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ORIGINAL PAPER

Long-term outcome of infants with positional occipital plagiocephaly

Paul Steinbok · David Lam · Swati Singh · Patricia A. Mortenson · Ashutosh Singhal

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Abstract
Background Despite much interest in positional (deformational) plagiocephaly, the natural history is unclear. The purpose of this study was to determine cosmetic and developmental outcomes at a minimum of 5 years of age in children diagnosed in infancy with positional plagiocephaly (PP) and the impact of cranial orthotic use.
Methods A questionnaire survey was sent to parents of children diagnosed with PP in infancy and now aged more than 5 years. A retrospective review of the child's clinic chart was performed of consenting families, and prospective follow-up was done when families agreed to return for assessment.
Results Of 278 eligible children with plagiocephaly, questionnaires were completed by 65 parents, and 27 brought their child for assessment. Participants and nonparticipants were similar. Cranial orthoses were used in 18 of 65 children. Parents perceived the cosmetic appearance of their child as "very abnormal" in 2, "mildly abnormal" in 25, and "normal" in 38. Residual asymmetry was noted by parents in 58%, but only 21% were concerned about appearance. In the last year, 7.7% of children commented about asymmetry of head, and 4.6% were teased occasionally. Thirty-three percent had received learning assistance, and 14% were in a special class. Long-term outcomes, as perceived by the parent or child, were no different between children with and without orthosis use.
Conclusions The results allow better counselling of parents about outcome of infants with PP, reducing anxiety, and allowing more rational selection of management modality.

Keywords Occipital plagiocephaly · Positional plagiocephaly · Outcome · Headband · Orthotic · Counter-positioning

P. Steinbok · D. Lam · S. Singh · A. Singhal
Division of Neurosurgery, Department of Surgery,
University of British Columbia,
Vancouver, British Columbia, Canada

P. Steinbok (✉)
Division of Paediatric Neurosurgery,
4480 Oak St, #K3-159,
Vancouver V6H 3V4 British Columbia, Canada
e-mail: psteinbok@cw.bc.ca

P. A. Mortenson
Department of Occupational Therapy,
Children's and Women's Health Centre of British Columbia,
Vancouver, British Columbia, Canada

Introduction

The incidence of positional plagiocephaly (PP) has been reported in the past at 1 in 300 live births [28]. However, since 1992, when the American Academy of Pediatrics recommended that infants sleep on their back to help prevent sudden infant death syndrome, the incidence of this condition has increased dramatically, and recent reports indicate an incidence of up to 15% in infancy [11, 16]. Interest in this condition has also increased among the medical profession and the lay public. Between 1980 and 1994, there were four published MEDLINE indexed articles containing occipital plagiocephaly or PP or posterior plagiocephaly as a keyword, but from 1995 to the end of November 2006, there were 79 such publications. The problem has been publicized widely in the lay media, particularly on the Internet. Indeed, a simple Google search on the term "plagiocephaly" in June 2006 led to more than 156,000 'hits', ranging from parent support groups, petitions, advertising orthotic companies, hospital pro-

grams, and companies selling related products, such as sleep positioners, T-shirts, and coffee mugs.

Although some studies have suggested impacts on infant/parent bonding, self-image, and development of motor skills [6, 19, 24], the problem is considered to be almost completely cosmetic. Nonetheless, stimulated in large part by the media and Internet attention to the disorder, significant anxiety exists among parents, many of whom demand some form of therapeutic intervention [4]. Reported interventions include preventative counselling, physiotherapy to assess and treat those with an associated torticollis, active counter-positioning, the use of orthotic helmets or headbands [25], and in rare severe cases, surgical reconstruction of the occiput [10, 27].

In active counter-positioning, parents are guided to position the baby "back to sleep," but to turn the baby's head to the side opposite the occipital flattening. Supported upright positioning and supervised tummy time are recommended as preferred awake and play positions. Active counter-positioning is the easiest therapy to institute, is the least traumatic to the child, and has no associated cost. Another more interventional approach is to utilize a custom made helmet or headband designed to apply pressure to the prominent aspects of the asymmetric head and room to grow for the flattened parts. The rationale for using such a cranial orthotic device is that the prominent areas of the occiput will become flatter, and with growth, the flattened parts will round out, and that this may be more effective than simple counter-positioning. Orthotic treatment typically requires that the infant wear the device for 23 or more hours a day for 3 to 6 months. The device may not be well tolerated by some infants and may cause some degree of discomfort to the child and anxiety to the family. In addition, there is a significant cost of the device, which varies from $1,000 to more than $3,000 US. Furthermore, correct fabrication of the headband or helmet requires an orthotist with specific expertise. The limited number of qualified orthotists may create difficulties in initial access and in obtaining appropriate follow-up to have adjustments made in the device and to make sure that excessive pressure is not being placed on the skin.

Despite the large amount of attention being given to the problem of PP, there have been no formal population studies investigating the natural history of the disorder, and studies investigating the effectiveness of interventions have revealed mixed results [4, 21, 28]. In particular, the effectiveness of orthotic devices has not been proven [4]. A variety of orthotic devices have been developed, including helmets, headbands, and many variations [1, 5, 7, 13, 15]. A number of studies [5, 8, 13, 14, 22, 26, 29–32] have presented weak evidence to indicate a benefit to the use of orthotic treatment in the short term. Other studies [9, 18, 21, 23, 27] have suggested similar improvements using repositioning strategies. Up to now, there have been no randomized trials or even open studies with controls [2, 4], and all studies have looked at relatively short periods of follow-up of a few months to 2 years.

There is no information available about what happens to the head shape in the longer term, with or without any intervention. As the problem is primarily cosmetic, parents are particularly anxious about what their child's head shape will be like by the time the child is ready to go to school at about 5 years of age. Parents wish to know whether the head will round out to a normal shape and, more importantly, whether or not the child will have a normal cosmetic appearance or have an unusual head shape and be at risk for teasing by peers. The lack of this type of information makes the decision-making process with respect to different therapies difficult for parents.

Another issue that has been raised intermittently has been the possible relationship of PP to developmental delay in the child [12, 19, 24]. This is of concern to parents and to physicians, but there is only limited information in this regard, and the issue tends to be avoided in discussions with parents.

The primary purpose of this study was to obtain information about cosmetic outcomes at a minimum of 5 years of age in children diagnosed in infancy with occipital PP. Additional objectives were to describe developmental outcomes for this population and determine if there was any difference in outcome at age 5 years or later between children treated with an orthotic device (helmet or headband) and those treated with counter-positioning.

Methodology

The research protocol was approved by the Ethics Committee of the University of British Columbia. The neurosurgical clinical database at BC Children's Hospital was searched to identify children with occipital PP seen by the neurosurgical service before January 1999. A retrospective review of these records was done to extract data from the time of the first initial neurosurgical assessments conducted on infants ranging in age from 3 to 16 months of age. This age range was chosen, as it represented the age range during which helmet/headband therapy might be instituted. Data were collected on two anthropometric measures of the extent of asymmetry: (1) the cranial vault asymmetry (CVA), which is measured from the lateral orbital margin to the contralateral occiput, and (2) the fronto-orbital asymmetry (FOA), which is measured from the lateral orbital margin to the contralateral tragus. The cosmetic appearance of the child, as documented in the records, from the front and top of the head was noted. In addition, data

were obtained as to whether or not the child was treated with helmeting or headbanding and if there were any comorbid developmental delays or risk factors for developmental delay.

A questionnaire and covering letter were mailed to parents of the children diagnosed previously with PP (Appendix 1 and 2). The parents were invited to participate in the study. If they agreed to participate, they were requested to sign the attached consent form and complete the questionnaire survey. A second and final mailing was made to those parents who did not return the questionnaire. The questionnaire addressed issues related to the cosmetic appearance of the child, the impact of any cosmetic abnormality on the child's life, and the intellectual and behavioral function of the child. Parents were asked to indicate whether they would be prepared to accept a follow-up telephone call by the investigators if further clarification of information was required as part of the study. Parents were also asked if they would be prepared to bring their child for assessment at a special clinic. Those parents who agreed to bring their children for a clinic appointment were contacted with a convenient clinic time, and the child was assessed in the clinic. At this clinic, anthropometric measurements, namely, CVA and FOA, were made of the child's head to determine the extent of asymmetry. These measurements were done using high-precision calibrated spreading and sliding GPM brand calipers. In addition, the cosmetic appearance from the front, side, and top of head was noted by an independent assessor, who was not part of the study. This assessor examined the child from five views: front, back, left profile, right profile, and from above and then documented the extent of asymmetry, using a three-point Likert scale.

Analysis

The data were entered into an Excel spreadsheet and then exported to SPSS. The primary and most important endpoint was the overall cosmetic appearance, as indicated by the parent at the time of completion of the questionnaire. For the subset of subjects who attended the long-term follow-up clinic appointment, the overall appearance, as assessed by the independent observer, was correlated with the parent's response on the questionnaire, to gauge whether the parental sense of the cosmetic issue was consistent with the impression of an independent (layperson) observer. Secondary outcomes included parent's assessment of residual asymmetry; parent's concern about the child's head shape; child's concern about his/her head shape; comments from others about the child's head shape; teasing about the child's head shape; asymmetry of CVA and FOA at latest assessment; the change in CVA and FOA and change in cosmetic appearance from infancy to latest follow-up; the impact of helmeting/headbanding and the initial severity of the plagiocephaly on late outcome; and the incidence of learning problems in this cohort. For the purposes of statistical analysis, CVA difference between the two sides of <3 mm was considered to be normal, CVA difference of ≤12 mm to indicate mild to moderate asymmetry, and CVA difference of >12 mm to indicate severe asymmetry. An FOA difference of ≤2 mm was considered to be normal and >2 mm to be abnormal.

For the second objective of the study, multivariate statistical analyses (such as logistic regression) were performed to determine the causal relationship between independent variables (such as treatment group) and each of the outcome measures, controlling for age and initial severity of plagiocephaly.

Results

Questionnaires were sent to parents of 278 children with plagiocephaly at their last known address. Of these, 85 were returned as undeliverable. One hundred and twenty-four questionnaires were not returned (presumably delivered), three parents declined participation in the study, and one child under the age of five was excluded, for a total of 128 nonparticipants. Questionnaires were completed by 65 parents (23% response rate), and of these, 27 also came with their child for an assessment at the neurosurgical clinic. In some of the questionnaires, some of the questions were not answered, so that the number of responses for some questions was fewer than the total number of questionnaires completed. There were no differences between the four groups (undeliverable, nonresponders, participants–questionnaire only, and participants–questionnaire and follow-up visit) with respect to the age at initial visit (ANOVA, $p=0.2173$), overall cosmesis at initial visit (Wilcoxon test, $p=0.1623$), initial CVA (ANOVA, $p=0.4188$), initial FOA (ANOVA, $p=0.4059$), or use of a helmet/headband (chi-square analysis, $p=0.1275$). These comparisons are summarized in Table 1. Relatively more of the participants who agreed to return to the neurosurgical clinic for follow-up had used helmets or headbands (39.3% vs 22.0% in patients who did not return to clinic). However, as noted above, this was not statistically different.

The mean age of the children at the time of the completion of the questionnaire was 8.9 years (standard deviation 3.8 years, range 5.0 to 18.5 years).

At the baseline assessment during infancy, the overall appearance for the 65 patients for whom questionnaires were returned, as judged by the treating neurosurgeon, was severely asymmetric in 7 and moderately asymmetric in the remaining 51 for whom information was available. Five of

Table 1 Comparison between undeliverable, nonresponder, questionnaire only, and questionnaire and follow-up visit groups

	Undeliverable	Nonresponders	Questionnaire only	Questionnaire and follow-up visit
Age (initial visit, in months, with standard deviations)	6.8 (2.4)	7.2 (2.8)	7.1 (2.8)	8.2 (3.2)
Median overall cosmesis (initial visit)	Category 2	Category 2	Category 2	Category 2
Mean CVA (mm, with standard deviations)	13.21 (4.66)	11.70 (4.92)	10.62 (4.66)	12.50 (4.77)
Mean FOA (mm, with standard deviations)	6.03 (5.61)	4.31 (4.23)	3.67 (2.46)	4.00 (2.77)
Use of orthotic (helmet or band)	22%	24%	17%	41%

the 7 with severe asymmetry (71%) and 12 (24%) with moderate asymmetry were treated with helmets or headbands. The rest of the children were managed by their parents according to advice provided at the clinic about positioning and developmental strategies.

The primary outcome was the overall cosmetic appearance of the child as judged by the parents. The parents perceived the overall cosmetic appearance of their child to be "very abnormal" in 2, "mildly abnormal" in 25, and "normal" in 38 (65 questionnaire responses). When asked if they had had any concerns about the shape of their child's head in the past year, of 64 responders, 4 indicated a high level of concern, 10 minimal concern, and 50 no concern. Residual asymmetry at the back of the child's head was felt to be significant in 3 patients, mild in 36 patients, and absent in 26 patients. In the last year, three children had commented to a parent about being teased at school because of the asymmetric appearance of the head, and all were teased only occasionally. Fifty-nine of the 62 responders to this question indicated that their child had not commented about being teased. Only 5 children out of 64 responders had commented to a parent about an asymmetric appearance of his/her head (all 5 indicated "occasionally" as the frequency). In the past year, 8 parents, out of 64 responses, indicated that they had received comments from others regarding head asymmetry (all 8 indicated "occasionally" as the frequency).

Helmets or headbands had been used in 18 of the 65 children for whom questionnaires were completed. Of the 18 with helmets or headbands, 14 parents perceived that the orthotic device had helped, "quite a bit" in 10 and "a little" in 4. There was no significant difference between children with and without helmet/headband use with respect to the parent's perception of the overall cosmetic deformity (Pearson chi-square statistic, 0.09 [p=0.7642]). There was no apparent difference (no statistical analysis done) between children with and without helmet/headband use with respect to the concern of the parent about the shape of the child's head, the likelihood of the child being teased, the extent of residual asymmetry as noted by the parent, or the likelihood of others commenting about the child's asymmetry of the head (Table 2). There were two children who were felt by the parent to have a "very abnormal" overall cosmetic appearance, and both had been treated with a helmet or headband. One of the 18 (6%) parents of children who had received helmets/headbands was "very concerned" about the child's appearance, and 4 of the 47 (9%) parents

Table 2 Comparison of questionnaire responses in patients with cranial orthotic vs counter-positioning

	Cranial orthotic used	Counter-positioning only
Parent concern for shape	14 (78%) none	36 (77%) none
	3 (17%) minimal concern	7 (15%) minimal concern
	1 (6%) significant concern	4 (9%) significant concern
Parent perception of deformity	10 (56%) normal	28 (60%) normal
	6 (33%) mildly abnormal	19 (40%) mildly abnormal
	2 (11%) very abnormal	0 (0%) very abnormal
Child teased at school	14 (82%) never	45 (100%) never
	3 (18%) occasionally	0 (0%) occasionally
	0 (0%) frequently	0 (0%) frequently
Residual asymmetry (as per parent)	8 (47%) none	17 (36%) none
	9 (53%) mildly asymmetric	27 (57%) mildly asymmetric
	0 (0%) significantly asymmetric	3 (6%) significantly asymmetric
Others commented upon asymmetry	14 (82%) never	42 (89%) never
	3 (18%) occasionally	5 (11%) occasionally
	0 (0%) frequently	0 (0%) frequently

of children with no helmet/headband use were "very concerned."

Twenty-seven children and parents returned to the neurosurgical clinic for assessment. The overall appearance of the child, as judged by an independent observer was felt to be normal in 16, moderately asymmetric in 7, and severely asymmetric in 1. Three children were not assessed by the independent observer. The one severely asymmetric child had been treated by a cranial orthotic. When comparing the observer's impression of appearance with the parent's perception of cosmesis, in 15/24 patients, there was agreement on the appearance (11 normal, 5 mildly asymmetric). In six patients, the parent thought that the child had mild asymmetry, and the observer thought the child was normal, and in two patients, the reverse was true. In one patient, the parents thought the child was mildly asymmetric, and the observer thought the child was severely asymmetric.

Among patients who participated in the questionnaire portion of the study, 27/65 had baseline CVA. The mean baseline CVA difference between the two sides was 11.6 mm (standard deviation of 4.7), with 1 patient <3 mm (normal), 14 patients with CVA 3–12 mm (mild to moderate asymmetry), and 12 patients with CVA >12 mm (severe asymmetry). The mean baseline CVA in the patients receiving orthotic treatment was 13.88 mm, compared to 10.63 mm for the group treated with counter-positioning only (unpaired t test: $t=1.685$ [$p=0.104$, two-tailed]). Sixteen of 26 children had abnormal baseline FOA measurements (>2 mm).

CVA and FOA were measured in all 27 children who returned for clinical assessment. The mean CVA difference between the two sides at follow-up was 8.29 mm (standard deviation 5.55, range 0–26.7 mm), with six patients <3 mm (normal), 16 patients 3–12 mm (mild to moderate asymmetry), and 5 patients >12 mm (severe asymmetry). The mean FOA difference was 2.78 (standard deviation 2.29, range 0.3–9 mm), with 12 children measured having an abnormal FOA difference of >2 mm.

In a smaller subset of children, data were available to allow a determination of the change in overall anthropometric measurements from baseline, including 14 children with initial and follow-up CVA measurements, and 13 children with initial and follow-up FOA measurements. In nine children, CVA measurements improved, with a range of 1–20.3 mm of improvement. In four children, CVA measurements worsened (range 1.6–4.6 mm), and in one child, the CVA measurement was unchanged. In eight patients, the FOA measurements improved (range 0.3–6.7 mm), in three, it worsened (range 1.3–7 mm), and in two children, the FOA measurement was unchanged. The mean improvement in CVA from baseline to last follow-up was 1.1 mm for the counter-positioning group compared to 6.57 mm for the children treated with orthotics (unpaired t test: $t=1.3988$ [$p=0.187$, two-tailed]).

In 27 children, it was possible to compare CVA with parental impression of cosmesis. In the 14 patients whom the parents considered "normal," with regards to head cosmetic deformity, the mean CVA was 6.84 mm (range 0–13.7 mm, standard deviation 4.32), compared with a mean CVA of 9.85 mm (range 1–26.7 mm, standard deviation 6.43) in patients in whom the parents indicated "mildly abnormal" head cosmesis (unpaired t test: $t=1.44$ [$p=0.162$, two-tailed]).

In 24 children, it was possible to compare CVA with the observer's impression of cosmesis. In the 16 patients whom the observer considered "normal," the mean CVA was 6.83 mm (range 0.3–13.7 mm, standard deviation 3.84), compared with a mean CVA of 12.03 mm (range 2–26.7 mm, standard deviation 7.08) in the 8 patients that the observer considered moderately asymmetric (7) or severely asymmetric (1; unpaired t test: $t=2.35$ [$p=0.028$, two-tailed]).

All children were or had been in school. The grade level varied from Grade 1 to Grade 12, and one child had completed Grade 12. Thirty-one children were at their appropriate grade level for age, and four were at a lower grade level. Nine children of 64 responses (14%) were in a special education class, and 21 children out of 62 responses (34%) had received learning assistance.

Retrospective review of possible infant developmental delays and risks factors was completed for 63 of the 65 patients. In total, eight patients (13%) were found to have identified delays or risk factors, with two of the eight having more than one risk factor. Of these, four were born prematurely at <36 weeks, three were diagnosed with neurological impairment (microcephaly, atrophic brain development, hydrocephalus), two were diagnosed with genetic syndromes, and one had suffered a possible episode of anoxia at birth. Two patients were identified at the time of initial assessment as having unexplained developmental delays.

Discussion

In a systematic review of the available literature on nonoperative treatments for PP, Bialocerkowski et al. [2] commented that for what is primarily a cosmetic problem, "outcome measurement should reflect parents' perceptions of the cosmetic appearance of their infants, rather than relying solely on clinically measured physical variables." They commented further that standardized validated outcome measures for cosmetic outcomes had so far not been developed. There have been some studies that have looked at parents' perception of cosmesis using one or other non-validated measure. [23, 26, 32], but these have all been

conducted at less than 2 years of age. The longer-term cosmetic outcome is what parents are more interested in and what should determine the utility of any intervention, as this is what will matter to the child as he/she goes to school and grows to be an adult.

In the present study, outcomes of children diagnosed in infancy with PP were assessed when the child had reached a minimum of 5 years of age and were in at least Grade 1 in school. It was remarkable that although parents perceived the overall cosmetic appearance of their child to be abnormal in 27 of 65 (42%) and recognized that there was still asymmetry of the head in 39/65 (60%), only 23% (15/65) of parents indicated that they had any concern about the cosmetic appearance, and only 5 of these 15 patients were very concerned. From the child's perspective, 3 of 62 (5%) indicated that they had been teased about their head shape (all teased only occasionally), and 8% (5/64) had actually commented to the parent that they had noted that their head was asymmetric. Thus, in the vast majority of the children, there was no or minimal concern about any residual plagiocephaly on the part of the child or parents. These findings are in keeping with the report of Hutchinson et al. [11] who studied 200 children with plagiocephaly and noted that no parent expressed concern about their child's head shape at 2 years of age.

In this study, one of the secondary questions was whether or not there was a difference in the outcome for those patients who were treated with a helmet or headband compared to those not treated in this manner. All parents of children with an orthotic device felt that the device had improved the head shape in the short term. However, at the time of the questionnaire survey, there was minimal or no difference between children with and without helmet/headband use with respect to the parent's or child's perception of the cosmetic outcome, likelihood of being teased, or degree of residual asymmetry. The only two children who were felt by the parents to have a "very abnormal" overall cosmetic appearance were in the group that had been treated with a helmet or headband. This almost certainly reflects the fact that there was a bias towards treating the more severely affected infants with orthoses. Indeed, the mean baseline CVA in the group treated with orthotics tended to be higher than in the group treated by counter-positioning (13.88 mm vs 10.63 mm), although this difference did not reach statistical significance. Only one parent of a child with helmet/headband was "very concerned" about their child's appearance. Parents of four children, not treated with an orthotic device, indicated that they were "very concerned" about their child's cosmetic appearance. One probable explanation for these findings is that parents, who opted for orthotic treatment, felt that they had done everything possible for their child and, therefore, accepted the residual cosmetic deformity, even when it was significant. On the other hand, parents who had not opted for orthotic treatment may harbor some guilt at not having done everything possible to help their child and, therefore, may continue to be concerned about any residual cosmetic abnormalities. These parents may think that the child may have been better had they opted for treatment with a helmet or headband. This possible explanation has to be considered in any discussion about the pros and cons of headbanding or helmeting.

If one accepts that, with few exceptions, parents and affected children express little concern about the cosmetic appearance by the time the child goes to school, one has to question why this might be the case. It could reflect an improvement in the degree of plagiocephaly, but it could be related to other factors, such as more hair on the head or a relative decrease in the ratio of asymmetry between the two sides with continued growth, although the CVA, measured as an absolute difference, has changed little. In this study, parents indicated that the appearance of their child's head still looked abnormal in 42% and that there was still asymmetry of the head in as many as 58%. In the small group of children, who came back to have anthropometric measurements done after the questionnaire had been completed, the absolute CVA (difference between the two sides) was a mean of 8.29 mm (range 0–26.7 mm). There was a trend toward more improvement in CVA measurements from infancy to the time of the study in those who had received a helmet or headband (mean of 6.57 mm better) than those not so treated (mean of 1.1 mm better). The difference did not reach statistical significance, but the number of patients under consideration was very small. One of the caveats about these data is that the interobserver variability for the measurements of CVA in infants was shown to be very large in a previous study in this center [20], with the absolute CVA varying between two observers by a mean of 2.2 mm with a standard deviation of 6.5 mm. Thus, the measurements used in these anthropometric assessments may be unreliable. It is possible that in the older children, who are more cooperative, measurements may be more precise. However, the posterior landmark for the measurement of CVA, namely, the most prominent part of the occiput vs the flattest part, is somewhat subjective.

The findings of residual asymmetry in this study are consistent with reports in the literature looking at this in the shorter term. Boere-Boonekamp and van der Linden-Kuiper [3] reported that 47% of 259 infants with plagiocephaly had persistent occipital asymmetry at 2 years of age. Hutchison et al. [11] followed a group of 200 "normal" children serially up to 2 years of age to determine the prevalence of plagiocephaly, defined by the ratio of the measurements between the two sides, as opposed to the absolute difference, as used in the current study. When they used a CVA ratio of ≥106% to indicate the presence of plagio-

cephaly, they noted a prevalence of 9% at 4 months of age, decreasing to 2% at 2 years of age. Thus, 22% of children diagnosed at 4 months with plagiocephaly had asymmetry with a CVA ratio of >106% at 2 years. When limits for plagiocephaly were changed such that a CVA ratio of ≥105% was said to indicate plagiocephaly, the prevalences at 4 months and 2 years were approximately 15 and 8%, respectively. Using these parameters for defining plagiocephaly, 53% of children diagnosed at 4 months with plagiocephaly continued to have asymmetry at 2 years. Losee and Mason [17] have indicated, without any detail, that they had evaluated a cohort of children aged 2 to 5 years, who presented with "persistent craniofacial asymmetries" and concluded that "not all children self-correct." It would appear from this study and from other reports [3, 11, 17] that residual asymmetry is present commonly, but the asymmetry measured as a ratio between the two sides decreases. This may, in part, reflect the fact that the head has increased in size, so that even if the absolute difference in measurements between the two sides was unchanged, the ratio would be less.

One of the other concerns that has been expressed is the possibility that PP may cause developmental delay. Miller and Clarren [19] reported that 25 of 63 children (39.7%) with persistent deformational plagiocephaly had received special educational services. By comparison, only 7 of 91 siblings (7.7%), serving as controls, required similar services. In another report, Panchal et al. [24] assessed infants with PP using Bayley Scales of Infant Development-II and noted that both the mental developmental index and the psychomotor developmental index scores were significantly different from the expected norms. More recently, Kordestani et al. [12] examined infants with PP and found that there were significant delays in mental and psychomotor development. Whether or not that persisted in later life is not known. The delays in the Kordestani study were almost entirely explained by "confounding" variables (e.g., premature birth, failure to thrive, illness, etc.—other factors that would explain a delay). In addition, they used the Bayley II norms as a comparison—this test has since been revised and has new norms (the Bayley III).

As this present study was a retrospective review, standardized developmental information is not available for the study subjects. However, five subjects (8%) had a comorbid diagnosis consistent with developmental delay at the time of their initial plagiocephaly assessment, and three others (5%) had risk factors for possible delays. Only two subjects (3%) had unexplained delays at initial assessment. At long-term follow-up, nine subjects (14%) were in a special education class. This is slightly higher than the average of 10.2% for children classified as requiring special needs assistance in schools in the province of British Columbia, where this study was carried out. The percentage of children requiring special assistance in this study was significantly lower than the 39.7% noted in a prior report by Miller and Clarren [19]. While this study was not primarily designed to answer questions regarding the relationship of developmental delay and plagiocephaly, it does raise questions of whether plagiocephaly is indeed related to developmental delays, and if so, which causes which. Does the plagiocephaly place children at higher risk for delays, or rather, are children with preexisting delays and risk factors more prone to plagiocephaly because they are not moving as quickly and are spending more time on their backs?

Limitations of study

There are a number of limitations in this study. The response rate to the questionnaire was low, and the number of children returning for follow-up was small. The small numbers in the study limited the ability to do statistical analyses and also creates a significant potential for a type II error in those statistical analyses that were done. Thus, although we were not able to show statistical significance in any of the analyses performed, it is possible that with a larger study group, there could be a statistically significant difference in some of the comparisons, particularly where there seemed to be a trend. Another limitation is the inaccuracy of the CVA measurements, which has been mentioned previously.

Importance

At present, advising parents of infants with occipital PP about the various options and what to expect in the future is difficult, as the long-term results of any management protocol are not known. The results allow better counselling of parents about outcome of infants with PP, reducing anxiety for these parents, and more rational selection of management modality.

Appendix

A.1 Cover letter to parents

Dear Parent,

According to our records, your child (name of Child) was assessed for positional plagiocephaly many years ago at the Neurosurgery Clinic at BC Children's Hospital. One of the questions that many parents asked was what to expect with the head asymmetry in the future. Up to now, we have no answer to the question, and we are now doing a

study to try and answer this question. The study will look at what has happened to children with positional plagiocephaly as infants after many years, when the child has reached school age. I hope that you will help to answer this important question by participating in the study. If you agree to participate, I would ask that you complete the attached questionnaire, which should take about 5 minutes to complete.

I would also ask that you sign the attached consent form, which indicates your agreement to participate in this study.

After completion, please mail the questionnaire and signed consent form back in the self addressed stamped envelope.

If you have indicated your agreement on the questionnaire form, my assistant or I may contact you by telephone to clarify any answers. If you have indicated on the questionnaire form your willingness to return to BC Children's Hospital for a special assessment of your child, my assistant will contact you to set up an appropriate appointment. At the visit to the hospital clinic, an independent observer will look at your child's head and chart whether or not the head shape looks normal, mildly asymmetric or significantly asymmetric. Dr Steinbok or his assistant will measure the extent of asymmetry using special calipers. The assessment should take about 10 minutes. If you do bring your child to BCCH for assessment as part of this study, we may be able to provide financial assistance of $75.00 for patients coming from Vancouver and the Lower Mainland, and up to $300.00 for those coming from outside of the Lower Mainland. This will depend on whether or not we have been successful in obtaining funding for this study.

A.2 Questionnaire

Plagiocephaly Questionnaire

Name of child: Date of Birth:

If you do not wish to complete this questionnaire or participate in this study, please circle NO here and return the uncompleted questionnaire in the stamped envelope, so that you do not receive a second mailing.

If you wish to participate, please answer the following questions and then return questionnaire in stamped envelope.

Qn1. In the last year have you had any concern about the shape of your child's head?

1-No 2-Yes-minimal concern 3-Yes-significant concern

Qn2. Indicate what the cosmetic appearance of your child's head is like to you

1-Normal 2-Mildly abnormal 3-Very abnormal

Qn3. Irrespective of how you answered the last question, do you think that there is any residual asymmetry at the back of your child's head, for example if you feel the back of the head?

1-No 2-Yes-mildly asymmetric Yes-significantly asymmetric

Qn4. In the last year, has your child commented to you about being teased at school because of the asymmetric appearance of his/her head?

1-Never 2-Occasionally 3-Frequently

Qn5. In the past year, has your child commented to you about an asymmetric appearance of his/her head?

1-Never 2-Occasionally 3-Frequently

Qn6. In the past year, has any one commented to you about an asymmetric appearance of your child's head?

1-No 2-Yes

Qn7. What grade is your child in at school?

Qn8. Is this the normal grade level for his age?

1-No 2-Yes

Qn9. Is your child in a special class?

1-No 2-Yes

Qn10. Has your child received learning assistance at school?

1-No 2-Yes

Qn11. Did your child receive treatment with a helmet or headband?

1-No 2-Yes

Qn12. If yes to helmeting or headbanding, do you think that the helmeting/headbanding improved your child's head shape?

1-No 2-Yes-a little 3-Yes-quite a bit

Qn12. Are you agreeable to having Dr Steinbok or his assistant telephone you if any additional information or clarification is required?

1-No 2-Yes-Best telephone number is:

Qn13. Would you be prepared to bring your child to a clinic at BCCH to have your child's head observed and measured with calipers?

1-No 2-Yes-Best telephone number for Dr Steinbok's assistant to contact you is:

Date completed:

Name of person completing questionnaire:

Relationship to Child:

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