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Six month-old infants with deformational plagiocephaly do not differ from unaffected infants with respect to vocal control

Kathleen Wermke [a, *, 1], Christian Linz [b, 1], Annette Hasenberg [a], Felix Kunz [c], Philipp Meyer-Marcotty [c, 2], Tilmann Schweitzer [d]

[a] Center for Prespeech Development & Developmental Disorders, Department of Orthodontics, University Hospital of Würzburg, 97070 Würzburg, Germany
[b] Department of Oral and Maxillofacial Plastic Surgery, Specialist for Oral and Maxillofacial Surgery, University Hospital of Würzburg, 97070 Würzburg, Germany
[c] Department of Orthodontics, University Hospital of Würzburg, 97070 Würzburg, Germany
[d] Department of Neurosurgery, Section of Pediatric Neurosurgery University Hospital of Würzburg, 97070 Würzburg, Germany

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ABSTRACT

Objectives: The recommendation of a supine sleeping position led to a significant reduction of SIDS, but increased positional skull deformities (DP). Here, a quantitative analysis of babbling aims to complement previous studies of language-relevant competence based on items of the Bayley-scales that suggested the presence of developmental language delays in DP infants. Measures of fundamental frequency variability as proxies for vocal control are well suited for testing this assumption, since the laryngeal neuromuscular system matures early and is coupled with brain function while working rapidly in coordinating the structures and mechanisms involved in infant sound production.

Methods: Sixty-six healthy, full-term infants with normal hearing and a monolingual background took part: (1) moderately asymmetrical DP group - N = 41; 21 male; (2) severely asymmetrical DP group - N = 10; 8 male; and (3) controls - N = 15; 5 male (group assignment based on stereophotogrammetric 360° scans). Fundamental frequency (fo) measures were taken as proxies for vocal control skills during babbling.

Results: A MANOVA revealed no significant multivariate effect for the shape group, Wilks' $\lambda = 0.86$, $F(2, 63) = 1.21$, $p = 0.30$, $\eta 2 = 0.07$ (medium effect-size). The results do not support previous findings based on Bayley scale evaluations that suggested a negative impact of DP on language development during infancy.

Conclusions: A strong link between DP and brain dysfunction affecting vocal control, which would cause deviations in otherwise healthy DP infants, was not observed. Objective long-term studies of sound production are necessary to identify and/or understand the potential consequences of DP on early language development.

1. Introduction

The most dynamic period of postnatal brain growth and, consequently, skull growth is the first year of life. During these first months, external molding forces on the malleable and growing cranium enable skull shape modifications. The tradition of *active* head shaping with bandages or boards is very old, possibly even going back to 45,000 BC in Homo neanderthalensis [1–3]. In contrast, we are confronted today with the phenomenon of non-intentional skull deformities because of external forces that arise due to supine positioning. While the recommendation of a supine sleeping position (back-to-sleep campaign) [4] led to a significant reduction of sudden infant deaths (SIDS), an increase of positional skull deformities such as deformational plagiocephaly (DP) became obvious [5]. DP describes an asymmetric skull, typically characterized by unilateral occipital flattening, an ipsilateral ear shift and

* Corresponding author. Center for Prespeech Development & Developmental Disorders, University Hospital of Würzburg, Pleicherwall 2, 97070 Würzburg, Germany.
E-mail address: wermke_k@ukw.de (K. Wermke).
[1] Contributed equally to the work.
[2] PMM moved; now: head of the Department of Orthodontics, University Medical Center, 37099 Goettingen, Germany.

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possible facial asymmetry (cf. Fig. 2).

The coincidence of a supine sleeping position and increased DP appears to be striking, but there are other biological and environmental risk factors of decisive significance than the supine sleep position alone. Factors like the male sex, maternal age, preterm delivery, sleeping in car seats and other individual specifics of the everyday care environment are among the identified risk factors [6]. The higher incidence of DP in male infants and other identified biological risk factors point to a potential neuro-physiological cause for the development of DP, with a supine sleeping position just being an amplifier gain or even an epiphenomenon.

The recognition of DP as a neuro-developmental at-risk condition was fostered by the finding of morphological brain specifics in several regions in DP infants; at the age of eight months, these children were found to have: asymmetry and flattening of the posterior brain and cerebellar vermis; shortening of the corpus callosum; and differences in the orientation of the corpus callosum [7,8].

Balan et al. even reported on developmental delays in school-aged children diagnosed as infants as suffering from plagiocephaly [9]. Using auditory ERPs to sound in infant patients to detect possible cognitive impairment, the authors reported that “infants with plagiocephaly exhibited smaller amplitudes of the P150 and the N250 responses to tones than healthy controls” (p. 520). ERPs (event-related brain potentials) are measurement of electrophysiological response to a stimulus. ERP measurement is a non-invasive tool in uncovering the time course and structure of sound or language processing in the infant brain [10]. By placing electrodes on the infant scalp, ERPs are identified within recordings of an ongoing electrical brain activity (EEG). A brain response to a frequently repeated auditory stimulus can be measured by separating event-locked neural firing (ERPs) from the spontaneous background neural firing pattern. In this case, the resulting ERP curve displays peaks labelled by their polarity (P – positive, N – negative) and latency [ms]. Balan et al. [9] found a smaller amplitudes of the P150 and the N250 peaks of the ERP curve in responses to sound stimuli among infants with plagiocephaly compared to healthy controls. This was suggested as an indication that a “plagiocephalic skull signals compromise of brain functioning” (p. 520) and as an elevated risk of auditory processing disorders in these children.

As a result, it might not come as a surprise that DP in infants is sometimes regarded as a “disease” causing developmental motor and cognitive (language) delay [11–15]. These findings are, however, controversial, and there is an ongoing debate on whether or not DP is just an aesthetic condition [9,16], as Obladen (2012), for instance, put it: “Fashion is subject to change, and occipital flattening, a regional aesthetic ideal for millennia, has gradually lost esteem and is now considered a disease.” (p. 677) [1].

The assumption of a neuro-physiological at-risk state in DP infants, resulting in developmental cognitive and language delays, as well as delayed motor performances, has been exclusively based on evaluations using the Bayley Scales of Infant and Toddler Development (BSID) - version II/III [11–14,17]. An uncritical application of these subjective scales when assessing language outcomes in young infants is methodologically problematic and may not be very reliable. The BSID subscales for cognitive and language skills for infants are based on a few examples that are not very reliable given the great variability in early language development.

Consequently, supplementary investigations in DP infants using objective speech (voice)-related performances are desirable and necessary. Efficient vocal control and pitch processing are crucial for language development and are strongly coupled with brain function [18–20]. Fundamental frequency (fo) and related measures are adequate to identify dysfunctions of the neuro-physiological mechanisms underlying phonation. It is essentially the short-time variability of fo (time function), which reflects the developmental refinement of respiro-laryngeal coordination and the mastering of adjusted phonatory-articulatory interaction in babbling. In view of ongoing vocal tract growth and brain maturation over the first months of life, the mastering of fo control in articulated sounds is a demanding neuro-physiological task required for undisturbed language acquisition. In consequence, dysfunction of neuro-physiological control underlying phonatory control in babbling will result in significant deviations of measures related to fo variability compared to an undisturbed condition.

Due to its strong connection to survival, the respiratory-laryngeal neuro-muscular system is characterized by early maturation and a strong link to brain function, while working rapidly in coordinating the orchestra of involved structures and mechanisms. If the assumption of a strong, direct link between a DP condition and brain dysfunction causing language delay is true, vocal control and so fo variability as its proxy should deviate in otherwise healthy DP infants compared to controls.

The present study examined whether fo measures in DP infants vary with the severity of skull asymmetry and in comparison to infants with a symmetrical head shape. The study aimed to assess vocal control capability in infants of babbling age, which is the common age for clinical helmet therapy [21]. Based on past reports of a potential at-risk situation for language development in DP infants, we investigated the hypothesis that DP infants would show higher fo variability reflecting less robust and stable vocal control and, hence, a less efficient phonatory-articulatory adjustment.

The participants exhibited different degrees of severity of skull asymmetry (plagiocephaly) assessed by objective measurements using 3D-photogrammetry (Fig. 1). Infants at our observation age (six months) were found to produce intonation patterns in babbling that are shaped by the ambient language [22,23]. This capability, which requires a well-developed neuro-physiological control system underlying phonation, motivated the present approach to search for potential signs of regulatory dysfunction in DP infants.

2. Materials and methods

2.1. Study design

We invited the parents of infants with skull deformation and those with unaffected infants to participate in the pilot study, with their agreement sought based on our institution's informed consent procedures. The study was approved by the university's ethics

Fig. 1. Acquisition of a 3D stereophotogrammetry of a dummy. The infant is placed in the center of five synchronized camera pods. Image acquisition takes less than 1.5 ms, meaning that it is free of movement artifacts.