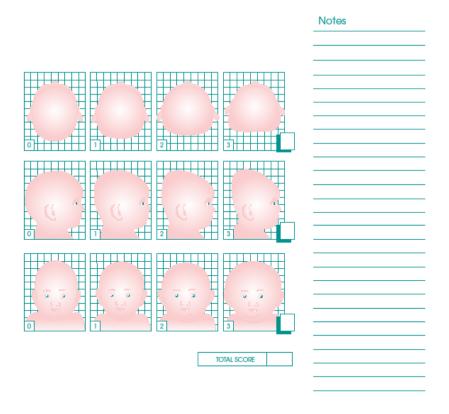
Appendix A

Cranial Technologies granted permission to publish the Severity Assessments.

Appendix B

Severity Assessment for BRACHYCEPHALY

Patient Name _____ Sex [M] [F]

Date _____

Date of Birth _____

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| 0 | 1 | 2 | 3 |

| 0 | 1 | 2 | 3 |

TOTAL SCORE [____]

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Appendix C

Prevention of acquired infant cranial asymmetry
Guidelines for child health nurses:

The recommendation to place infants on their backs for sleep led to a decrease in Sudden Infant Death (SIDS), so this sleep position recommendation must always be followed for infant safety. However, the back-to-sleep position can easily lead to a flat area developing on the back of infants' formable heads in the early months since infants cannot turn themselves. Re-positioning strategies initiated in early infancy help to prevent this flattening.

1. Begin informing parents at the first home visit. Show parents the information on page 14 of the Child's Health Book and talk about it.

2. Assess back of infant's head shape regularly for at least 6 months
   - Observe head from above and looking down – bird's eye view
     Is there centered flattening? one-sided flattening? ear misalignment?
   - Observe and palpate back of head for flattening
     A bald spot on one side could be a sign of one-sided flattening.
   - Observe left and right profile view for flattening.
     Does the head peak?
   - Observe the infant's face.
     Does the face seem wide despite average head circumference?
     Does one side of the forehead bulge?

Signed parental consent to publish photographs has been obtained.

3. Assess for head positional preference and head tilt
   - Ask parent at first visit if their infant has a head positional preference. This can be counteracted by placing the infant's head in another direction.
   - Ask parent if infant always looks in the same direction? This can be counteracted by encouraging the infant to look in the other direction.
   - Assess the cervical range-of-motion with the neonate laying down.

- When infant can hold head upright, observe from the back for head tilt.

4. Monitor head shape carefully if infant has following risk factors: premature, firstborn, twin, side preference, torticollis, only bottle-fed, vacuum-assisted delivery, born with a flat spot, cephalohematoma, decreased movement or level of activity, and brace for hip dysplasia.

5. Refer to physician: if asymmetry does not improve within 2 months; if craniosynostosis is suspected; if torticollis is suspected.

Recommendations for parents to prevent cranial asymmetry

Infants should sleep on their back and lie with their head alternatingly in the right and left direction.

Minimize pressure on the back of infant's head whenever infant is awake.
- Begin getting infant accustomed to tummy time from week one.
- For safety, the infant should always be awake and under surveillance during tummy time.
- Begin with short periods of tummy time several times daily and increase the time successively.
- Make tummy time fun by being on the floor together or placing a toy in front of your infant.
- Pick up your infant before the child becomes too upset.
- An infant can also become accustomed to tummy time by laying on the chest of a reclining parent or by laying on a parent's lap.
- If you place your infant's elbows close to their body, this will provide a more stable base when your infant begins to lift his/her head.

Photographs and signed consent have been obtained from parents.

- Only have infant in car seat during car rides.
- Minimize time infant spends in bouncer and swing.
- Minimize time back of infant's head is placed on hard surfaces, such as on the floor in a stationary activity center.

- Alternate arms when bottle-feeding.
- Carry your infant.

Recommendations for parents to reverse flattening that has developed

The major principle is to alleviate pressure on the back of infant's head whenever infant is awake.

- Increase tummy time under surveillance as much as possible when infant is awake.
- Carry your infant in your arms or in a carry shawl.
- The Bumbo chair is useful once your infant has begun to sit, but your infant needs to be watched and should only sit as long as he/she can hold his/her head upright.
- Avoid longer periods in car seats, bouncers, and infant swings where your infant's head is not supported, otherwise a side preference can develop quickly.
- For infants who are bottle-fed and have developed a one-sided flattening, change arms when feeding so that the flattened side is upwards.

To alleviate pressure on the flattened area while the infant sleeps, use Delta Baby side pillow (wedge pillow) in the crib and carriage. This wedge pillow provides safe side positioning. If it is one-sided flattening, the flattened side of the head should be upward when placed for sleep. If the flattening is in the middle, alternate left and right side upward when placed for sleep.

Signed parental consent to publish photographs has been obtained.

Developed by Freda Lennartsson, Skaraborg, Sweden 2018 in consultation with Professor Göran Wennergren.