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REGULAR ARTICLE

Characteristics, head shape measurements and developmental delay in 287 consecutive infants attending a plagiocephaly clinic

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Brachycephaly, Cephalometry, Deformational plagiocephaly, Infant care, Torticollis

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Abstract

Aim: To describe the characteristics, developmental status and severity of head shape deformation in infants presenting to a plagiocephaly clinic.

Methods: Head shape was measured and neck function assessed in 287 consecutive infants presenting to a plagiocephaly outpatient clinic. Information was obtained on demographic and obstetric factors, plagiocephaly history and current positioning strategies. Development was assessed by the Ages and Stages Questionnaire (ASQ).

Results: After clinical examination, craniosynostosis was suspected in seven infants (2%) and a skull computed tomography (CT) scan was performed; five showed suture abnormalities. Fifty-eight percent of cases had a history of limitation of neck function; this was more likely in plagiocephalic infants than brachycephalic infants. Males, firstborn infants, instrument-delivered infants, supine sleep position and right-sided flattening were predominant. One or more delays on the ASQ were seen in 36% of infants.

Conclusion: Physical examination of infants with head shape deformities is essential in order to rule out craniosynostosis. Infants with deformational plagiocephaly frequently have neck muscle dysfunction. We postulate that the higher than expected number of developmental delays may be related to the effects of supine sleep position, low or variable tone, lower activity levels, male gender and neck muscle dysfunction.

BACKGROUND

Deformational plagiocephaly and brachycephaly, or positional head deformities, are common in infants since the widespread use of the supine sleep position to combat sudden infant death syndrome (SIDS) (1). In plagiocephaly, there is asymmetrical posterior flattening of the head, often with compensatory bossing of the contralateral occipital area and ipsilateral frontal area. Brachycephaly refers to more central occipital flattening, where the head is short in the anteroposterior dimension and wide between the biparietal eminences. Positional preference and plagiocephaly have been associated with an imbalance of neck muscle function (2), a failure of parents to regularly vary the head position (3), altered tone (4) or activity levels (3), persistent feeding positions (5) and the placement of environmental stimuli encouraging preferential head orientation. Most mild deformations improve spontaneously over time (6).

For the parents of young infants who are developing an unusual-shaped head, the problem can be very distressing and most are very keen to know how they can help it improve. For the health professional, it is essential to recognize the occasional case of craniosynostosis or premature sutural fusion, which usually requires surgical intervention, among the much more common cases of positional deformity.

A plagiocephaly clinic was commenced in May 2005 in conjunction with a developmental paediatric outpatient clinic at Starship Children's Hospital, Auckland, New Zealand. It was the main venue for referrals of infants with positional head shape deformation in the Auckland region, which has an annual birth rate of approximately 22 000 (7). Referred cases do not represent all cases in the region as many are managed by general practitioners (GPs), child health nurses or neurodevelopmental therapists. This study is a retrospective review of all children attending the plagiocephaly clinic from its conception in May 2005 until August 2007. This study covers only data recorded at the first clinic visit, and does not cover follow-up head measurements or compliance with positioning strategies.

Abbreviations
ASQ, Ages and Stages Questionnaire; CI, cephalic index; CT, computed tomography; OCLR, oblique cranial length ratio; OFC, occipitofrontal circumference; ROM, range of motion; SAS, statistical analysis software; SD, standard deviation; SIDS, sudden infant death syndrome.

AIMS

The aims of this study were to:

(1) review the types and severity of plagiocephaly and brachycephaly cases;
(2) describe the demographic, maternal and infant characteristics of the infants;
(3) assess the study population for developmental delays.

METHODS

Infants with head shape deformation were referred to the clinic by their GP, well-child nurse, specialist or physiotherapist. No patient was referred to the clinic with a diagnosis of craniosynostosis. The parents answered a questionnaire covering demographic information, obstetric history, history of the plagiocephaly and current positioning strategies.

The parents also completed an Ages and Stages Questionnaire (ASQ) Second Edition (8) appropriate to the infant's age. The ASQ is a parent-completed developmental screening assessment tool for children 4 months to 5 years and assesses age-appropriate development in the communication, gross motor, fine motor, personal/social and problem-solving domains. Validation of the ASQ has shown that sensitivity for the 4-month ASQ was 51%, while specificity was 84%. For the 8-month ASQ, sensitivity was 78% and specificity 88% (8).

All assessments were performed by the same researcher (BLH). At the clinic visit, the infant was examined and an objective measurement of head shape made. The infant's weight, length and head circumference were recorded. Neck range of motion (ROM) was assessed by having the infant watch a colourful musical toy as it was taken to each side at eye level, from midline to above each shoulder, and noting if there was any limitation in active ROM. Passive ROM was also assessed, particularly in younger infants who were not yet visually fixing and following. The presence of a head tilt was assessed visually. Parents were also asked if they had earlier noted a limitation of neck function, as sometimes by the time of the clinic visit, the neck dysfunction had resolved. Neck dysfunction was defined as an observed head tilt or limitation in ROM or a parental report of a past definite difficulty turning in one direction.

The technique used to objectively measure the head shape is called HeadsUp™ and has been developed and used extensively by the authors. It has been fully described elsewhere (9). Briefly, a small stocking cap is placed on the infant's head to hold the hair down, and a soft stretchy headband is placed around the maximum occipitofrontal circumference (OFC). The band has movable markers to indicate the nose and ear positions. The band clearly defines the head shape in the OFC plane. A digital photograph is taken from above, and a custom-written computer programme called HeadsUp™ processes the resulting image. The midline (head length) is established by drawing from the nose position through the point that bisects the line between the ear positions and extending the line to the posterior of the head. The oblique cranial diameters are measured from above the frontozygomatic points cross-diagonally to points situated 40 degrees either side of the posterior midline (see Fig. 1). The main measurements used are the cephalic index (CI, the ratio of the maximum head width to the head length), which measures brachycephaly or central occipital flattening, and the oblique cranial length ratio (OCLR, the ratio of the longest to the shortest oblique cranial diameters), which measures plagiocephaly, or asymmetric head shape. If the CI is below 93% and the OCLR is below 106%, the head shape is deemed to be in the normal range. These cut-off points were established earlier (6,9).

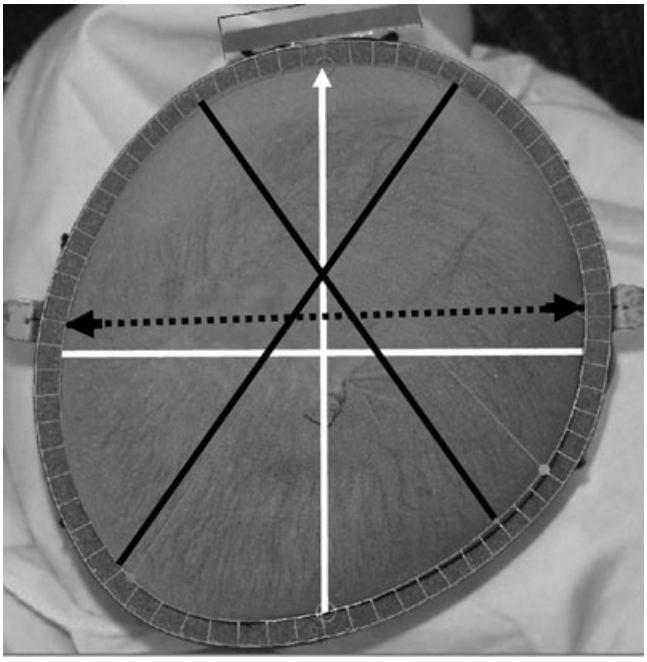

Figure 1 Example of heads-up photo. The centre of the nose is indicated by white arrow at top of picture; ear positions are indicated by black arrows at each side. White lines indicate head length and maximum width, dotted black line is ear alignment indicator and black lines indicate oblique cranial lengths. In this example, OCLR = 115.0 and CI = 96.0.

Cranial orthotic devices are rarely used in New Zealand to correct deformational plagiocephaly, and so parents were counselled regarding positioning strategies and were given written positioning protocols. Where necessary for diagnostic purposes, referrals were made to radiology services. Infants with torticollis were referred if necessary to paediatric physiotherapy services. Follow-up appointments for further head shape measurements or for developmental concerns were arranged as necessary.

Statistical analysis was performed using SAS (version 9.1, 2002–2003, SAS Institute Inc., Cary, NC, USA). The study was approved by the Northern X Regional Ethics Committee.

RESULTS

Over the first 27 months of clinic operation, 287 children were seen, having a median age of 22 weeks (interquartile range 16–29 weeks). The median age for the deformity being first noted by parents was 6 weeks (interquartile range 4–10 weeks).

In seven infants (2.4%) a computed tomography (CT) head scan with three-dimensional (3D) reconstruction of the skull was ordered on clinical grounds based on overall appearance and suspicious sutural ridging. Four showed coronal synostosis, and another had extra sutures within the parietal bone on the more prominent side (os parietale

Table 1 Characteristics of infants

Variable	n	% (% at NWH*)
Gender		
Male	197	68.6 (51.5)
Female	90	31.4 (48.5)
Ethnicity		
NZ European	171	59.6 (51.6)
Maori	38	13.2 (8.3)
Pacific	32	11.2 (14.1)
Asian	41	14.3 (22.7)
Other	5	1.7 (3.3)
Parity (missing = 1)		
Firstborn	178	62.2 (47.2)
Later born	108	37.8 (52.8)
Delivery (missing = 1)		
Normal vaginal delivery	139	48.6 (52.9)
Caesarean	90	31.5 (33.1)
Breech	2	0.7 (0.7)
Instrument assisted	55	19.2 (13.3)
Referred by (missing = 1)		
General practitioner	125	43.7
Specialist	27	9.4
Plunket nurse	105	36.7
Physical therapist	29	10.1
Type (missing = 2)		
Normal ('noncase')	62	21.8
Brachycephalic	47	16.5
Plagiocephalic	107	37.5
Both	69	24.2
Side of flattening (missing = 3)		
Right	181	63.7
Left	103	36.3
Neck dysfunction (missing = 3)		
Yes	165	58.1
No	119	41.9

*Compared with births at National Women's Health in 2006 (10).

divisum). The latter case was severe and progressive and was complicated by severe torticollis. Two of the seven infants had normal CT scans.

The infants' characteristics are listed in Table 1, and show a preponderance of males at 68.6% and firstborn infants at 62.2%. Fewer than half (48.6%) were born by normal vaginal delivery, and 19.2% were delivered by forceps or ventouse. Those with right-sided flattening comprised 63.7%. There was a wide range of head measurements ranging from normal to very severe deformities (Fig. 2). Although deformity is on a continuum, we suggest cut-off levels to indicate mild, moderate, severe and very severe cases. Those on the upper and outer extremities of the chart are the most severe cases. Thus, 21.6% of head shapes were within normal limits ('noncases'), 15.3% were mild cases, 24.7% were moderate, 18.8% were severe and 18.8% were very severe.

Cephalic index ranged from 73.8 to 107.6; the mean being 91.2 (SD 6.8). OCLR ranged from 100.2 to 121.0, with a mean of 107.6 (SD3.9). The mean diagonal difference was 10.0 mm (SD 4.9, range 0–28.7 mm). Of the cases (i.e. those with measurements outside the normal range), 21% were brachycephalic only, 48% were plagiocephalic only and 31% were both brachycephalic and plagiocephalic. The mean and standard deviation CI and OCLR for each group are shown in Table 2.

In the first 6 weeks of life, 92% of infants had slept only on their backs. There was a significant difference between the cases and the noncases, with 94% of cases sleeping on their backs and 84% of noncases doing likewise ($p = 0.0009$).

Neck muscle dysfunction was recorded in 58% of the infants (Table 1). These were either documented in clinic as having a current neck muscle dysfunction, which ranged from congenital muscular torticollis to a subtle imbalance of sternocleidomastoid function, or the parent gave a history of such neck problems in the infant's past. Seventy-two percent of the plagiocephaly-only group and 64% of the both plagiocephaly and brachycephaly group had a limitation of neck function, while only 26% of the brachycephaly-only group were similarly affected ($p < 0.0001$).

In infants aged 4 months or over, an ASQ was completed by the parent. For 64% of the children, there were no delays on the ASQ, but one delay was seen in 17% of infants and two or more delays were seen in a further 19%; hence, 36% had one or more delays. In those with a neck dysfunction, 41% had one or more delays, compared to 29% of infants with no neck problem who had one or more delays ($p = 0.08$). The activity level of those with one or more delays was significantly more likely to be described by parents as 'somewhat inactive' or 'average', rather than 'quite active' or 'very active', compared with those having no delays ($p = 0.008$).

The greatest number of delays was seen in the gross motor domain (18% of infants), followed by problem solving (17%), personal–social (15%), fine motor (14%), and communication (7%). Neither severity nor type of abnormality (i.e. plagiocephaly, brachycephaly or both) was associated with the presence of delays.

We found no significant difference in neck dysfunction for cases of male infants, instrument-delivered infants or firstborn infants. Those who had had an instrument-assisted delivery were not significantly different in age at referral, side of flattening or severity. No difference in delays was found between cases and noncases, or between instrument-delivered infants and others.

DISCUSSION

No child was referred with a diagnosis of craniosynostosis and yet we found five cases of abnormal sutural formation that had been missed by the referrer. Identification of craniosynostosis using a physical examination has been well described in the literature (11–13) and can be confirmed if necessary using a plain skull X-ray or CT scan. We feel CT scans are not needed for the majority of children with plagiocephaly, and thus concerns about the unnecessary adverse effects of radiation can be avoided with careful clinical examination (14).

Many other studies of deformational plagiocephaly have not differentiated between plagiocephaly and brachycephaly, or have only reported plagiocephalic cases, and this

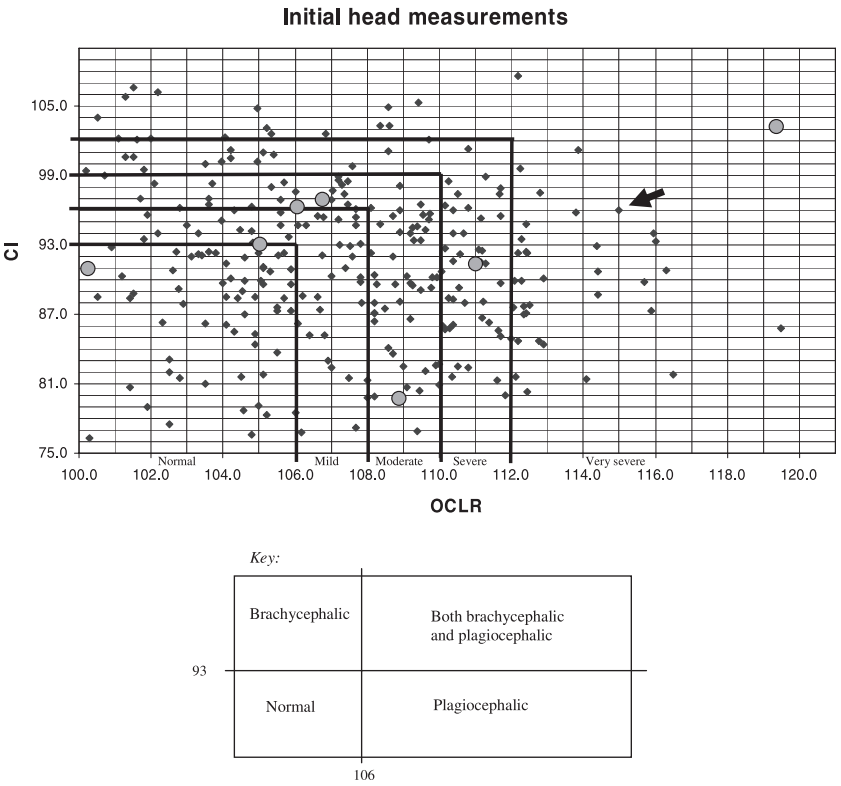

Figure 2 Head measurements of 287 infants. The black arrow points to the infant in Figure 1. Greyed circles are infants who had CT scans.

Table 2 Head measurements by type of deformity

Type	Mean CI (SD)	Mean OCLR (SD)
Brachycephaly only (n = 47)	98.7 (3.8)	103.4 (1.6)
Plagiocephaly only (n = 107)	86.7 (4.2)	110.3 (2.7)
Both (n = 69)	97.1 (3.2)	109.6 (2.6)

study has shown that parents who are concerned enough to seek a referral includes more than 20% whose infants are purely brachycephalic. This is similar to Teichgraeber et al.'s 2004 study (15) showing that 22% of cases were brachycephalic.

Compared with National Women's Health (NWH), the main obstetric hospital in the region, males, firstborns and instrument-delivered infants were more frequently seen. Other studies (1,3,5,16–18) have reported similar results. Right-sided flattening was more common than left sided and this accords with many other studies that have also demonstrated this characteristic (3,17–20). Various explanations have been offered regarding these phenomena, such as the effects of foetal constraint in utero and males having larger, faster-growing heads, but no study has yet confirmed these hypotheses.

More than half of our infants had a limitation of neck function, this being significantly more likely in those with plagiocephaly than those with brachycephaly (72% vs. 26%, respectively), and this confirms the strong association we and others have noted between plagiocephaly and neck dysfunction. Golden et al. (2) found a comparable proportion of plagiocephalic infants with neck dysfunction, while Captier et al. (21) similarly showed that brachycephalic infants demonstrated fewer neck problems than plagiocephalic infants. We did not collect hard data to show when the neck problems became manifest and thus we are unable to postulate on primary versus secondary neck dysfunction. We are of the impression that while congenital muscular torticollis is the initiating factor in the development of many cases of plagiocephaly, in other cases positional preference arising from repetitive positioning during feeding, sleeping and play may bring about a unilateral weakness in neck musculature that perpetuates the postural preference (2).

We had a higher number than expected with developmental delays on the ASQ, with 36% of children having delays in one or more domain. This was significantly higher ($p = 0.013$) than the percentage with one or more delays documented during validation of the ASQ (8). There may be several explanations for our result. Firstly, the ASQ was developed in 1980, with revisions in 1991, 1994 and 1997, before the widespread use of the supine sleep position. Nearly all of our infants were back sleepers, and it has been shown (22–24) that back-sleeping infants achieve motor milestones later than prone or side-sleeping infants, although there is some evidence they catch up by about 18 months of age. Lack of experience in prone play has been shown to affect developmental scores at 6 months of age (25). It is possible that these factors may help account for some of the

discrepancy. Other characteristics of the sample, such as different ethnicities, could possibly also contribute to these differences, but this effect is likely to be small.

Secondly, Schertz et al. (26), using the Alberta Infant Motor Scale, found that infants with torticollis had a high rate of gross motor delays, although most normalized by the age of 1 year. Infants with plagiocephaly are also more likely to have low or variable tone (4) or to be less active (3,6). Thirdly, gender may have an effect. More than two-thirds of our infants were male, and males have been shown to score lower on the ASQ than females in all except the gross motor domain (27).

To summarize, although others have noted developmental problems in infants with positional plagiocephaly (28–30) and have suggested that delays may be caused by the plagiocephaly, we suggest that in some infants developmental delay is the initiating factor. Further, our impression is that in many other infants with plagiocephaly there is a combination of events, such as supine sleeping, lack of prone play experience, low tone, low activity levels, male gender and neck muscle dysfunction that impacts not only their ability to reposition their heads, but also their early development, resulting in delays on developmental screening tools.

Most infants (92%) had slept supine for the first 6 weeks of life. This contrasts with 65% of infants in a 2005 New Zealand survey of normal children who slept only supine (31). That survey also showed that parents do change the sleep position to riskier positions due to head shape concerns and that a common reason for parents changing the sleep position from supine to side or mixed positions is their fear of head shape deformation. It is vitally important to maintain the advice to parents that they should sleep their infants on the back to help prevent SIDS. It is also important to educate parents about varying the head position from day 1 of the infant's life. If there appears to be a restriction or if the head returns repeatedly to a preferred position, then they should seek an opinion on the neck function, and commence physiotherapy if necessary. Most plagiocephaly improves with time, but prevention is important.

We acknowledge some limitations to this study. Being a retrospective review of clinic patients, we did not have a control group, although we have compared our results with those of others, such as ASQ validation studies and NWH birth data. Additionally, head shape measurements lie on a continuum and therefore our categories describing normal to very severe were arbitrarily assigned. We feel that in most cases they accurately describe the visual impression obtained on clinical examination.

Finally, as with many other studies using cephalometry, we acknowledge a lack of standardization of measurement technique in assessing infant head shape. HeadsUp™ has been developed by our study group in an attempt to obtain an objective measurement of head shape that is quick, well accepted by infants and their parents and easily used in the field. It is hoped that in the future, simple, safe and affordable 3D scanning systems will eventually provide better standardization of head shape measurement.

CONCLUSIONS

Physical examination of infants with head shape deformities is essential in order to rule out craniosynostosis. Infants with deformational plagiocephaly frequently have a neck muscle dysfunction. Deformational brachycephaly is common but is less likely to be associated with neck dysfunction. We suggest that developmental delays may be related to the effects of supine sleep position, low or variable tone, lower activity levels, male gender and neck muscle dysfunction. Further study is needed to determine whether these delays continue to be manifest in the long term.

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CONFLICT OF INTEREST

There are no conflicts of interest to declare.

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