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Photometric evaluation of cranial and facial symmetry in hemicoronal single suture synostosis treated with surgical fronto-orbital remodeling



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ABSTRACT

Objective: Evaluation of frontal vault symmetry and progressive facial symmetrization in a cohort of patients with hemicoronal single suture synostosis treated with a standardized cranioplasty and rigid fixation.

Patients and methods: Fifty-four patients with hemicoronal synostosis operated between 1999 and 2014 were reviewed retrospectively. Pre, immediately postoperative and yearly photographs from the top of the skull and frontal views of the face were taken with the same head position and projection. A photogrammetric method was applied to quantify the pre and postoperative contour changes. The anterior skull hemispheres were traced, divided into two equal parts and the areas were compared. Angular measurements obtained by the intersection of the interpupillary line and the glabella perpendicular vertical line were calculated. The average photographic follow-up was 6.8 years. Range 1–14 years.

Results: The average advancement on the affected side was 18 mm (range: 16–23 mm). The pre-surgical cranial area on the affected side was increased on average 14.6 + 2.4% (range: 10–18%). The angular measurements documented the frontal symmetry obtained and the progressive improvement of facial symmetry.

Conclusion: Cranioplasty with rigid fixation achieved a stable correction of anterior plagiocephaly leading to subsequent symmetrical facial growth. Photogrammetry allowed for a quantitative long-term validation.

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1. Introduction

The incidence of unilateral coronal synostosis (UCS) is about 1 per 10,000 (Cohen, 2000). In a recent epidemiological study, the global incidence of craniosynostosis was reported to be stable, affecting 1 in 2000/2500 live births (Selber et al., 2008a, 2008b). Unilateral coronal synostosis represents 15–20% of all single suture craniosynostosis and is the third most common type, following scaphocephaly and trigonocephaly (Slater et al., 2008; Wilkie et al., 2010; Di Rocco et al., 2012; Di Rocco et al., 2009). Hemicoronal

single suture synostosis can be isolated or a symptom of Muenke syndrome. Patients affected by Muenke syndrome, in addition to plagiocephaly, may present with mild abnormalities of the hands or feet, hearing loss, and seldom mental or developmental delay and learning disabilities (Agochukwu et al., 2014; Keller et al., 2007; Sabatino et al., 2004). The craniofacial morphologic alterations are characterized by superior, posterior, and lateral displacement and deformity of the ipsilateral fronto-orbital area and cranial base. Nose deviation, asymmetrical palpebral rim, vertical zygomatic dystopia, and difference in midfacial height and shape produce a midfacial twisting and, later on, a bimaxillary and occlusal cant (Pelo et al., 2011; da Silva Freitas et al., 2010; Nagasao et al., 2010; Abramson et al., 1996; Loomis et al., 1990; Marsh et al., 1986). Controversy exists regarding functional problems associated with

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hemicoronal synostosis, such as mental development interference (Becker et al., 2005; Speltz et al., 2004; Kapp-Simon, 1998), eye movement disorders (Tarczy-Hornoch et al., 2008; Levy et al., 2007; Weiss and Phillips, 2006), and lateralized head posture (Gosain et al., 1996; Raco et al., 1999). This controversy leads to the question of the true goals of cranioplasty in hemicoronal single suture synostosis. The true effects on long-term neurocognitive development, vision, oculomotion, head posture, and progressive facial symmetrization remain unclear. In the literature, the most important studies on hemicoronal single suture synostosis report the presence of intracranial hypertension only in 16% of cases, with no effects on neurocognitive development (Mathijssen et al., 2006). However, above all, there is no scientific evidence of the beneficial effects on neurocognitive development of cranioplasty, as well as most unisutural craniosynostosis (Da Costa et al., 2013; Starr et al., 2007). On the other hand, several studies confirm that the facial deformities cause a reduction of self-esteem by hindering psychosocial relationships (Sarwer et al., 1999; Thoma and Ignacy, 2012). In plagiocephaly, there is a large secondary component that determines a twisting of the face. Skull base asymmetry affects the growth of facial bones, as growth of the neurocranium occurs in the first 30 months of life, guiding the subsequent facial growth which is completed at 15 years of age (Kreiborg, 1981; Arvystas et al., 1985; Marsh et al., 1986; Captier et al., 2003). It is therefore fundamental to adopt a technique that will enable us to correct morphological defects of the fronto-orbital area, thereby creating the conditions to allow symmetrical growth of the face. Craniofacial malformations are genetically determined and tend to recur after surgical treatment, especially asymmetrical deformities (Reinhart et al., 1996; Meazzini et al., 2012a, 2012b). Since the late 1960s, when Paul Tessier described surgical cranioplasty with the preparation of a frontal bone segment and fronto-orbital bandeau, several alternatives were proposed: variations of the osteotomy design (Guzman et al., 2011), the application of distractors (Taylor et al., 2014; Choi et al., 2010) or springs (Shen et al., 2015), and endoscopic techniques with post-surgical helmets (Sauerhammer et al., 2014; Stelnicki et al., 2009). Part of the re-expression of the phenotype may be due to inadequate procedures (Shen et al., 2015). The aim of this study was to evaluate the overall efficacy on the facial appearance and facial growth of our procedure of fronto-orbital remodeling, advancement, and stabilization with over-correction of the affected side and rigid fixation of unicoronal suture synostosis.

2. Material and methods

2.1. Patient population

In this retrospective study, 54 consecutively treated patients affected by anterior plagiocephaly were selected. Thirty-two underwent genetic testing. Six were found to carry Muenke's syndrome mutation. All patients underwent open surgical cranioplasty, with fronto-orbital advancement, between January 1999 and April 2014. Eight patients were excluded from this study because the photographic documentation was inadequate. The sample included 18 male patients and 36 female patients. The right suture was affected in 22 and the left suture in 32 cases. A family history of craniosynostosis was present in two patients. The mean age of the first examination was 5 months (range: 1.5–14 months). The preoperative study included a multidisciplinary assessment by a maxillofacial surgeon, neurosurgeon, geneticist, neuropsychologist, and ophthalmologist. After 5 months of age, a craniofacial computed tomography (CT) scan with three-dimensional (3D) reconstruction was performed to confirm the clinical diagnosis (Seidenbusch et al., 2008), as this is the right time in order to reduce

the risk of radiation exposure to the brain and to plan the correct treatment. All patients had clinical photographs preoperatively, intraoperatively, and postoperatively. The mean age of the surgery was 8.9 months (range: 6–17 months). This study did not include patients who were operated on after 2 years of age.

The average weight of the child at time of surgery was 8.8 kg (range: 7–13 kg). The operating time ranged from 174 to 248 min (average: 215 min). Follow-up ranged from 1 month to 14 years with an average follow-up of 5.8 years. During the follow-up, none of the patients underwent a CT scan.

2.2. Surgical technique

Under general anesthesia, after trichotomy and infiltration with local anesthetic and vasoconstrictor to reduce bleeding, a zig-zag incision is made in the coronal area and a galeocutaneous flap is harvested through supraperiosteal dissection of the anterior parietal, frontal, and temporal region bilaterally. Once the pericranial flap has been harvested and lifted to allow a view of the frontal, temporal, anterior parietal, and orbitonasal bones and of the open and fused coronal sutures, the osteotomic lines of the frontal bone segment and the fronto-orbital bandeau are designed. The design includes the coronal and all other pathological sutures and all of the deformed areas. The fronto-orbital bandeau is designed keeping an average height of approximately 12 mm from the upper orbital frame and should be extended for approximately 20–25 mm in the temporal fossae bilaterally to include the anterior third of the orbital roofs. Then, a frontal single block bone flap is removed with a diamond drill and an osteotomy of the frontal bone segment using a pediatric craniotome or a reciprocal saw. After the dissection of the anterior cranial base, including, laterally, the great wings of the sphenoid to allow the protection of the frontal lobes and the anterior temporal poles, the osteotomies of the fronto-orbital bandeau are made using a reciprocal saw and bone scalpels. After having removed the fronto-orbital bandeau, the perimeter of the bandeau is traced with a pen on a sterile sheet in order to later compare the final results of the molding with the original deformed morphology. On the basis of the movements required, the site and direction of the corticotomies are drawn on both the external and internal side of the bandeau. In particular, the internal corticotomies are needed to curve the bandeau, whereas the external ones are needed to straighten it. The inclination and the position of the corticotomies affect the final shape of the bone segments during the molding step. At this time, long titanium plates with a thickness of 1.3 mm and 24 holes are applied, involving all the areas harboring the corticotomies (Fig. 1): two plates on the affected side, to obtain torsional movements, one plate in the middle, and two short plates on the healthy side. The plates are long enough to extend beyond the lateral borders of the bandeau to allow a modulation of the advancement during the repositioning of the bandeau. The bandeau and the titanium plates, weakened by the corticotomies and strengthened by the plates, creates an easily plastic and malleable “bone-plate unit” so that the shape can be changed using Tessier forceps without risking fragmentation. During the molding step, the initial drawing of the deformed morphology is compared with the new profile, taking care to observe the symmetry and the number of stepwise corrections until a final symmetrical structure is obtained. The goal of the reshaping is to give an adequate curvature to the affected flattened side of the bandeau and to correct the twisting (Fig. 2). Once the desired shape has been achieved, repositioning and fixation of the bone segments in the required position can begin, using the previously applied plates and obtaining a bone-to-bone contact to allow bone fusion. The unaffected side is repositioned in the

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