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Statistical Shape Analysis of Metopic Craniosynostosis: A Preliminary Study

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Abstract- **This preliminary study was conducted to explore different analytical shape methods for use in evaluating children born with cranial vault deformities. Twenty skull outlines from patients with metopic craniosynostosis were ascribed landmarks. Scale, location, and rotational factors were removed using Procrustes Analysis. A single index of severity from 0-5, with 5 being the most severe, was developed using Procrustes Distance in shape space. Skull 20 had the highest score in our data set. Principal Component Analysis was performed to determine areas of large shape variability. Principal Component 1 and 2 accounted for 86 % of the shape variability which was attributed to early closure of the metopic suture. Procrustes Analysis used in combination with Procrustes Distance and Principal Component Analysis are powerful tools for the evaluation of cranial vault deformities and can be used to objectively categorize the severity of the skull deformity and outcome from surgical reconstructive surgery.**

Key words- **Procrustes Analysis, Craniosynostosis, Shape Analysis, Principal Component Analysis, Procrustes Distance**

I. INTRODUCTION

Shape is defined as "the geometrical information that remains when location, scale, and rotational effects are filtered out from an object [1]." In the field of plastic surgery, where the biological shape is intentionally altered, it is often up to the surgeon to determine what will look "normal" and what will look pleasing to the eye. There is a need to develop objective measurements and evaluations to evaluate the outcome from treatment. Among them, shape analysis is crucial for aesthetics and normality. Statistical shape analysis in the field of plastic surgery is to quantify "normalcy" and to quantify "aesthetics"

An example is illustrated in children born with craniosynostosis where the normal cranial vault shape is deformed as a result of early closure of one or more of the various skull sutures [2]. Figure 1 shows the varying types of craniosynostosis compared with a normal skull shape.

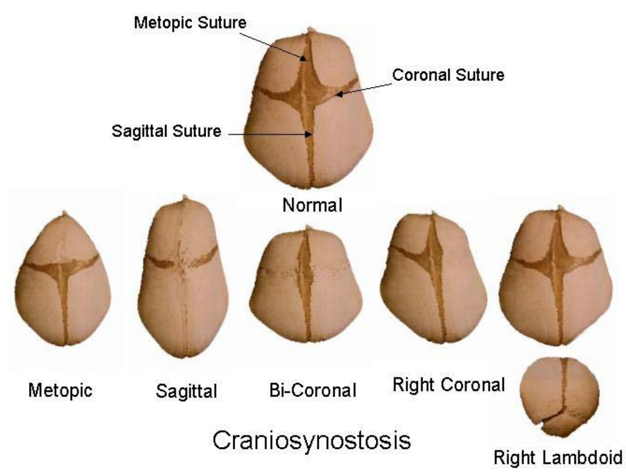

Fig. 1. Varying skull shapes caused by craniosynostosis

The presence of open sutures is critical to accommodate the rapidly growing brain in the infant. Surgery then involves dissembling the cranial vault and then reassembling the skull in a new shape to allow normal growth and development to occur. For the craniofacial surgeon, determining what the 'new' shape is and how far it deviates from normalcy frequently relies on subjective criteria. Thus an objective measure is needed to quantify the severity of the deformity and to measure the outcome from surgery.

The goal of this study is to develop a single index to quantify the severity of the cranial deformation based upon statistical shape analysis of the skull. Furthermore, if we can also determine where the greatest variation from norm occurs on the skull, the surgeon will be able to find out where to focus the largest amount of effort. As a preliminary study, we focused on the planar shape analysis in this study.

II. METHODOLOGY

We used a comprehensive methodology involving Procrustes Analysis, Principal Component Analysis, and Procrustes Distance in Shape Space. These methods will allow a more complete quantification of shape and normalcy [3].

A. Data Acquisition

The CT scans of 20 skulls were taken and 2D planar slices were used. The outlines of these planar slices were drawn. Landmarks and their coordinates on the outlines were extracted using Microscribe 3D (Immersion Corporation, San Jose, CA) in a Cartesian coordinate system. A set of 36 landmarks were ascribed to each of the 20 skulls by defining the centroid visually and marking off 10 degree increments along the outline of the skull. The center of the first skull was arbitrary since Procrustes Analysis will remove any location factors.

B. Generalized Procrustes Analysis

Procrustes Analysis (PA) is a method used for comparing two or more sets of data through least squares orthogonal mapping. It filters out all the translational, rotational, and scale effects between the shapes, leaving only the shape variance through minimizing the sum of squared deviations between landmarks. The set of landmarks associated with a shape is called a configuration. The configuration matrix X is the (k × m) matrix consisting of the Cartesian coordinates of the k landmarks in m dimensions (with m = 2 in this preliminary study). The configuration matrices are ordered sets with the i^{th} row of the X matrix corresponding to the coordinates of the i^{th} landmark on X shape [1, 4].

With more than two or more landmark matrices, the process is known as Generalized Procrustes Analysis (GPA). The goal of GPA is minimize the Procrustes sum of squares (given by the following equation). Parameters β_i, γ_i, Γ_i are for scale, translation, and rotation, respectively [1].

$$G(X_1,\ldots,X_n) = \frac{1}{n}\sum_{i=1}^{n}\sum_{j=i+1}^{n}\left\| \begin{matrix}(\beta_1 X_1 \Gamma_1 + 1_{\kappa\gamma_i}{}^{\mathbf{T}}) \\ -\,(\beta_j X_j \Gamma_j + 1_{\kappa\gamma_i}{}^{\mathbf{T}})\end{matrix} \right\|^2 \qquad (1)$$

The free software package PAST (available at http://folk.uio.no/ohammer/past/index.html) was used to perform Procrustes Analysis as well as Principal Component Analysis.

C. Overall Procrustes Distance

We used the Procrustes distance between two shapes to give a more overall view of shape variability. It is the square root of the sum of squared differences in the positions of the landmarks of the two shapes.

$$P_d^2 = \sum_{j=1}^{n}[(x_{j1} - x_{j2})^2 + (y_{j1} - y_{j2})^2] \qquad (2)$$

The Procrustes Distance was taken between the Procrustes normalized data sets. This was then indexed by difference from norm through the use of standard deviations or variance in data [5].

D. Developing an Index

We theoretically developed a single index of severity of deformation by utilizing the standard deviation of the data set. The number of standard deviations from a mean normal skull shape can be rated from 0-5, with 5 being the most severe. This Likert-type scale will allow a surgeon to quickly assess the need of patient in terms of work involved in the surgery [6].

Since there was no "normal skull" data set to use to determine the average skull shape, we chose Skull 1, which is visually less deformed to be the "normal" skull for comparison based on a simple visual approximation of normalcy. Procrustes distance measurements were taken in relation to the skull 1 for each of the other 19 configurations.

E. Principal Component Analysis

Principal Component Analysis (PCA) was used for identifying patterns in data sets. Data is expressed in a way to highlight differences and similarities. For our purpose, PCA was used to determine where the greatest variance in shape occurs. Performing PCA on a 2 dimensional data set follows the proceeding algorithm. Principal Component Analysis was run on the Procrustes normalized coordinate data sets.

1. Acquire Procrustes normalized data
2. Subtract the mean from each data point
3. Calculate the covariance matrix in 2 dimensions
4. Calculate the eigenvectors and eigenvalues of the covariance matrix
5. Choosing components and defining a feature vector
6. Derive new data set

The loadings of the principal components are important for explaining the correlation of each PC. They show the weight of the principal component on different landmarks, allowing for a more physical description of variability. Higher loadings indicate that the principal component has a higher effect on that particular landmark. By observing on which landmarks the principal component has the largest effect, the physical correlation of the principal component can be inferred.

Using PCA, we determined which landmarks of the skull have the most significant deviation from norm. Areas of no significance can be ignored or filtered out. This allows for a more precise view of shape variability. Principal Component Analysis gives us principal components and orders them by significance [7, 8].

III. RESULTS

After Procrustes analysis is performed on landmark data, size, rotation, and location factors are removed from all the skull shapes leaving difference between each landmark configuration based on shape alone. Examples of the old landmark data and new Procrustes coordinates are listed in Table 1. Figure 2 is a plot of the landmark data after Procrustes analysis. The single index score is listed in Table 2.

TABLE 1
LANDMARK DATA

Normal Coordinates	x1	y1
Skull 1	334.6798	-244.327
Skull 2	321.2901	-234.674
Procrustes Coordinates	x1	y1
Skull 1	-0.03546	0.117596
Skull 2	-0.03083	0.115699

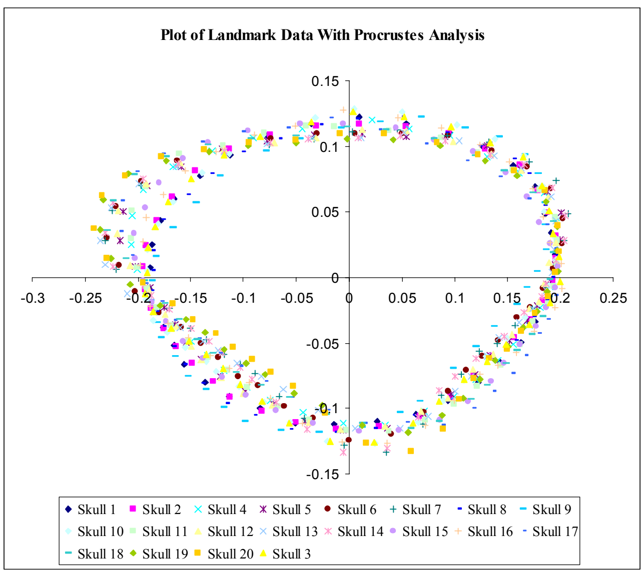

Fig 2. Plot of landmark data after Procrustes Analysis

Near 95 % of the shape variability in the landmark data can be attributed to the first 5 principal components. Of those components, principal component 1 accounts for 66.02 % of the shape variability. Although there are 2k components (k being dimension), the first five can be considered the most important since they account for 95 % of the shape variability. More components can be incorporated for a more exact study. However, though only 5 principal components are used, it is fairly accurate because of the large percentage of shape variability they account for.

TABLE 2
SINGLE INDEX OF SEVERITY

	Index Score (0-5)
Skull 2	0.603
Skull 3	1.212
Skull 4	1.661
Skull 5	1.704
Skull 6	2.608
Skull 7	2.781
Skull 8	0.708
Skull 9	2.374
Skull 10	1.192
Skull 11	1.662
Skull 12	1.812
Skull 13	2.814
Skull 14	2.696
Skull 15	1.923
Skull 16	1.571
Skull 17	3.281
Skull 18	2.960
Skull 19	3.343
Skull 20	3.718

Since the location of the early suture closure is known, principal component analysis can be used to describe where the changes occur. The first principal component, PC1, describes approximately 66% of the shape variability in the sample size. The largest loadings occur around landmark 6 and around landmark 15. Large loadings occur plus or minus 4 landmarks from those 2 landmarks. From the original skull outlines, these landmarks correspond to areas of large deformation, including the most obvious triangular deformation of the skulls near the bottom of the outlines. From looking at the loadings in both the x and y directions, it can be seen that at the landmarks of highest loadings, the x loading is usually higher than the y loading. Therefore, PC 1 has more of an effect on the variability in the x direction on these landmarks. The landmark data was obtained from metopic synostosis patients. Since PC 1 describes approximately 66% of the shape variability in the sample size, PC 1 can possibly be correlated to the early closure of the Metopic suture.

The second principal component, PC2, represents approximately 20% of the shape variability. The largest loadings occur around landmarks 2, 7, 23 and 31. From the large distribution of these loadings, it can be inferred that PC 2 describes a more detailed portion of the configurations. Like PC 1, PC 2 has more of an effect in the x direction as the loadings in those directions are usually greater than the loadings in the y direction. These findings indicate that these two principal components describe early closure of vertical sutures including the Metopic suture. Although the most prominent change in metopic synostosis is the formation of the V-shape structure near the forehead, changes in skull patterns also occur near the back of the skull. PC 2 can possibly describe changes in the back of the skull where PC 1 describes the formation of the V-shape structure near the forehead.

Of the first 5 principal components, the first 2 describe the largest portion of the shape variability, accounting for approximately 86% of the variability. PC 1 and PC 2 are attributed to the broadest changes in shape. PC 3, 4, and 5 describe more specific changes in shape that account for around 10% of shape variability. Each of those principal components is only important for very specific descriptions of shape.

IV. DISCUSSION

Craniosynostosis, where there is premature fusion of a cranial vault suture, occurs in 1 in every 1,800 to 2,200 births [2]. While surgery can release the fused suture and correct the cranial deformation, correcting deformity is technically challenging because of the inherent difficulty in "normalizing" a skull. Being able to quantify the degree of the skull deformity will allow the surgeon to tailor the reconstruction and to assess the outcome of surgery.

Developing a single index of severity is important for quantifying the severity of the skull deformation of a child with craniosynostosis. This study has developed a method for developing the index of severity with the given skull data sets. Procrustes Distance is utilized to find overall shape deviation from norm. From the Procrustes Distance data, the largest values correspond to the largest deviations from norm (Skull 1). The largest Procrustes Distance was associated with Skull 20. This would be interpreted as the most deformed. On the single index scale from 1-5 using standard deviations from norm, this skull would correspond to a score of 3.71. Using standard deviations in relation to the single index is favorable because it takes into account the range of Procrustes Distances.

There are some drawbacks inherent in the use of a single index with Procrustes Distance. The index only gives a limited view of a patient. The index yields information only about the overall difference between two different skulls. Although severity is often based on difference from norm, it is also useful to determine where the largest variation occurs. For this reason, it would be recommended that the single index be utilized in conjunction with Principal Component Analysis. These methods allow for a better quantification of deformity. Utilizing these two tools will allow a surgeon to isolate the landmark of greatest deformity.

The index of severity developed in this study was made according to a sample set of patients with metopic craniosynostosis. Various other types of craniosynostosis exist including multiple suture closures. The index of severity utilized was tested only in a limited patient database of children with metopic synostosis. For future study, it is important that different craniosynostosis types are utilized.

As with Procrustes Distance, there are some drawbacks to the use of PCA. In the opposite view of the single index, PCA gives a more narrow view of patient's diagnosis. Principal Component Analysis finds trends in the data and accounts for large shape variability but it still gives us multiple variables. Principal Component Analysis succeeds in reducing the number of variables significantly but the remaining variables must still be interpreted.

Another drawback of PCA is that the principal components obtained do not necessarily have biologically interpretations because they are obtained statistically [5]. Usually the first few principal components are the only ones used to describe biological correlations in data. Landmarks corresponding to biologically or anatomically significant points on a shape are better suited to biological interpretation of principal components.

Further study of deformed skulls with PCA will allow for a better understanding of the methodology used and its clinical relevance. As with Procrustes distance, a larger sample size of skull outlines with different types of craniosynostosis will allow for a more comprehensive study.

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