

# Deformational plagiocephaly: a follow-up of head shape, parental concern and neurodevelopment at ages 3 and 4 years

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## ABSTRACT

**Objectives** To compare head shape measurements, parental concern about head shape and developmental delays in infancy with measurements obtained at follow-up at ages 3 and 4 years.

**Design** Longitudinal cohort study.

**Setting** Initial assessments were conducted at a plagiocephaly clinic; follow-up assessments were conducted in the children's homes.

**Participants** 129 children with a mean age of 4 years (range 3 years 3 months to 4 years 9 months), all of whom were diagnosed in infancy with deformational plagiocephaly or brachycephaly.

**Main outcome measures** Head shape measurements of cephalic index and oblique cranial length ratio; level of parental concern about head shape; and delays on parent-completed age-appropriate Ages and Stages Questionnaires.

**Results** 61% of head shape measurements reverted to the normal range; 4% remained severe at follow-up. Brachycephaly improved more than plagiocephaly. Facial and frontal asymmetry reduced to almost nil. Most had good improvement, but 13% were categorised as having 'poor improvement'. Initially, 85% of parents reported being 'somewhat' or 'very' concerned; this decreased to 13% at follow-up. The percentage of children with  $\geq 1$  delay decreased from 41% initially to 11% at follow-up.

**Conclusions** Overall, head shape measurements, parental concern and developmental delays in infancy showed a dramatic improvement when re-measured at 3 and 4 years of age.

## INTRODUCTION

A persistent resting head orientation either in the antenatal or in the postnatal period, or both, may expose the infant's rapidly growing and malleable cranial bones to uneven compressive forces resulting in positional or deformational plagiocephaly or brachycephaly. Since the practice of supine sleeping for sudden infant death syndrome (SIDS) prevention has been more widely adopted, the condition has become more prevalent,<sup>1</sup> with some infants developing alarmingly flat occipital areas in association with ear misalignment and compensatory flattening in other areas of the skull.

Spontaneous improvement can occur in the first 2 years.<sup>2-3</sup> Risk factors include: male sex, firstborn child, difficult delivery, supine sleep position, multiple birth, prematurity and torticollis or other imbalance of neck muscle function.<sup>4-5</sup> Some feel that the disorder is purely cosmetic,<sup>6</sup> that the condition is 'clinically unimportant'<sup>7</sup> and that there are no negative neurologic consequences,<sup>8</sup> but

## What is already known on this topic

- ▶ Deformational plagiocephaly and brachycephaly are common conditions arising in early infancy as a result of compressive forces acting on the growing infant cranium, but there has been a lack of evidence to show how the deformity persists into later childhood.
- ▶ Infants with positional head deformities are more likely to be developmentally delayed, particularly in the gross motor domain.

## What this study adds

- ▶ Most positional head deformities improve by the age of 3–5 years, with the majority returning to the normal range.
- ▶ By age 3–5 years, developmental delays reduce to what might be expected in the normal population.

others have suggested that if not treated early the deformity may persist<sup>9-11</sup> and that there may also be associated developmental difficulties in the longer term.<sup>12-13</sup>

Longer-term outcomes in infants with deformational plagiocephaly have rarely been studied. The few studies that have researched the topic have been limited by small samples and poor follow-up rates<sup>7-13-14</sup> or did not use an objective measurement of head shape.<sup>7-15-16</sup> No study has yet evaluated children treated exclusively with positioning strategies, including physiotherapy if warranted, and compared head shape measurements in infancy with those taken more than 2 years later.

Infants with deformational plagiocephaly/brachycephaly have higher rates of developmental delay than non-plagiocephalic children,<sup>5-12-17-22</sup> but whether these delays persist is uncertain. Some authors<sup>12-13</sup> have shown an increased need in later childhood for further educational help, but this is a controversial area in which there is sparse data and poor follow-up rates. No study showing delayed development in deformational plagiocephaly has determined cause and effect.

The main aims of this study were to compare head shape measurements at age 3–5 years with

those obtained in infancy and to investigate factors common to those children whose head shapes had not improved. In addition, we aimed to determine the level of parental concern about the children's head shapes and to assess the extent of developmental delays.

## METHODS

Infants with positional head shape deformities from throughout the Auckland region, New Zealand, were referred by general practitioners, community child health nurses, physiotherapists and paediatricians to an outpatient clinic at Starship Children's Hospital for assessment and advice. This was the only dedicated plagiocephaly clinic in the region. Parents answered a questionnaire covering demographic information, obstetric history and a history of the plagiocephaly and current positioning strategies. The infant was examined, neck function assessed and an objective measurement of head shape was made. Neck muscle dysfunction was defined as an observed head tilt or a limitation in active or passive range of lateral movement, or if there was a parental report of a past definite difficulty in turning the head in one direction that had resolved by the time of the clinic visit. All parents were counselled fully about how to perform counterpositioning strategies and the infant was referred for physiotherapy if indicated for neck muscle dysfunction. All assessments were performed by the same researcher (BLH).

Parents completed an Ages and Stages Questionnaire (ASQ),<sup>23</sup> assessing age-appropriate development in the communication, gross motor, fine motor, personal/social and problem solving domains. ASQ second edition questionnaires were used at both assessments, but cut-offs from the ASQ third edition were used as the questions are almost identical but the cut-offs are now based on a much larger validation sample of >12 000.<sup>24</sup> Minor differences in wording between the two versions of the questionnaires were deemed not to influence the results.

The technique used to objectively measure the head shape, HeadsUp, is fully described elsewhere<sup>25</sup> and is further discussed at <http://www.fmhs.auckland.ac.nz/som/paed/plagiocephaly/default.aspx>. In brief, a soft stretchy headband is placed around the head circumference, thus outlining the head shape in that plane. A digital photograph of the band is taken from above and a custom-written computer program takes measurements from the resulting image of the band shape. The main measurements used are the cephalic index (CI) and the oblique cranial length ratio (OCLR). CI is the ratio of the width of the head to the length of the head. A CI $\geq$ 93 is considered outside the normal range and indicates brachycephaly or short wide head shape with central occipital flattening. OCLR is the ratio of the longer to the shorter oblique cranial diameters, and indicates plagiocephaly or asymmetrical head shape. An OCLR $\geq$ 106 is considered abnormal.

All children diagnosed in the clinic with deformational plagiocephaly or brachycephaly were included in this study if they were at least 3 years 3 months old during the follow-up period (July 2009 to March 2010). Children older than 12 months at initial assessment, those living outside the region, those with significant medical problems or neurodevelopmental conditions such as Down's syndrome, families who did not speak English and children who were in state care were excluded.

A letter inviting participation in the study was sent to the families, followed up by a phone call to further explain the study and to make an appointment to reassess the child at home. At the home visit, the parents signed a consent form and answered a short questionnaire. HeadsUp measurement

photographs were taken of the child's head, and an age-appropriate ASQ was completed by the parent with the child.

Head shape measurements at follow-up were compared with those obtained at the initial assessment. The severity of the head flattening was categorised using cut-offs established earlier.<sup>5</sup> Previous and current ASQ scores were compared. Infants were classified as having poor improvement at follow-up if they were worse than at the initial assessment, if they were still in the severe category or if they had not reduced the equivalent of one severity level, as per an algorithm established earlier.<sup>3</sup>

The parents' occupations were rated using the New Zealand Socioeconomic Index and were categorised into high, medium and low socioeconomic status.<sup>26</sup> The score of the higher-rated parent was used. Statistical analysis was performed using SAS (V.9.1, 2002–2003, SAS Institute, Cary, North Carolina, USA).  $\chi^2$  Analysis was used for categorical variables and paired t tests and Wilcoxon signed rank tests were performed on continuous variables.

The study was approved by the Northern X Regional Ethics Committee and the Auckland District Health Board Research Office.

## RESULTS

### Characteristics of study population

There were 161 eligible children of the 220 children with deformational plagiocephaly seen in the clinic from May 2005 to August 2007. Those children who were not eligible were classified as follows: not yet attained the age of 3 years 3 months, 28; aged >12 months at initial assessment, 11; living out of the region at time of initial assessment, 9; other, 11. Of the 161 eligible children, 129 (80%) were followed up. Five families declined to participate and 27 had moved away or could not be found. No child had moulding helmet therapy. Two children refused to have their head measured at the follow-up assessment; other data for these two children are included in the results, but head measurement data is missing (table 1).

### Head shape measurements

Initially, 47% were in the severe range, 31% were in the moderate range and 22% were in the mild range. By follow-up, 77 (61%) of the measured children had achieved the normal range for head shape, and only 5 (4%) were in the severe range (figure 1).

Reductions in severity occurred over all severity levels and there were many children who had been severe initially whose follow-up measurements fell into the normal range (figure 2). At follow-up only two children (2%) had frontal asymmetry and four (3%) had facial asymmetry, compared to 57 (44%) and 38 (29%), respectively, with frontal and facial asymmetry at initial assessment. Although 62% had a history of neck muscle dysfunction at initial assessment, no child had problems with neck function at follow-up.

Table 2 lists types of head shapes and the mean CI and OCLR of all infants. The mean CI in children who were initially brachycephalic fell from 98.5 (SD 4.2) to 90.4 (SD 3.4); in initially plagiocephalic children the mean OCLR fell from 110.0 (SD 2.4) to 106.1 (SD 2.3). Similar levels of reduction were seen in the combination group.

### Poor improvement

There were 17 (13%) children who were classified with poor improvement, 12 who were initially plagiocephalic and 5 with

**Table 1** Characteristics of study children

Variable	n (%)
Mean age at assessment	
Initial	5.8 (SD 2.3) months
Range	2.0–12.0 months
Follow-up	47.4 (SD 4.5) months
Range	3 years 3 months to 4 years 9 months
Mean age difference between assessments	3.5 (SD 0.4) years
Range	2.6–4.4 years
Gender	
Male	91 (70.5)
Female	38 (29.5)
Parity	
Firstborn	90 (69.8)
Later-born	39 (30.2)
Gestation	
<37 weeks	22 (17.1)
Term	107 (82.9)
Multiple birth (missing=5)	
Yes	8 (6.4)
No	116 (93.6)
Difficult delivery	
Yes	30 (23.3)
No	99 (76.7)
Breastfeeding	
Never or <1 month	34 (26.3)
≥1 month	95 (73.7)
Infant's ethnicity	
New Zealand European	82 (63.6)
New Zealand Maori	14 (10.8)
Pacific	15 (11.6)
Asian	17 (13.2)
Other	1 (0.8)
Socioeconomic status	
Low (0–40)	9 (7.0)
Medium (41–60)	58 (45.0)
High (>60)	62 (48.0)
Maternal qualifications	
Low (none or school certificate)	23 (17.8)
Medium (sixth form or Bursary)	23 (17.8)
High (professional or tertiary qualification)	83 (64.3)
Age plagiocephaly first noticed (weeks)	
Mean (SD)	6.9 (4.7)
Range	0–22.0
Age first seen in clinic	
<6 months	77 (59.7)
≥6 months	52 (40.3)
Neck dysfunction at initial assessment (missing=1)	
Yes	80 (62.5)
No	48 (37.5)

combination head shapes. Seven of the 17 were in the mild category by follow-up. No brachycephaly-only child was in the poor improvement group at follow-up. Those in the poor improvement group had a higher mean OCLR of 110.2 (SD 2.4) at initial assessment compared to those in the well-improved group with a mean initial OCLR of 109.2 (SD 1.8) ( $p=0.02$ ).

### Parental concern

Most parents (85%) were 'very' or 'somewhat' concerned about the head shape at the initial assessment, with 15% being 'not very' concerned. At follow-up, 13% were 'very' or 'somewhat'

concerned, 28% were 'not very' concerned and 59% were 'not at all' concerned.

### ASQ delays

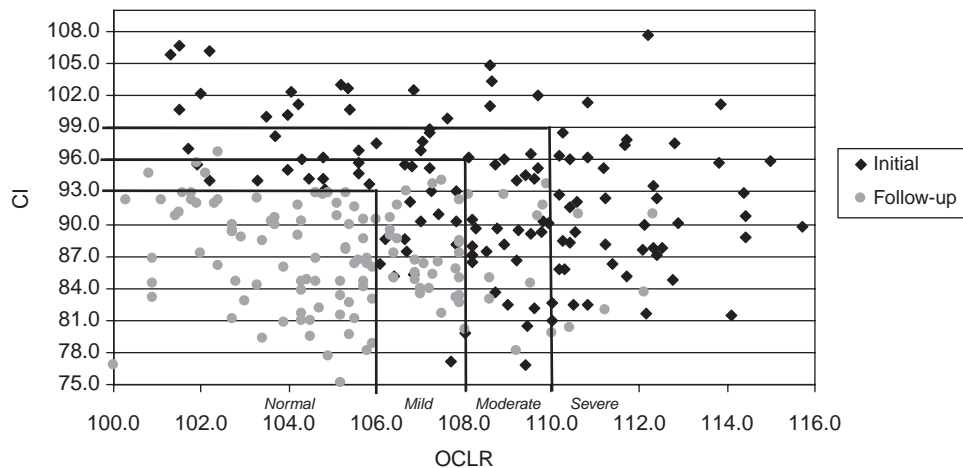
Seven infants did not have an ASQ assessment at the initial clinic visit as they were <15 weeks of age. Of those who had both assessments, at the initial assessment 41% had one or more delays on the ASQ and 22% had two or more delays. At follow-up only 11% of these had one or more delays and 4% had two or more delays. Using ASQ-3 validation data, one would expect around 15% to show one or more delays at each age, and around 5–8% to have two or more delays.<sup>24</sup> The difference between observed and expected was significant at the initial assessment ( $p<0.0001$ ) but not at the follow-up assessment ( $p=0.26$ ). The mean drop in the number of delays between the two assessments was 0.6 ( $p<0.0001$ ). Delays at initial assessment were predominantly in the gross motor domain, with 29% of all delayed children having problems in this area. There was a moderate level of delays in the fine motor (17%), problem solving (15%) and personal/social (17%) domains, but few in the communication domain (4%). At follow-up, there were fewer than 5% in each domain with delays (expected: 6–7%).

Initially, there was no difference in delays between the good and poor improvement groups. However, at the follow-up assessment, five (29%) of the poor improvement group exhibited delays, compared with nine (8%) of the good improvement group ( $p=0.02$ , Fisher's). These delays were primarily in the communication domain, with 18% of the poor improvement group showing delays in this domain compared with 3% in the well-improved group ( $p=0.02$ ). However, there were small numbers in each group and this difference should be interpreted with caution. Variables found not to be significantly related to poor improvement included: gender, ethnicity, socioeconomic status, age at first assessment, prematurity, side of flattening, neck dysfunction initially and being firstborn.

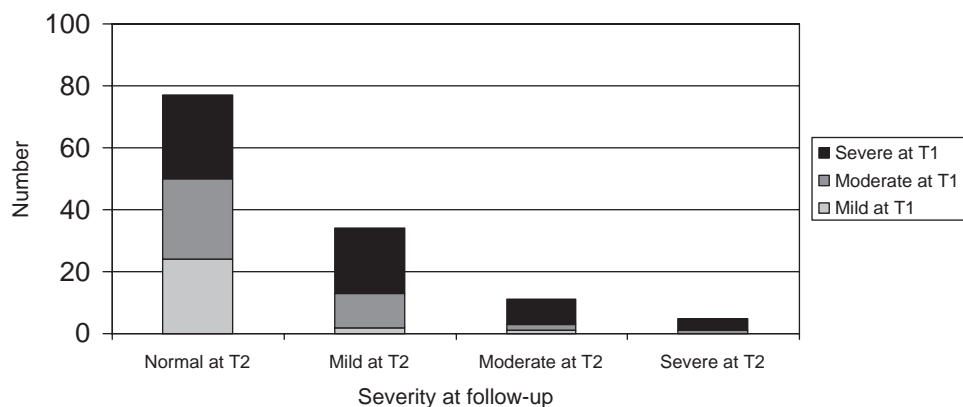
### DISCUSSION

In accordance with our previous work on younger children,<sup>2,3</sup> in this study we have shown good improvement over time in 87% of preschoolers who had exhibited deformational brachycephaly and plagiocephaly as infants. Importantly, 61% improved enough to fall into the normal range 3.5 years later, with only 4% still in the severe range by then. In an earlier study with 1 year of follow-up, these figures were 42% and 8% respectively,<sup>3</sup> indicating that improvement continues to occur as the child ages. The small group who improved less well tended to be those who were initially more severely plagiocephalic. This is to be expected as those with an OCLR of 110 or greater at follow-up were all categorised as poor improvement and many of these had started with a very high OCLR measurement that had not yet dropped below 110. Other studies of longer-term outcomes are few and are limited by subjective evaluations of head shape<sup>7</sup> or by poor response rates.<sup>13</sup> A few shorter-term studies of positional head deformities have also shown improvement in head shape in infants treated conservatively.<sup>3, 27, 28</sup>

Brachycephaly improved more than plagiocephaly over time. Teichgraeber *et al*<sup>29</sup> showed significant improvements in brachycephaly in a subgroup of brachycephalic infants followed for 4.5 months on average, but they concluded that brachycephalic children rarely revert to normal using either positional strategies or moulding helmets. However, the authors used norms that were set in the 1980s by Farkas,<sup>30</sup> at a time when supine sleeping was not common. Cephalic configurations in



**Figure 1** Before and after head measurements. Severity categories: normal ( $CI < 93$  and  $OCLR < 106$ ); mild ( $93 \leq CI < 96$  and  $106 \leq OCLR < 108$ ); moderate ( $96 \leq CI < 99$  and  $108 \leq OCLR < 110$ ); severe ( $CI \geq 99$  or  $OCLR \geq 110$ ). CI, cephalic index; OCLR, oblique cranial length ratio.



**Figure 2** Severity compared over time. Change in severity between initial assessment (T1) and follow-up (T2).

**Table 2** Head types and measurements, initially and at follow-up

Head measurements	Initial (mean (SD))	Follow-up (missing=2) (mean (SD))
Oblique cranial length ratio	108.6 (3.3)	105.4 (2.6)
Cephalic index	92.6 (6.6)	87.0 (4.7)
Type	Initial (n (%))	Follow-up (missing=2) (n (%))
Plagiocephaly	64 (49.6)	42 (33.1)
Brachycephaly	27 (20.9)	4 (3.1)
Combination	38 (29.5)	4 (3.1)
Normal		77 (60.6)

infants are strongly influenced by sleep position,<sup>31</sup> and CI is higher in supine sleeping populations.<sup>2</sup> It is possible that our brachycephaly cut-offs for 3- and 4-year-old children should be lower than the level of 93 previously set for infants. There is a need for a large population-based study to establish CI norms for western supine-sleeping populations from infancy through childhood.

Parental concerns about the head shape reduced markedly over time. This tendency has also been noted by others.<sup>13–15</sup> Indeed, many mothers commented that they had all but forgotten about the head shape issue until they received our letter about the study. It seems that full hair growth by this age hides any residual asymmetry unless the hair is wet or very short.

As in other studies<sup>22</sup> a high percentage of the children in this study exhibited developmental delays as infants, but these

were predominantly gross motor in nature. Although no previous study has shown cause and effect, it is possible that early gross motor delays may be on the causal pathway that leads to positional head deformation, that is, developmental delays lead to lack of movement and therefore more likelihood of positional head flattening. Delays may be related to the effects of the supine sleep position,<sup>32</sup> especially where prone play experience is inadequate,<sup>33–34</sup> thus limiting the development of strength and upper body coordination. Neck muscle dysfunction,<sup>35</sup> low tone,<sup>21</sup> lower activity levels<sup>36</sup> and the tendency for the condition to be more prevalent in males<sup>36</sup> may also impact on developmental scores. Nevertheless, this study has shown that development catches up over time. In contrast, others<sup>12–13</sup> have suggested that delays continue into later childhood; however, these studies were hampered by small numbers in the follow-up



groups. Further research into long-term outcomes for development is needed to clarify these issues, especially in children whose head shapes do not revert to normal.

A limitation of this study is the two-dimensional nature of HeadsUp measurements in what is essentially a three-dimensional problem. It is also possible that cut-offs to indicate brachycephaly may need to be revised for older children, and we await population-based norms to indicate the appropriate level. The strengths of the study were the good rates of follow-up and the use of the same tools at both assessments to measure head shape and development. No child had orthotic helmet therapy; therefore, the results are useful in determining the natural history of positional head deformities after positioning advice and physiotherapy have been provided.

Another limitation was that there was no local control group and out of necessity we used cut-offs established from the ASQ normative population based on >12 000 children. It is possible that cut-offs for New Zealand children may be different but we have no reason to believe this to be so. It is difficult to say how representative of the general population this group of children was, but it is to be noted that they attended the only dedicated plagiocephaly clinic in the region at the time and had been referred by a wide range of practitioners. We note the relatively large number of families in the study from high socioeconomic status and with higher maternal qualifications, but have insufficient data to determine why this was so.

Supine sleeping has been largely responsible for an impressive reduction in SIDS<sup>37</sup> and it is vitally important to maintain the supine sleeping message. If deformational plagiocephaly or brachycephaly does occur, one should provide the parents with positioning strategies and reassure them that in all likelihood improvement will happen with time. In New Zealand, the recommendation to avoid the prone sleeping position has been in place since 1991,<sup>38</sup> although the decline in prevalence of prone sleeping position started in 1989.<sup>39</sup> Thereafter, an increase in deformational plagiocephaly was soon noticed,<sup>1</sup> with some describing the increase as being of 'epidemic' proportions.<sup>40–41</sup> We have not noticed an epidemic of older children and teenagers with unusual head shapes in our clinical practices and in our communities. We suspect that improvement continues into older childhood, and we look forward to further studies describing much longer-term outcomes of this commonly occurring infant condition.

## CONCLUSIONS

Most, but not all, positional head shape deformities improved with time, some dramatically. Brachycephaly showed better improvement than plagiocephaly, and facial and frontal asymmetry all but disappeared. Parental concern about the head shape also reduced markedly. Although there was a high level of developmental delays in infancy, this had reduced to an expected level by preschool age.

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**Competing interests** None.

**Patient consent** Obtained.

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