A Longitudinal, Statistical Study of Reoperation Rates in Craniosynostosis

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A prospective, statistical study of reoperation rates was done in the treatment of 167 consecutive children with nonsyndromic and syndromic craniosynostosis over a 6-year period at Scottish Rite Children's Medical Center in Atlanta, Georgia. Mean length of follow-up was 2.8 years, with a range of 3 months to 6 years. Reoperation equal to or exceeding the magnitude of the original procedure occurred in 7 percent of cases. Multiple regression analysis revealed several factors associated with reoperation: Females and children with syndromic synostoses were more likely to require reoperation. Total reoperation rates for syndromic and nonsyndromic synostoses were 27.3 and 5.9 percent, respectively. Age at initial surgery, length of operation, and estimated blood loss did not predict a higher reoperation rate. (*Plast. Reconstr. Surg.* 100: 305, 1997.)

The rate of reoperation is an important outcome variable in the surgical treatment of craniosynostosis. Although quantitative changes in 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 6-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 of 1989 to January of 1995, 167 patients with craniosynostosis were managed surgically at the Center for Craniofacial Surgery at the 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 nonsyndromic and syndromic patients are shown in Tables I and II. 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). Because of the recent controversy regarding diagnosis and treatment of posterior skull deformities, a review of patients with a diagnosis of lambdoidal synostosis is currently underway at our institution. Therefore, this group was excluded from the study. There were 12 children with bicoronal synostosis (7.2 percent), 39 with metopic synostosis (23.4 percent), 18 with unicoronal synostosis (10.8 percent), 46 with sagittal synostosis who had strip craniectomies (27.5 percent), 31 with sagittal synostosis undergoing total cranial vault reconstruction (18.6 percent), 9 with multiple synostoses (5.4 percent), and 12 with syndromic synostoses (7.2 percent) (Fig. 1). Only the neurosurgeon was involved if a strip craniectomy was carried out, but for the remaining patients, four combinations of teams conducted the operation. The length of operation (total anesthesia time) ranged from 81 to 570 minutes (mean 245 minutes). The mean estimated blood loss was 203 ml. In all, 23 percent of patients required no transfusion, 16 percent required less than 100 ml, and 61 percent required more than 100 ml. Thirty-four percent of patients were sent to the intensive

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TABLE I
Treatment of Nonsyndromic Synostosis

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 to 6 months of age

care unit after surgery, while 67 percent were transferred to the floor. Mean length of hospital stay was 4.67 days. The children were followed either directly through clinic visits or indirectly through personal phone communication. No patients were lost to follow-up.

Reoperation was defined as total if the reoperation equaled or exceeded the magnitude of the original procedure and partial if it did not (Table III). Partial reoperations were almost always for recontouring with or without hardware removal and were excluded from this review. Hardware removal was recorded as a separate partial reoperation.

Statistical Analysis

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 percent).[6]

Interval variables such as operating times in minutes or length of hospital stay were analyzed with *t* tests. In some cases, Wilcoxon rank-sum tests appeared more valid given the skewed distributions of the interval variables.

TABLE II
Complications Requiring Reoperation

Sagittal		
	Early strip craniectomy	(<7 mos)—None
	Late strip craniectomy	(>7 mos)—None
	Early vault remodeling	(<7 mos)—None
	Late vault remodeling	(>7 mos)—Significant relapse
Metopic		
	Suboptimal cranial contouring	
Bicoronal		
	Suboptimal cranial contouring	×2
	Significant relapse	×2
Unicoronal		
	Residual plagiocephaly	
	Suboptimal cranial contouring	
Multiple		
	Significant relapse	
Syndromic		
	Marked brachioturricephaly	
	Significant relapse	
	Significant relapse	

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 percent of the number of reoperations ($n = 12$) or nonreoperations ($n = 149$). This suggests that multivariable analysis is not possible. This criterion can be stretched in order to identify important relationships among the variables, but the results must be interpreted cautiously. Also, the power of the statistical test is not high due to the limited number of operations.

Results

Twelve of the 167 patients required a total reoperation (Table IV), for a reoperation rate of 7 percent (see 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 statistically significant difference in reoperation between the synostoses. However, when reoperation rates in syndromic patients (27 percent) were compared with those in nonsyndromic patients (6 percent), a statistical difference was found ($p = 0.037$, Fisher's exact test, two-tailed).

Five of the 12 reoperative patients (41.7 percent) were done for significant relapse, as demonstrated clinically and radiographically (Fig. 2). Neither early nor late sagittal strip craniectomies required reoperations; only one patient in the late cranial vault remodeling group for sagittal synostosis (>7 months) demonstrated relapse and reoperation. Relapse was seen in two patients with bicoronal synostosis requiring reoperation and two more patients in this group underwent a second procedure for suboptimal cranial contouring. Three patients with syndromic craniosynostosis required reoperation, two for relapse and the third for marked brachiturricephaly.

The rates of total reoperation significantly differed by gender (Fisher's exact test, $p = 0.029$). Of the female patients, 13.1 percent required reoperation, compared with 3.8 percent of the males. Other variables that initially appeared to be associated with a significantly increased risk of reoperation included the operating surgeon, but this trend did not persist with multiple variable analysis.

Descriptive analysis for the interval variables