Coronal and lambdoid suture evolution following total vault remodeling for scaphocephaly

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OBJECTIVE Different types of surgical procedures are utilized to treat craniosynostosis. In most procedures, the fused suture is removed. There are only a few reports on the evolution of sutures after surgical correction of craniosynostosis. To date, no published study describes neosuture formation after total cranial vault remodeling. The objective of this study was to understand the evolution of the cranial bones in the area of coronal and lambdoid sutures that were removed for complete vault remodeling in patients with sagittal craniosynostosis. In particular, the investigation aimed to confirm the possibility of neosuture formation.

METHODS CT images of the skulls of children who underwent operations for scaphocephaly at the Hôpital Femme Mère Enfant, Lyon University Hospital, Lyon, France, from 2004 to 2014 were retrospectively reviewed. Inclusion criteria were diagnosis of isolated sagittal synostosis, age between 4 and 18 months at surgery, and availability of reliable postoperative CT images obtained at a minimum of 1 year after surgical correction. Twenty-six boys and 11 girls were included, with a mean age at surgery of 231.6 days (range 126–449 days). The mean interval between total vault reconstruction and CT scanning was 5.3 years (range 1.1–12.2 years).

RESULTS Despite the removal of both the coronal and lambdoid sutures, neosutures were detected on the 3D reconstructions. All combinations of neosuture formation were seen: visible lambdoid and coronal neosutures (n = 20); visible lambdoid neosutures with frontoparietal bony fusion (n = 12); frontoparietal and parietooccipital bony fusion (n = 3); and visible coronal neosutures with parietooccipital bony fusion (n = 2).

CONCLUSIONS This is the first study to report the postoperative skull response after the removal of normal patent sutures following total vault remodeling in patients with isolated sagittal synostosis. The reappearance of a neosuture is rather common, but its incidence depends on the type of suture. The outcome of the suture differs with the incidence of neosuture formation between these transverse sutures. This might imply genetic and functional differences among cranial sutures, which still have to be elucidated.

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KEYWORDS neosuture; heterogeneity; single sagittal synostosis; craniosynostosis; scaphocephaly; total vault remodeling

DIFFERENT types of surgical procedures are utilized to treat craniosynostosis. In most procedures, the fused suture is removed. In 1973, Shillito¹ reported for the first time the reappearance of a suture radiologically identical to a normal suture on postoperative control radiographs after strip craniectomy. Despite this early observation, only a few reports on the evolution of sutures after surgical correction of craniosynostosis have been published in the past decades. In some instances, the occurrence of a neosuture with almost the same characteristics of a normal suture has been described at the site

of osteotomy.^{1–5} These studies reported experiences with endoscope-assisted procedures, strip craniectomies, or spring-assisted cranioplasties. Although secondary fusion of patent coronal and lambdoid sutures after cranial vault remodeling for scaphocephaly has been described,^{6–8} to our knowledge no study has described this phenomenon after total cranial vault remodeling.

The objective of this study was to study the evolution of the cranial bones in the area of coronal and lambdoid sutures removed for complete vault remodeling in patients with sagittal craniosynostosis. In particular, this investiga-

tion aimed to confirm the possibility of neosuture formation and whether such an event could differ at the sites of coronal and lambdoid sutures or vary in relation to the known heterogeneity of sagittal suture synostosis.⁹

Methods

CT images of the skulls of children who underwent operations for scaphocephaly at the Hôpital Femme Mère Enfant, Lyon University Hospital, Lyon, France, from 2004 to 2014 were retrospectively reviewed. Inclusion criteria were diagnosis of isolated sagittal synostosis, age between 4 and 18 months at surgery, and availability of postoperative CT images at a minimum of 1 year after surgical correction.

This study was approved by our ethical committee according to French regulations for medical studies.

Population

Of 186 children who underwent surgical correction for isolated scaphocephaly with open total vault remodeling in the period considered, only 37 underwent a postoperative CT scan that could be analyzed for this study. Indeed, only patients who had a postoperative CT scan for a head traumatism or to control a residual bone defect could be included because no systematic postoperative CT scans were routinely performed.

There were 26 boys and 11 girls. The mean age at surgery was 231.6 days (range 126–449 days). No perioperative surgical complications or postoperative infections were reported in the medical files. The mean interval between total vault reconstruction and CT scan was 5.3 years (range 1.1–12.2 years). No repeat surgeries for any reason were necessary.

Operative Technique

All children underwent correction of sagittal synostosis with the following technique. The patient was placed prone. We made a bicoronal incision, lifted the periosteum flap, and created two parietal bone flaps using a high-speed craniotome. We removed a minimum of 5 mm of bone on each side of both the coronal suture (down to the pterion) and lambdoid suture (down to the asterion) with straight bone-cutting forceps (Fig. 1). We removed the bone strip with the fused sagittal suture and cut its longitudinal midline into two strips. These two bone strips were then fragmented into three to four segments, which were used to reconstruct the vertex (the bone pieces were placed perpendicular to their initial orientation between the upper parietal edges in order to enlarge the space that resulted from the removal of the fused sagittal suture). Additional occipital and frontal osteotomies were performed, if needed, to reshape the occipital pole and the forehead. With linear osteotomies, we also enlarged the transversal diameter at the level of the temporal bone below the squamosal suture. The final reconstruction was performed using resorbable plates and stitches. The periosteum was replaced over the bony pieces.

CT Scan Analysis

The bone at the site of the removed coronal and lamb-

FIG. 1. Early postoperative 3D reconstruction CT scans illustrating the osteotomies performed with our open total vault remodeling technique. Note that both the lambdoid (A) and coronal (B) sutures are largely removed.

doid sutures was evaluated on 3D reconstruction CT images and native 2D images. According to the literature,^{20–22} neosuture was defined as the reformation of a new suture with normal radiologic appearance on the 3D CT scan (“zigzag” pattern). Therefore, it was clearly a different radiologic morphology than the straight bony osteotomy line that is classically seen after craniectomy.

Statistical Analysis

IBM SSPS (version 20, IBM Corp.) and ANOVA were used for statistical analyses. Significance was assumed for $p \leq 0.05$.

Results

No subject developed a neosagittal suture. Despite removal of both the coronal and lambdoid sutures, neosutures were detected on the 3D reconstructions of the coronal sutures in 22 patients (59%) and of the lambdoid sutures in all but 5 patients (86%). All findings were bilateral. These neosutures were seen as early as 1.1 years after the removal of the suture, but also as late as over 12 years in a patient who received late follow-up CT. All combinations were seen (Fig. 2): visible lambdoid and coronal neosutures ($n = 20$) (Fig. 3); visible lambdoid neosuture with frontoparietal bony fusion ($n = 12$) (Fig. 4); frontoparietal and parietooccipital bony fusion ($n = 3$) (Fig. 5); and visible coronal neosuture with parietooccipital bony fusion ($n = 2$) (Fig. 6). ANOVA did not find any significant differences in the mean interval between total vault reconstruction and CT scan ($p > 0.05$) or in the mean age at surgery ($p > 0.05$) between groups.

Discussion

Since the first description of the appearance of a so-called neosuture after surgical correction of craniosynostosis in 1973, the literature remains surprisingly scarce on the response of the skull after the removal of cranial sutures. Though the importance of such sutures has been well understood since the work of Otto⁹ on skull growth perpendicular to the suture, and further refined by Virchow,¹⁰ the fate of sutures after surgical treatment of craniosynostosis has been poorly studied.

FIG. 2. Bar graph showing the different postoperative combinations of neosutures and bone fusion.

In fact, in 1973, Shillito¹ reported a series of 164 patients who underwent strip craniectomies for different kinds of craniosynostosis. In 16% ($n = 26$) of their patients, they found suture reformation on control plain radiographs. Forty-three years later, in 2006, a further study investigated the reformation of sutures.¹¹ In this paper on 42 strip

craniectomies for isolated sagittal synostosis, 16.7% ($n = 7$) of patients presented with a new sagittal suture.¹¹ Today, the main criticism of this series is that reappearance of sutures was evaluated on simple plain radiographs, which could have underestimated or overestimated the real incidence of neosuture formation compared with CT.

FIG. 3. Late postoperative 3D reconstruction CT scan obtained 5 years after surgery, showing lambdoid and coronal neosutures.

FIG. 4. Late postoperative 3D reconstruction CT scan obtained 8.5 years after surgery, showing lambdoid neosuture and frontoparietal bony fusion.

FIG. 5. Late postoperative 3D reconstruction CT scan obtained 6 years after surgery, showing frontoparietal and parietooccipital bony fusion.

More recently, some reports have been published on postoperative skull evolution after suturectomies performed using different techniques. In 2014, Sauerhammer et al.⁵ reported on 17 patients with unicoronal synostosis who underwent endoscope-assisted suturectomy and postoperative helmet therapy. Overall, at an average of 31.9 months after surgery, 88.2% ($n = 15$) of patients had a persistent craniectomy gap, evidence of neosuture formation, or both without bony union. Only 17.6% ($n = 3$) of children in this series had complete neocoronal suture reformation. The remaining 2 patients showed evidence of neosuture formation and craniectomy gap, as well as areas of bony union or healing across the craniectomy gap. Sauerhammer et al.⁵ also reported that the rate of neosuture formation increased as time passed, while the rate of craniectomy gap decreased. This could also be interpreted as the growth of the bone on both sides of the craniotomy rather than actual suture formation.

Interestingly, Sauerhammer et al.⁵ noted a difference in the evolution of frontosphenoidal sutures. Indeed, only patients with evidence of bony union had a closed frontosphenoidal suture, while the others still surprisingly showed an open suture.⁵ In 2016, Salehi et al.³ also reported different results with the same operative technique. In a large series of 96 patients with sagittal ($n = 73$), unicoronal ($n = 11$), bicoronal ($n = 7$), and lambdoid ($n = 5$) synostosis, they found an overall rate of neosuture formation of 23.9% ($n = 23$); however, rates differed between patients with sagittal (22%), unicoronal (36%), bicoronal (29%), and lambdoid (20%) synostosis. Also, if only complete neosutures were considered, the overall rate dropped to 14.6% ($n = 14$). The most recent experience was reported by Säljö et al.⁴ in 2019. They used spring-assisted cranio-plasty and checked for the presence of neosutures on CT

FIG. 6. Late postoperative 3D reconstruction CT scan obtained 3.5 years after surgery, showing coronal neosuture (A) and parietooccipital bony fusion (B).

prior to spring removal and 6 months after initial surgery in 84 patients with sagittal ($n = 70$), lambdoid ($n = 3$), bicoronal ($n = 3$), unicoronal ($n = 1$), or multiple ($n = 7$) synostosis. In total, 19% ($n = 16$) had complete neosuture formation, and an additional 8% ($n = 7$) had partial neosuture formation at the site of the removed fused suture on preoperative CT.⁴

To our knowledge, no studies have reported neosuture formation after total vault remodeling involving patent sutures. Because our surgical technique included the removal of both coronal (down to the pterion) and lambdoid (down to the asterion) sutures, we were able to examine postoperative changes after the removal of nonpathological sutures for the first time, in addition to those that occurred at the level of the fused sagittal suture. We evaluated cranial response to the removal of both pathological and nonpathological sutures, with a mean follow-up of more than 5 years after operations for cranial remodeling. None of our patients had a sagittal neosuture. This is different from all reported data on other surgical techniques.^{1,3,4,11} A possible explanation for the different results could be related to the use of the bone segments interposed between the two superior parietal edges in our technique. In our children, we reconstructed the vertex of the skull by placing the interposed bone pieces into an orientation perpendicular to the normal orientation of the sagittal suture, thereby possibly preventing the formation of a new suture.

Conversely, we found that a neosuture was present in the region of the coronal or lambdoid sutures in the great majority of the children. Nevertheless, it is worth noticing the discrepancy between the reappearance of a neosuture at the level of the coronal and lambdoid sutures. Indeed, almost all subjects showed a neolambdoid suture, but only half showed a neocoronal suture. Although we cannot elucidate the cause of this discrepancy on the grounds of this study, we certainly can affirm different incidences of postoperative neosuture formation at the level of the coronal and lambdoid sutures.

Several hypotheses may explain neosuture formation. One hypothesis considers the two conflicting theories regarding the pathogenesis of craniosynostosis (primary abnormality of the cranial base vs anomaly of the sutures)^{12,13} and the higher rate of resynostosis in syndromic craniosynostosis compared with that of nonsyndromic craniosynostosis. According to this hypothesis, in nonsyndromic cases

without skull base abnormalities, removal of the pathological suture is curative and allows the skull to resume normal growth. Animal models confirm this hypothesis. It has been shown that after neonatal craniectomy in rabbits, normal calvarial and sutural development may occur.^{14,15} A second hypothesis emphasizes the osteogenic potential of the dura and pericranium. The same authors who showed normal calvarial and sutural development after calvariectomy in rabbits pointed out the need for dural and periosteal integrity to achieve such an osteoblastic response.^{14,15} Sauerhammer et al.⁵ used this hypothesis to explain the benefit of endoscope-assisted procedures that minimize soft-tissue disruption compared with open procedures, as other animal model have shown less effective redevelopment of calvaria and worse patency of cranial sutures in the case of dural alteration.¹⁶ However, according to Salehi et al.,³ the dura is preserved in endoscope-assisted procedures, but the pericranium over the suturectomy site may be disrupted. We have shown that reappearance of suture is common following total vault remodeling, and in our technique, we stress the importance of replacing and maintaining the periosteum over the bony pieces of the reconstructed calvaria with accurate sutures. Sauerhammer et al.⁵ also argued that the use of a helmet increases the rate of neosuture appearance. Indeed, the pressure applied to the growing brain and cranial base may be transmitted to the dura, thereby influencing sutural bone deposition and patency.^{17–19} Sutural bone formation would vary in response to mechanotransduction, resulting in alteration of dural signals at the sutures.^{13,20–26} Therefore, Sauerhammer et al.⁵ claim that the action of the helmet associated with suturectomy would accelerate bone formation along the craniectomy gap by altering the dural signals at the sutures. The combination of these factors would result in inhibition of bony deposition at the level of the bone gap, leading to reappearance of the suture. Even if this hypothesis is true, in this series we have shown a high incidence of neosuture formation in children who underwent total cranial vault remodeling without a postoperative helmet.

A third theory hypothesizes that because the function of the cranial sutures ends at different times in different subjects, this function could also start at different times. Therefore, the premature closure of a suture does not represent the premature end of function, but rather a default in the start of function.⁴ So, function might be “switched on” at the time of surgery, and a new anatomically and functionally normal suture could form after the fused suture is removed (or close when the function is “switched off”). This on/off switch may be mediated by dural signals.⁴ In our series, normal coronal and lambdoid sutures were present in all patients prior to surgery, i.e., the start function was already “switched on” and allowed the regrowth of neosutures. According to this hypothesis, failure of the suture to reappear would be abnormal. Indeed, the disappearance of patent coronal and lambdoid sutures has been described after surgical correction of scaphocephaly that spared these sutures. Secondary closure of these sutures may be explained by changes in mechanical stress forces.⁶

Regardless of the mechanism responsible for neosuture formation after the removal of normal sutures, as occurred following our skull-remodeling procedure, the reappearance

of nearly all lambdoid sutures but only little more than half the coronal sutures remains to be explained.

Conclusions

This is the first study to report the postoperative skull response after the removal of normal patent sutures following total vault remodeling in patients with isolated sagittal synostosis. Our results show that the reappearance of neosutures is rather common, but its incidence depends on the type of suture. Lambdoid sutures reformed considerably more frequently than coronal sutures. Our results show that the outcome of the suture differs with the incidence of neosuture formation between these transverse sutures. This might imply genetic and functional differences among cranial sutures, which still need to be elucidated. A better understanding of this phenomenon is also necessary before interpreting the results of endoscopic or spring treatments of simple craniosynostosis and the use of postoperative helmet therapy.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Di Rocco, Beuriat. Acquisition of data: Beuriat. Analysis and interpretation of data: Di Rocco, Beuriat. Drafting the article: all authors. Critically revising the article: all authors. Reviewed submitted version of manuscript: Di Rocco, Szathmari, Chauvel-Picard, Gleizal, Paulus, Mottolese. Statistical analysis: Beuriat. Study supervision: Di Rocco.

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