ORIGINAL RESEARCH

Cosmetic and Cognitive Outcomes of Positional Plagiocephaly Treatment

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Abstract

Purpose: Positional plagiocephaly is an acquired deformation of an intrinsically normal infant skull by sustained or excessive extrinsic forces. Non-surgical techniques include counter-positioning, supervised prone time and orthotic molding for more refractory cases. Long-term effects of positional plagiocephaly on development remain undefined, and this study evaluated cosmetic and cognitive outcomes of plagiocephaly management.

Method: Surveys were administered to parents of patients treated for positional plagiocephaly through the Children's Hospital of Eastern Ontario. Categorical responses interrogated cosmetic outcome, school performance, language skills, cognitive development and societal function. Pearson coefficient analysis tested outcomes dependency on gender, age, and plagiocephaly side at the 0.05 level of significance.

Results: Eighty respondents (51 male, 29 female) were divided as 58 right- and 22 left-sided pathology. Positional therapy was uniformly applied, and a helmet orthosis was utilized in 36% of cases. Median follow-up age was nine years with normal head appearance in 75% of cases. Only 4% of parents and 9% of patients observed significant residual asymmetry. These results did not vary by gender, age or deformity side. Left-sided disease predicted poorer language development and academic performance. Expressive speech abnormality occurred in twice as many patients with left-sided disease (36% versus 16%, p=0.04) along with three-fold greater special education requirements (27% versus 10%, p=0.04).

Conclusions: Non-surgical plagiocephaly management achieved good cosmetic outcome among patients in this study. Children with left-sided disease frequently encountered difficulties with cognitive and scholastic endeavors, although the roles of the underlying disease and the treatment measures in this delay cannot be differentiated.

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Positional plagiocephaly (PP) is an acquired cranial deformational malformation observed in children of normal development and in whom presentation of craniosynostosis has been excluded. The most consistent presentation is one of occipital flattening, with advancement of the ipsilateral ear and ipsilateral frontal bossing; features that provide differentiation from the isolated and rare lambdoid suture synostosis. The incidence of this abnormality has been climbing in the last two decades owing to the initiatives against sudden infant death syndrome (SIDS) with consequent supine positioning during infant sleeping. While such maneuvers are effective to decrease the likelihood of SIDS by approximately 40%, a remarkable increase in the incidence of positional posterior plagiocephaly (PP) has been consistently observed [1]. The deformity is more common among males and more frequently right-sided. While PP occurred in 1 in 300 live-births in 1974, the reported incidence in 1996 was 1 in 60 [2]. Other common causes of PP include congenital torticollis, fetal positioning in utero, abnormal birth position, and a low level of activity have also been implicated in the development of PP [3].

Positional plagiocephaly is usually diagnosed early in life, with most cases being mild and self-limited, resolving with conservative management. Active re-positioning during sleep and play, physiotherapy to address torticollis and stretch otherwise tight musculature, and orthotic molding helmets can improve the cosmetic outcome, with surgery almost never required. In more severe cases, orthotic molding can provide for more complete and rapid resolution of PP than with repositioning alone, with more favorable cosmetic outcomes observed when such treatment is applied earlier in the disease course between 6 and 12 months [4]. Mulliken and coworkers [5] describe the natural history of plagiocephaly without intervention to yield residual deformity among 45% of patients at 24 months of age. The literature surrounding the cognitive outcomes among patients treated for positional plagiocephaly remains scarce and ill-defined.

Objective

The objective of this work was to define the cosmetic and cognitive outcomes observed following management of positional plagiocephaly at the Children's Hospital of Eastern Ontario. An extension of this analysis was to evaluate if the side of plagiocephaly could impact on the child's neurocognitive development and outcome. Such information remains uncertain in the literature surrounding positional plagiocephaly, far more defined among patients with underlying structural craniosynostosis.

Methods

This study was a retrospective analysis of patients managed for plagiocephaly at CHEO from 1996 to 2000 after 2000, please add "...in a dedicated clinic for patients with positional plagiocephaly, developed and coordinated by the senior author (M.V.) and managed by a nurse practitioner. Research ethics board approval was obtained to initiate data accumulation and patient contact. Patient consent was not required for the retrospective component of this work with de-identified subjects. Consent for the questionnaire data was implicit on parents completing and returning the survey, although, again, patients have been de-identifed. For baseline analysis, the charts of all patients treated for positional plagiocephaly were reviewed and, from them, information regarding gender, age at presentation, side of pathology, course of treatment, and baseline cognitive function (subjective parental concern and physician-defined developmental delay) was obtained. The severity of each patient's plagiocephaly was prospectively recorded in the chart according to Argenta's five-point plagiocephaly severity score, [6] previously validated to have moderate interobserver reliability. Following identification of the patients, a survey questionnaire was mailed to each patient's guardian to assess various cosmetic and cognitive outcome variables. Examples of these questions included cosmetic enquiry into residual asymmetry (Q3: "Do you think that there is any residual asymmetry at the back of your child's head?"), parental or child concerns about head shape, and teasing behavior at school (Q4: "In the last year, has your child commented to you about being teased at school because of asymmetric appearance of his head?") Cognitive enquiry included whether the child was at age-appropriate grade at school (Q7: "What grade is your child in at school?" Q8: "Is this the normal grade level for their age?"), utilization of special education resources, as well as gross and fine motor skill development and ease of communication for both expressive and receptive language (Q13e "Do you have concerns about your child's development in the area of language comprehension?").

Exclusion criteria included those patients with craniosynostosis and those for whom chart information was incomplete.

Patients were classified by both gender and side of pathology. Demographic differences between groups were analyzed by $\chi 2-$test for categorical variables and Mann-Whitney test for age at presentation. All cosmetic and cognitive outcome variables were categorical and tested by $\chi 2-$test. All analyses were performed at the $\alpha = 0.05$ level of significance.