Journal of Plastic, Reconstructive & Aesthetic Surgery (2014) 67, 900–905

JPRAS
An International Journal of Surgical Reconstruction
www.JPRASurg.com

Three-dimensional orbital dysmorphology in metopic synostosis

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Received 7 December 2013; accepted 8 March 2014

KEYWORDS
Metopic;
Synostosis;
Trigonocephaly;
Orbital dysmorphology;
Materialise

Summary *Background:* Metopic synostosis is characterized by trigonocephaly, lateral supraorbital retrusion, and hypotelorism. Most phenotypic evaluations have focused on the forehead without much emphasis on the orbits. The study seeks to explore differences in orbital dysmorphology for metopic and control patients, along with different degrees of metopic synostosis.
Methods: Demographic and craniometric data were compiled. CT scans were digitized (Materialise) and metopic and control groups were compared. Degree of trigonocephaly was classified into moderate and severe cases based on endocranial bifrontal angle. Orbital plane angle, width, depth, volume, and corneal projection were measured. Statistical two-paired *t*-tests were used, with significance determined as $p < 0.05$.
Results: Forty-six CT scans were analyzed (23 affected, 23 controls). Mean ages (6 months metopic, 7 months control) and genders (18 males metopic, 10 males control) were determined. Orbital plane angle measurements showed differences between the metopic and the control ($p = 0.0002$), along with a correlation to trigonocephaly ($p = 0.0097$). Orbital width and height were insignificant between controls and overall metopics, though height was less in severe metopics ($p = 0.046$ left, $p = 0.0337$ right). Orbital Depth was significant between control and metopics ($p = 0.0106$ left, $p = 0.0025$ right), and pronounced in severe cases $p = 0.0349$ left, $p = 0.0071$ right). Corneal Projection correlates with metopic severity ($p < 0.01$ left, right), while orbital volume showed insignificant change between control and metopic cases.

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http://dx.doi.org/10.1016/j.bjps.2014.03.009

Conclusions: Orbital dysmorphology worsens with increasing degree of trigonocephaly, presenting additional functional defects. The true exorbitism most directly correlates with worsening trigonocephaly. Expanding and advancing the lateral orbital wall is a critical treatment element in correction.
Level of evidence: III.

Introduction

Trigonocephaly ensues following a premature fusion of the metopic suture before or within a time period shortly after birth. The timing and degree of suture fusion, is responsible for the degree of detectable phenotypic characteristics, such as a keel-shaped frontal upper forehead, bitemporal narrowing, and a compensatory widening of the posterior cranial vault.[1–3] Orbital dysmorphology, including ethmoidal hypoplasia, orbital hypotelorism, teardrop-shaped orbits, and bilateral depressed orbital rims can be seen in metopic synostosis. Despite these anatomic findings, most treatment methods focus mostly on the forehead, with less emphasis on the superolateral orbital deflection. Less emphasis and understanding is placed on the relationship between the bony orbital framework and its contents in metopic synostosis. As a corollary, addressing the orbital dimensions is not a strongly featured component of most surgical maneuvers, though implicit effort may exist. To the author's knowledge, there is no study focusing on the orbital dysmorphology in metopic synostosis, especially in light of the degree of trigonocephaly, and when compared to the normal population. The purpose of this study therefore is to characterize the three-dimensional orbital dysmorphology with worsening metopic classification, and compared to an age-matched normal population. This may shed light on the underpinnings of the disease process and impact the various current treatment strategies.

Materials and methods

Yale University Institutional Review Board approved this study (protocol: HIC# 1101007932). Computed tomographic (CT) scans of both recent metopic synostosis infants, and age-matched controls presenting to the Plastics and Craniofacial program at Yale were included. Control subjects were chosen without confounding any craniofacial pathology. CT images were digitized for two- and three-dimensional analysis using surgical planning software (Materialise, Leuven, Belgium).

Anatomical landmarks were identified and used for craniometric measurements (Table 2). Craniometric measurements focused on orbital dysmorphology in metopic synostosis, indicated in Table 2, were then compared across groups (control, moderate, and severe metopic synostosis). Subjects with metopic synostosis were classified as moderate or severe, based on the endocranial bifrontal angle (EBA) (Table 2).[4] Correlations were analyzed statistically by means of a simple *t*-test of comparative means with *p*-values <0.05 considered significant (Figure 1).

Results

46 CT scans were included for analysis, 23 controls and 23 metopic synostosis. The control group contained 10 males and 13 females, with an average age of 7 months. In the metopic group, 14 were moderate, with 18 males and 5 females, and a mean age of 6 months (Table 1). The endocranial bifrontal angle differed significantly among the groups, using statistical tests ($p < 0.05$). Tables 3 and 4 summarize our findings (Figure 2).

The Orbital Plane Angle was significantly different between control and metopic groups ($p = 0.0002$). Comparing

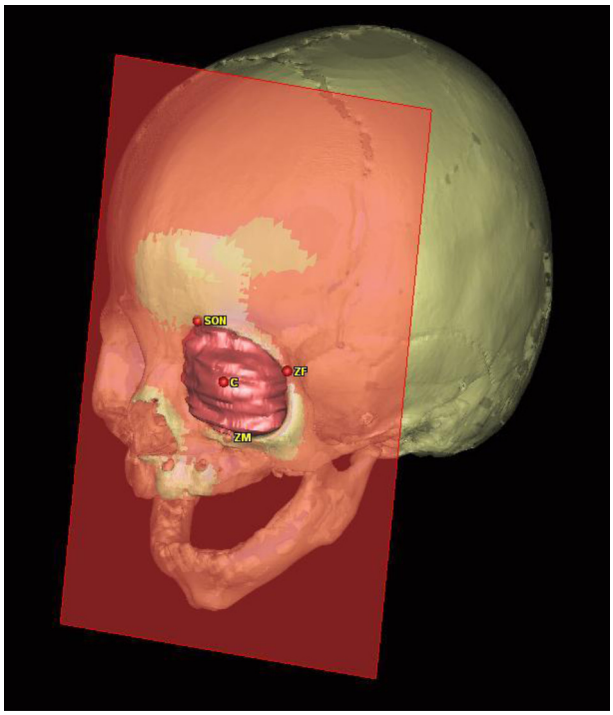

Figure 1 Craniometric evaluation of the orbit (oblique view). Orbital plane, ZF: Zygomaticofrontal suture; ZM: Zygomaticomaxillary suture; SON: Supraorbital Notch; C: Corneal projection. This oblique view of a severely metopic patient demonstrates our definition of the left orbital plane, along with the relative exophthalmos of the orbital soft tissue. A metopic ridge is visible, along with a lateral orbital wall retrusion.