

Evaluation of Long-term Outcomes of Facial Sensation following Cranial Vault Reconstruction for Craniosynostosis

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Background: Cranial vault reshaping to correct craniosynostosis in infants may injure terminal branches of the trigeminal nerve, namely the supraorbital, supratrochlear, zygomaticofacial, and zygomaticotemporal nerves, especially if a fronto-orbital advancement is performed. Despite numerous studies demonstrating successful esthetic outcome after FOA, there are no long-term studies assessing facial sensation after possible damage to these nerves as the result of surgery.

Methods: A cross-sectional case-control research design was used to evaluate facial sensory threshold in the trigeminal branches after cranial vault reconstruction in children with isolated, nonsyndromic metopic, and unicoronal craniosynostosis, compared with those with sagittal craniosynostosis and age-matched nonaffected controls. Study participants were recruited from the Hospital for Sick Children between the ages of 6 and 18 years. Sensory outcome was determined using the Weinstein Enhanced Sensory Test, the Ten Test, and self-reported facial sensibility function questionnaire.

Results: The sensory outcomes of 28 patients and 16 controls were examined at an average age of 9.6 years and 10.3 years, respectively. No subjective or objective sensory deficit was noted in supraorbital, supratrochlear, zygomaticofacial, or zygomaticotemporal nerve distributions between groups. Qualitative reports of facial sensibility function indicated no difference in subjective sensation, protective sensation, or motor behavior between groups.

Conclusions: These results suggest that while sensory nerve injury during routine FOA may occur, it does not result in a quantifiable nor clinically significant long-term sensory deficit threshold. Patients do not develop long-term neuropathic pain following surgical intervention. (*Plast Reconstr Surg Glob Open 2019;7:e2135; doi: 10.1097/GOX.00000000002135; Published online 11 March 2019.*)

BACKGROUND

Craniosynostosis is the premature fusion of one or more of the sutures that make up the cranial vault and cranial base.¹ Growth restriction perpendicular to the

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The authors have no financial interest to declare in relation to the content of this article.

Copyright © 2019 The Authors. Published by Wolters Kluwer Health, Inc. on behalf of The American Society of Plastic Surgeons. This is an open-access article distributed under the terms of the Creative Commons Attribution-Non Commercial-No Derivatives License 4.0 (CCBY-NC-ND), where it is permissible to download and share the work provided it is properly cited. The work cannot be changed in any way or used commercially without permission from the journal. DOI: 10.1097/GOX.00000000002135 fused sutures, and compensatory growth in the remaining patent sutures in response to brain growth, results in suture-specific patterns of cranial vault dysmorphism. The majority of craniosynostosis cases are single-suture, nonsyndromic in nature, affecting between 1 in 2,000 and 1 in 5,000 infants.² The most commonly involved sutures are the sagittal suture, producing a scaphocephaly deformity, followed by the metopic suture and the coronal suture (unilateral), producing trigonocephaly and plagiocephaly, respectively. The primary drive for surgical intervention in children with single-suture, nonsyndromic craniosynostosis is improvement of facial form, thereby curtailing the negative psychosocial impact associated with visible facial difference and improving the quality of life.^{3,4} Functional risks of craniosynostosis also prompt surgical intervention, including elevated intracranial pressure and hydrocephalus, which can lead to neurodevelopmental delay and visual abnormalities but are rare in these cases.

The majority of single-suture nonsyndromic cases of craniosynostosis are managed with a single cranial vault reconstruction operation during the first 12 months of life.¹

Supplemental digital content is available for this article. Clickable URL citations appear in the text. Fronto-orbital advancement (FOA) is the gold standard to correct malformations such as trigonocephaly seen in metopic synostosis and anterior plagiocephaly seen in unicoronal synostosis.^{5–7} The surgical procedure involves exposure of the cranial vault and removal, reshaping, and repositioning of the fronto-orbital skull segments to correct the underlying deformity and allow cranial growth in the desired direction.^{6,8,9}

During exposure of the bony orbits, several terminal sensory branches of the trigeminal nerve are invariably manipulated. These include the supraorbital (SO), supratrochlear (ST), zygomaticotemporal (ZT), and zygomaticofacial (ZF) nerves (Fig. 1). The SO nerve, a branch of the frontal nerve which supplies sensation to parts of the forehead skin,¹⁰ exits through the SO foramen or notch. Traditionally, identification and isolation of this nerve is recommended. More recently, however, surgeons have advocated transection of this nerve to avoid partial damage due to intraoperative manipulation, which might result in dysesthesias of the forehead.11 The ST nerve, another branch of the frontal nerve, supplies sensation to the skin and soft tissues of the glabella, lower medial portion of the forehead, upper eyelid, and conjunctiva.¹² It runs along the medial roof of the orbit and exits the forehead through the frontal foramen. The ZT and ZF nerves also have the potential to be injured. The ZT and ZF nerves are terminal branches of the zygomatic nerve and supply sensation to the skin over the temporal bone. The zygomatic nerve enters the orbit via the inferior orbital fissure and runs along the floor of the orbit along the infraorbital sulcus, where it bifurcates into the ZT and ZF nerves.¹³ The ZT and ZF nerves then run along the lateral wall of the orbit and pass through the ZT and ZF foramina, respectively.¹⁴

During intraorbital dissection, these nerves are often seen and coagulated.

Despite numerous studies demonstrating successful esthetic outcome after FOA,⁶⁻⁹ the sequelae of surgical manipulation and potential transection of the SO, ST, ZT, and ZF nerves is not well understood. The aim of this study was to evaluate long-term sensory outcomes in the SO, ST, ZT, and ZF nerve distributions following cranial vault reconstruction with and without FOA in children undergoing surgical treatment for isolated nonsyndromic craniosynostosis.

METHODS

A cross-sectional research design approved by our institution's Research Ethics Board (Hospital for Sick Children) was used to evaluate facial sensory threshold of the SO, ST, ZT, and ZF nerve distributions in 3 groups of patients: (1) children who had undergone anterior cranial vault reconstruction with bandeau for metopic and unicoronal craniosynostosis, (2) children who had undergone total cranial vault reconstruction without bandeau for sagittal craniosynostosis, and (3) age-matched controls. Operative management differs in these craniosynostosis groups, with the metopic and unicoronal groups undergoing more extensive stripping of bone to remove the bandeau after frontal orbital osteotomy, which potentially sacrifices one or more of the SO, ST, ZT, or ZF terminal branches of the trigeminal nerve. Cranial vault reconstruction for sagittal craniosynostosis, on the other hand, only risks injury to the SO nerve.

The database at the Hospital for Sick Children was searched to identify children who had undergone cranial

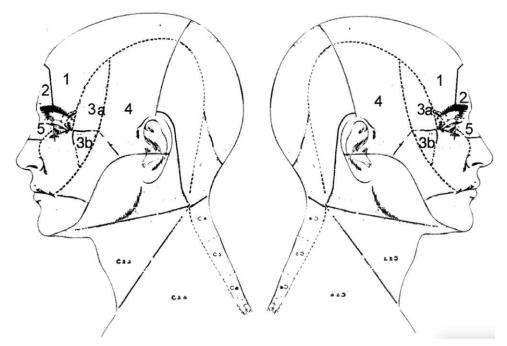


Fig. 1. Sensory nerve distribution of the face. Area 1 corresponds to supraorbital nerve distribution; area 2 corresponds to supratrochlear nerve distribution, and areas 3a and 3b correspond to zygomatic nerve distribution (zygomaticotemporal and zygomaticofacial, respectively).

vault reconstruction between January 2002 and December 2008 for sagittal, metopic, or unilateral coronal nonsyndromic craniosynostosis. Sensory evaluation is reliable in children older than 6 years,¹⁵ thus children under the age of 6 were not included in our study. Children with significant developmental delay, who underwent revision surgery, or who were non-English speaking were also excluded. Healthy age-matched children who had not undergone surgery and had no known global developmental diagnosis were recruited as controls. Written informed consent was obtained from the parents, and assent was obtained from children. The following data were retrospectively collected: sex, date of birth, diagnosis, operative procedure, surgeon (C.R.F., J.H.P.), age at surgical procedure, and intraoperative or perioperative complications.

Children were scheduled for a follow-up visit to evaluate sensory disturbance of the SO, ST, ZT, and ZF nerve distributions using 2 measures: the Ten Test and the Semmes Weinstein Monofilament [Weinstein Enhanced Sensory Test (WEST)] assessment. The Ten Test is a patient-reported test of moving sensation. This test has been found to be valuable in the adult population due to its ease of administration and reported good sensitivity.¹⁶ It is suitable for children over 5 years of age.¹⁷ Clinically, the Ten Test is important as it quantifies the child's perceived discomfort and sensory changes after an injury. Children were asked to rate the feelings of all 4 sensory nerve distributions (Fig. 1) on a scale of 1 to 10 (10 being normal), and the ratio of subjective sensation was recorded.¹⁸ The Semmes Weinstein Monofilament (WEST) assessment has been recognized as the most reliable and valid test of sensory thresholds.¹⁹ It is a noninvasive sensitivity test composed of 5 standardized nylon monofilaments of increasing stiffness that are applied to the skin until they bend, applying a predetermined pressure. The filaments are numbered according to the force they apply; the stiffer and thicker filaments apply more pressure and have a higher number. This allows an objective and repeatable measurement of light-touch sensation.²⁰ Its testing methodology is also well documented.20

Patient self-reports are an important component of nerve injury assessment, as changes in sensation and the extent to which these changes cause discomfort or problems with daily life may not correlate with sensory testing results.²¹ When sensory nerves are damaged, there is a potential for neuroma formation, chronic pain, and paresthesias. It is, therefore, important to address not only lack of or reduced sensitivity (anesthesia or hypesthesia) but also abnormal sensation (paresthesias) and pain (dysesthesias). There are 6 dimensions that must be addressed in a patient self-report of sensory change²¹: (1) Does the patient perceive any change in sensation, (2) Where, (3) When: spontaneously, evoked, both, (4) How does the patient describe change, hypesthesia/hyperesthesia, paresthesia, dysesthesia, (5) What does the patient perceive as the functional or behavioral sequelae in daily life, and (6) How much of a burden do these sequelae cause. These 6 dimensions were examined using a facial sensibility functional questionnaire (see figure, Supplemental Digital Content 1, which displays the facial Sensibility Function

Questionnaire, distributed to participants to complete before their sensory threshold assessment, *http://links.lww. com/PRSGO/B10*).

The questionnaire was piloted to 3 children using cognitive debriefing techniques before study commencement.²²

Statistical analysis was performed using SPSS Statistics (IBM). The ratios of subjective sensation from the Ten Test were analyzed using one-way analysis of variance. Distribution of objective WEST assessment sensory thresholds for all groups was compared using Fisher's exact test. Comparison of the thresholds at the numbest point of the face (highest threshold) was done between the 3 groups using Kruskal-Wallis test. Descriptive statistics were used to analyze the questionnaires. Chi-square analysis was used to compare the distribution of responses on the ordinal scale between groups. A *P*-value of ≤ 0.05 was considered to be significant.

RESULTS

We identified 129 patients who had undergone cranial vault remodeling at the Hospital for Sick Children between January 2002 and December 2008 who were eligible for our study (Fig. 2). We successfully recruited 28 patients, 15 with metopic or unicoronal craniosynostosis and 13 with sagittal craniosynostosis (Table 1). An additional 16 unaffected age-matched controls were recruited (Table 1). The average age at surgery (±SD) was 365 ± 71 days for the metopic and unicoronal group, and 231 ± 157 days for the sagittal group (Table 1). All operations were performed by 1 of 2 senior staff craniofacial surgeons (C.R.F., J.H.P.). Although attempts are made to preserve the sensory nerves to the face, surgery proceeds to expose the bony areas of interest at the potential expense of these nerves. The fate of these nerves is not generally noted in the operative records. No intraoperative or postoperative complications were encountered in these patients. Average age at follow-up assessment was 10.1 ± 2.5 years for the metopic and unicoronal group, 9.1 ± 1.3 years for the sagittal group, and 10.3 ± 2.8 years for the control group (Table 1).

No sensory deficit could be identified using the Ten Test (Fig. 3) or the Weinstein monofilaments (Fig. 4) when comparing patients who had undergone anterior cranial vault remodeling with bandeau for metopic or unicoronal craniosynostosis with patients who had undergone total cranial vault remodeling without bandeau for metopic craniosynostosis, and to age-matched controls. This was true for all 4 nerve distributions: SO, ST, ZT, and zygomaticofacial (ZF). Moreover, there was no difference in the highest threshold between all 3 groups.

A self-reported questionnaire was used to examine facial sensibility function (see **figure, Supplemental Digital Content 1**, which displays the facial Sensibility Function Questionnaire, distributed to participants to complete before their sensory threshold assessment, *http://links.lwww. com/PRSGO/B10*). Children were asked multiple questions about the sensations, movements, and appearance of their forehead and face. Compared to age-matched

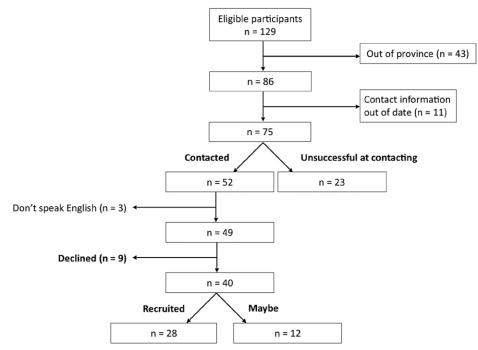


Fig. 2. Patient recruitment. Patients (n = 129) had undergone surgery for isolated metopic, unicoronal, or sagittal craniosynostosis at the Hospital for Sick Children between January 2002 and December 2008. After excluding those who lived out of province (n = 43), those whose contact information was out of date (n = 11), those that we were unsuccessful at contacting (n = 23), those who did not speak English (n = 3), and those who declined (n = 9), 28 patients were successfully recruited to participate.

Table 1. Patient Demographics

Demographics	Metopic/ Unicoronal	Sagittal	Controls
No. patients	15	13	16
Age at surgery (d), mean \pm SD	377 ± 60	231 ± 157	_
Age at assessment (y), mean \pm SD	10.1 ± 2.5	9.1 ± 1.3	10.3 ± 2.8
Sex (M:F)	8:7	12:1	9:7
Intraoperative complications	None	None	_
Postoperative complications	None	None	

Diagnosis, average age at surgery, average age at assessment, sex, intraoperative and postoperative complications are shown for all recruited patients, including controls.

F, female; M, male.

controls, we did not identify any significant difference in subjective sensation, no decrease in protective sensation, and no altered motor behavior (see **table, Supplemental Digital Content 2**, which displays the facial sensibility function results for all patients. Q1, Q2, Q3, and so on correspond to the questions of the Facial Sensibility Function Questionnaire, *http://links.lww.com/PRSGO/B11*).

Several children who had undergone cranial vault reconstruction did comment on cold sensitivity (when it is cold, the skin on my forehead/face bothers me) and perceived difference in appearance (the skin on my forehead or face looks different than in other kids), though neither was clinically significant.

DISCUSSION

In an attempt to preserve sensation to the forehead during FOA, identification and isolation of the SO nerve

from the SO foramen is generally recommended.23-25 However, few studies have evaluated the long-term sensory outcomes following cranial vault reconstruction. The present work examines sensory outcomes of 28 patients in a 9-year follow-up study following surgery for craniosynostosis. No subjective or objective sensory deficit was noted in any of the SO, ST, ZT, or ZF nerve distributions in children who had undergone surgery that risks injury to all 4 of these nerves (metopic, unicoronal craniosynostosis), nor in children who had undergone surgery that risks injury only to the SO nerve (sagittal craniosynostosis), when compared with age-matched controls. Similar findings were reported by Engel et al.²⁸ In a series of 36 children treated with FOA, no sensory deficits were found in the SO nerve distribution with an average follow-up of 6 years, even in 3 children where the SO nerve was noted to be transected. A study by Wiewrodt and Wagner¹¹ described 2 patients with bilateral frontal dysesthesia of 63 and 78 months following partial SO nerve preservation during FOA. This study suggested deliberate anatomical transection of the SO and ST nerves was fully compensated over time and was thus favorable over nerve preservation, which may lead to partial injury during dissection and result in dysesthesias of the forehead. However, follow-up sensory measurements were short term, and it is unknown whether sensory disturbances may have recovered in the long-term had patients been followed for longer. Notably, 1 patient in the control group had significantly lower sensory thresholds on monofilament testing (patient 43; Fig. 4). This participant's Ten Test scores were within the

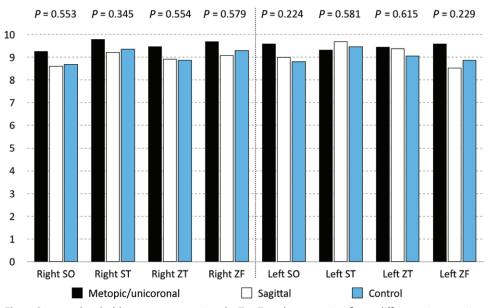


Fig. 3. Sensory threshold measurement using the Ten Test shows no significant difference in sensation in the right/left supraorbital (SO), supratrochlear (ST), zygomaticotemporal (ZT), or zygomaticofacial (ZF) nerve distributions between patients who underwent cranial vault remodeling for metopic/unicoronal craniosynostosis (n = 15), sagittal craniosynostosis (n = 13), and age-matched controls (n = 16). *P*-values from one-way analysis of variance are shown above bars.

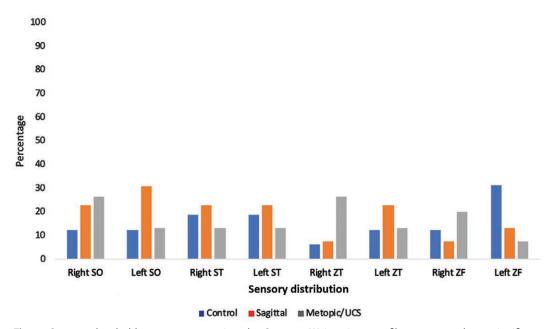


Fig. 4. Sensory threshold measurement using the Semmes Weinstein monofilaments reveals no significant difference in sensation in the right/left supraorbital (SO), supratrochlear (ST), zygomaticotemporal (ZT), or zygomaticofacial (ZF) nerve distributions between patients who underwent cranial vault remodeling for metopic/ unicoronal craniosynostosis (UCS) (n = 15), sagittal craniosynostosis (n = 13), and age-matched controls (n = 16). Values shown indicate the percentage of patients with abnormal values in each sensory distribution. Statistical significance is determined by chi-square analysis (P-values between 0.2 and 0.8).

normal range. When this participant's data were removed from the statistical analysis, the results were not affected.

This study provides the longest sensory follow-up after cranial vault reconstruction in the literature to date. Moreover, this study evaluates not only the SO nerve but also the ST, ZT, and ZF nerves, which have not been previously assessed. We show here that as with previous literature on the SO nerve, no long-term sensory deficits result from injury to these nerves. Not only is there no significant difference in sensibility between children who had undergone surgery with age-matched controls, these values are also in keeping with normal values provided in the literature for the SO, ST, and ZT distributions in the adult population.^{27–29} To the best of our knowledge, normative values of sensory threshold in the zygomatic nerve distribution do not exist in the pediatric literature. Although a relatively small cohort of control patients were used, these data here can serve as a starting point for providing normative values in the pediatric population.

This study examines subjective and objective sensory thresholds. Subjective moving sensation via the Ten Test is suitable for children over 5 years of age,¹⁷ and quantifies the child's perceived discomfort and sensory changes after an injury with good sensitivity.¹⁶ The Semmes Weinstein Monofilament is known to be the most reliable and valid test of sensory thresholds.¹⁹] In addition to sensory testing, changes in sensation (paresthesias and dysesthesias) and the extent to which these changes cause discomfort or problems with daily life is also important to determine. Moreover, nerve damage has the potential to result in a lack of protective sensation and thereby increase the risk of injury. It has been suggested that damage to branches of the trigeminal nerve affects the ability to translate patterns of altered nerve activity into functionally meaningful motor behaviors.²¹ As such, a patient-reported questionnaire of facial sensibility function was included in this study. Some patients did report cold sensitivity, though this was not statistically significant. Neuropathic pain is known to be rare following peripheral nerve injury before the age of adolescence,^{30,31} which is in keeping with our findings. This phenomenon is thought to be a result of neuroimmune activity suppression.³⁰ Studies on neuropathic pain have shown, however, that peripheral nerve injury in early life can lead to neuropathic pain once adolescence has been reached.³² Unfortunately, our study did not include adequate follow-up to determine whether late-onset neuropathic pain may result. Several children in this study stated that they perceived their faces looking different than those of other children. Many studies report successful esthetic outcome after FOA.⁶⁻⁹ These studies are based on expert (surgeon) opinion of surgical results and anthropometric measurements of cranial indices.^{9,33} This highlights the need for patient-reported outcome measurement in assessment of surgical outcomes, which has not been established for pediatric craniofacial deformities.34

In keeping with these findings of normal sensation following both deliberate nerve transection and functional nerve injury are reports of spontaneous return of sensation in noninnervated radial forearm flaps used for head and neck reconstruction³⁵ as well as in face transplantations despite suboptimal sensory nerve repairs.^{36,38,39} Although the exact mechanism of sensory return is not completely understood, it is thought that spontaneous reinnervation of flaps may be a result of nerve ingrowth from the recipient bed to flap margins.³⁷ A similar mechanism may be responsible in this population, where cranial vault reconstruction does injure sensory branches of the trigeminal nerve.

A potential limitation of this study is that despite starting with a significantly large cohort of eligible participants (n = 129), only 22% were successfully recruited (n = 28), raising the possibility that this study was not sufficiently powered. Another limitation of this study is that nerve injury during initial surgery was not documented in the operative note on a routine basis. As such, it is possible any injury incurred could have been a neuropraxia. However, both senior surgeons acknowledge the lack of hesitancy of transecting a sensory nerve if it is felt to impair adequate exposure of the craniofacial skeleton or compromise the integrity of the bandeau. A study on the immediate postoperative assessment of sensation is not possible due to the age of these patients. As such, we feel the findings of this article indicate satisfactory return of function of facial sensation in this patient population, and this in turn can be used in preoperative counseling for parents of affected children undergoing surgery.

CONCLUSIONS

In this study, we have demonstrated that potential functional injury to the SO, ST, ZF, or ZT nerves during FOA does not result in a quantifiable nor clinically significant long-term sensory deficit. Moreover, surgical intervention for craniosynostosis during infancy does not seem to result in significant neuropathic pain. This information may be useful in preoperative counseling of patients and families.

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