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ORIGINAL PAPER

Cranial molding helmet therapy and establishment of practical criteria for management in Asian infant positional head deformity

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Abstract
Background The growing number of infants with deformational plagiocephaly (DP) has raised clinical questions about which children, at what age, and how molding helmet therapy (MHT) should be performed especially in Japan.
Methods A total of 1,011 Japanese pediatric head deformity infants had undergone MHT after being diagnosed with non-synostotic DP. Three ratios of left to right comparison (anterior, posterior, and overall) were created and analyzed comparing age of starting treatment, helmet wearing period, and severity of skull deformity before with after MHT.
Results The averages of head symmetry ratios after treatment in all groups (for the occipital region) showed apparent improvement; $t(930)=-60.86$, $p=0.000$. ($t(932)=-57.8$, $p=0.000$.) In the "severe" deformation group, the earlier the treatment was started, the higher symmetry ratio recovery was obtained. Treatment was especially effective when started in 4-month-old infants. In contrast to the "severe" group, the "mild" deformation group showed that MHT was most effective if treatment started before 6 months of age. Again, the earlier the treatment was started, the higher symmetry ratio was achieved, but compared to the "severe" group, it had a modest effect when treatment was started in infants older than 8 months.
Conclusion This is the first large-scale molding helmet study reporting the method and efficacy in Japanese infants. It demonstrated that despite the structural and physiological differences from infants of other races, molding helmet therapy is effective in Asian-born infants, provided that intervention timing and recognition conditions are met.

Keywords Positional plagiocephaly · Brachycephaly · Scaphocephaly · Cranial molding helmet therapy · Non-synostotic · Japanese infants

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Introduction

Deformational plagiocephaly (DP) is a multi-planar deformity of the cranium occurring either pre- or post-natally in infants and is one of the most frequently used terms among many others used for positional head deformity (PHD). This, as the many other "non-closed cranial suture" head deformity descriptions and reported entities resulting from prolonged intentional infant head positioning, if left untreated, may lead to significant cosmetic and functional–neurological and psychological consequences. For example, the pressure exerted on the intraorbital muscles and nerves, among others, can result in sensory and motor disturbances [1]. As a consequence, infants with head deformities attempt to compensate for the head's abnormal orientation in space which can result in ocular and vestibular impairment [2]. Given that the skull undergoes 85 % of its postnatal growth [3] within the first year of life, early recognition and treatment of PHD within this small window of opportunity is paramount [4]. However, relevant information on the cranial shape of Japanese infants in relation to definition, recognition, and treatment of cranial deformities has not been reported until now [5]. After the "back to sleep" campaign in the USA [2], the number of PHD cases worldwide increased, and for the first time in Japan, we have evaluated and treated 1,011 patients. Documentation of the effectiveness of deformity treatment requires feasible and reproducible methods to quantify head shape and any existing asymmetry. This study aimed to demonstrate that

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despite structural and physiological differences, molding helmet is effective in Asian-born babies, and this non-surgical intervention is successful in improving cases provided that timing and recognition conditions are met.

Materials and methods

Three-dimensional head shape capturing and quantifying was performed in a clinical setting using the STAR scanner (Orthomerica, Orlando, FL) laser data acquisition system [6]. The software programs provided with the STAR scanner were used to capture three-dimensional images, quantify and compare head shape changes, and then design and produce the individual cranial orthosis in all infants with positional head deformity.

The three-dimensional image was divided into 12 equal levels or cross sections (Fig. 1). Level 0 represented a cross section running through the sellion and right and left tragions. The software reconstructed 10 equal cross sections of the skull superior to level 0 and 2 inferior to it. The height of each cross section or level was determined by dividing the overall height of the child's head above the 0 plane into those 10 equal levels, and two levels with the same height were added below. The software used the image of these 12 levels to evaluate growth on each scan comparing cross sections over time and by that quantified the dynamics of growth. Each cross-section was divided into four quadrants (quadrant 1, anterior left; quadrant 2, anterior right; quadrant 3, posterior right; quadrant 4, posterior left), which was created by the software, defining a y-axis from the midpoint between the two tragion landmarks (on the x-axis) and sellion. Based on these x- and y-axes were built the two planes rising above the skull base and dividing the skull into four quadrant volumes. Q1, Q2, Q3, Q4 default volumes were based on level volumes from 2 to 8 added together (Fig. 2). This method allowed clinicians to assess symmetry at multiple cross sections and as a total for the quadrants of each group. Quadrant volumes would change relative to changes in growth, symmetry, and proportion. Quadrant volumes were also used to attempt defining quantitatively symmetry by specific ratios, as *anterior symmetry ratio* (ratio of Q1 volume to Q2 volume or vice versa, whichever is less than one), *posterior symmetry ratio* (the ratio of Q3 volume to Q4 volume or vice versa, whichever is less than one), and *overall symmetry ratio* (Fig. 3). This last parameter resulted from averaging the anterior (left and right) and posterior (left and right) volume compartments using specific proprietary software method of volume definition. The clinical significance of these parameters might be obvious for the anterior and posterior symmetry ratios, where the bossing area has bigger volume than the opposite side, and in the process of successful treatment and achieving symmetry, left and right volumes will come closer to each other (approaching 1.0). As it might be expected, the changes and corrections in these ratios will be more significant in plagiocephaly and less in brachi- and scaphocephaly. We have defined the goal of treatment at 0.9 or higher ratio values. The overall symmetry ratio as a global symmetry formula including all quadrants is less specific in expressing particular deformities and their corrections, and can indicate the anterior to posterior proportion in general, that is influenced by the prolonged face-up head position.

Molding helmet treatment method

In mild plagiocephaly, the helmet was built according to the initial diagram, aiming contact with the whole head surface except the quadrant of reduced volume, while in moderate and severe cases, the helmet contacted the two bossing areas. In mild to moderate brachycephaly, contact was maintained in the frontal and anterior parietal areas. Severe brachycephaly required similar attitude with special attention to the parietal area, where additional buildup should take place (Figs. 4 and 5).

Patient population

A total of 1,011 infants diagnosed by a pediatrician, neurosurgeon, or craniofacial plastic surgeon over a 4-year period (2007–2011) with moderate to severe craniofacial positional deformity were referred to Tokyo Women's Medical University (TWMU) for a cranial remolding orthosis. The male/female ratio was 721:290. Plagiocephaly was diagnosed in 964, brachycephaly in 44, and scaphocephaly in 3 patients. The age range was from 3 to 12 months. We excluded 80 patients lost to follow-up (74 with plagiocephaly, 5 with brachycephaly, and 1 with scaphocephaly) (Table 1).

All patients had molding helmet therapy for approximately 3–6 months. We divided the patients according to the severity of deformity into severe deformity group (SDG)—both overall symmetry ratio under 85.5 % and posterior symmetry ratio under 80.5 % and mild deformity group (MDG)—above these values.

The patients were divided into groups according to the age in months at presentation (3, 4, 5, 6, 7, 8, and 9-month-old groups), showing the distribution as in Table 2.

On the other hand, we analyzed the results according to the age when treatment for plagiocephaly was started, obtaining the following groups: at 4 months 184, at 5 months 198, at 6 months 140, at 7 months 109, at 8 months 88, and older than 8 months 125.

The 3 months of age group was excluded at statisticians' recommendation due to its lower number of patients and their distribution, compared to the other groups, which would have compromised the results of the analysis of variance.

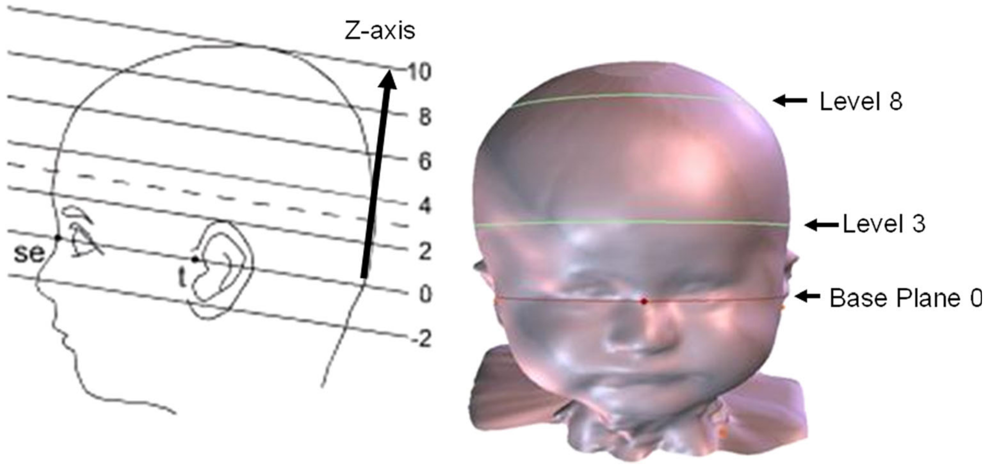

Fig. 1 Cross-section software divides the head into 12 cross-sectional levels and defines the 0 cutting plane by driving a plane through the sellion (se) and tragion landmarks. The height of each cross section or level is determined by dividing the overall height of the child's head above the 0 cutting plane into equal levels. Ten levels are above the 0 level, and two levels are below the 0 level. This provides a way to compare cross sections over time and addresses the dynamic of growth when comparing two scans. Level 3 is used as the default level for the summary report. *Z*-axis is perpendicular to the base plane

Analysis

We evaluated the overall and posterior symmetry ratios for significant improvement after treatment. We also evaluated the role of age when treatment was started relative to outcome. To compensate for the deformation ratio factor before treatment, considering the likelihood of a ceiling and floor effect in each age group, we performed analysis of variance of three factors (overall/posterior head deformity ratio before and after treatment, age in months when treatment started (4, 5, 6, 7, 8, or older), and degree of deformity (mild or severe) (Tables 3 and 4), applying a level of significance 0.01. We used the standard Windows software SPSS ver. 18.00 to perform the analysis.

Results

Treatment effect on overall symmetry ratio

(1) Paired sample *t* test results before and after treatment for the overall symmetry ration in all children.

Averages of overall symmetry ratio values for all participants, comparing before (M=86.3 %, SD=1.14) and after treatment (M=92.0, SD=1.71), showed a statistically significant difference—$t(930)=-60.86$, p=0.000 and indicated improvement after treatment.

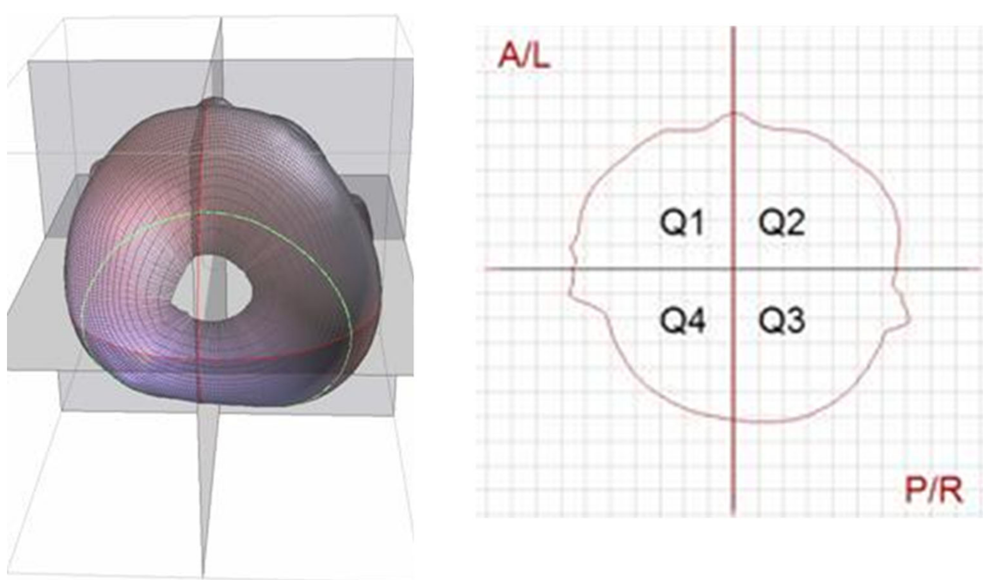

Fig. 2 The software creates the *y*-axis by finding the midpoint or origin between the two tragion landmarks and driving a plane through the sellion and origin. Q1, Q2, Q3, Q4 default volumes are based on levels 2–8 added together. Quadrant 1: Anterior left quadrant, Quadrant 2: Anterior right quadrant, Quadrant 3: Posterior right quadrant, Quadrant 4: Posterior left quadrant

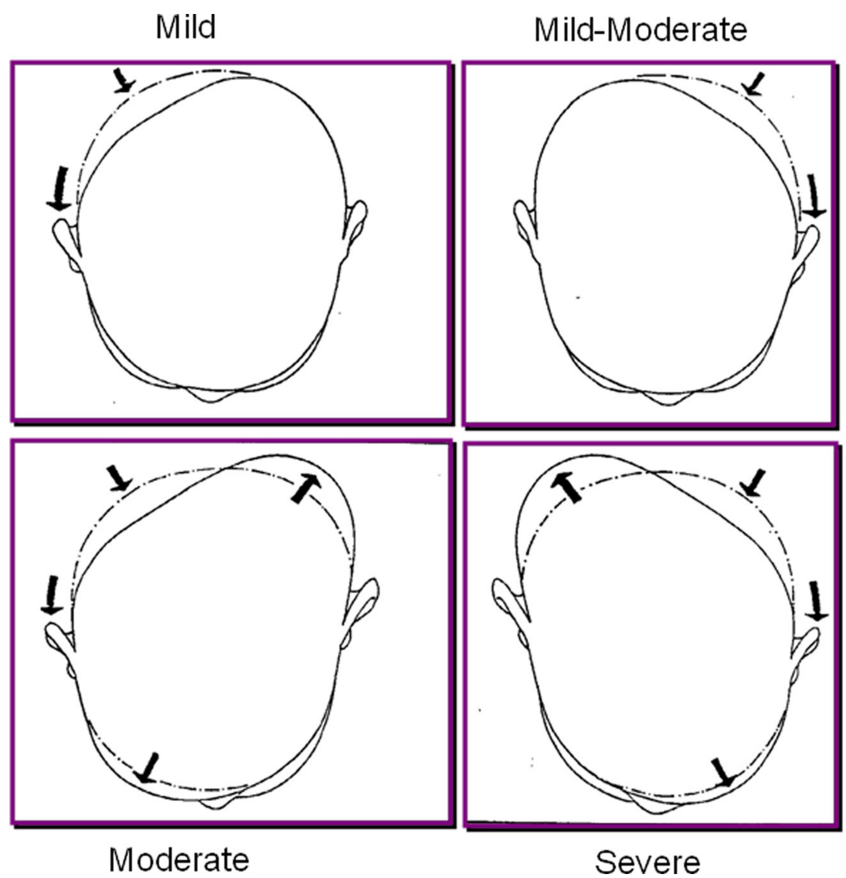

Fig. 3 Asymmetry grading. Mild—one posterior quadrant involvement is flat with minimal ear shift. Mild-moderate—one posterior quadrant is flat with moderate ear shift on same side as posterior flattening. Moderate—one posterior quadrant, and contralateral anterior quadrant is involved with moderate ear shift. Facial asymmetry may be present. Two quadrant involvement and ear shift. Severe—All four quadrants are either flat or bossed with severe ear shift, forehead, and orbital asymmetry, and asymmetry of the cheek and jaw

(2) Analysis of variance (ANOVA) for three variables of the overall symmetry ratio in plagiocephaly children.

We analyzed symmetry ratios before and after treatment regarding the following variables: age in months when treatment started (4, 5, 6, 7, 8, 9, or older) and degree of deformity (mild or severe), and the results are shown in Table 3.

Comparing pre- and post-treatment, overall symmetry ratio results in these children showed statistically significant difference ($F[1, 832]=4743.60$, $p<0.0001$), indicating a higher

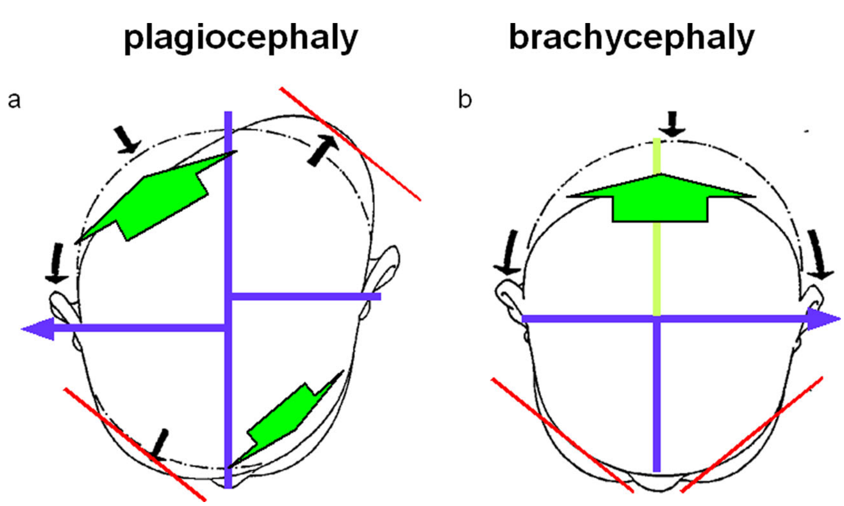

Fig. 4 The mechanism of the Helmet therapy which improves the head shape of babies with deformational plagiocephaly and brachycephaly. (**a**) The flattened areas are built up with plaster in the posterior-lateral quadrant to obtain symmetry. The flattened frontal area is also built up with plaster to obtain symmetry. Contact will be maintained over the prominent or bossed areas to deter growth in those areas. (**b**) Primary buildup on the positive mold occurs across the central occipital region to obtain improved proportions of the head. Contact is maintained over the frontal and parietal regions to deter growth

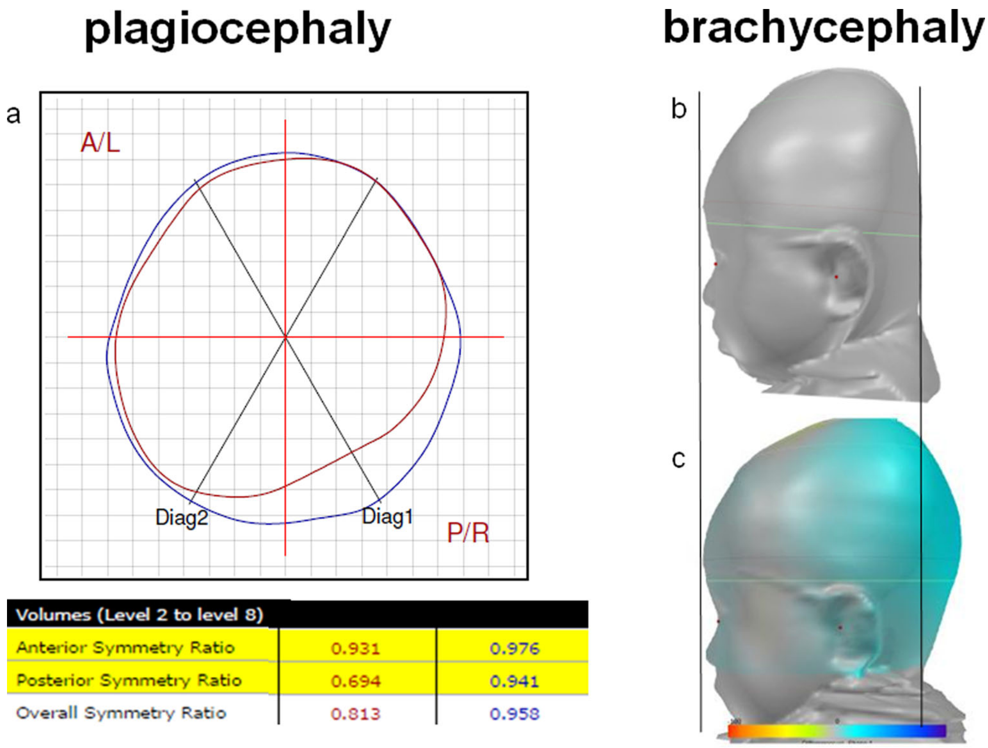

Fig. 5 Analyzing measurement sheet of plagiocephaly pre- (*red shape*) and post-treatment (*blue shape*) head shape. This provides a way to compare cross sections over time and addresses the dynamics of growth when comparing the two scans. (**a**) Three-dimensional skull view pre- (**b**) and post-molding (**c**) helmet therapy for brachycephaly. The part of *blue color* shows the part of remodeling

symmetry ratio after treatment. The role of age of initiating treatment ($F[5, 832]=21.76$, $p<0.0001$) and degree of deformity ($F[1, 832]=943.88$, $p<0.0001$) were also significant. The three-way interaction was also significant ($F[5, 832]=4.01$, $p=0.001$) While the effect of age when initiating treatment was not significant before treatment for both severity groups (severe—$F[5, 832]=1.584$, $p=0.162$; mild—$F[5,832]=2.97$, $p=0.011$), it was after treatment for both of them (severe—$F[5, 832]=29.25$, $p<0.0001$; mild—$F[5,832]=27.74$, $p<0.0001$).

The application of the multiple comparison of Bonferroni method to post-treatment data showed that for severe deformity group, symmetry ratio of 4-month olds was significantly higher than that of other age groups (4–5, 4–6, 4–7, 4–8, 4–older than 8—MSe=10.53, $p<0.01$ for each), symmetry ratio of 5-month-olds was also significantly higher than that of 7-month-olds and those older than 8 months (5–7, 5–older than 8—MSe=10.53, $p<0.01$ for each), and symmetry ratio of 6-month-olds was significantly higher than that of older than 8 months group (6–older than 8 month—MSe=10.53, $p<0.01$). These results indicated that the earlier the treatment started, the higher the symmetry ratio rises in the severe deformity group, and especially most effective when started in 4-month-olds (Fig. 6a).

The same method was applied to the post-treatment data and showed that for the mild deformation group, symmetry ratio of 4-month-olds was significantly higher than that of 7-month-olds, 8-month-olds, and older than 8 months (4–7, 4–8, 4–older than 8—MSe=10.53, $p<0.01$ for each), symmetry ratio of 5-month-olds was also significantly higher than that of 7-month-olds, 8-month-olds, and those older than 8 months (5–7, 5–8, 5–older than 8—MSe=10.53, $p<0.01$ for each), symmetry ratio of 6-month-olds was also significantly higher than that of 8-month-olds and older than 8 months (6–8, 6–older than 8—MSe=10.53, $p<0.01$ for each), and symmetry ratio of 7-month-olds was significantly higher than that of the group older than 8 months (7–older than 8 months—MSe=10.53, $p<0.01$). Again, these results indicate that the earlier the treatment is started, the higher the symmetry ratio rises in both deformity group, but in the mild deformity group, it was most effective if started even before the age of 6 months (Fig. 6b).

Table 1 Excluded cases in each skull deformity type

Exclusion		
	Whole head	Occipital region
Plagiocephaly	74	72
Branchycephaly	5	5
Scaphocephaly	1	1
Total	80	78

Treatment effect on occipital symmetry

(3) Evaluation of results before and after treatment in the occipital region of all children.

Table 2 Age grouping by sex and case attributes in the study

Age (month)	Number	Sex (male/female)	Plagiocephaly	Brachycephaly	Scaphocephaly
3	50	(34:16)	48	2	0
4	205	(145:60)	196	9	0
5	223	(167:56)	212	10	1
6	159	(116:43)	151	8	0
7	127	(81:46)	120	7	0
8	99	(70:29)	95	3	1
9~	148	(108:40)	142	5	1
Total	1,011	(721:290)	964	44	3

Paired sample *t* test

The comparison of the posterior symmetry ratio before and after treatment for all participants resulted in a significant difference in the averages of this ratio before (M=81.8 %, SD=1.29) and after treatment (M=91.3, SD=2.72); $t(932)=-57.8$, p=0.000. These results indicated that after treatment, the occipital region in all participants showed apparent improvement.

(4) Analysis of variance (ANOVA) for three variables in the occipital region of plagiocephaly children.

Symmetry ratio data before and after treatment were analyzed by a three-way analysis of variance (ANOVA) with the age of starting treatment (4, 5, 6, 7, 8, or older) and degree of deformity (mild or severe) in the posterior quadrants in children with plagiocephaly. The results of pre-post treatment are shown in Table 4.

The change of the posterior symmetry ratio compared before and after treatment was higher after treatment ($F[1, 833]=3716.8$, $p<0.0001$), being significant. The effect of age when initiated treatment ($F[5, 833]=20.2$, $p<0.0001$), the deformation severity ($F[1, 833]=251793.8$, $p<0.0001$), and the three-way interaction were also significant ($F[5, 833]=3.18$, p=0.008). As in the case of the overall symmetry ratio, the effect of age was not significant before treatment for both severity groups (severe—$F[5, 833]=1.81$, p=0.108; mild—$F[5, 833]=1.08$, p=0.369); however, it was significant after it (severe—$F[5, 833]=25.5$, $p<0.0001$; mild—$F[5, 833]=20.8$, $p<0.0001$).

The multiple comparison of Bonferroni for post-treatment data showed that in the severe deformation group, symmetry ratio of 4-month-olds was significantly higher than that of 7, 8, and older than 8 month of age (4–7, 4–8, 4–older than 8—MSe=28.63, $p<0.01$ for each), symmetry ratio of 5-month-olds was also significantly higher than that of 7, 8, and older than 8-month-olds (5–7, 5–8, 5–older than 8—MSe=28.63, $p<0.01$ for each), and symmetry ratio of 6-month-olds was significantly higher than that of 7, 8, and older than the 8-month-old group (6–7, 6–8, 6–older than 8 months—MSe=28.63, $p<0.01$). These results indicated that the earlier the treatment started, the higher the symmetry ratio rises in the severe deformation group, and it is most effective if started before the age of 6 months (Fig. 6c).

The use of the same multiple comparison of Bonferroni method for post-treatment data also showed that in the mild deformity group, posterior symmetry ratio of 4-month-olds was significantly higher than that of 7, 8, and older than

Table 3 Mean value (according to the initial severity) of the overall symmetry ratio after helmet therapy in each starting age

Overall ratio

	Mild group		Severe group	
Age(month)	Pre-M (SD)	Post-M (SD)	Pre-M (SD)	Post-M (SD)
4	89.2 (±2.7)	95.5 (±2.4)	81.9 (±2.9)	92.6 (±2.3)
5	88.9 (±2.7)	95.0 (±2.8)	81.9 (±2.5)	90.9 (±2.0)
6	89.7 (±2.5)	94.6 (±2.8)	81.9 (±3.0)	90.2 (±2.3)
7	89.1 (±3.1)	93.6 (±2.8)	81.7 (±2.7)	88.2 (±2.3)
8	88.5 (±1.7)	92.8 (±2.5)	83.3 (±2.2)	89.0 (±1.8)
9~	88.6 (±2.2)	92.1 (±2.4)	82.5 (±2.2)	87.3 (±2.4)

M molding

Table 4 Mean value (according to the initial severity) of right and left occipital heteromorphic rates and Posterior symmetry Ratios pre and post helmet therapy by starting age

Occipital heteromorphic rates

	Mild group		Severe group	
Age(month)	Pre-M (SD)	Post-M (SD)	Pre-M (SD)	Post-M (SD)
4	87.5 (±5.0)	96.7 (±3.3)	73.6 (±5.0)	91.3 (±2.7)
5	87.0 (±5.1)	96.6 (±3.4)	73.4 (±4.3)	89.2 (±2.5)
6	87.5 (±5.0)	95.7 (±4.3)	75.0 (±4.3)	89.6 (±3.1)
7	87.2 (±6.1)	94.7 (±4.7)	73.9 (±4.2)	85.5 (±3.5)
8	86.9 (±4.8)	93.8 (±5.1)	74.7 (±4.7)	85.3 (±3.8)
9~	86.4 (±5.0)	91.9 (±5.2)	73.8 (±4.1)	82.6 (±3.9)

M molding

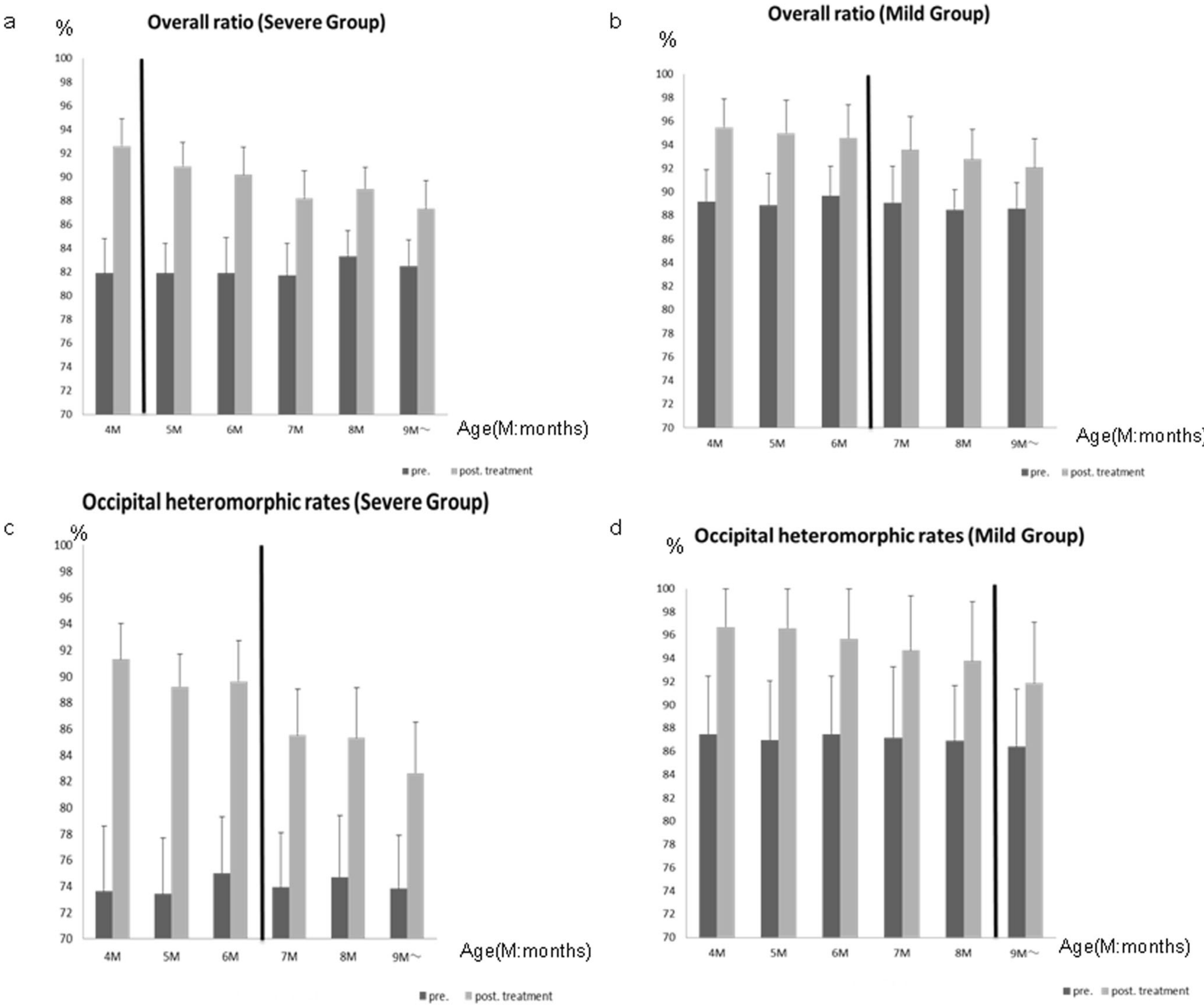

Fig. 6 (**a–d**). **a** In the severe deformity group, the overall deformity ratio improved with a statistically significant difference comparing before and after therapy. Mean values after treatment indicated strong corrective effect for children with plagiocephaly, being the highest from multiple comparisons after the treatment for those started at 4 months of age. **b** In plagiocephaly cases, the overall symmetry ratio improved with a statistically significant difference comparing before with after therapy in the mild deformity group. From the multiple comparisons after treatment, it was shown that the earlier the age of starting therapy, the higher the mean of symmetry ratio; however, maximal correction effect was obtained if treatment started by the 6 months after birth. **c** Posterior symmetry ratio improved with a statistically significant difference comparing before and after therapy for the severe deformity plagiocephaly group. If molding helmet therapy could be started before 6 months of age, its efficacy was significantly higher. **d** Posterior symmetry ratio improved with a statistically significant difference comparing before and after therapy in the mild deformity plagiocephaly group. Multiple comparison analysis after the treatment indicated that the overall symmetry ratio after the helmet therapy was higher in the group where the start of therapy was early. It was shown in particular that the correction effect significantly lowered when treatment started after the age of 9 months

8 months of age (4–7, 4–8, 4–older than 8—MSe=28.63, $p<0.01$ for each), symmetry ratio of 5-month-olds was also significantly higher than that of 7, 8, and older than 8 months of age (5–7, 5–8, 5–older than 8—MSe=28.63, $p<0.01$ for each), and symmetry ratio of 6, 7, and 8-month-olds were also significantly higher than that of older than 8 months (6–older than 8, 7–older than 8, 8–older than 8—MSe=28.63, $p<0.01$). Again, the meaning of these results is that the earlier the treatment started, the higher the symmetry ratio rises in both deformity groups, but the effect of helmet treatment fell markedly in older than 8 months of the mild deformity group (Fig. 6d).

Discussion

"Positional" plagiocephaly, among the other PHDs, is the most common condition and is defined by specific pathophysiology and biomechanical factors and not determined only by deformity description as a result of prolonged supine head position. It has specific management related to its pathophysiology and biomechanics, and its incidence demands the establishment of principles and goals of treatment. As the child starts free head movement in the early months of life, particularly after 4 months of age (when able to roll), the application of "molding" forces on the cranium gradually becomes impossible, and treatment might become difficult [7], [8]. Therefore, an oversimplified concept of being a temporary cosmetic effect on the head shape because of prolonged head positioning is not acceptable. Analysis of the factors at the background of this condition and those related to the effective treatment can bring proper solution to this problem. [9]

Predisposition to positional plagiocephaly

Many factors—both *prenatal* and *postnatal*—have been related to the development of positional plagiocephaly. It is increased in firstborns, twins, and other multiples, even when cesarean delivery is performed. This was demonstrated to be the more frequent scaphocephalic head shape in these infants. That is also more often seen in breech positions and prolonged vaginal deliveries, exerting overload to the otherwise natural head deformability in newborn. If a difficult labor, particularly after forceps or vacuum extraction delivery, leads to birth trauma, the resulting motor deficit can create conditions for poor or restricted head movements and increase the positional inconveniences. Premature born infants have also higher deformity rates for the same reason.

In general, the supine sleeping position exacerbates deformation process; however, positional plagiocephaly is only one component of this deformation process. Often, other medical factors accompany this deformity. Factors accompanying direct deformation include facial deformation, mandibular asymmetry, congenital and/or acquired muscular torticollis, abnormal eye placement, external ear deformity and misalignment, orbital asymmetry resulting in strabismus and other ocular problems, and epicanthal fold on the side of flatness. [10] The indirect factors (often diagnosed in infants with positional plagiocephaly) are positional foot deformities, developmental hip dysplasia, middle ear infections, and migraine headaches [5, 11].

Cosmetic vs. reconstructive procedures

In plagiocephaly, the deformation of one element leads to compensatory deformation and displacement of all other connected elements of the system, and as a result, other cranial asymmetries are commonly identified as abnormal cranial height, abnormal cranial width/breadth, and occipital flattening with ipsilateral forehead bossing, significant ear misalignment, abnormal alignment and asymmetry of the orbits, among others. This compensation for the head's abnormal orientation in space results in ocular and vestibular impairment and distortion of the orbits with pressure on the extraocular muscles and nerves, resulting in sensorimotor disturbances [11].

A cranial remolding orthosis has a direct effect on frontal, parietal, sphenoid, temporal, and one of the occipital bones of the neurocranium. Indirectly, it affects the entire facial alignment (i.e., viscerocranium) due to the direct transfer of forces through the neurocranial structures. Throughout the orthotic treatment program, measurable changes in the cranial base, cranial vault, orbitotragial depth, and cephalic index have been documented. By returning the cranial and facial bones to a normal alignment, long-term dysfunction to hearing, vision, and mandibular mechanics could likely be avoided [10] together with other medical conditions and development difficulties in cognition.

Criteria for management

The presence of anthropometric data was verified for moderate to severe plagiocephaly [7]. Several studies have already pointed out on the maximum efficacy of orthotic treatment during the window of rapid head growth and its proportional decrease with the increase of cranial rigidity concomitant with age. The pace of re-formation relates to the rate of brain growth, which is much more rapid during the first 6 months than later in infancy. In a prospective study of 114 infants, those managed with helmet (51 infants) did significantly better than those with only head positioning in the crib (63 infants) Therefore, timing, recognition/grading, and early treatment were the main criteria for success. Our results indicate that the same tendencies of effective "time window" combined with importance of proper attitude of the medical community exist with Japanese infants. Clarren et al. documented the safety and efficacy of cranial remolding orthosis for positional plagiocephaly. Rekate et al. reported that cranial remolding orthosis should be prescribed for all infants with positional plagiocephaly when asymmetry persists after repositioning attempts and also prior to the consideration of surgical intervention for infants less than 12 months of age [12]. Littlefield et al. reported that significant correction of cranial deformation was achieved through the use of cranial remolding orthosis and was maintained after the discontinuation of the orthotic treatment program. Kelly et al. also identified the need for early intervention. Statistically significant increases in cranial growth were associated with concomitant reductions of the cranial asymmetries in deformational plagiocephaly. Joganic et al. documented the use of cranial remolding orthosis to facilitate post-surgical outcomes. Graham found that early intervention relates to both the length and success of the orthotic treatment program. All these studies are very similar to our findings, indicating that Asian infants respond in a similar way to the molding helmet therapy. But definite Asian infants' skull bone deformity data has not been available until now [5].

Developmental positional plagiocephaly

A common concept on positional plagiocephaly is that the infant's head "will round out on its own as the child becomes more active, begins to roll over, and learns to sit up." This misconception is based in part on outdated scales of motor development and a lack of understanding on the effect of supine sleep positioning. The pattern of early motor development is affected by sleep position. On average, supine sleepers attain common motor milestones later than prone sleepers [13]. Prior to 1992, infants' heads often corrected in the first few months of life because infants that were placed prone to sleep were generally in a variety of positions during the day, thus avoiding prolonged time in one position. Now that supine is the position of choice and there is a 4–6-week delay in the acquisition of head and trunk control, infants' heads often do not "round out" as they did previously. As noted previously, torticollis may be the cause or effect of positional plagiocephaly. Binder et al. (1987) studied the long-term effects of torticollis and found significant "persistent functional asymmetry of the involved body side despite mild or moderate severity, early diagnosis, and complete resolution of the torticollis," leading to long-term complications [14].

Treatment regimens

Optimal beginning for positional helmet therapy was reported to be at the age of 5 to 6 months in the European countries [1]. The evidence obtained from Japanese infant clinical data shows clearly decreased efficacy of molding helmet therapy just after the age of 6 months [15]. The weight of the helmet, particularly that made in the USA, created a problem with the still unstable neck at an earlier age and could not be applied in our studies. A lighter helmet could bring down the treatment initiation age. [16] A further improvement might be to design different helmets, proportional to the physical characteristics of Caucasian, African American, Asian, and so on infants. The different groups may have also different starting treatment age.

Current situation in Japan and the international community

Currently, there is no study estimating the prevalence of positional skull deformity in infants in Japan as compared with American and European countries. [17, 18] At the same time, the interest of the Japanese medical community on positional plagiocephaly, particularly the pediatricians on general practice, has been very low and not based on any guidelines, leaving the problem without established criteria between pediatric plastic surgeons and pediatric neurosurgeons [9]. That often leads to the recommendation of only observation on a huge number of infants with deformities. As a result, we currently do not have any data on patients suitable for comparison or control group.

van Wijk et al. reported a recent randomized trial on helmet therapy which showed that no effect of helmet therapy can be shown in infants with moderate to severe positional skull deformation [19]. Regarding our cases, even though skull deformation was moderate to severe, the earlier the treatment was started, the higher symmetry ratio recovery was obtained. Treatment was especially effective when started in 4-month-old infants. In contrast to the "severe" group, the "mild" deformation group showed that MHT was most effective if treatment started before 6 months of age. Again, the earlier the treatment was started, the higher symmetry ratio was achieved, but compared to the "severe" group, it had a modest effect when treatment was started in infants older than 8 months.

In our study, treatment was started since the initial diagnosis of each case as soon as possible and in a significant portion of patients before the age of 5 months. As a result, the time of consultation (diagnosis establishment) and start of therapy were correlating.

The difference of results between our study and the HEADS could be related to the younger infant age at diagnosis. For those reasons, earlier diagnosis and start of treatment was related to different quantitative expression of the deformity because of the early age, a major degree of parents' apprehension and disapproval regarding their child's head shape and all that leading to early entry into the study.

Last, but not least, the patients population is different, as considering Asian patients, compared to European ones. Some cultural influences, as head shape perception and parents' considerations of importance if minor deformities continue to exist, might have influenced results. Another point of difference with the HEADS study is the rate of complications of the helmet group, which are in very significant proportions. Being a "pragmatic study," that factor might have influenced the compliance, which has not been recorded in details, something that a future study should address.

On correcting the cranial deformity of infants, pediatricians should be aware of the critical time period within which the initiation of orthotic treatment is able to correct effectively the cranial deformity as our data indicate. In a case of severe skull deformity when we have to undertake immediately corrective orthotic management, consenting with the parents can be extremely important. In addition, awareness of positional deformities and their management should be extended to the obstetricians, pediatricians, midwives, and nursing staff [18].

Mental distress

Skull deformities are well known for inducing inferiority complex in childhood [20]. The parents of more than 80 %

of our treated skull deformity infants had some degree of inferiority complex because of skull deformity from which both their fathers and mothers had difficulties in wearing glasses and auricular asymmetries, hair style problems, temporo-mandibular joint asymmetries, and teeth alignment problems, even occasionally have been unable to wear motorbike helmets. Their own experience was a motivating factor for the management of their child.

Future grading system

While similar reports exist for American and European infant populations, this is the first one able to demonstrate efficacy of the helmet orthotic treatment for Japanese infantile skull deformity. However, some problems in the evaluation methodology remain. Plagiocephaly can be diagnosed and graded based on the existing asymmetry or as we have applied the symmetry ratios. Brachycephaly diagnosis and grading however remain more difficult as different criteria using the ratio of occipito-frontal length and skull breadth or the overall symmetry ratio averaging anterior and posterior quadrant with the purpose of comparison have been applied. The existing differences in measuring methodology might have influenced interpretations among different institutions [21–23]. The accumulation of Japanese infant skull data form a database using a single standard method and creating reference values became an urgent task [2, 24]. While at the moment only height, weight, and head circumference are routinely recorded in Japan for infant development evaluation, considering American and European sources, we are intending to promote that the index of the Japanese infantile skull deformity rate be routinely included in the future [25, 26].

Conclusion

We introduced a Japanese infant positional skull deformity evaluation system and described the efficacy of helmet therapy for its treatment, discussing the critical periods for intervention. Its application showed that similar to European and American PHD infants' management, it is an effective treatment according to specifically established criteria. Analysis of results showed the optimal time and deformity criteria values for obtaining adequate treatment. The application of this approach traces the possibility of a wider application of deformity evaluation and management for the whole Japanese infant population. In conjunction with data from other geographical regions, it may provide more precise guidelines for countries with existing racial diversity in their infant populations.

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Hitoshi Dairoku Yoshikazu Okada—Mr Dairoku designed the data collection instruments and coordinated and supervised data collection. Dr. Okada critically reviewed the manuscript.

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