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CASE REPORT

Pediatric/Craniofacial

Neosagittal Suture Formation after Endoscopic Sagittal Strip Craniectomy: A Case Report and Literature Review

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Summary: The fate of the excised synostotic suture in craniosynostosis remains relatively understudied. The purpose of this report is to describe a case of neosagittal suture formation following endoscopic excision of a pathology-proven synostotic suture, with CT demonstration of complete reossification in the areas adjacent to the neosagittal suture. We additionally review the existing literature on neosuture formation that has been published over the past 50 years. We conclude that continued investigation is warranted, both through histological comparison of normal and neosutures and through studies to determine clinical risk factors, as this may improve our understanding of the underlying mechanism of pathologic premature suture fusion in craniosynostosis. *(Plast Reconstr Surg Glob Open 2021;9:e3368; doi: 10.1097/GOX.0000000000003368; Published online 22 January 2021.)*

Although there is a large volume of clinical and animal studies devoted to investigating the pathogenesis of premature suture fusion in craniosynostosis,[1] the behavior of the synostotic suture following surgical excision is poorly understood. Theoretically, several actions during suturectomy, whether performed open or endoscopically, should impair refusion. Suturectomy consists of removal of bone with pericranium and coagulation of dural bleeding, which should impede both pericranial and dural osteogenic potential.[2,3] In addition, there are 2 main competing theories regarding the development of craniosynostosis: Babler hypothesizes that the abnormality is confined to the affected suture, likely related to intrauterine compression and other gene–environmental interactions,[4] whereas Moss and Young suggest that pathologic fusion is related to abnormalities of the cranial base.[5] If the former is correct, suture reformation or refusion would be unlikely, as the underlying pathology is addressed with ostectomy, though this is not the case with the latter theory.

Despite these theoretical factors, there have been documented instances of both suture refusion and reformation after surgical treatment for craniosynostosis. Refusion can lead to recurrent craniosynostosis and poor morphologic improvement, often requiring reoperation.[6–8] In contrast, instances of neosuture development in place of the excised synostotic suture are likely underappreciated due to a potentially asymptomatic presentation. We present the case of a sagittal suture reforming in its entirety following endoscopic suturectomy, and discuss this finding in the context of what is currently known regarding neosuture formation following surgical management of craniosynostosis.

CASE

A 3-week-old male child born following an uncomplicated gestation via vaginal delivery was referred for abnormal head shape. Clinical examination revealed features consistent with sagittal synostosis, including a palpable sagittal ridge, biparietal narrowing, frontal bossing, mild occipital protuberance, and a cranial index of 0.66. The anterior fontanelle was closed. Coronal, metopic, and lambdoid sutures were normal to palpation. No genetic or additional physical abnormalities were identified.

At the age of 3 months, the patient underwent endoscopic sagittal suturectomy and biparietal osteotomies. Synostectomy was performed via 2 incisions, 1 just posterior to the anterior fontanelle and a second at lambda. The fused sagittal suture and additional bone were excised to yield a 5-cm wide gap in the midline. Barrel stave osteotomies were then made anterior to the lambdoid sutures and posterior to the coronal sutures to facilitate outward

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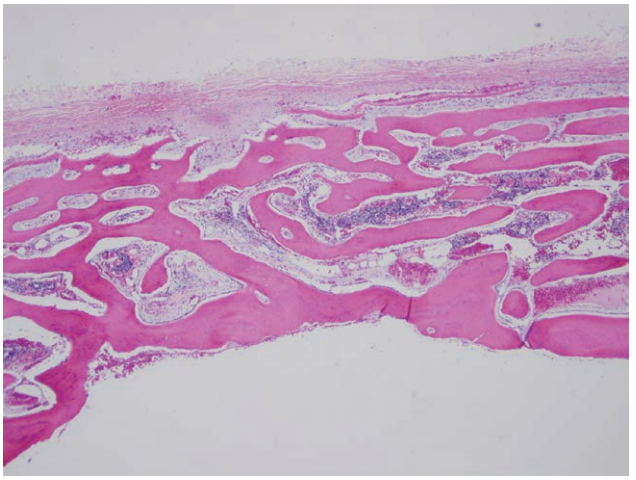

Fig. 1. Histology of synosed sagittal suture. Histologic examination of excised bone submitted to pathology at the time of sagittal suturectomy demonstrates white-pink osteocartilaginous cut surfaces upon sectioning, consistent with a fused suture.

movement of the parietal bones. The fused sagittal suture was sent to pathology for evaluation; pathological analysis confirmed suture fusion consistent with sagittal synostosis (Fig. 1). Postoperatively, the patient recovered uneventfully. He underwent 5 months of helmet therapy, beginning 1 week postoperatively. Cranial index at 1 year postoperatively was 0.80.

The patient re-presented at the age of 2 years due to parental concern regarding a new "lump" at bregma. Ophthalmology examination at the time did not show papilledema. He did not demonstrate any signs of increased intracranial pressure. Occipitofrontal circumference was 53.5 cm and tracking above and parallel to the head growth curve. Clinical examination demonstrated mild scaphocephaly. CT scan image was obtained demonstrating a patent sagittal suture (Fig. 2). There were no radiographic signs of increased intracranial pressure. Given that the patient was asymptomatic, no intervention was taken. The patient is currently doing well without any stigmata of arrest of cranial growth.

DISCUSSION

We present a case of complete neosagittal suture formation following endoscopic suturectomy with parietal osteotomies in a case of isolated sagittal synostosis. This case adds to Kinsella et al's similar case report in 2011, in which the authors reported parasagittal suture formation with a persistent small bony defect in a 7-year-old boy after extended strip craniectomy with biparietal wedge osteotomies at the age of 3 months, also for isolated sagittal synostosis.[9] However, unlike this prior report, we demonstrate pathology-proven premature suture fusion at the time of suturectomy, and complete reossification in the areas adjacent to the neosagittal suture. Furthermore, the timeline for development of the neosuture was significantly more accelerated; in our case, the neosuture was detected at the age of 2 years, compared with the age of 7 years in Kinsella et al's report.

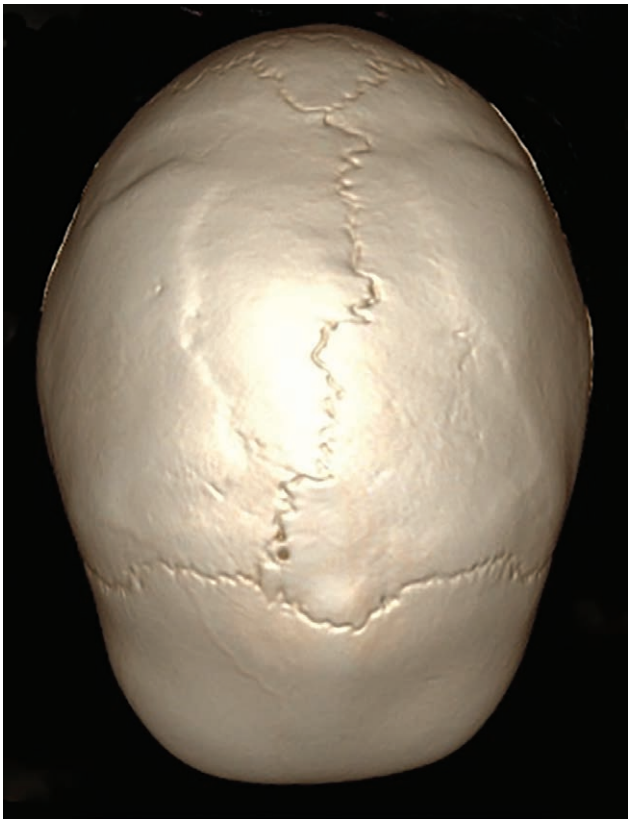

Fig. 2. Postoperative CT scan image demonstrating neosagittal suture. CT scan at approximately 2 years postoperatively demonstrates a patent sagittal suture with slight rightward deviation of the anterior sagittal suture at its confluence with the coronal suture. There is age-appropriate patency of the coronal and lambdoid sutures, in addition to closure of the metopic suture.

Since the initial observation of neosuture formation following suturectomy in the 1960s, there have been a limited number of case series exploring this finding (Table 1).[2,9–14] Early reports relied upon plain radiographic evaluation, whereas more recent case series have employed postoperative CT to examine the behavior of the area of the excised synostotic suture. In comparison with plain radiograph, CT can detect finer details, such as bony interdigitation, that characterize normal cranial sutures.[11,12]

In all of the cases in Table 1, no patients with partial or complete neosuture formation required additional operative interventions. This is in contrast to premature suture refusion or secondary suture fusion (ie, fusion of an initially patent suture), in which secondary ossification can lead to unfavorable head shape with sequelae of restricted brain growth and increased intracranial pressure.[15–17] In our case, the patient initially presented due to the parents' perceived cranial abnormality, though clinical examination revealed this to be a normal phenotypic variant of bregma. Thus, as in the previously reported cases of neosuture formation, repeat surgical intervention was not required to address any clinically significant sequelae of craniosynostosis.

Table 1. Neosuture Formation following Surgery for Craniosynostosis

Study	No. Cases of Neosuture	Suture	Syndromic	Primary Craniectomy Technique	Diagnostic Method
Shillito, 1973	26 (13%)	11 sagittal 10 coronal 1 metopic 4 multiple	NA	Open strip craniectomy	XR
Agrawal et al, 2006	7 (16.7%)	Sagittal	No	Open strip craniectomy with biparietal wedge osteotomies	XR
Kinsella et al, 2011	1	Sagittal	No	Open strip craniectomy with biparietal wedge osteotomies	CT
Sauerhammer et al, 2014	12 (70.6%) partial 3 (17.6%) complete	Coronal	No	Endoscopic suturectomy and helmet therapy	CT
Salehi et al, 2016	9 (9.4%) partial 14 (14.6%) complete	9 sagittal 4 coronal 1 lambdoid	No	Endoscopic suturectomy ± wedge osteotomies and helmet therapy	CT
Saljo et al, 2019	7 (8%) partial 16 (19%) complete	20 sagittal 2 lambdoid 1 multiple	NA	Spring-assisted cranioplasty	CT
Persad et al, 2020	10 (31.3%) partial	Sagittal	N	Endoscopic suturectomy with biparietal wedge osteotomies and helmet therapy	CT

CT, computed tomography; NA, not available; XR, radiograph.

There are several take-aways from the collective consideration of these studies and our reported case. First, the factors governing neosuture formation remain unclear because some patients developed partial versus complete neosutures, whereas others did not. Second, the role of syndromic conditions in neosuture development is not illustrated in these case reports and series because the majority concerned isolated nonsyndromic synostoses. Two studies included patients with multisutural synostoses, though the occurrence of an associated syndrome was not specified.[10,13] Lastly, the clinical significance of neosuture formation is worthy of continued investigation, as long-term implications remain unknown. Continued study of this occurrence, both through histological comparison of normal and neosutures, and through studies to determine clinical risk factors, may ultimately help elucidate the underlying mechanism of pathologic premature suture fusion in craniosynostosis.

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