

# Outcome Assessment in Craniosynostosis: a Prospective, Statistical Analysis of Reoperation Rates

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## SUMMARY

A prospective statistical study of reoperation rates were reviewed in the treatment of 167 consecutive children with nonsyndromic and syndromic craniosynostosis over a six year period at Scottish Rite Children's Medical Center in Atlanta, Georgia. Reoperation equal to or exceeding the magnitude of the original procedure occurred in 7% of cases. Multivariable statistical analysis revealed several factors associated with reoperation. Females and children with syndromic synostoses were more likely to require reoperation. Age at initial surgery, length of operation, and estimated blood loss did not predict a higher reoperation rate.

## INTRODUCTION

The rate of reoperation is an important outcome variable in the surgical treatment of craniosynostosis. Although quantitative changes of craniofacial remodeling are critical to understanding operative results, the decision to reoperate on a particular child is determined primarily from subjective measures of outcome, most commonly aesthetic appearance. Longitudinal studies of reoperation rates from a variety of centers are beginning to appear in the literature [1-5]. Herein, a prospective, six year study of reoperation rates in the treatment of 167

children with nonsyndromic and syndromic craniosynostosis is presented. Preoperative, operative and postoperative variables are analyzed to determine if clinical factors associated with reoperation can be identified.

## MATERIALS AND METHODS

From January 1989 to January 1995, 167 patients with craniosynostosis were surgically managed at the Center for Craniofacial Surgery at Scottish Rite Children's Medical Center in Atlanta, Georgia. All children were seen by a multidisciplinary group and operated on by a team of craniofacial and neurosurgeons. Our protocols for management of non-syndromic and syndromic cases are shown in Tables 1 and 2. Sixty-one children were females and 106 were males, whose ages at the initial operation ranged from 2 weeks to 6 years (mean= 1 year).

**Table 1:** Treatment of Nonsyndromic Synostosis.

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Sagittal synostosis (< 7 weeks, mild to moderate deformity) - Strip craniectomy

Sagittal synostosis (>7 weeks, severe deformity) - Total cranial vault reconstruction at presentation.

Unicoronal, bicoronal, metopic synostosis - Fronto-orbital remodeling, floating forehead at 4-6 months of age.

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**Table 2:** Treatment of Syndromic Craniosynostosis.

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Fronto-orbital advancement, cranial reshaping at 4-6 months of age, earlier if increased ICP present.

\* Redo Fronto-orbital advancement, cranial reshaping as required at approximately 2 years of age.

Monoblock or LeFort III with or without bipartition at 4-7 years of age.

Orthognathic surgery at adolescence (post-growth period).

\* Reoperations evaluated in this study include this procedure.

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Patients treated with lambdoid disorders were excluded from the study. There were 12 children with bicoronal synostosis (7.2%) 39 with metopic (23.4%) 18 with unicoronal (10.8%) 46 with sagittal synostosis who had strip craniectomies (27.5%); 31 with sagittal

synostosis undergoing total cranial vault reconstruction (18.6%); 9 with multiple synostoses (5.4%) and 12 with syndromic synostoses (7.2%). Only the neurosurgeon was involved if a strip craniectomy was carried out, but for the remaining patients 4 combinations of teams conducted the operation. The length of operation (total anesthesia time) in minutes ranged from 81 to 570 (mean= 245 minutes). The mean estimated blood loss was 203 ml. Twenty-three percent of patients required no transfusion, 16% required less than 100 ml and 61 % required more than 100 ml. Thirty-four percent of patients were sent to the intensive care unit after surgery, while 67% were transferred to the floor. Mean length of hospital stay was 4.67 days.

Reoperation was defined as total if the reoperation equaled or exceeded the magnitude of the original procedure and partial if it did not. Partial reoperations were almost always for recontouring with or without hardware removal.

Hardware removal was recorded as a separate partial reoperation.

### Statistical Analysis [6]

Crude statistical analysis of reoperation rates following primary repair of craniosynostosis was carried out using Fisher's Exact

Tests or Generalized Fisher's Exact Tests because of the relatively small number of reoperations (7%).

Interval variables such as operating times in minutes or length of hospital stay were analyzed with T-tests. In some cases the Wilcoxon Rank Sum tests appeared more valid given the skewed distributions of the interval variables.

Options for multiple logistic regression analysis of total reoperation rates using some of the variables that appeared significant on crude, univariate analysis were limited in this study. The rule of thumb is that the total number of variables in the logistic regression should be no greater than the smaller of 10% of the number of reoperations (n=12) or non-reoperations (n=149). This suggests that multivariable analysis is not possible. We can stretch this criterion a bit, however, in order to identify important relationships among the variables, but the results must be interpreted cautiously.

### RESULTS

Twelve of the 167 patients required a total reoperation for a reoperation rate of 7% (Fig. 1). The mean length of follow-up was 2.8 years with a range of 3 months to 6 years. When compared individually, there was no statisti-

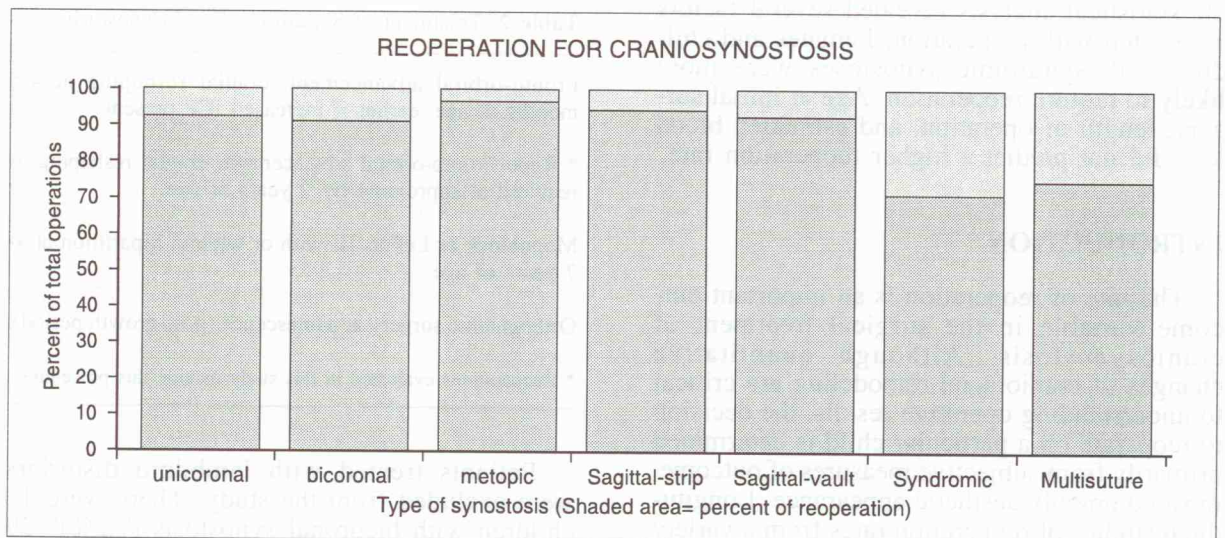


Fig. 1: Reoperation for Craniosynostosis.

cally significant difference in reoperation between the synostoses. However, when reoperation rates in syndromic cases (27%) were compared to nonsyndromic cases (6%), a statistical difference was found ( $p=.037$ , Fisher's exact test, 2-tail). The rates of total reoperation significantly differed by gender (Fisher exact test  $p=.029$ ). Of the female patients 13.8% required reoperation compared to 3.8% of males. Other variables significantly associated with an increased risk of reoperation were operating surgeon and whether or not a complication occurred.

Descriptive analysis for the interval variables also revealed several statistically significant indicators of reoperation. We found the average length of hospital stay was longer in those patients who required a reoperation (Wilcoxon Rank Sum  $p=.018$ ). Similarly, the median surgical minutes (total anesthesia time) in those requiring and not requiring reoperation were 298 minutes and 242 minutes, respectively (Wilcoxon Rank Sum  $p=.052$ ).

To date, 12 of the 167 (7%) patients underwent bony recontouring with hardware removal, while 3 (2%) underwent hardware removal only.

### Multivariable Analysis of Total Reoperation Rates

To implement a multiple regression analysis we compared variables using three groups: syndromic; saggital (strip plus vault reconstruction); and nonsyndromic. One control variable (i.e. diagnosis, gender, age at operation, EBL, transfusion, minutes of surgery, ICU stay, length of hospital stay, complication, and length of follow-up) was added at a time. This permitted investigation into the joint effects of variables, demonstrating the degree to which specific findings did or did not change when controlling for one factor at a time.

Total reoperation rates for syndromic, saggital strip/vault and nonsyndromic cases were 27.3%, 6.5% and 5.9%, respectively. Generalized Fisher Exact Test indicated that the reoperation rates differed in a statistically significant fashion ( $P=.05$ ). Using odds ratios, we estimate that the odds of having a total reoperation were six times larger in the syndromic compared to the other groups.

The effect of female gender on increasing the risk of total reoperation is statistically sig-

nificant even after controlling for diagnostic group ( $p=.04$ ). Using the adjusted odds ratio, females were 3.9 times more likely to require total reoperation than males. The multivariable analysis also indicates that gender and diagnostic group are both independent variables influencing reoperation rates. Another words, there were not more females in the syndromic group negating the significance of the diagnostic category.

Age did not appear to have an effect on reoperation rates. A one year increase in age at operation was not statistically significant after controlling for diagnostic group. The effect on reoperation rate of a 100 ml increase in estimated blood loss was not significant after controlling for diagnostic group. Similarly, length of hospital stay, length of surgery, intensive care unit admission, and the amount of transfusion were not statistically significant after controlling for diagnostic category.

### DISCUSSION

Outcome analysis functions to determine the efficacy and reasonability of a service or a product. This model is complicated in health care by the balance of cost containment to patient satisfaction and quality. Even so, application of outcome analysis provides the medical community a method of comparing and adjusting practice modalities to meet specific demands. It can also provide a means of establishing and communicating standard of care levels to those involved in the health care industry.

This study was completed to evaluate the effectiveness of current treatment methods for craniosynostosis at our institution. It also provided additional information to an increasing amount of data regarding the reasonable expectations of reoperation as it relates to the nature of the dysmorphism and characteristics of the patient. Analysis was limited to clinical outcomes: cost analysis was not included.

Effective use of outcome analysis within and between various centers requires well-defined, comparable treatment modalities. At our institution, fronto-orbital remodeling with a floating forehead was completed at 4-6 months of age for non-syndromic synostosis other than saggital synostosis (Table 1) [7, 8]. This approach is similar to treatment of isolated synostosis in several centers [1, 9, 10]. Bilateral fronto-orbital remodeling has been shown

to be comparable to unilateral remodeling and was done in all cases [2, 8]. Strip craniectomies were limited to sagittal synostosis with mild to moderate deformities. Total cranial vault remodeling was completed for severe deformities if greater than 7 weeks old.

Patients with syndromic craniosynostosis underwent fronto-orbital advancement and cranial reshaping at 4-6 months unless increased cranial pressures required decompression (Table 2). In this review, only frontal bone and skull deformities requiring reoperations were reviewed. Monoblock or Lefort III advancements were done at 4-7 years old. Orthognathic surgery was then completed in adolescence. Again, reoperations for midface deformities were not included in this review.

Whitaker's classification of clinical results after a craniofacial procedure includes category III (C-III) requiring major bone grafting or other osteotomies and category IV (C-IV) requiring duplication of the craniofacial procedure [2].

Total reoperations in our review were classified into the latter group. The increase in reoperation rates of syndromic synostosis (or symmetrical dysmorphisms) were consistent with previous reviews. Whitaker et al. showed a C-IV reoperation rate of 3% for asymmetrical lesions (isolated synostosis) and 64% for the symmetrical lesions (95% in Apert's syndrome). Excluding strip craniectomies, McCarthy had a 6.7% reoperation rate for isolated synostosis and 28.3% for syndromic deformities [7, 8]. Again, the highest reoperation rate was found in Apert's syndrome (37.5%). Finally, Wall and associates demonstrated a reoperation rate of 5.1 % for single suture synostosis and 10.8% for syndromic cases [1]. Surgical approaches similar to ours were utilized in each of the aforementioned studies. Other studies demonstrated similar differences in the reoperation rate between the two groups but were not comparable in surgical technique [5, 10].

In most studies, no differences were seen in reoperative rates for treatment of single suture stenosis as related to age; recommendations for age of primary intervention ranged from 2- 18 months [2, 10, 4]. High reoperative rates in symmetrical deformities led to recommendations from Whitaker and associates for late surgery unless psycho-social issues demanded earlier intervention. Wall and associates showed an increased reoperation rate of 20% in nonsyndromic synostosis when primary treat-

ment was at less than 6 months of age compared to 5.6% in patients greater than 6 months old. In syndromic disorders, patients less than 6 months had a 30.2% reoperation rate, compared to 9.1 % when greater than 6 months (40.9 months follow-up) [1]. Other reviews have shown no relationship of age to reoperative rates [11, 12]. In our study, multivariate analysis demonstrated no relationship between age of initial treatment and need for reoperation in either group (Table 3). We agree that parents should be advised of the possibility of reoperation for any remodeling procedure.

Table 3: Age - Related Reoperation.

	Total Reoperations	
	Yes	No
Number of patients	11	144
Mean age	0.57 yrs	1.0 yrs
Standard Deviation	0.52 yrs	1.27 yrs
Standard Error	0.16 yrs	0.11 yrs
P=NS		

Though descriptive indicators of reoperation were identified with univariate analysis (length of hospital stay, surgical time, complication rates), these indicators were not significant when scrutinized under multivariate analysis. Only diagnostic category and gender (females over males) were found to be independent predictors of increased reoperation rates. The reason for the increased odds ratio associated with females is unknown. Since multivariate analysis has not been done for this variable in other studies, comparison cannot be made.

Finally, there did not appear to be an increase in reoperation rates over longer periods of follow-up. Although this number may change as the mean length of followup increases, it is conceivable that only short follow-up times are necessary to reveal a very important outcome measurement-reoperation.

Outcome analysis of craniofacial procedures is an important tool for establishing standards of care, comparing results between centers, and allowing effective interaction with third party payers. We found the probability of reoperation in the treatment of isolated synostosis to be 6% and was unaffected by the age of treatment.

