

## Best Evidence Statement (BESt)

**Date posted:** 6-3-11

### **Title: Prognosis of Infant Development with Plagiocephaly, Torticollis**

Will my child's head shape affect his/her development?

#### **Clinical Question**

- (Population) Among infants with torticollis and positional plagiocephaly  
(Outcome) does severe plagiocephaly compared to less severe plagiocephaly predict developmental delay?

#### **Target Population** Infants with torticollis and positional plagiocephaly

Definitions:

1. Positional Plagiocephaly is an asymmetric head shape caused by positioning, as opposed to an asymmetric head shape caused by craniosynostosis. This is also called deformational plagiocephaly (DP). Many infants who are referred to physical therapy for torticollis also have positional plagiocephaly.
2. Severe Plagiocephaly is plagiocephaly measurements that warrant recommendation for helmeting

#### **Relevant CCHMC policies / procedure:**

CCHMC Guideline 33, Congenital Muscular Torticollis, pages 1-13

#### **Recommendation** (See Table of Recommendation Strength following references)

It is recommended that parents wishing to know if their child's head shape will affect development, be provided the following prognostic information:

- that plagiocephaly alone is not a predictor of developmental delay
- in very young infants (average age 22 weeks), developmental delay appears related to sleep position, muscle tone, activity level, male gender and neck dysfunction

(Hutchison, L. B., Hutchison, L. A., Thompson, J. M., & Mitchell, E. A. (2004[2a]; Hutchinson, B. L., Stewart, A. W., & Mitchell, E. A. (2009) [4a]; Hutchinson B. S., 2010 [2a]).

#### **Discussion/summary of evidence:**

Developmental outcomes in the short term (from 6 weeks until 2 years of age) and in the long term (from 2 years through 18 years of age) for children with plagiocephaly were reported in ten articles from 2000 until 2010.

Short term effects:

In 2001, Panchal et. al [4a] found that of 42 consecutive infants diagnosed with DP, no infants tested in the accelerated range as opposed to 11% seen in the general population, yet more of these infants than expected tested within the normal range using the Bailey Scales of Infant Development, 3<sup>rd</sup> Edition (BSID III). Kordestani et. al [4a] found similar results when prospectively studying 110 infants between 1997 and 2003. In a 2004 prospective cohort study by Hutchinson et al [2a], 181 infants were assessed with the Revised Denver Prescreening Questionnaire. The only age at which the percentage of infants with developmental delays were higher in infants with DP (6.3%) compared to those without DP (1.8%) was at 6 weeks of age.

In 2008 Fowler et.al [4b] compared 49 infants with plagiocephaly with 50 infants without plagiocephaly. The average age of both infants was 8.1 months. Development was assessed with the ASQ; muscle tone and behavioral state was assessed with the Hammersmith Infant Neurological Assessment. Infants with DP scored lower on the ASQ, but this was only statistically significant for personal/social skills. Infants with DP were also more likely to have abnormal muscle tone than those without DP. In 2009 Kennedy et. al [4a] paired 27 infants with DP to 27 age, gender and race-matched infants without DP. The Alberta Infant Motor Scale (AIMS) and the Peabody Developmental Motor Scale (PDMS) were carried out in the home. In addition, the parents filled out a diary recording time spent in prone, supine and other positions. Scores on both tests were found not to be significantly different between those with DP and those without DP, however increased awake time in prone was associated with higher test scores on the AIMS for both groups. In 2009 Hutchinson et. al [4a] performed a retrospective review of the records of 287 patients attending a plagiocephaly clinic. Parents had completed a questionnaire covering demographic information, history, and current positioning strategies. They had also completed the Ages and Stages Questionnaire 2<sup>nd</sup> Edition (ASQ2) developmental screening questionnaire. Twenty-one percent were classified as non-cases of plagiocephaly, and these were compared to children with plagiocephaly. Developmental delay appeared to be related to sleep position, low muscle tone, low activity, male gender and neck muscle dysfunction, but not to plagiocephaly. In a 2010 study in which 235 infants (average age 6 mos.) with deformational plagiocephaly were compared with 237 controls, Speltz et. al [4a] found that infants with plagiocephaly performed worse than controls on the BSID III. They concluded that deformational plagiocephaly is associated with but does not necessarily cause developmental delay in infants. All of these studies used a standardized measure (either directly testing the infants or using a standardized parent questionnaire).

#### Long term effects:

Miller and Clarren (2000 [4a]) conducted a descriptive study of 63 families of children who had been diagnosed as infants with DP. Families were interviewed to determine if the children (now 8-19 yrs.) had received special services in school. Siblings who had not been diagnosed with DP served as controls (they were not age or sex-matched). Almost 40% of cases had received special help as compared with approximately 8% of their siblings (no standardized testing performed). Steinbok et al (2007[4b]) studied cosmetic and developmental outcomes in 65 children who had been diagnosed with plagiocephaly in infancy and who were now between 5 and 18 years of age using a parent questionnaire. Of the 64 children for whom these data were available, 14% were in a special education class, and 34% had received learning assistance. Limitations of this study are that development was not directly measured, sample size was small, and other risk factors for development were not eliminated. Hutchinson et. al (2010[2a]) performed a longitudinal cohort study of 129 children with a mean age of 4 years who had been diagnosed with DP as infants were assessed with the Ages and Stages Questionnaire. In 61% of those studied, head shapes reverted to within normal limits. The percentage of children with more than 1 delay in infancy decreased from 41% to 11% (close to that found in the general population). It is worth noting that no children had been helmeted.

Grade of the body of evidence: High

#### **Health Benefits, Side Effects and Risks:**

##### Benefits:

1. Health benefits of sharing the information about plagiocephaly and developmental delay with parents: reassures parents that their child's head shape is not causing a developmental delay; aids parents in making a decision about the plan of care

2. Health benefits of using a standardized tool to detect developmental delays: can insure that delays are identified and addressed in a timely manner

**Risks:**

1. Sharing information about the possibility of developmental delays in their infant can heighten parental anxiety about their child's condition.
2. Because the ASQ is a screening tool, not all skills in prone are assessed; a child can score within normal limits on the ASQ and still have delays in selected gross motor skills in prone such as pivoting. If the ASQ is used exclusively and not supplemented by the therapist's clinical observations, opportunities for building strength and postural control in the neck and upper extremities could be missed.

**Reference List**

- Fowler, E. A., Becker, D. B., Pilgram, T. K., Noetzel, M., Epstein, J., & Kane, A. A. (2008). Neurologic Findings in Infants With Deformational Plagiocephaly. *Journal of Child Neurology*, p. 742-747 [4b].
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- Steinbok, P., Lam, D., Singh, S., Mortenson, P. A., & Singhal, A. (2007). Long-term outcomes of infants with positional occipital plagiocephaly. *Child's Nervous System*, 23, 1275-1283 [4b].
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Note: Full tables of evidence grading system available in separate document:

- [Table of Evidence Levels of Individual Studies by Domain, Study Design, & Quality](#) (abbreviated table below)
- [Grading a Body of Evidence to Answer a Clinical Question](#)
- [Judging the Strength of a Recommendation](#) (abbreviated table below)

**Table of Evidence Levels** (see note above)

<i>Quality level</i>	<i>Definition</i>
1a† or 1b†	Systematic review, meta-analysis, or meta-synthesis of multiple studies
2a or 2b	Best study design for domain
3a or 3b	Fair study design for domain
4a or 4b	Weak study design for domain
5	Other: General review, expert opinion, case report, consensus report, or guideline

†a = good quality study; b = lesser quality study

**Table of Recommendation Strength** (see note above)

<i>Strength</i>	<i>Definition</i>
“Strongly recommended”	There is consensus that benefits clearly outweigh risks and burdens (or visa-versa for negative recommendations).
“Recommended”	There is consensus that benefits are closely balanced with risks and burdens.
No recommendation made	There is lack of consensus to direct development of a recommendation.

**Dimensions:** In determining the strength of a recommendation, the development group makes a considered judgment in a consensus process that incorporates critically appraised evidence, clinical experience, and other dimensions as listed below.

1. Grade of the Body of Evidence (see note above)
2. Safety / Harm
3. Health benefit to patient (*direct benefit*)
4. Burden to patient of adherence to recommendation (*cost, hassle, discomfort, pain, motivation, ability to adhere, time*)
5. Cost-effectiveness to healthcare system (*balance of cost / savings of resources, staff time, and supplies based on published studies or onsite analysis*)
6. Directness (*the extent to which the body of evidence directly answers the clinical question [population/problem, intervention, comparison, outcome]*)
7. Impact on morbidity/mortality or quality of life

## Supporting information

### Introductory/background information

Deformational plagiocephaly is a condition that has become much more prevalent since the Back to Sleep campaign began in 1992. This condition is often seen in infants who are referred to physical therapy for treatment of torticollis (a type of neck muscle dysfunction characterized by unilateral tightness of the neck muscles, usually the sternocleidomastoid). Currently, infants referred to physical therapy for treatment of torticollis are also screened for the presence of deformational plagiocephaly and referred to Plagiocephaly Clinic/Plastic Surgery if they demonstrate any amount of plagiocephaly (see CCHMC Guideline 33, pages 1-13). When discussing with parents the referral of infants to Plagiocephaly Clinic for consideration of helmeting, parents often ask the physical therapist whether the infant’s head shape will affect their development. Until recently, there was no literature that could answer this question.

### Group/team members

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## Search strategy

**Key words:** position\$ and plagiocephaly and child development; position\$ and plagiocephaly and development; cognitive delays and skull; cognitive delays and plagiocephaly; developmental delays and plagiocephaly; flattened heads; positional plagiocephaly

**Limits:** English, Children, Infants, all dates inclusive

**Databases:** Canchild, Cinahl Plus with Full Text/Ebsco Host, Cochrane of Systematic reviews, Ovid, Pub Med, Pedro

**Dates of retrieval:** retrieved between 7/29/10 and 4/28/11

**Applicability issues:** Because infants with torticollis often have deformational plagiocephaly, it has been difficult to tease out the influence of each of these conditions on child development. Current literature suggests that head shape is not directly related to developmental delay; delays appear to be related to neck tightness, low muscle tone, positioning, and male gender and sleep position. Furthermore, there is evidence to suggest that head shape corrects without helmet intervention (Hutchinson 2010). This information can guide parents to make an informed decision regarding helmet therapy for their children. There is a need to develop a means for giving information to parents that plagiocephaly is not directly related to developmental delay. This can be done by training physical therapists to share this information with parents during the assessment of the infants head shape at the physical therapy evaluation.

Because developmental delays have been seen in young infants with neck dysfunction, it is appropriate that children referred to physical therapy for neck dysfunction be screened using a standardized screening tool to guide physical therapy treatment and referral to other appropriate services.

Copies of this Best Evidence Statement (BEST) and related tools (if applicable, e.g., screening tools, algorithms, etc.) are available online and may be distributed by any organization for the global purpose of improving child health outcomes.

Website address: <http://www.cincinnatichildrens.org/svc/alpha/h/health-policy/best.htm>

Examples of approved uses of the BEST include the following:

- copies may be provided to anyone involved in the organization's process for developing and implementing evidence based care;
- hyperlinks to the CCHMC website may be placed on the organization's website;
- the BEST may be adopted or adapted for use within the organization, provided that CCHMC receives appropriate attribution on all written or electronic documents; and
- copies may be provided to patients and the clinicians who manage their care.

Notification of CCHMC at [EBDMinfo@cchmc.org](mailto:EBDMinfo@cchmc.org) for any BEST adopted, adapted, implemented or hyperlinked by the organization is appreciated.

Please cite as: Cincinnati Children's Hospital Medical Center: Best Evidence Statement Title, <http://www.cincinnatichildrens.org/svc/alpha/h/health-policy/best.htm>, BEST number, pages 1-number, Date.

This Best Evidence Statement has been reviewed against quality criteria by 2 independent reviewers from the Cincinnati Children's Hospital Medical Center (CCHMC) Evidence Collaboration.

*Additionally for more information about CCHMC Best Evidence Statements and the development process, Center for Professional Excellence/Research and Evidence-based Practice office at [CPE-EBP-Group@cchmc.org](mailto:CPE-EBP-Group@cchmc.org)*

## Note

**This Best Evidence Statement addresses only key points of care for the target population; it is not intended to be a comprehensive practice guideline. These recommendations result from review of literature and practices current at the time of their formulation. This Best Evidence Statement does not preclude using care modalities proven efficacious in studies published subsequent to the current revision of this document. This document is not intended to impose standards of care preventing selective variances from the recommendations to meet the specific and unique requirements of individual patients. Adherence to this Statement is voluntary. The clinician in light of the individual circumstances presented by the patient must make the ultimate judgment regarding the priority of any specific procedure.**