Nonsynostotic Plagiocephaly

Subjects: Pediatrics

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The dissertation, comprising a clinical intervention and three supporting studies, aimed to assess if it is possible to prevent nonsynostotic plagiocephaly while promoting safe infant sleeping practices. Five individuals were trained to assess cranial asymmetry and then reliability-tested; the interpreted results indicate substantial strength of rater-agreement. Intervention participants were allocated to group. Only intervention group nurses participated in the continuing education on plagiocephaly developed for nurses. A survey compared information intervention and control group parents received from nurses; intervention group parents were significantly more aware of recommendations than the control group parents. The nurse education was evaluated by asking intervention and control group nurses and parents two open-ended questions; the intervention group nurses and parents reported new re-positioning strategies. The effect of the intervention on cranial shape was evaluated by assessing asymmetry at 2, 4, and 12 months (176 intervention group; 92 controls). It was nine times more common that cranial asymmetry 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. An infant's risk of asymmetry persisting until 12 months was significantly reduced in the intervention group (RR = 0.35 [0.13; 0.94], p = 0.03). Preventing brachycephaly was difficult. Conclusions: the assessors were considered reliable; educating nurses promoted the integration of new recommendations in practice; the intervention was associated with early reversal of nonsynostotic plagiocephaly.

Keywords: assessments ; child health ; education ; infants ; intervention ; nonsynostotic plagiocephaly ; nurses' instruction ; parents ; prevention ; reversal

1. Introduction

Nonsynostotic plagiocephaly (NSP) is acquired cranial asymmetry that develops from pressure which occurs when an external force is regularly applied to an area of an infant's cranium over a period of time [1]. The contact force generated between the cranium and resting surface resists growth in the area where there is contact and displaces growth to areas with no resistance. This process is similar to how a pumpkin flattens as it grows—it cannot expand into the ground and therefore grows along it [2]. There are three main groups of NSP: plagiocephaly-skewed occipital flattening, brachycephaly-symmetric occipital flattening, and combined plagiocephaly/brachycephaly [3].

Prevalence is difficult to determine. In the first weeks postpartum, it is difficult to differentiate between pre-natal NSP and cranial molding from the birth process; no study has established when cranial molding stops and post-natal NSP begins [4]. In addition, prevalence is a measure which is calculated at one point in time, but NSP is not static. It can develop, reverse, and develop again [5]. The prevalence seems to rise during the first four months and then gradually decreases [6], so age differences are important to consider when calculating prevalence. According to a systematic review, the point of prevalence may be as high as 22.1% at seven weeks and as low as 3.3% at two years [7].

Coinciding with the "back to sleep" campaign in the early 1990s, when parents were recommended to place their infants supine for sleep to prevent Sudden Infant Death Syndrome (SIDS), there was a sharp increase in NSP referrals to craniofacial centers [8][9][10]. A noticeable asymmetric face is often considered less attractive, which can lead to psycho-social developmental consequences [11]. Severe NSP in the childhood years can lead to teasing, poor self-conception, and teacher bias [12]. Possible sequelae of NSP beyond the psycho-social concerns are being researched. A study evaluating neurologic profiles of infants 4 to 13 months old found significantly more altered tone—deflecting abnormally high and low tone—in infants with NSP compared to infants without [13]. A study comparing three-year-old children who had been operated on and not operated on for their NSP, found that 25% of the 12 children who had not been operated had severe receptive language skill problems. [14]. A study comparing the development of 36-month-old children found that the 224 children diagnosed with NSP at seven months scored lower on all of the Bayley Scales of Infant and Toddler Development than the 231 children without NSP at seven months. The largest differences were seen in cognition, language, and parent-reported adaptive behavior. Even children with at least mild NSP at 36 months, which had not previously been detected—control group children—had lower developmental scores than unaffected children. However,

these findings do not imply that NSP causes developmental problems [15]. In a follow-up study of 129 infants diagnosed in infancy with NSP, 11% had one or more delays in the parent-completed age-appropriate Ages and Stages Questionnaires at age 3–4, and 13% of parents reported concern [16]. A positive association between NSP and developmental delay was found in 13 of the 19 studies in a systematic review [17]. However, the severity of NSP cannot be used to predict the presence or degree of developmental delay according to findings of a prospective, nonrandomized study of 27 infants referred to a cranial facial clinic [18]. Thus, the relationship between NSP and early developmental delays remains poorly understood [19].

In a systematic review of 22 studies with a total population of 27,782 children, 60 risk factors for NSP were identified. 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 [20]. A prospective cohort study of 200 infants in the first two years of life found that three factors deter recovery: supine sleep position, limited head rotation, and lower infant activity level [6].

In 2008, a project was initiated in Skaraborg, Sweden in an attempt to prevent NSP. Sweden has a National Child Health Care Program, and the attendance rate was nearly 100% in 2005 [21]. The primary health care providers at the child health clinics are public health and/or pediatric nurse specialists. They are responsible for monitoring infants' growth and development and informing parents about the Swedish Board of Health and Welfare's recommendations, including recommendations on safe infant sleep positioning. Since nearly all infants in Sweden attend the child health clinics, these clinics provide an ideal venue for monitoring infant cranial shape and providing NSP prevention recommendations to parents.

The project commenced with a literature search on NSP prevention practices to develop evidence-based guidelines for the nurses [22]. The idea was to provide a working tool on NSP prevention for the busy nurses. The guidelines were tested in a pilot study [23] and revised. A continuing education on NSP, which included the revised guidelines, was developed for the nurses, and a clinical intervention was planned.

2. Discussion

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. Findings indicate that, while the intervention helped reverse NSP which developed in early infancy, it did not succeed in preventing NSP from developing. Examining what turned out to be successful or less successful in the project provides some useful insights for further prevention and reversal efforts.

Several strategies worked well. Motivation: Both nurses and parents were motivated to try to prevent NSP. Of the child health nurses employed at the time, 79% agreed to participate in the studies, and 93% of the participating nurses followed through. All parents followed through unless the family moved—278 of 284 parents (98%) followed through. 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.

Imparted knowledge to parents: Findings of the 4-month survey indicate that, while both groups of nurses worked to inform parents about NSP prevention, educating child health nurses about NSP did increase parents' awareness of recommendations. Intervention group parents reported significantly more recommendations from their nurse than control group nurses during the early months of infancy when parents' knowledge can influence infants' head shape. Findings of the 12-month qualitative inquiry indicate that intervention group nurses imparted both regular and new re-positioning strategies to parents, including how to accomplish occipital pressure relief when infants are awake, asleep, and being fed.

There are similarities between our 4-month survey and a qualitative nursing study from the UK [24]. That study was similarly conducted alongside an intervention study, and the nurses were also allocated to an intervention group that received an education or a control group which did not. Furthermore, these researchers also found that nurses in the intervention group actively applied their new knowledge and that nurses in the control group aimed for positive change by using their existing skills and experience.

Integrated new knowledge into practice: Intervention group parents' responses to the open-ended questions regarding infant care included details of NSP prevention that were introduced in the continuing education for nurses; intervention group parents who perceived severe cranial asymmetry at 3–4 months reported utilizing newly introduced positioning strategies.

Another indication of integration of new knowledge into practice is the intervention group's early reversal success of combination plagiocephaly/brachycephaly cases. This could be due to the continuing education for nurses including specific reversal recommendations while the national recommendations did not. Another reason could be that intervention group nurses learned how to assess cranial asymmetry, while control group nurses did not. A further indication is that intervention group nurses seemed to have integrated cranial asymmetry assessments in daily practice using the Severity Assessments because they provided assessments for 179 infants of the 184 intervention group infants at 2 months, and 180 at 4 months. However, we do not know if the nurses continue to make recommendations and do assessments now that the study is over.

Joint reversal efforts: Findings of the process-oriented approach in the qualitative inquiry indicate that nurses and parents collaborated in their attempts to reverse incipient NSP.

Decreased risk for persistent asymmetry: The risk for persistent asymmetry at 12 months was significantly lower for the intervention than the control group infants (RR = 0.35, [0.13; 0.94], $p = 0.03$) in the subgroup of infants who had NSP at two months. This indicates that intervention group nurses' and parents' collaboration was effective in decreasing infants' risk of having persistent asymmetry at 12 months, although the numbers were low, i.e., six intervention group and nine control group infants.

Assessing cranial asymmetry: The findings indicate the substantial strength of assessor agreement when assessors were trained how to assess cranial asymmetry using Severity Assessments and then tested. Additionally, assessors showed excellent ability to detect NSP in the clinical setting. This indicates that their assessments in the clinical intervention can be considered reliable. In a wider clinical context, results indicate that child health nurses can also be trained to assess NSP, which can be helpful for early detection.

However, not all strategies were as successful. Intervention group nurses' cranial asymmetry assessments did not always agree with assessors', although both were trained to assess cranial shape in the same way. A sensitivity analysis of data intervention group nurses provided from their 2-month cranial asymmetry assessments showed a 65% sensitivity in detecting NSP when using assessors' 2-month assessments as the gold standard. Intervention group nurses failed to detect 59% of cases detected by assessors. However, it is worth noting that intervention group nurses detected at least mild asymmetry in 31 of the 37 2-month assessor-detected cases, but mild asymmetry did not meet our rating system's criteria for NSP. Moreover, comparing nurses' assessments with those of the assessors is not completely fair because the assessors' only duty was assessing cranial asymmetry, just one small part of the nurses' job during a visit. Furthermore, the Severity Assessments are not precise tools.

Recommendations did not "get through" to all intervention group parents. Although all parents received the "This is Your Child's Health Book" from their nurse, which included the regular recommendations, only 68% of intervention group parents reported having received written recommendations from their nurse during their infants' first four months. Recommendations on infant positioning devices did not seem to 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. Parent-estimated minimum–maximum time spent in a bouncer daily was 0-480 min. 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, important safety aspects when providing an infant pillow. 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 even aware of the need for surveillance during tummy time, because few reported this safety aspect.

In a qualitative study examining parents' views of NSP prevention in Australia, researchers reported that some parents were more concerned about preventing NSP than SIDS because NSP was more real to them. Once NSP occurred, the majority of parents stopped following Australia's SIDS guidelines on safe infant sleep [25]. In contrast, we did not observe parent incompliance to SIDS guidelines in our study. Few parents in our study reported unsafe infant sleep positions; parents who reported placing their infants prone for sleep provided explanations which indicate *unawareness* of SIDS guidelines, not incompliance [26].

Prevention was difficult in both groups, especially brachycephaly, despite nurses' and parents' high motivation to participate and follow through, and intervention group parents' seemingly good knowledge about NSP prevention at four months. In the sensitivity analysis of the intervention group nurses' 2-month assessments using assessors as the gold standard, nurses failing to detect about three in five cases at two months is one possible explanation for early prevention failure. In the subgroup of infants who were non-cases at T1 and subsequently developed brachycephaly, overall brachycephaly prevention failure in the intervention group (25%) was ≥6 times more common than overall plagiocephaly prevention failure (4%).

An Italian cohort study including 283 infants, reported that an estimated 38% of infants had plagiocephaly at two to three months and 12% had combination plagiocephaly/brachycephaly [27]. This is in contrast to our findings where the proportion of infants with plagiocephaly at two months in the intervention and control groups were 13% and 14%, respectively, and the proportion with combination plagiocephaly/brachycephaly were 5% and 7%. Although the results of a cohort study conducted at 2 to 3 months should not be compared with an intervention study conducted at 2 months, our control group results at least give an indication that the Swedish child health program provides a good starting point for NSP prevention.

In a randomized controlled trial (RCT) evaluating early intervention, recommendations were provided directly to parents by a neonatologist in a 15-min private guidance session and in written form before discharge from the maternity unit; NSP was assessed using 2D and 3D craniofacial imaging [28]. In this RCT, the prevalence of NSP was 11% in the intervention group and 31% in the control group in a 2D analysis at *three* months. In our study, the prevalence of NSP was 23% in the intervention group and 32% in the control group at *four* months using the Severity Assessments. When comparing net results, this age difference is important to consider, since NSP peaks at about four months [6]. In their follow-up study, where all parents concerned about their infant's head shape received advice on repositioning regardless of previous group allocation, the head shapes of infants from three to 12 months were investigated. When sorted according to original group allocation, 13% of intervention group infants and 20% of control group infants in that study had NSP at 12 months [29], while 13% of intervention group and 16% of control group infants had NSP at 12 months in ours. Since the Severity Assessment is not nearly as accurate as 3D and 2D analyses, the results of these studies cannot be accurately compared. However, at least we seemed to do just as well when child health nurses provided parents with NSP prevention and reversal recommendations.

Early identification of head positional preference was missed in the continuing education and guidelines for nurses. Intervention group nurses were trained to evaluate the cervical range-of-motion in infants who were old enough to support their heads and were only instructed to ask parents about side preference. However, 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 early—i.e., with neonates lying supine [30]. Asking parents specifically about head positional preference could help in the early identification of risk for developing brachycephaly, which turned out to be difficult in both prevention and reversal

3. Clinical Implications

The main principle of NSP prevention—to relieve pressure on the infant's malleable occiput—is simple but important since newborns sleep a lot and lack muscle strength to change their own head position. Yet the supine sleep position which puts consistent pressure on an area of the infant's occiput is recommended as the safest infant sleep position, and no infant should ever come to harm from NSP prevention and reversal efforts. Thus, infants' vulnerability for NSP and the supine sleep position are both here to stay. Therefore, nurses need to intensify efforts to help parents understand the importance of reducing pressure on the occiput whenever infants are awake.

Interestingly, parent awareness of written recommendations from their nurse helped reverse NSP regardless of group, yet parents in the study seemed to remember receiving verbal information more than written information. Consequently, it seems as though both written and verbal information from their nurse are important for parents in NSP prevention efforts. Synthesizing these findings infers that nurses discussing recommendations when providing printed material could improve parents' recall and understanding. However, recommendations need to be tailored to parents understanding and the situation at hand, so good communication skills are important.

Preventing NSP is a continuing challenge for several reasons: supine sleeping young infants will always be vulnerable to NSP; the flow of information from a nurse education to the actual integration of the many small recommendations into daily infant care is long, so information can get lost during the process; communication is complex.

4. Conclusions

Assessors were considered reliable; educating nurses on NSP increased parental awareness of recommendations and promoted integration of newly introduced re-positioning recommendations in practice; the intervention was associated with early NSP reversal and reduced infants' risk that NSP at two months persisted at 12 months. However, prevention was difficult, especially brachycephaly prevention. More research on NSP prevention is needed.

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