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Three-dimensional analysis of cranial and facial asymmetry after helmet therapy for positional plagiocephaly

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Abstract

Purpose Helmet therapy is a non-surgical option for treating positional plagiocephaly, and its effectiveness has been validated by various researches. In addition to cranial flattening and asymmetry, ipsilateral prominence of the mid-face and relative anterior transposition of the ipsilateral ear is also common. Hence, we investigated the impact of helmet therapy on mid-facial asymmetry.

Methods Ninety-nine patients diagnosed with positional plagiocephaly and treated by helmet therapy between September 2005 and July 2012 were enrolled. Therapy was initiated at various ages: group I, <6 months (n=35); group II, 6 months to 1 year (n=43); group III, >1 year (n=21). A cranial vault asymmetry index was measured at the levels of the inferior orbital rim (CVAI_{IOR-MF}) and superior orbital rim (CVAI_{SOR-LC}) and midway from the superior orbital rim to the vertex (CVAI_{MID-UC}). Anterior transposition of the ipsilateral ear was verified by measuring the distance (D_{EAR}) between the actual position of the ear and its expected position relative to the contralateral ear. All variables were compared before and after helmet therapy and were categorized by age at treatment initiation.

Results $\text{CVAI}_{\text{IOR-MF}}$ and $\text{CVAI}_{\text{SOR-LC}}$ were lower in all three age groups after helmet therapy, confirming therapeutic efficacy. $\text{CVAI}_{\text{MID-UC}}$ (upper level cranial asymmetry) and D_{EAR}

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(mid-facial soft tissue asymmetry) also improved significantly in groups I and II which were younger than 1 year old. *Conclusion* In positional plagiocephaly, helmet therapy is effective in correcting both cranial and mid-facial asymmetries. Outcomes were optimal in patients <1 year of age, but older patients also showed some improvement.

Keywords Positional plagiocephaly · Helmet therapy · Mid-facial asymmetry · Ear position

Introduction

Since 1992, the American Pediatric Society has advocated a supine sleeping position for infants to prevent sudden infant death syndrome. Subsequently, a significant increase in the incidence of positional plagiocephaly has been observed [1]. Positional plagiocephaly has been linked with a variety of predisposing factors (e.g., primiparity, assisted vaginal delivery, prolonged labor, multiple birth, male sex, and unusual birth position) [2–5], associated conditions (e.g., intrauterine constraint, supine sleeping position, positional preference, and torticollis) [6–9], and potential consequences (e.g., mandibular asymmetry, auditory disorders, strabismus, and developmental problems) [10–14].

Positional plagiocephaly, which refers to deformed head shape without true synostosis of cranial sutures, has specific characteristics. The primary features include occipital flatness, frontal bossing, and the contralateral forehead may be retruded. The fronto-orbital-zygomatic region and cheek on the contralateral side may be retruded as well, and the ipsilateral ear typically is transposed anteriorly. In an overhead view, the shape of the head forms a parallelogram, which unilateral coronal or lambdoidsynostosis is less apt to produce. The chin

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point is also deviated to the contralateral side. Clinical asymmetry in positional plagiocephaly thus extends to mid-facial and orbito-zygomatic regions and is not limited to the cranium. Although contralateral frontal flatness was once emphasized, current descriptions of positional plagiocephaly now center on ipsilateral occipital flatness given that pressure on the ipsilateral side is implicated in the pathogenesis [15]. Of note is a recent report which indicates that children with leftsided plagiocephaly frequently experience difficulties with cognitive, linguistic, and scholastic development [16].

Helmet therapy is one of the non-surgical options for treating positional plagiocephaly, and its utility is backed by research. An earlier systematic review (2008) [17] found this method to be 1.3 times more effective than postural repositioning. Those previous researches on helmet therapy have been confined to cranial asymmetry. However, in addition to cranial asymmetry, the mid-face asymmetry is a major concern in patients with this condition. Our study was conducted to evaluate the impact of helmet therapy on the mid-facial asymmetry that occurs in patients with positional plagiocephaly. Cranial and facial asymmetry was assessed via three-dimensional computed tomography (3D CT), and the outcomes of treatment were analyzed. Soft tissue asymmetry was measured based on the distance in side-to-side positioning of the ears.

Materials and methods

For this retrospective review, we enrolled 99 patients diagnosed with positional plagiocephaly and treated by helmet therapy between September 2005 and July 2012. Inclusion criteria were nonsynostotic plagiocephaly, completion of helmet therapy, and regular follow-up during therapy with complete documentation. The Institutional Review Board of Yonsei University Health System approved this study. All researches involving humans adhered to the Declaration of Helsinki. Each subject underwent 3D CT scanning (Somatom Sensation 64; Siemens Medical Systems, Erlangen, Germany) of facial bones before starting helmet therapy to exclude synostosis of cranial sutures. The second 3D CT scan was also performed following the period of helmet therapy to evaluate post-treatment state. The parents and guardians were informed on the objective of CT scan previously.

All helmet orthotics were custom-manufactured by a single company. Each unit consisted of a plastic outer shell and a foam liner. The liner had voids in areas of cranial flattening, enabling growth without interference, but otherwise rested directly on the scalp to limit expansion (Fig. 1).

Patients were stratified by age at the start of therapy as follows: group I, <6 months (n=35); group II, 6 months to 1 year (n=43); group III, >1 year (n=21).



Fig. 1 Helmet orthotics, which were custom-manufactured, consisted of a plastic outer shell and a foam liner. The liner had space in areas of cranial flattening, enabling growth without interference, however otherwise making direct contact with the scalp to limit expansion. Each orthotics was designed to affect the lateral malar region, zygomatic arch, temporo-mastoid, and occipital areas

Measurement of cranial and facial asymmetry

The cranial vault asymmetry index (CVAI) was calculated according to Loveday and de Chalain [18] from dual cranial diagonal diameters (A and B) as follows: CVAI = difference in cranial diagonal diameters/short cranial diagonal ×100.

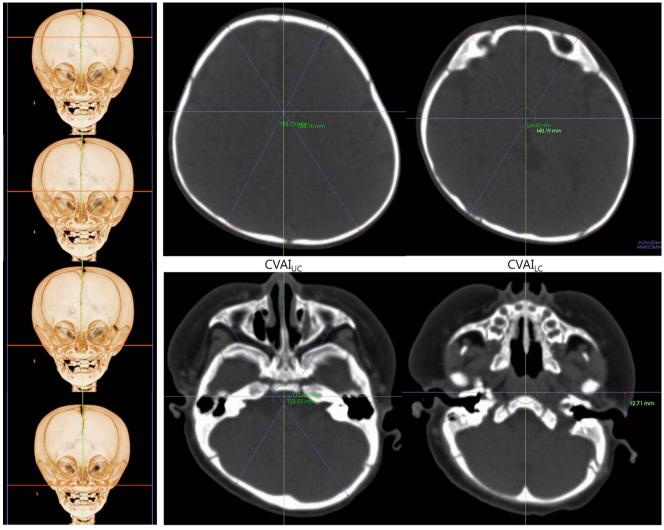
These measurements were carried out after standardization of CT scan with image analysis program (Invivo5, Anatomage Inc., San Jose, CA, USA). We adjusted the midline of face into vertical axis and Frankfrut horizontal line into horizontal axis. All measurements were derived from axial views of facial bone CT scans at multiple levels, including mid-facial level at the inferior orbital rim (CVAI_{IOR-MF}), lower cranial level at the superior orbital rim (CVAI_{SOR-LC}), and upper cranial level at midway from the superior orbital rim to the vertex (CVAI_{MID-UC}).

Anterior transposition of the ipsilateral ear was verified by measuring the distance (D_{EAR}) between the position of the ipsilateral auricular tragus and its expected position relative to the contralateral ear. Both horizontal ($D_{EAR-HOR}$) and vertical ($D_{EAR-VER}$) distances were assessed in axial and coronal CT views, respectively (Fig. 2).

All parameters calculated before and after helmet therapy were compared and categorized by age at the initiation of helmet therapy.

Statistical analysis

A paired *t*-test was used to compare degrees of asymmetry before and after helmet therapy, and the parameters were categorized by age at the initiation of helmet therapy. Statistical significance was evaluated with 95 % confidence interval. For statistical analysis, SAS (version 9.1.3; SAS Institute Inc., Cary, NC, USA) was used.



CVAIMF

 $\mathsf{D}_{\mathsf{EAR}}$

Fig. 2 Cranial vault asymmetry index (*CVAI*) was calculated from facial bone computed tomography imaging in axial views at multiple levels: upper cranial level (midway from the superior orbital rim to the vertex, *CVAI_{MID-UC}*), lower cranial level (the superior orbital rim level, *CVAI_{SOR-LC}*), and mid-facial level (the inferior orbital rim level, *CVAI_{IOR-MF}*).

Results

The mean age at the initiation of helmet therapy was $4.6\pm$ 0.9 months in group I, 8.5 ± 1.6 months in group II, and 22.5 ±13.8 in group III. The mean duration of therapy was $7.8\pm$ 3.9 months in group I, 10.0 ± 6.0 months in group II, and $9.8\pm$ 4.6 months in group III; the resulting period for group I exhibited a shorter duration than group II with statistical significance.

The mean CVAI_{MID-UC} decreased from 10.61±2.08 to 2.58±0.61 in group I, 10.1±2.2 to 3.77±1.8 in group II, and 9.77±2.45 to 8.31±2.75 in group III. These reductions in mean CVAI_{MID-UC} indicated a significant statistical improvement (p<0.05) in groups I and II only (Fig. 3). The mean CVAI_{SOR-LC} decreased from 11.64±3.02 to 2.5±0.57 in group

Anterior transposition of the ipsilateral ear was verified by measuring the distance between the actual position of the ipsilateral auricular tragus and the expected position relative to the contralateral ear (D_{EAR}). Horizontal ($D_{EAR-HOR}$) and vertical ($D_{EAR-VER}$) distances were assessed in axial and coronal CT views, respectively

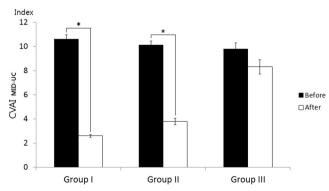


Fig. 3 The mean cranial vault asymmetry index at upper cranial level (midway from the superior orbital rim to the vertex, $CVAI_{MID-UC}$) decreased from 10.61 ± 2.08 to 2.58 ± 0.61 in group I, 10.1 ± 2.2 to 3.77 ± 1.8 in group II, and 9.77 ± 2.45 to 8.31 ± 2.75 in group III. Reductions in CVAI_{MID-UC} indicate significant clinical improvement (p<0.05) in groups I and II only

I, 10.65±3.22 to 4.38±2.61 in group II, and 9.43±2.23 to 8.11±1.98 in group III. These reductions in mean CVAI_{SOR-LC} indicated a significant statistical improvement (p<0.05) for all three groups (Fig. 4). The mean CVAI_{IOR-MF} decreased from 11.88±2.19 to 3.19±0.74 in group I, 11.89±3.18 to 5.46±3.4 in group II, and 10.01±2.94 to 7.48±3.22 in group III. Again, significant therapeutic benefit (p<0.05) was evident in all three groups (Fig. 5).

As for the distance of ear position, the mean $D_{\text{EAR-HOR}}$ changed from 2.1±1.6 to 0.85±0.93 mm in group I, 1.8±1.1 to 1.49±0.86 mm in group II, and 2.5±2.2 to 2.39±2.02 mm in group III. The mean $D_{\text{EAR-VER}}$ decreased from 1.2±0.69 to 0.84±0.56 mm in group I, 1.38±0.77 to 1.16±0.71 mm in group II, and 1.82±1.82 to 1.67±1.77 mm in group III. Reductions in horizontal and vertical ear asymmetry were significant (p<0.05) in groups I and II only (Figs. 6 and 7) (Table 1).

Case I

A 4-month-old male visited Severance Cranioplasty Clinic and was diagnosed with left-sided positional plagiocephaly. A cranial molding helmet was designed based on a 3D CT craniofacial bone scan. The helmet was applied at 4 months of age and was withdrawn at 13 months of age. This equaled a 9month therapeutic period with the average maintenance duration being 20 h/day. Pre-treatment indices were as follows: $CVAI_{MID-UC}$ 13.71, $CVAI_{SOR-LC}$ 13.57, $CVAI_{IOR-MF}$ 5.48, $D_{EAR-HOR}$ 7.83, $D_{EAR-VER}$ 5.30. Post-treatment indices were improved to the following values: $CVAI_{MID}$ -UC 8.18, $CVAI_{SOR-LC}$ 6.58, $CVAI_{IOR-MF}$ 5.26, $D_{EAR-HOR}$ 2.21, $D_{EAR-VER}$ 1.43 (Fig. 8).

Case II

Index 14

12

10

8

6

4

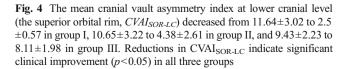
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Group I

CVAI SOR-LC

An 8-month-old male visited Severance Cranioplasty Clinic and was diagnosed with right-sided plagiocephaly. A cranial molding helmet was designed based on a 3D CT craniofacial



Group II

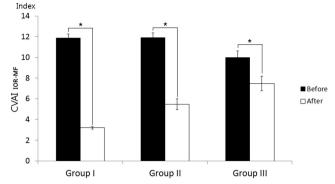


Fig. 5 The mean cranial vault asymmetry index of mid-facial level (the inferior orbital rim, $CVAI_{IOR-MF}$) decreased from 11.88 ± 2.19 to 3.19 ± 0.74 in group I, 11.89 ± 3.18 to 5.46 ± 3.4 in group II, and 10.01 ± 2.94 to 7.48 ± 3.22 in group III. Reductions in $CVAI_{IOR-MF}$ indicate significant clinical improvement (p<0.05) in all three groups

bone scan. The helmet was applied at 9 months of age and was withdrawn at 22 months of age. This equaled a 13-month therapeutic period with the average maintenance duration being 15 h/day. Pre-treatment indices were as follows: CVAI_{MID}_{UC} 14.92, CVAI_{SOR-LC} 11.59, CVAI_{IOR-MF} 10.64, $D_{EAR-HOR}$ 20.27, $D_{EAR-VER}$ 12.14. Post-treatment indices were improved to the following values: CVAI_{MID-UC} 8.54, CVAI_{SOR-LC} 6.67, CVAI_{IOR-MF} 6.96, $D_{EAR-HOR}$ 14.57, $D_{EAR-VER}$ 5.80 (Fig. 9).

Discussion

Before

□After

Group III

This retrospective study confirms that helmets are therapeutic for both cranial and mid-facial asymmetry in younger patients (<1 year old) with plagiocephaly. The positive impact that cranial molding has on the mid-face may be due to the fact that these customized helmets cover the zygomatic, temporal, and occipital areas, in addition to covering the entire calvarium. In particular, the lateral malar region, zygomatic arch, and mastoid are selectively subjected to pressure. Relative

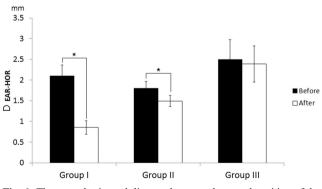


Fig. 6 The mean horizontal distance between the actual position of the ipsilateral auricular tragus and the expected position relative to the contralateral ear ($D_{EAR-HOR}$) changed from 2.1±1.6 to 0.85±0.93 in group I, 1.8±1.1 to 1.49±0.86 in group II, and 2.5±2.2 to 2.39±2.02. Reductions in $D_{EAR-HOR}$ indicate significant clinical improvement (p<0.05) in groups I and II only

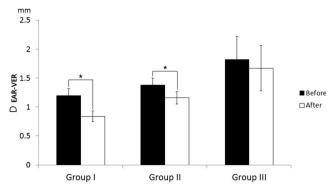


Fig. 7 The mean vertical distance between the actual position of the ipsilateral auricular tragus and the expected position ($D_{\text{EAR-VER}}$) decreased from 1.2 ± 0.69 to 0.84 ± 0.56 in group I, 1.38 ± 0.77 to 1.16 ± 0.71 in group II, and 1.82 ± 1.82 to 1.67 ± 1.77 in group III. Reductions in $D_{\text{EAR-VER}}$ indicate significant clinical improvement (p<0.05) in groups I and II only

compression of the ipsilateral malar and contralateral mastoidoccipital areas is thereby achieved, whereas the contralateral malar and ipsilateral mastoid-occipital areas undergo relative decompression. As previously shown, helmet therapy also helps in re-establishing the symmetry of the ears, indicating the capacity of this treatment to alter soft tissues as well as skeletal structures [19].

An inverse correlation between the degree of cranial and facial asymmetry correction and the age at helmet application has been noted by others [20]. Previous research suggested that success with helmet therapy was caused by the growing brain so that longer treatment was needed as infants were getting aged and brain growth decelerated. Indeed the size of a newborn's brain increases by 200 % during the first 6 months of life, adding only another 50 % of its mass over the next 24 months [21]. By 2 years of age, the brain is approximately

70 % percent of its adult size. Any further growth then occurs slowly over the next 4 years [22].

In our cohort, infants younger than 1 year old showed a greater improvement in cranial vault asymmetry and midfacial asymmetry, midway from the vertex to the supraorbital rim and in the distance of the ear position when compared with infants older than 1 year old. Hence, early recognition of infants in need of helmet therapy is essential. The age of 5-6 months is generally recommended as optimal for starting helmet therapy, although there are different points of view about the age of starting helmet use. One obvious concern is that delays will entail longer treatment periods. Clearly, delays do undermine outcomes, but some improvement may still be achieved in infants older than 12 months [20].

In our analysis, younger infants from group I required less time to correct cranial and mid-facial asymmetries when compared with older infants from groups II and III (mean duration of therapy: group I, 7.8 ± 3.9 months; group II, 10.0 ± 6.0 months; group II, 9.8 ± 4.6 months), with group II registering the longest mean duration. Similarly, post-treatment CVAI values were <3.5 in group I (CVAI_{MID-UC}, 2.58 ± 0.61 ; CVAI_{SOR-LC}, 2.5 ± 0.57 ; CVAI_{IOR-MF}, 3.19 ± 0.74), <5.5 in group II (CVAI_{MID-UC}, 3.77 ± 1.8 ; CVAI_{SOR-LC}, 4.38 ± 2.61 ; CVAI_{IOR-MF}, 5.46 ± 3.4), and >7 in group III (CVAI_{MID-UC}, 8.31 ± 2.75 ; CVAI_{SOR-LC}, 8.11 ± 1.98 ; CVAI_{IOR-MF}, 7.48 ± 3.22), underscoring that outcomes of helmet therapy are truly age dependent.

In this retrospective study, 3D CT scans were undergone before and after treatment states. The second CT scan was the most appropriate to evaluate the degree of alteration accurately, and imaging studies were conducted after obtaining informed consents. However, 3D image scanning equipment with laser shape digitizer has been developed currently,

Table 1 Cranial and mid-facial asymmetry indices before and after helmet therapy <i>CVAI</i> cranial vault asymmetry index, <i>CVAI_{MID-UC}</i> CVAI midway from the superior orbital rim to vertex at upper cranial level, <i>CVAI_{SOR-LC}</i> CVAI at the superior orbital rim, lower cranial level, <i>CVAI_{IOR-MF}</i> CVAI at the inferior orbital rim, mid-facial level, <i>D_{EAR}</i> distance between the actual position of the ipsilateral auricular tragus and the expected position (relative to the contralateral ear)		Index	Pre-treatment	Post-treatment	<i>p</i> value
	Group I (<6 months), <i>n</i> =35	CVAI _{MID-UC}	10.61±2.08	2.58±0.61	<0.0001*
		CVAI _{SOR-LC}	11.64 ± 3.02	2.5±0.57	< 0.0001*
		CVA _{IOR-MF}	11.88 ± 2.19	$3.19{\pm}0.74$	< 0.0001*
		$D_{\text{EAR-HOR}}$	2.1±1.6	$0.85 {\pm} 0.93$	< 0.0001*
		$D_{\text{EAR-VER}}$	1.2 ± 0.69	$0.84{\pm}0.56$	< 0.0001*
	Group II (6 months–1 year), n=43	CVAI _{MID-UC}	10.1 ± 2.2	3.77±1.8	< 0.0001*
		CVAI _{SOR-LC}	10.65 ± 3.22	4.38±2.61	< 0.0001*
		CVAI _{IOR-MF}	11.89 ± 3.18	5.46±3.4	< 0.0001*
		$D_{\text{EAR-HOR}}$	1.8 ± 1.1	$1.49 {\pm} 0.86$	0.049*
		$D_{\text{EAR-VER}}$	1.38 ± 0.77	1.16 ± 0.71	0.044*
	Group III (>1 year), n=21	CVAI _{MID-UC}	9.77±2.45	8.31±2.75	0.056
		CVAI _{SOR-LC}	9.43±2.23	8.11±1.98	0.005*
		CVAI _{IOR-MF}	10.01 ± 2.94	7.48±3.22	0.009*
		$D_{\text{EAR-HOR}}$	2.5±2.2	2.39±2.02	0.184
* <i>p</i> <0.05 (significant intergroup difference on paired <i>t</i> -test)		$D_{\rm EAR-VER}$	1.82±1.82	1.67±1.77	0.082

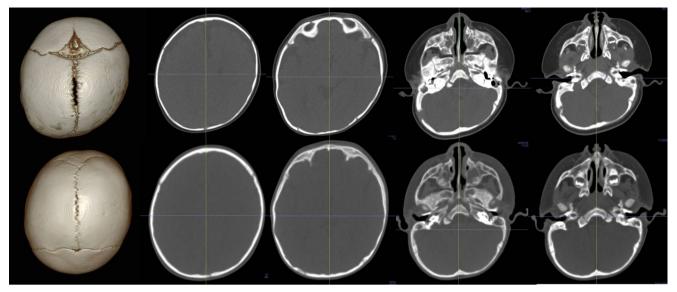


Fig. 8 Computed tomography images of a 4-month-old male infant with left-sided plagiocephaly. *Upper row* before treatment; *lower row* after treatment. From *left* to *right*: 3D CT image above vertex, upper cranial level, lower cranial level, midface level, and ear tragus level. 3D CT image was flipped as a mirror image to make it easy for understanding. Left occipital flattening, frontal bossing, and anterior displacement of left ear were noted before treatment. Significant improvement was achieved

with regard to asymmetry of cranium and midface. Discrepancy of ear position was also reduced. Pre-treatment indices were as follows: CVAI_{MID-UC} 13.71, CVAI_{SOR-LC} 13.57, CVAI_{IOR-MF} 5.48, $D_{EAR-HOR}$ 7.83, $D_{EAR-VER}$ 5.30. Post-treatment indices were improved to the following values: CVAI_{MID-UC} 8.18, CVAI_{SOR-LC} 6.58, CVAI_{IOR-MF} 5.26, $D_{EAR-HOR}$ 2.21, $D_{EAR-VER}$ 1.43

avoiding exposure to X-ray [23]. Furthermore, adverse effects of ionizing radiation were reported in literatures [24, 25]. Since 2014, authors have performed helmet therapy on the basis of anthropometric measurement with calibration instruments and attempted 3D image scanning additionally. CT scans are recommended in case clinical manifestations of craniosynostosis are presented, and synostotic plagiocephaly should be ruled out.

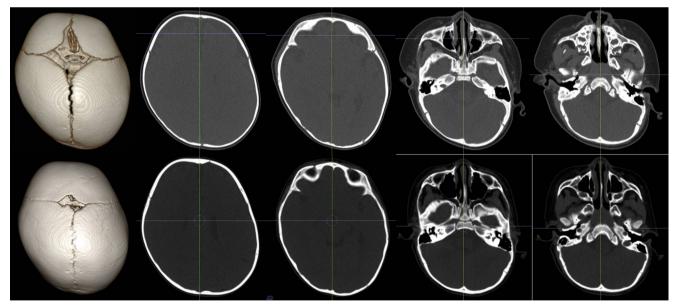


Fig. 9 Computed tomography images of an 8-month-old male infant with right-sided plagiocephaly. *Upper row* before treatment; *lower row* after treatment. From *left* to *right*: 3D CT image above vertex, upper cranial level, lower cranial level, midface level, and ear tragus level. 3D CT image was flipped as a mirror image to make it easy for understanding. Right occipital flattening, frontal bossing, and anterior displacement of right ear were noted before treatment. Significant

improvement was achieved with regard to asymmetry of cranium and midface. Discrepancy of ear position was also reduced. Pre-treatment indices were as follows: CVAI_{MID-UC} 14.92, CVAI_{SOR-LC} 11.59, CVAI_{IOR-MF} 10.64, $D_{EAR-HOR}$ 20.27, $D_{EAR-VER}$ 12.14. Post-treatment indices were improved to the following values: CVAI_{MID-UC} 8.54, CVAI_{SOR-LC} 6.67, CVAI_{IOR-MF} 6.96, $D_{EAR-HOR}$ 14.57, $D_{EAR-VER}$ 5.80

Positional plagiocephaly bears an association with other morphologic and functional disorders, including congenital muscular torticollis, facial hemihypoplasia, superior oblique palsy, and ocular torticollis [9, 26]. In instances of left-sided plagiocephaly, poor cognitive ability and language development have been documented, necessitating special education [16]. Some have attributed these phenomena to compressive and gravitational forces [26]. Cranial molding helmets have been utilized extensively to correct positional plagiocephaly and have been found superior to active repositioning [17–20, 27].

A recent literature demonstrated comparative analysis on helmet therapy and the natural course of skull deformation revealing equivalent outcomes, although the research excluded patients with very severe deformation showing oblique diameter difference index>113 % (CVAI>13) [28]. Studies regarding helmet therapy, however, proved its effectiveness in plagiocephaly with various degrees [29]. Our results exhibited a significant improvement in severe cases as well (Figs. 8 and 9). Consequently, helmet therapy should be considered as one of the therapeutic options, relying on the proven efficacy [19, 20]. Furthermore, an immediate decision on helmet therapy is crucial since the time period to achieve satisfactory outcome is limited [29, 30].

Nevertheless, care must be taken to avoid inherent side effects such as pain, sweating, pressure sores, localized ethanol erythema, unsatisfactory prosthetic fit, skin infection, corrective failures, and subcutaneous abscess [30, 31]. In addition, economic burden has been presented as a possible disadvantage. The cost of helmet therapy is about \$2000 in South Korea, and the amount is comparable in the majority of countries [28]. Parents and guardians of affected children, however, usually agree with the necessity of helmet therapy, although related procedures are not supported by national health insurance. Differential diagnosis and decision followed by strategic application are essential to achieve improvement and thus minimize adverse effects [29, 30].

Conclusion

Helmet therapy is a proven non-surgical treatment for plagiocephaly. The results of this study confirm the value of helmet therapy in correcting both cranial and mid-facial asymmetries. Outcomes are optimal in younger patients (<1 year old), but older infants may still benefit from this treatment.

Author contributions IS Yun performed all therapeutic procedures, analyzed the data, and wrote one draft of the paper. MC Lee analyzed the data, wrote the paper in part, and edited figures—all commensurate with the contributions of IS Yun. J Hwang analyzed the data and edited the manuscript. YO Kim discussed therapeutic procedures and formulated strategies. KW Shim and EK Park played roles in therapeutic managements. DH Lew supervised the progress of this report, which included extensive editing and overseeing related communications.

Conflict of interest None of the authors have any conflicting or potentially influential interests with respect to this research.

Financial Disclosure The authors have no proprietary or commercial interest in any materials discussed in this article.

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