Reformation of the sagittal suture following surgery for isolated sagittal craniosynostosis

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Object. Data from animal studies have shown that in experimentally induced craniosynostosis, removal of the involved calvaria results in the formation of new calvaria with time, and sutures redevelop in their normal anatomical positions. However, the pattern of suture reformation following surgery in humans with craniosynostosis remains ill-defined. The aim of this study was to determine the pattern of postoperative suture reformation in children who have undergone surgery for isolated sagittal synostosis and assess possible factors related to suture reformation.

Methods. Records were retrospectively reviewed for 42 consecutive infants who had surgery for isolated sagittal synostosis between 1987 and 2000 and for whom postoperative skull radiographs were available. The radiographs were evaluated for sagittal suture morphology and patency of the coronal and lambdoid sutures. Surgery involved at a minimum 1) a vertex craniectomy, characterized by removal of the sagittal suture and a 1.5- to 2.5-cm piece of adjacent parietal bone with the attached pericranium bilaterally, and 2) parietal osteotomies and/or craniectomies.

The median age at surgery was 3.9 months (range 1.9–7.6 months). The mean duration of follow up was 32.2 months (range 6–144 months). The sagittal suture had reformed in only seven (16.7%) of the children at follow up. In the other 35 (83.3%), the craniectomized bone defects had reossified without any part of the sagittal suture being visible on the radiographs.

Conclusions. There is a very low incidence of suture reformation in children after surgery for isolated sagittal craniosynostosis. Genetic predisposition, inclusion of undiagnosed syndromic patients, and current operative techniques may be some of the factors responsible for the low incidence of suture reformation seen in this series.

KEY WORDS • craniosynostosis • suture reformation • osteogenesis • sagittal suture • dura mater • pericranium • radiography • outcome analysis • pediatric neurosurgery

THE estimated incidence of craniosynostosis ranges from 0.4 to 1 per 1000 live births, with sagittal craniosynostosis being by far the most common form, comprising 56 to 58% of the cases. Surgical correction is usually recommended for treatment of sagittal synostosis, and many operative procedures have been described. These vary from simple linear strip craniectomy of the involved suture and adjacent parietal bone to a similar strip craniectomy with the addition of parietal craniectomies and/or osteotomies, such as barrel staving, with or without occipital or frontal remodeling. Most techniques used in infants do not create a fixed construct of the cranial vault, but rely on the immediate widening of the skull with release of the sagittal suture, supplemented by further remodeling with rapid brain growth or by external pressure with an orthosis for a good result.

The basis of the management of isolated sagittal craniosynostosis is that it typically occurs as an isolated entity in an otherwise healthy child, with operative intervention usually being offered to improve cosmesis. It has been recognized that about 10 to 15% of children with sagittal synostosis have increased intracranial pressure, and some physicians have recommended surgery in the hope that it might prevent the adverse effects of raised intracranial pressure.

In spite of the relatively high incidence of patients with sagittal craniosynostosis, most of whom undergo surgical correction, follow-up studies on the fate of the sagittal suture following surgical intervention are lacking. The purpose of this study was to determine the incidence of reformation of the sagittal suture following surgical procedures for sagittal synostosis that involved a sagittal strip craniectomy, and to discuss factors that may have a bearing on suture reformation.

Clinical Material and Methods

In this retrospective study, the database of the neurosurgical division at British Columbia’s Children’s Hospital was searched to identify children who underwent surgery for isolated sagittal craniosynostosis between January 1, 1987 and December 31, 2000. Surgery involved at a minimum 1) a vertex craniectomy, characterized by removal of the sagittal suture plus a 1.5- to 2.5-cm piece of adjacent parietal bone with the attached pericranium on each side,
and 2) parietal osteotomies and/or craniectomies. Children who had the vertex bone flap replaced were excluded from the study. Postoperative radiographs were sought, and the study group comprised those patients in whom such radiographs were available for review. The radiographs were evaluated for sagittal suture morphology and patency of the coronal and lambdoid sutures. In our review, we found that postoperative skull radiographs were not obtained uniformly in this population and their availability in large part reflected the practice patterns of the two senior surgeons. Whereas one surgeon requested skull radiographs routinely at 1 to 2 years after surgery, the other obtained skull radiographs only in symptomatic children.

The clinical charts were reviewed to assess the cosmetic outcome, defined as the surgeon’s and/or parents’ subjective perception of cosmetic outcome at the last follow-up examination.

**Results**

We identified 114 children who had undergone surgery for isolated sagittal craniosynostosis in the specified period. Of these, 42 children had undergone a minimum of vertex and parietal craniectomies, had no bone replaced over the sagittal sinus, and had postoperative skull radiographs available. These 42 patients composed the study group.

The median age at surgery of the patients in the study group was 3.9 months (range 1.9–7.6 months) and the mean duration of follow up was 32.2 months (range 6–144 months). At least part of the sagittal suture was found to have reformed in seven children (Fig. 1), and in 35 (83.3%) of the children, the craniectomized bone defects had ossified without any part of the sagittal suture being visible on the radiographs. Four children developed concomitant coronal synostosis postoperatively, and the sagittal suture did not reform in any of these four. The cosmetic outcome in these four patients was not good, whereas the other 38 children were considered to have a good cosmetic outcome.

**Discussion**

Literature on the fate of involved sutures in isolated non-syndromic craniosynostosis following surgical intervention is extremely sparse. Although isolated cases of pansynostosis after surgery for single-suture craniosynostosis have been reported, in only one previous study have investigators attempted to determine the incidence of reformation of the sagittal suture in children who have undergone surgery for isolated craniosynostosis. The results of our study indicate an extremely low rate of suture reformation in infants after strip craniectomy for isolated, nonsyndromic sagittal craniosynostosis. These results add to the discussion about the cause and management of isolated sagittal craniosynostosis.

The debate regarding the cause of craniosynostosis has primarily revolved around two competing theories: Moss’s hypothesis of a primary abnormality at the cranial base and Babler’s hypothesis that the abnormality is in the affected calvarial sutures. Concerning syndromic craniosynostosis, it is often thought that the primary abnormality involves the cranial base, and that cranial base alterations affect the dural reflections, thereby somehow predisposing to premature closure of the sutures. As surgery does not correct the underlying pathology, it is not surprising that synostosis can recur in these cases.

In contrast, nonsyndromic, single-suture synostosis is usually ascribed to compressive intrauterine forces that act on individual sutures; recurrent synostosis is said to be less likely in these cases because the surgery usually removes the pathological suture. Support for this hypothesis comes from animal experiments that have shown that if the calvaria is excised and discarded, a new calvaria forms with time and sutures redevelop in their normal anatomical positions. These neosutures initially do not have the interdigitations of normal sutures, and the interdigitations form after a variable time period. Reformation of sutures in humans is believed to follow the same process. In all children who experienced suture reformation in this study, the neosuture did not have the normal interdigitations (Fig. 1), but it is possible that such interdigitations would have developed in the longer term.

In animal studies, the reskeletonization of the calvaria and reformation of the sutures have been found to depend on the continuity of the dura mater and the age of the animal. In fact, it has been unequivocally shown that both dura mater and pericranium have osteogenic properties, and that dura appears to be the source of central new bone, whereas pericranial contact appears to enhance peripheral new bone formation. If one considers single-suture craniosynostosis as an isolated event, removal of the pathological suture should similarly result in reformation of a normal
suture. However, as our study shows, reformation of the suture occurs only in a minority of the cases, the reasons for which remain poorly defined. We hypothesize that a number of factors, including (but not limited to) operative techniques, genetic predisposition, and the inclusion of unrecognized syndromic patients, may be responsible for the low rate of suture reformation in this study.

Sagittal craniosynostosis surgery usually involves removal of the central strip of bone with the attached pericranium; removal of this pericranium could potentially impair bony regeneration as well as suture reformation. It is also a common practice to coagulate the bleeding points on the dura over the sagittal sinus after removal of the bone flap, which again could impair the osteogenic capacity of the dura, resulting in an uneven bony contour due to areas of poor ossification being interspersed with areas of excessive bone formation. As the primary purpose of surgery in sagittal craniosynostosis is cosmetic, the persistence of bony defects and uneven contour of the bony regrowth may result in patient dissatisfaction and occasionally in repeated surgery. Although unproven, limiting coagulation on the dura and replacing the pericranium could potentially result in consistent bone regeneration with smooth contour and reformation of a normal suture. However, further prospective studies would be required to prove this hypothesis.

The fact that reformation of part of the sagittal suture did occur in seven children in this study could also indicate that isolated sagittal suture synostosis is not a single disease entity, but a spectrum of disorders. In those children who experienced suture reformation after surgical removal of the fused suture, there may be a primary suture abnormality, perhaps caused by calvarial deformation in utero. However, in the remainder, in whom reossification of the cranietomized bone defects occurred, it is possible that the sutureal abnormality was secondary to a problem at the cranial base. There is yet another group of patients, who appear initially to have isolated sagittal synostosis, but in whom synostosis of other sutures evolves after surgery for the sagittal suture craniosynostosis. Four children developed coronal synostosis postoperatively in our study, and these children may have a primary genetic defect that predisposes them to suture closure, similar to that seen in patients with syndromes such as Crouzon. In all four of these children, the sagittal suture did not reform after surgery.

Thus, operative technique, genetic factors, and inclusion of undiagnosed syndromic patients could be some of the factors responsible for the extremely low suture reformation rate seen in our study. Changes in operative practice and larger prospective studies could clarify the role of each of these factors and possibly lead to a better understanding of this seemingly simple, but actually complex disorder.

Conclusions
After surgery for isolated sagittal craniosynostosis in children, there is a very low incidence of reformation of the sagittal suture. The variability in reformation of the suture after surgery suggests a heterogeneous etiology and pathogenesis for isolated sagittal synostosis. Genetic predisposition to synostosis, inclusion of undiagnosed syndromic patients, and current operative techniques may be some of the factors responsible for the low incidence of reformation after surgery for isolated sagittal synostosis.

References