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Spectrum of positional deformities – Is there a real difference between plagiocephaly and brachycephaly?

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ABSTRACT

Aim: This study analyses pathomorphological and physiological head shapes and classifies the pathomorphology in positional plagiocephaly and brachycephaly.

Patients and methods: 78 infants with a positional plagiocephaly (5.99 months) and 32 infants with a positional brachycephaly (6.53 months) with a Cephalic index > 94% were investigated in this study and compared to a matched control group of 35 infants. The head shapes were analysed by stereophotogrammetry 3D data.

Results: The cephalic index, the total width, and coronal circumference were the highest values in patients with brachycephaly and the lowest values in the control group. The asymmetry of the head showed that the diagonal difference in brachycephalic patients more than doubled, and in patients with plagiocephaly almost tripled compared to the controls. A significantly higher total volume and vertex height was found for the patients with plagiocephaly and the patients with brachycephaly compared to the controls.

Conclusion: The cephalic index is a valuable and reliable parameter in order to differentiate positional deformities from unaffected skulls. Pathomorphology of a plagiocephaly is associated with the most severe asymmetry of the head. Plagiocephaly and brachycephaly overlap in several criteria. Therefore it seems justified to speak of a continuum rather than to differentiate between plagiocephaly and brachycephaly.

1. Introduction

There has been an increase in the incidence of positional plagiocephaly since the “back to sleep campaign” in 1992 (American Academy of Pediatrics, 1992), which was successfully implemented to reduce the risk of sudden infant death syndrome (1992). The term “plagiocephaly” might be misleading as symmetric positional brachycephaly can also be a consequence of supine sleeping position. Positional deformities usually develop due to postnatal positioning for different reasons (Kane et al., 1996). Additional cofactors for developing skull asymmetries such as intrauterine restriction are described (Pogliani et al., 2011; Rossi et al., 2003). In healthy infants younger than 1 year a range from 16% to 48% is given for the incidence of a positional plagiocephaly (Hutchison et al., 2004). This wide range could be explained because of the diagnostic instruments and the sensitivity used to set the diagnosis as well as the lack of reference data of a physiologically formed head (Hutchison et al., 2004; Meyer-Marcotty et al., 2012). Although the descriptions of the physical findings have been fairly consistent a clear demarcation of the physiological morphology of the infants head versus an abnormal shaped head is still missing. In order to evaluate the degree of deformity, Argenta et al., (2004) outlined a classification method based on the observer’s clinical inspection. He and others have emphasised the need for a better classification of the spectrum of malformed heads in order to individualize patients according to the degree of their deformity, which is essential for correct diagnosis and therapeutic considerations (Argenta et al., 2004; McGarry et al., 2008; Xia et al., 2008).

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This is even more important in the rare cases of synostotic skull abnormalities that have to be diagnosed and thoroughly followed (Pagnoni et al., 2013).

Understanding the complexity of cranial deformities, the function and architecture is important for testing hypotheses on different aspects of craniofacial growth and its variations (Lieberman et al., 2002; Engel et al., 2013) Therefore craniometric data have been used to quantify the diversity of the cranium and to differentiate between morphology and pathomorphology (Roseman and Weaver, 2004). A clear classification is necessary within the context of increasing neurocranial asymmetries due to positioning (Argenta et al., 2004).

Initial differentiation between these two types of positional deformation is possible by a general description of the head's shape:

Positional plagiocephaly is characterized by unilateral flattening of the occipital region, whereas brachycephaly is characterized by bilateral almost symmetric flattening of the whole occiput (Robinson and Proctor, 2009; Wilbrand et al., 2012a, 2012b; Xia et al., 2008). The asymmetric appearance of the positional plagiocephaly may be more obvious if it accompanied by an ipsilateral ear shift as well as a bulging forehead on the flattened side (Argenta et al., 2004; Sakurai et al., 2001).

Nevertheless there are only holistic descriptions of a positional plagiocephaly or brachycephaly without analysis of the head shape in detail. Beside these clinical aspects there are no quantitative, scientific variables to distinguish between these pathomorphologies. A clear quantitative, scientific analysis is necessary to allow the clinician a correct communication in between an interdisciplinary team. The fact that health insurance providers call for objective values in order to decide the cost coverage sheds another light on the need of an exact analysis. Furthermore reliable clinical variables together with the longitudinal analysis enable the evaluation of different therapeutic options currently available.

The aim of this study was:

- to generate three-dimensional data of deformational plagiocephaly and physiologically normal infants heads using a non-invasive approach,
- to differentiate pathomorphological and physiological head shapes by quantitative, scientific variables,
- to classify the pathomorphology in positional plagiocephaly and brachycephaly.

2. Patients and method

2.1. Patients

In this study 110 Caucasian infants (70 male and 40 female) were investigated. All infants were referred from Paediatricians or Physiotherapists to the Centre for Craniofacial Anomalies at the University Hospital of Würzburg, because of an assumed deformational asymmetry of the head.

Patients were classified into two groups: those with a unilateral positional plagiocephaly (plagiocephaly group) and those with a bilateral positional flattening of the occiput (brachycephaly group) (Fig. 1a and b). To differentiate these two entities from each other we used the cephalic index (CI):

To define a threshold for a significantly higher CI than the average the normative data of physiological formed head shapes were used. These three-dimensional data of normal growth of the infant's head were established in the Craniofacial Centre of Würzburg in a previous study (Meyer-Marcotty et al., 2012). The mean of the CI in a normal population of Caucasian infants was 84% ± 5.0%. Therefore it was possible to define a confidence interval around the mean of the CI with a probability of 95% (Mean of the CI $+ 1.969 \times$ SD $=$ 95% of the variability is included) (Ramm et al., 1976). Related to this study the threshold of a CI $> 94\%$ was established for diagnosis of a brachycephaly.

Mean age of the patients was 5.99 ± 0.23 months in the brachycephaly group and 6.53 ± 0.18 months in the plagiocephaly group. Examination of every infant by an experienced paediatric neurosurgeon, as well as an ultrasound work up of the sutures, ruled out any synostotic abnormality.

The study inclusion criteria were a birth date between the 37th and the 42nd week of pregnancy (mean of brachycephaly group: 39.33 ± 1.32; plagiocephaly group: 39.07 ± 1.38 weeks of pregnancy), a normal head circumference (brachycephaly group: 44.12 ± 1.47; plagiocephaly group: 44.02 ± 1.89 cm), and a positioning-induced abnormality of the occipital region. The presence of any congenital anomaly, syndromes, previous operations or any neurological or musculoskeletal deficits were exclusion criteria.

2.2. Control group

In order to provide morphometric parameter of a physiological head shape the data of 35 infants (24 female, 11 male) with no visible cranial asymmetry were recruited from paediatric practices by means of an information brochure (Fig. 1c). The mean age of the children in this control group was 6.30 ± 0.09 months. The inclusion criteria were a birth date between the 37th and the 42nd week of pregnancy (mean of 39.11 ± 1.32 weeks of pregnancy), had a normal head circumference 43.44 ± 1.37 cm and had no visible deformity of the head. Infants with a congenital anomaly or syndromes, with previous operations or with any neurological or musculoskeletal deficits were excluded.

2.3. Methods – 3D data acquisition

The study was based on a clinical study design, examined and approved by the Ethics Committee of the Medical Faculty of the University Wuerzburg (ethics number 143/09). The study was carried out according to the Declaration of Helsinki, and written consent was obtained from each infant's parents.

For the analysis of the head shapes 3D data were generated of the entire head of every infant. The 3D data were generated using a standardized recording protocol: The infants were seated in a special chair, preventing artefacts by movements and cantered in the 3D-Scanner. To avoid artefacts due to hair, each infant was fitted with a tight nylon cap before recording. The data acquisition was performed by a specially developed scanner for infant recordings (3dMD®, Atlanta, GA, USA). Five synchronized cameras based on the stereophotogrammetric method were used to get a virtual data set. By using this procedure non-invasive 360° data were generated with only one scan with a recording time of 1.5 ms.

2.4. Methods – 3D data analysis

After triangulation and editing, the scans were converted to a common 3D data format (.stl) and analysed with Cranioform Analytics 4.0 3D software (Cranioform®, Alpnach, Switzerland). To align the 3D data sets in virtual space, a coordinate system was established based on four anatomically defined reference points (tragion left (Tl), tragion right (Tr), nasion (N) and subnasale (Sn)). The procedure of aligning had been performed in a previous study (Meyer-Marcotty et al., 2012): First the midpoint of the coordinate system was defined as the intersection of the tragus connection line. According to the midpoint (M) a line through the nasion (N) represents the *Y*-axis. To construct the *X*-axis in a first step a sagittal reference plane through the midpoint (M), nasion (N) and