DEFORMATIONAL BRACHYCEPHALY IN SUPINE-SLEEPING INFANTS

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Objectives **Medical dictionaries and anthropologic sources define brachycephaly as a cranial index (CI = width divided by length × 100%) greater than 81%. We examine the impact of supine sleeping on CI and compare orthotic treatment with repositioning.**

Study design **We compared the effect of repositioning versus helmet therapy on CI in 193 infants referred for abnormal head shape.**

Results **Eighty percent of the infants had a pretreatment CI > 81%. Their initial mean CI at mean age 5.3 months was 89%, and after treatment, their mean CI was 87% (±2 SE = 0.9%) at mean age 9.0 months. For 92 infants with an initial CI at or above 90%, their initial mean CI of 96.1% was reduced to a mean of 91.9%.**

Conclusions **Post-treatment CI was 86% to 88%, CI in neonates delivered by cesarean section was 80%, and CI in supine-sleeping Asian children was 85% to 91%, versus 78% to 83% for prone-sleeping American children. Repositioning was less effective than cranial orthotic therapy in correcting severe brachycephaly. We recommend varying the head position when putting infants to sleep. *(J Pediatr 2005;146:253-7)***

Brachycephaly refers to a head, which is shortened in the anteroposterior dimension and wide between the biparietal eminences, with a cranial index (CI = width divided by length × 100%) > 81% (Figure 1).[1] Dolichocephaly is present when the CI is less than 76%, so the normal CI ranges from 76% to 81%. The most frequent cause of brachycephaly is constant supine positioning during infancy, which must be distinguished from bilateral coronal synostosis, which is comparatively rare. The increasing prevalence of both plagiocephaly and brachycephaly in recent years is a consequence of the success of efforts to prevent sudden infant death syndrome (SIDS).[2] The prone sleeping position has a protective effect for both positional preference and deformational brachycephaly.[3] In less than a decade, the normal head shape of the American infant has changed from normocephalic (CI = 76% to 81%) to brachycephalic, causing many families to consult their primary care providers for guidance in correcting unusual head shapes. In Asia, where infants have traditionally been positioned for sleep on their backs, brachycephaly is a normal head shape. We provide new normative data for current infant head shapes, present results from treating infants with excessive brachycephaly, and compare these head shapes with those in other populations.

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METHODS

Longitudinal data were collected on 193 normal infants with no malformations or neurologic problems who were referred and treated for abnormal head shapes at Cedars-Sinai Medical Center between 1997 and 2001. Institutional review board review was obtained for use of anonymized patient care data. Cranial measurements were made with anthropometric precision metal cranial calipers (B. Braun Medical Products, Aesculap Division, Tuttingen, Germany). Each measurement was repeated 3 times by one of two pediatric nurse practitioners and averaged. Comparisons between both practitioners measuring the same patient demonstrated no significant differences in measurement techniques.[2] The CI was compared before and after treatment, and we examined the

CI	Cranial index	SIDS	Sudden infant death syndrome

Table I. Initial and final means for age and CI in 193 infants by treatment group

	Reposition	Helmet	All
No. of infants	96	97	193
Initial mean age (mo)	4.6	6.0	5.3
Initial mean CI (confidence interval)	86.3% (84.8%–87.9%)	91.5% (89.8%–93.2%)	89.0% (87.8%–90.1%)
Final mean age (mo)	7.7	10.3	9.0
Final mean CI (confidence interval)	85.7% (84.3%–87.0%)	88.4% (87.1%–89.6%)	87.0% (86.1%–87.9%)
Reduction of brachycephaly (confidence interval)	0.7% (0.08%–1.4%)	3.2% (2.3%–4.0%)	1.9% (1.3%–2.5%)
P value for change in CI	.08	.0001	.0001

Figure 1. At 10.5 months, this male infant presented with a CI of 103% (*left and center frames*); after 5 months of orthotic helmet therapy, his CI was brought into the normal range (*right two frames*).

impact of treatment on positional brachycephaly. Practitioners instructed parents in how to perform neck physical therapy, if there was any positional preference, and repositioning was initiated at the onset of treatment. Statistical *t* tests, using the SAS software package, were used to compare the effect of repositioning versus helmet therapy. For those infants treated with cranial orthotic therapy, custom-designed plastic helmets were individually fitted by using methods previously described by Clarren in 1979 and 1981.[4,5]

RESULTS

We determined the mean and median post-treatment CI for the entire group of 193 infants who were treated with either repositioning or orthotic therapy during infancy (Table I). The initial mean CI at a mean age of 5.3 months for this entire group was 89%, and the mean final CI was reduced to 87% at a mean age of 9.0 months ($P < .0001$). Among a subgroup of 96 infants treated by repositioning from an average age of 4.6 months to 7.7 months, the mean final CI was 0.86. Among a subgroup of 97 infants treated with helmets from an average age of 6.0 months to 10.3 months, the mean final CI was 0.88. The change in CI for the subgroup of 96 infants treated with repositioning was not significant, whereas the change in CI with helmet therapy was significant (Table I). For infants treated with helmet therapy, treatment

Table II. Reductions in CI in helmet-treated infants by age group

Age group	N	Reduction in CI	*P* value
3–4.5 mo	16	5.1%	.0007
4.5–6.0 mo	41	3.2%	<.0001
>6 mo	38	2.9%	<.0001

at a younger age resulted in more improvement in the CI. Changes in CI with helmet therapy were significant at each age group, but results decreased in magnitude with advancing age (Table II). Within a subgroup of 92 infants with an initial CI at or above 90% (Table III), their initial mean CI of 96.1% at a mean age of 5.2 months was reduced to a mean of 91.9% at a mean age of 8.8 months.

By using "cephalic index" as a search term, we examined the medical and anthropologic literature to determine the CI for children from different cultures to examine how CI varied with different sleeping practices (Table IV).[6-18] Generalized linear model analysis comparing the mean CI among cultures with prone sleepers (80%), supine sleepers (84%), and newborn infants (80%) yielded a *P* value of .0033. Pairwise *t* tests showed that supine sleepers have a significantly higher mean CI than both prone sleepers and newborn infants, which were not significantly different from each other

DISCUSSION

In 1912, Franz Boas[17] suggested that exposure to the American environment resulted in significant differences in CI between same-age American and European children, though a recent re-analysis of Boas' data found these differences to not be significant.[18] The average CI for this population of 4677 children who slept prone was 79.8%, which is not different from the neonatal CI of 80.0%.[6] Brachycephaly refers to a head, which is short in the anteroposterior dimension and wide between the biparietal eminences, with a CI above 81%.[1] Comparison of CI between prone-sleeping cultures and supine-sleeping cultures (Table IV) indicates a significantly higher CI in supine-sleeping cultures.

The human infant cranium is highly malleable, and mechanical forces have been used by many cultures to deform

Table III. Initial and final means for age and CI in 92 brachycephalic infants (CI ≥ 90%)

	Reposition	Helmet	All
No. of infants	37	55	92
Initial mean age (mo)	4.5	5.7	5.2
Initial mean CI (confidence interval)	94.0% (92.8%–95.0%)	97.5% (96.1%–98.9%)	96.1% (95.1%–97.1%)
Final mean age (mo)	7.6	9.6	8.8
Final mean CI (confidence interval)	91.4% (90.0%–92.9%)	92.2% (91.2%–93.2%)	91.9% (91.1%–92.7%)
Reduction of brachycephaly (confidence interval)	2.5% (1.3%–3.8%)	5.3% (4.3%–6.3%)	4.2% (3.4%–5.0%)
P value for change in CI	.0003	$<.0001$	$<.0001$

Table IV. Cephalic index in different populations

Population Group	CI	Year (Reference)
Birth measurements		
Italy (108 term infants by CIs)	80.0%	1985 (6)
India (20 neonates)	80.6%	1972 (7)
Supine sleepers		
India (20 3-mo infants)	83.3%	1972 (8)
India (20 6-mo infants)	83.1%	1972 (8)
India (20 12-mo infants)	87.7%	1972 (8)
India (20 18-mo children)	83.4%	1972 (8)
India (20 24-mo children)	82.0%	1972 (8)
India (20 30-mo children)	83.9%	1972 (8)
India (20 36-mo children)	81.3%	1972 (8)
India (20 48-mo children)	81.4%	1972 (8)
Korea (430 7–10-y-old boys)	91.0%	1986 (9)
Korea (850 students)	84.8%	1986 (9)
Japan (Kyushu school girls)	87.0%	1986 (10)
Japan (62 boys >40 y)	82.1%	1992 (11)
Japan (62 girls >40 y)	83.3%	1992 (11)
Japan (Kyushu female adults)	81.0%	1962 (10)
Pakistan (757 Punjab adults)	90.4%	1978 (12)
Prone sleepers		
Nigeria (45 1-y-olds)	78.5%	1990 (13)
Nigeria (50 2-y-olds)	75.3%	1990 (13)
Nigeria (67 3-y-olds)	74.2%	1990 (13)
Nigeria (85 4-y-olds)	74.1%	1990 (13)
USA (98 5–11-y-olds)	81.0%	1952 (14)
Canada (age 4-y students)*	84.9%	1983 (15)
Canada (age 8-y students)*	83.7%	1983 (15)
Canada (age 12-y students)*	82.0%	1983 (15)
Canada (age 16-y students)*	81.2%	1983 (15)
Hawaii (475 white)	78.3%	1986 (16)
Hawaii (548 Chinese)	81.4%	1986 (16)
Hawaii (721 Filipino)	82.9%	1986 (16)
Hawaii (3881 Japanese)	81.6%	1986 (16)
European/USA (4677 adults)	80.0%	1910 (17)

*Eighty-five percent of Burlington, Ontario 3-year olds (Anglo-Saxon) followed longitudinally.

the shape of an infant's head, resulting in a wide variety of head shapes (Figure 2).[19,20] Positioning infants in hard wooden cradles created brachycephalic heads in some Middle Eastern cultures, due to inability of a swaddled infant to reposition itself, combined with constant supine positioning on a firm surface.[20] Thus, CI is heavily influenced by infant sleep position, and constant supine positioning is a frequent cause of deformational brachycephaly during infancy.

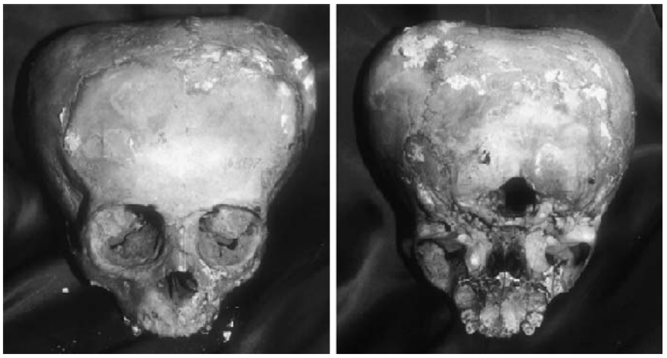

Figure 2. This Peruvian skull from the Field Museum in Chicago (collected in Carete Cerrodel Oro) was purposefully deformed into a brachycephalic shape using boards and binding. Highland Peruvians apparently applied circumferential bandages to the calvarium to achieve a conical head shape, whereas Coastal Peruvians applied boards to the occiput and pads to the frontal region, along with circumferential banding.[19]

The increasing prevalence of brachycephaly among American infants during recent years is a consequence of the success of efforts to prevent SIDS. The "Back to Sleep" campaign was initiated by the American Academy of Pediatrics in June of 1992 with the initial recommendation to place infants to sleep on their sides or backs to prevent SIDS. After 1996, the more stringent recommendation for only supine sleep positioning was made, when it was recognized that some side-sleeping infants were still dying from SIDS after assuming a prone sleeping position during the night.[21] The end result of this public education effort has been a dramatic decline in the prevalence of prone sleeping position from 70% in 1992 to 10.5% in 1997, with a concordant reduction in the rate of SIDS from 2.6 per 1000 in 1986 to 1.0 per 1000 in 1998.[22] When an infant remains in a persistently supine position without the developmental benefits of the head turning from side to side during consistent periods of tummy-time, the occiput becomes progressively flattened through the impact of gravity and persistent occipital mechanical pressure. A 1995 study of 7609 Dutch infants, who were screened for positional preference before age 6 months, revealed that 10% manifested occipital

Figure 3. This 7-month male infant (*left*) with right torticollis had a CI of 93% with a diagonal difference of 1.5 cm. After 5 months of helmet orthotic therapy (*right*), his CI was improved to 91% and his diagonal difference was reduced to 0.5 cm.

flattening. Of these infants, 45% showed persistent asymmetric occipital flattening at age 2 to 3 years.[3] An increase in prevalence of deformational posterior plagiocephaly has been documented,[2,3] and our culture is gradually learning how to prevent this unintended consequence.[23] The development of positional brachycephaly, with or without plagiocephaly, may indicate that parents are not providing their infants with tummy-time.

Cultures who put their infants to sleep in supine position are more brachycephalic CI than cultures who put their infants to sleep in prone position.[7-17] The CI was 80% for 108 term neonates delivered by cesarean section, which is similar to that of prone-sleeping cultures.[6] The CI during infancy in India (where infants sleep supine) is higher than later childhood.[15] Of note, child-rearing practices in India promote frequent tummy-time when the infant is awake and under observation. Prone-sleeping cultures have a normocephalic CI (mean CI = 80%, with a range from 76% to 81%), whereas school children in Japan and Korea (supine-sleeping cultures) are brachycephalic (85% to 91%).[8-10]

With the continued success of the "Back to Sleep Campaign," infants will have a more rounded head shape than cultures that previously put their infants to sleep on their stomachs. The current normative CI is 86% to 88%, and it is relatively rare to encounter a dolichocephalic infant in cultures whose infants sleep supine. Parents who become concerned about their infant's brachycephaly from prolonged supine positioning sometimes position their infants in a side-sleeping position, which is a dangerous sleep position for SIDS in an infant who has had little or no tummy-time. Infants who have no periods of consistent tummy-time become very distressed when placed in prone position, and their parents readily agree that their infants have never tolerated being on their tummies. The best management is to institute regular periods of tummy-time from early infancy while the infant is awake and under direct observation.

One prospective study from 1998 through 1999 randomized the treatment of 74 infants with positional plagiocephaly between repositioning (45 infants) and cranial orthotic therapy (29 infants).[29] Some infants were initially repositioned and then later put into helmets when they failed to respond; their subsequent responses to helmets were included among the helmeted group. Outcomes were similar in both groups, but repositioning took 3 times as long as helmet therapy, and the groups were too small for statistical analysis.[29] Cranial orthotic therapy has proven to be effective in correcting deformational posterior plagiocephaly,[2,24-29] and it has also been used to correct brachycephaly.[29] With extreme occipital flattening, a helmet can often be difficult to fit or may require prolonged treatment; thus, prevention is of paramount importance.[23] At the first sign of occipital flattening, repositioning and tummy-time should be promptly initiated to correct brachycephaly, and routine use of these practices during the first 6 weeks and thereafter should prevent this deformity. For those infants who do not make progress with repositioning and tummy-time, with severe persistent brachycephaly (CI > 90%) at age 5 months, use of an orthotic helmet will correct the brachycephaly (Figure 1) and any associated plagiocephaly (Figure 3). Reassurance is appropriate for those infants with CI < 90 at age 5 months, since brachycephaly is unlikely to develop or worsen after this age. Limitations to this study (and all previous studies of infant head deformation) include the lack of long-term observations into later childhood. There are also no natural history studies of positional brachycephaly in supine-sleeping infants, which extend beyond age 2 to 3 years. Anthropologic data (Table IV) were analyzed in the absence of such naturalistic observations.

It is important to distinguish deformational brachycephaly from craniosynostosis, since therapy and treatment are very different for each condition. Brachycephaly may also be associated with various syndromes that affect the pliability of the infant's skull or an infant's tendency to remain recumbent for prolonged periods of time. Thus, conditions associated with skull demineralization, such as osteogenesis imperfecta or hypophosphatasia, can lead to severe deformational brachycephaly because the cranium is demineralized. Conditions resulting in prolonged hypotonia, such as Down syndrome, will also result in brachycephaly, as will conditions associated with limited neck mobility, such as Klippel-Feil sequence. Infants with macrocephaly are at increased risk for positional head deformity, especially when the macrocephaly is accompanied by hypotonia. In all of these circumstances, variation of the infant's supine sleeping position from one side of the occiput to the other, along with tummy-time, can prevent the need for further corrective therapy.

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