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Review

Review of quantitative outcome analysis of cranial morphology in craniosynostosis

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Summary Outcome measures in craniosynostosis surgery have progressed from those based on the need for surgical revision to linear anthropometric measurements, 2D CT vector analysis and 3D CT vector analysis. However, finding an objective means to assess postoperative cranial morphological improvement remains challenging.

A critical review of previous studies used to measure craniosynostosis surgery outcomes is presented. We also introduce and briefly discuss the key features of the computational algorithm that is being utilized in our center for evaluating craniosynostosis surgical outcomes. This has addressed a number of the previous challenges encountered in quantitative measurement of cranial morphological change.

Point cloud representation and 3D stereophotogrammetry have made it possible to compare pre and post-operative images of children undergoing surgical correction for craniosynostosis. These pre- and post-operative images can also be compared to age, sex and race-matched controls throughout the patient's lifetime allowing longitudinal changes to be measured on follow up.

Contents

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Introduction

The goal of this paper is to provide a historical review of the various methods used to provide a quantitative outcome analysis of cranial morphology in craniosynostosis. Novel techniques are then described showing the future direction of quantitative outcome analysis. Hankinson et al. set the scene — "Pediatric craniofacial surgeons have not, however, agreed upon objective means to assess postoperative cranial morphological improvement. We should therefore endeavor to agree upon objective craniometric tools for the assessment of operative outcomes, allowing us to accurately compare the various surgical techniques that are currently available".[1]

Outcomes based on need for surgical revision

The Whittaker categorization was the first system to be used by several studies to quantitatively look at outcome measures. Recognition was made of how "the excellence of correction is ultimately a subjective aesthetic judgment to which both the surgeon and the patient or family contribute".[2] An attempt was made to move away from subjective measurement. A total number of 164 patients were categorized into those with a symmetrical versus asymmetrical deformity. The authors subdivided the end points of correction into four categories based on whether the patients warranted further revision surgery. Category I included those patients in whom no refinements or surgical revisions were considered advisable or necessary by the surgeon or the patient. In category II, soft-tissue or lesser bone-contouring revisions were desirable whether performed or not. Category III included patients in whom major alternative osteotomies or bone grafting procedures were needed or performed for example further repositioning of the orbits to improve residual exorbitism or onlay bone grafts. Category IV included patients in whom a major craniofacial procedure was necessary essentially duplicating or exceeding in extent the original surgery.[2]

In a landmark post-operative review of patients with non-syndromic and syndromic synostosis, McCarthy et al. used the Whittaker classification to analyze results in 104 patients treated over a 20-year period.[3] According to this study 87.5% were in categories I—II and 11.5% were in categories III—IV. The patients included in the study included those with diagnoses of bicoronal synostosis (10 cases), unilateral coronal synostosis (57 cases), metopic synostosis (29 cases), and sagittal synostosis (8 cases).

Seruya et al. in their study of 212 patients acknowledged the paucity of outcomes studies for the primary management of craniosynostosis over the most recent decade. Bi-coronal synostosis patients had a higher frequency of Whittaker class III/IV distribution, which was similar to the studies by Pearson and Sloan.[4] It also highlighted the problem in comparing cranial form between different surgical techniques such as endoscopic to total cranial vault remodeling. Sloan et al. modified the Whittaker system into a seven-category outcome classification system to allow recognition of more subtle differences in surgical results. Further studies highlighted the difficulties in analysis of "cranial form" in craniosynostosis and the complexity of the deformities.[5—8] The studies which used the Whittaker categorization were all retrospective with the data being derived from the notes but not from photos or by serial examination of the patient themselves. There were no pre-operative assessments made of cranial form, so comparison could not be made between before and after results. The success of the eventual outcome could also not be stratified by severity of pre-operative deformity.

Outcomes based on linear anthropometric measurements

The next development in outcome analysis has come through the use of two dimensional (linear) direct anthropometric measurements, with the most common being the cranial-index.

Fearon et al. analyzed 296 patients with single suture craniosynostosis retrospectively to assess long term growth using anthropometric measurements taken up to 11 years post-operatively with the mean follow up time of 4 years.[9,10] This study raised the important question of how authors select and justify the specific points and indices used for measurements. Fearon et al. stated, "four specific measurements were chosen for this analysis, for they most accurately defined the dimensions of the cranial vault that are involved in the single sutural synostoses. These measurements were minimum frontal breadth (ft-ft), head circumference, maximum cranial length (g-op) and maximal cranial breadth (eu-eu). However, it is well documented that the selection of cephalometric landmarks incurs errors that authors may not account for in their final calculations.[11] The major limitation of direct patient

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