Sudden infant death syndrome (SIDS) and Plagiocephaly: A Selected Annotated Bibliography

Hutchison L, Stewart A, Mitchell E.
**Infant sleep position, head shape concerns, and sleep positioning devices.**

AIM: The Back To Sleep campaign has successfully promoted the use of the supine sleep position for infants, with a corresponding decrease in sudden infant death syndrome death rates around the world. The aim of this study was to survey current infant sleep position practices, concerns about plagiocephaly, and the use of sleep positioning devices.

METHODS: A postal survey of 400 mothers of infants aged 6 weeks to 4 months was carried out in Auckland, New Zealand. RESULTS: Of the 278 (69.5%) respondents, the supine position was usually used in 64.8%, the prone position in 2.9%, with 32.3% using the side position or a combination of side and back positions. Approximately one-third had a concern about their infant's head shape, and 80% described practices to help prevent head deformation. Thirty per cent reported they had changed their infant's sleep position because of head shape concerns. A third of the mothers used some sort of positioning system to maintain the infant's sleep position. CONCLUSIONS: Anxieties about plagiocephaly, aspiration of vomit, and poor quality sleep are the main concerns that parents have about sleeping their infants on their backs. Further education is needed to inform mothers about these issues and to alleviate their fears.

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Plank LH, Giavedoni B, Lombardo JR, Geil MD, Reisner A.
**Comparison of infant head shape changes in Deformational Plagiocephaly following treatment with a cranial remodeling orthosis using a noninvasive laser shape digitizer.**

Deformational Plagiocephaly (DP) is a multi-planar deformity of the cranium occurring either pre-or postnatally in infants. In the last decade, the incidence of DP has increased substantially due to a number of factors, including supine sleeping positioning to reduce Sudden Infant Death Syndrome and the use of child carriers that increase supine positioning. Clinical questions persist about which children should be treated for DP and how to intervene, questions that are difficult to answer without accurate documentation of three-dimensional (3-D) head shape. This study explored a method for quantifying head shape and used that method to evaluate the success of orthotic treatment. Two hundred twenty-four infants who were diagnosed with DP received either a cranialremolding
orthosis or a repositioning program with no orthotic intervention. Data from 25 head shape variables were collected using a noninvasive laser shape digitizer. Only variables attributable to growth showed significant differences in the control population, while the treatment population showed significant differences in pre-and post-treatment values for all variables. The study identified four variables as particularly important in assessing the head shape of infants with plagiocephaly: the cranial vault asymmetry index, radial symmetry index, posterior symmetry ratio, and overall symmetry ratio. Ninety-six percent or more of subjects in the treatment group showed improvement in each variable. These data document the utility of a 3-D scanning device and the effectiveness of treatment with a cranial remolding orthosis.

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Positional plagiocephaly is a deformation resulting from intrauterine constraint or postnatal positioning leading to asymmetrical cranial growth. There has been a steady increase in referrals for positional plagiocephaly following the release of the American Academy of Pediatrics recommendation of supine infant sleeping position to prevent Sudden Infant Death Syndrome (SIDS) in 1992, largely because of poor parent education on the risks of prolonged occipital pressures. While this deformity is fairly easy to manage when diagnosed early, treatment can become more difficult and complicated with prolonged course. Because of this, it is essential that primary care physicians and parents be educated on recognition of positional plagiocephaly, prevention strategies, and treatment options. In milder cases, where diagnosis is made early, the deformation can be managed by stretching exercises and regular prone positioning; while in more severe cases molding helmets may be needed. Following appropriate treatment, success rates for acceptable cranial shape may be as high as 92%.


Infant sleep position impacts the development of head shape. Changes in infant sleep position, specifically the movement toward supine sleep, have led to a redefinition of normal head shape for infants in the United States. Historically, a dolichocephalic (elongated) head shape was the norm. Currently the norm has changed to a more brachycephalic (shorter and broader) shape. Since the American Academy of Pediatrics' Back to Sleep Campaign, the incidence of positional plagiocephaly has increased dramatically with a concurrent rise in the incidence of torticollis. Infants who require newborn intensive care, particularly premature infants, are more prone to positional...
plagiocephaly and dolichocephaly. Both can be prevented or minimized by proper positioning. The infant with an abnormal head shape requires careful evaluation; treatment varies according to the etiology. Craniosynostosis, a less common but pathological etiology for plagiocephaly, should be considered in the diagnostic process. Successful treatment of positional plagiocephaly and dolichocephaly includes systematic positioning changes to overcome the mechanical forces of repetitive positioning, physical and/or occupational therapy to treat underlying muscle or developmental challenges, and in some cases, molding helmet therapy.

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There has been a major decrease in the incidence of sudden infant death syndrome (SIDS) since the American Academy of Pediatrics (AAP) released its recommendation in 1992 that infants be placed down for sleep in a non prone position. Although the SIDS rate continues to fall, some of the recent decrease of the last several years may be a result of coding shifts to other causes of unexpected infant deaths. Since the AAP published its last statement on SIDS in 2000, several issues have become relevant, including the significant risk of side sleeping position; the AAP no longer recognizes side sleeping as a reasonable alternative to fully supine sleeping. The AAP also stresses the need to avoid redundant soft bedding and soft objects in the infant's sleeping environment, the hazards of adults sleeping with an infant in the same bed, the SIDS risk reduction associated with having infants sleep in the same room as adults and with using pacifiers at the time of sleep, the importance of educating secondary caregivers and neonatology practitioners on the importance of "back to sleep," and strategies to reduce the incidence of positional plagiocephaly associated with supine positioning. This statement reviews the evidence associated with these and other SIDS-related issues and proposes new recommendations for further reducing SIDS risk.

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The "Back to Sleep" campaign has dramatically decreased the incidence of sudden infant death syndrome; however, its sequelaes of deformational plagiocephaly have today reached epidemic proportions. In the last decade, we have learned to distinguish deformational plagiocephaly clinically from craniosynostosis, thereby preventing its unnecessary surgical correction. Primary care providers must increasingly be aware of
this condition and, in turn, educate new parents about its prevention. Should preventative measures fail and infants develop persistent sleep patterns that result in craniofacial deformities, deformational plagiocephaly can be treated successfully with behavior modification or cranial molding-helmet therapy.

Littlefield TR, Saba NM, Kelly KM.  
**On the current incidence of deformational plagiocephaly: An estimation based on prospective registration at a single center.**  

In 1992, the American Academy of Pediatrics (AAP) recommended supine sleeping to reduce the risk of sudden infant death syndrome. Although the incidence of deformational plagiocephaly is unknown, the consensus is that it has increased since this recommendation was made. To estimate the current incidence of plagiocephaly, we examined 342 infants for signs of deformational plagiocephaly, including occipital flattening, ear misalignment, frontal bossing, and facial asymmetry. Noticeable occipital flattening was documented in 15.2% of the infants (95% confidence interval, 11.6% to 19.5%); 1.46% had significant cranial deformities that also affected the skull base and face. Significant cranial asymmetry, defined as occipital flattening with concomitant skull base involvement and facial asymmetry, was observed in almost 1 in 68 infants. Adding to a growing body of evidence, our findings suggest significant increases in clinical deformational plagiocephaly since initiation of the AAP's "Back to Sleep" campaign.

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de Chalain T.  
**The Safe-T-Sleep device: Safety and efficacy in maintaining infant sleeping position.**  

Aims: The issue of infant sleeping position has socio-political ramifications. Current recommendations endorse supine sleeping as an aid to reducing the risk of sudden infant death syndrome (SIDS). Persistent sleeping of a newborn infant in the same position may induce plagiocephaly without synostosis (PWS). Parents in our craniofacial clinic, whose children present with PWS, often feel torn between apparently conflicting goals--avoiding SIDS and avoiding PWS. The Safe-T-Sleep device, a form of infant sleep wrap, purportedly allows safe semi-supine positioning, thus ameliorating PWS (by preventing the infant from lying on the cranial 'flat spot') while not increasing the risk of SIDS. Before recommending the device to parents in our plagiocephaly clinics, we designed a prospective, hospital-based trial to assess the safety and efficacy of the device in maintaining selected sleeping positions. This was not a trial of the efficacy of the Safe-T-Sleep device in treating plagiocephaly. Methods: The devices were trialed on 31 babies, between birth and 11 months of age. A total of 396 hours of observations were recorded. Results: The device maintained the selected body position in 94% of recorded observations and head position in 87%. There were no significant adverse events or complications associated with the use of the Safe-T-Sleep device. Conclusions: The device appears to be safe and effective. It is now being advocated in our clinic as an aid
to active counter-positioning strategies to passively correct incipient or established positional plagiocephaly in younger babies.


Hutchison BL, Thompson JM, Mitchell EA.
**Determinants of nonsynostotic plagiocephaly: A case-control study.**

Objective: There has been a large increase in reported cases of nonsynostotic plagiocephaly in infants since the adoption of supine sleeping recommendations to prevent sudden infant death syndrome. The objective of this study was to identify and quantify the determinants of nonsynostotic plagiocephaly in infants. Methods: One hundred infants who received a diagnosis of having nonsynostotic plagiocephaly were recruited as case patients and compared with 94 control subjects who were selected from a citywide database of infants. The infants all were aged between 2 and 12 months. Information concerning sociodemographic variables, obstetric factors, infant factors, and infant care practices was obtained by parental interview. Results: Case patients were significantly more likely to be male (adjusted odds ratio [aOR]: 2.51; 95% confidence interval [CI]: 1.23-5.16), to be a firstborn (aOR: 2.94; 95% CI: 1.46-5.96), and to have been premature (aOR: 3.26; 95% CI: 1.02-10.47). In the first 6 weeks, they were more likely to have been sleeping in the supine position (aOR: 7.02; 95% CI: 2.98-16.53), not to have had the head position varied when put down to sleep (aOR: 7.11; 95% CI: 2.75-18.37), and to have had <5 minutes a day of tummy time (OR: 2.26; 95% CI: 1.03-5.00). Mothers of case patients were more likely to perceive their infants as less active (aOR: 3.23; 95% CI: 1.38-7.56), to have a developmental delay (aOR: 3.32; 95% CI: 1.01-10.85), and to have had a definite preferred head orientation at 6 weeks (aOR: 37.46; 95% CI: 8.44-166.32). Case mothers were more likely to have no or low educational qualifications (aOR: 5.61; 95% CI: 2.02-15.56), although they were more likely to have attended antenatal classes (aOR: 6.61; 95% CI: 1.59-27.47). Conclusions: Early identification of a preferred head orientation, which may indicate the presence of neck muscle dysfunction, may help prevent the development or further development of nonsynostotic plagiocephaly in infants. Plagiocephaly might also be prevented by varying the head position when putting the very young infant down to sleep and by giving supervised tummy time when awake.

Full-text available at: [www.pediatrics.org](http://www.pediatrics.org) (not a U.S. Government site)

Persing J, James H, Swanson J, Kattwinkel J;
**Prevention and management of positional skull deformities in infants. American Academy of Pediatrics Committee on Practice and Ambulatory Medicine, Section on Plastic Surgery and Section on Neurological Surgery.**
Cranial asymmetry may be present at birth or may develop during the first few months of life. Over the past several years, pediatricians have seen an increase in the number of children with cranial asymmetry, particularly unilateral flattening of the occiput. This increase likely is attributable to parents following the American Academy of Pediatrics "Back to Sleep" positioning recommendations aimed at decreasing the risk of sudden infant death syndrome. Although associated with some risk of deformational plagiocephaly, healthy young infants should be placed down for sleep on their backs. This practice has been associated with a dramatic decrease in the incidence of sudden infant death syndrome. Pediatricians need to be able to properly diagnose skull deformities, educate parents on methods to proactively decrease the likelihood of the development of occipital flattening, initiate appropriate management, and make referrals when necessary. This report provides guidelines for the prevention, diagnosis, and management of positional skull deformity in an otherwise normal infant without evidence of associated anomalies, syndromes, or spinal disease.

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Biggs WS.
**Diagnosis and management of positional head deformity.**

In children with positional head deformity (posterior plagiocephaly), the occiput is flattened with corresponding facial asymmetry. The incidence of positional head deformity increased dramatically between 1992 and 1999, and now occurs in one of every 60 live births. One proposed cause of the increased incidence of positional head deformity is the initiative to place infants on their backs during sleep to prevent sudden infant death syndrome. With early detection and intervention, most positional head deformities can be treated conservatively with physical therapy or a head orthosis ("helmet").

Full-text available at: www.aafp.org/afp.xml (not a U.S. Government site)

Loveday BP, de Chalain TB.
**Active counterpositioning or orthotic device to treat positional plagiocephaly?**

Active counterpositioning and orthotic helmets are the two main nonsurgical management options for positional plagiocephaly. The purpose of this study was to compare these two management regimens. We included a random sample of infants referred between January 1, 1998 and October 31, 1999 to Middlemore Hospital and Auckland Surgical Center, for management of positional plagiocephaly. Two-dimensional head tracings were taken for each infant, every 3 to 12 months. From these tracings, we obtained Cranial Index and Cranial Vault Asymmetry Index. Seventy-nine infants were assessed during an average of 48.2 weeks. Five infants had normal head tracings, and were therefore excluded from the study. Of the 74 infants included in this study, 45 were managed with active counterpositioning, and 29 with orthotic helmets.
Average management time for active counterpositioning was 63.7 weeks, and 21.9 weeks for orthotic helmet treatment. For infants managed with active counterpositioning, the average change in Cranial Vault Asymmetry Index was 1.9%. In the orthotic group, average change in Cranial Vault Asymmetry Index was 1.8%. Orthotic helmets have an outcome comparable to that of active counterpositioning, although the management period is approximately three times shorter. Active counterpositioning generally had a slightly better outcome than orthotic management after the management period.

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Paulova M, Blaha P, Vignerova J, Riedlova J. 
Influence of positioning of infants on long-term changes of cephalic dimensions. 

The submitted investigation describes long-term changes of 3 main cephalic dimensions (head circumference, maximal length and maximal width of the head) and analyses the possible influence of positioning of infants after birth (prone, supine and side sleeping position) on these changes. Information about children aged 6 months to 3.99 years, where the need of up-to-date data is greatest, were collected as part of an extensive anthropological survey implemented in 1995 to 1997 in the entire Czech Republic. The authors confirmed the trend of brachycephalization, which is manifested by a statistically significant increase of the maximal length of the head and a statistically significant decrease of the maximal width of the head, as compared with children examined in the anthropological survey in 1956 to 1962 (1). These changes were established in the group of boys (200 boys) as well as in the group of girls (167 girls). The differences of the magnitude of long-term changes between boys and girls were not significant. Evaluation of the long-term changes of the head circumference in the entire group of 366 children aged 0.5-3.99 years (the head circumference of one girl was not measured) revealed a statistically significant increase of this dimension. During the period from 1956/62 till 1996, the influence of positioning on the magnitude of long-term changes of head circumference was not proved. We can say the same about the maximal length of the head of boys and girls and about the maximal width of the head of girls. Only between three differently positioned groups of boys (prone, side, supine) statistically significant differences in the magnitude of long-term changes of the maximal width of the head were found (p < 0.05). Highly significant changes of the maximum width and maximum length of the head occurred as compared with a reference group in all three groups of positioning of infants and in both sexes. The trend of brachycephalization seems to be thus a more potent factor, which affects long-term changes in the shape of the head, then the predominating sleeping position during the first months after birth. This conclusion is supported by the persisting trend of brachycephalization, although the supine position is now preferred.

An assistive device for the treatment of positional plagiocephaly. 
An assistive device (AD) was used to treat 122 infants with moderate-to-severe positional plagiocephaly. Clinical evaluations indicate that the AD provided the most benefit when applied to 2- to 8-month-old children. Our results suggest that the AD may join molding helmets and physical therapy as a treatment for moderate-to-severe positional plagiocephaly in infants under the age of 1 year.

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**Helmet versus nonhelmet treatment in nonsynostotic positional posterior plagiocephaly.**

A total of 105 infants with nonsynostotic posterior plagiocephaly were treated using a helmet or by head positioning. Effect of treatment was scored using a cosmetic outcome score (0-10 points) assigned by the parents. The onset of the observed skull deformity correction was not different for the helmet vs. non helmet treatment. Improvement was significantly better and faster in the helmet group compared with non helmet treatment (p < 0.01 and p < 0.001, respectively).

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Neufeld S, Birkett S.
**What to do about flat heads: preventing and treating positional occipital flattening.**

Across Canada there has been an increasing incidence of positional occipital flattening. This increase appears to be related to the recent change in infant sleep position to supine. In this paper, two patterns of positional occipital flattening, positional plagiocephaly and positional brachycephaly, are outlined. While there is no evidence of long-term developmental or neurological problems that result from positional occipital flattening, the infant's appearance can be distressing to parents who will then seek treatment. Prevention of positional occipital flattening requires a community approach with timely screening and early intervention should the infant's skull appear flat. Treatment involves repositioning the infant coupled with physiotherapy if there is neck muscle involvement. Should repositioning alone be ineffective, a helmet or headband program may be implemented. Neuroscience nurses can work in partnership with the community to ensure prevention strategies are implemented and timely interventions initiated.

Miller RI, Clarren SK.
**Long-term developmental outcomes in patients with deformational plagiocephaly.**

Objective: To determine whether there was an increased rate of later developmental delay in school-aged children who presented as infants with deformational plagiocephaly
without obvious signs of delay at the time of initial evaluation. Methods: A retrospective medical record review of 254 patients evaluated at the Craniofacial Center of the Children's Hospital and Regional Medical Center in Seattle, Washington, from 1980 through 1991 was completed. Consenting patient families were interviewed via telephone to determine what, if any, special medical or educational problems had occurred for the children who had had plagiocephaly in infancy or their siblings with normal head shapes. Results: A total of 181 families from the medical record review could be notified about the study and 63 families agreed to participate in a telephone interview. The sample of participants for the telephone interview was random to and representative of the group as a whole. The families reported that 25 of the 63 children (39.7%) with persistent deformational plagiocephaly had received special help in primary school including: special education assistance, physical therapy, occupational therapy, speech therapy generally through an Individual Education Plan. Only 7 of 91 siblings (7.7%), serving as controls, required similar services (chi\(^2\) = 21.24). Delays could not be specifically anticipated at the time of the diagnosis of deformational plagiocephaly from any simple set of factors including treatment with helmet therapy, although effected males with reported uterine constraint were at the highest risk for subsequent school problems. Conclusions: Infants with deformational plagiocephaly comprise a high-risk group for developmental difficulties presenting as subtle problems of cerebral dysfunction during the school-age years. There is a need for additional research on the long-term developmental problems in infants with deformational plagiocephaly. plagiocephaly, facial asymmetry, torticollis, developmental delay.

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