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Visuomotor function in school-age children with single-suture craniosynostosis

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Abstract

Objective—Previous studies have shown that infants and young children with single-suture craniosynostosis (SSC) perform more poorly on tests of visuomotor function than children without SSC. However, prior studies are limited by small sample sizes and little is known about the persistence of visuomotor problems into the school-age years. The aim of this study was to compare visuomotor function in children with and without SSC at the beginning of elementary school.

Methods—The study included 179 children with SSC (cases) and 183 children without SSC (controls). Visuomotor function was measured by the NEPSY-II Arrows, the Purdue Pegboard Test, and the Beery-Buktenica Developmental Test of Visual-Motor Integration. Case-control differences were estimated by using linear regression, adjusted for age, sex, socioeconomic status, and maternal IQ.

Results—Cases scored more poorly on all measures of visuomotor function, although the magnitude of case-control differences varied across measures. The greatest differences were observed for the Purdue Pegboard test, with an average adjusted difference of −0.2 to −0.4 standard deviation (SD) points (p-values ranged from 0.008 to 0.05). Case-control differences

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were small in magnitude for other measures of visuomotor function, ranging from −0.01 to −0.1 SD points (p-values ranged from 0.22 to 0.88).

Conclusion—Children with SSC experienced deficits in manual dexterity into the school-age years but were similar to children without SSC on measures of visual processing. These findings advocate for the assessment of fine motor function as part of school readiness evaluations in children with SSC.

INTRODUCTION

Single-suture craniosynstosis (SSC) is a congenital anomaly involving premature fusion of one of the cranial sutures (sagittal, metopic, coronal, or lambdoid). Surgery to release the fused suture and reshape the deformed calvarium is typically performed within the first year of life with the intent of reducing intracranial pressure and allowing for normal brain growth and neurodevelopment. However, several studies show that, even with surgery, infants with SSC are at increased risk for developmental delays/deficits relative to unaffected children [1,2] and school age children with SSC are more likely than controls to exhibit learning problems and reduced scores on measures of intellectual ability.[3,4] Most of the existing studies have used broad measures, such as measures of global IQ.[3] There are fewer studies of the association between SSC and specific neuropsychological domains, and the few existing studies have been limited by small sample sizes.[1,5]

Tasks involving the integration of visual and fine motor skills (i.e., visuomotor tasks) are of theoretical interest in children with SSC, because SSC can affect vision from an early age. This is particularly true for children with unicoronal and metopic synostoses, which result in deformities that affect the orbits and ocular muscles. Deficits in visuomotor function may, in turn, affect children's development in other domains, such as reading, writing, and other aspects of academic achievement.[6]

In this study, we examined visuomotor abilities in a large prospective cohort of school-age children with and without SSC. As we have previously reported on this cohort, children with SSC had lower average scores than demographically similar children on tests of motor development and neurocognitive functions from infancy through age 36 months.[2,7] These deficits carried over into the early school-age years and were evident on measures of academic achievement (e.g., reading, math) and global intelligence.[3] In addition to these global measures, we also administered standardized assessments of visuospatial processing, visual-motor integration, and hand-eye coordination. Based on our previous observations and on the anticipated effects of SSC on children's vision, we hypothesized that children with SSC would score lower on tests of visuomotor function than unaffected children. As secondary aims, we assessed whether magnitude of differences in visuomotor test scores depended on the location of the affected suture or on the presence or absence of vision problems.

PATIENTS AND METHODS

Study Design

This cross-sectional analysis includes surgically treated school-age children with SSC ("cases") and unaffected children ("controls") followed since infancy as part of a multi-center longitudinal study. In the original study, we approached eligible infants with SSC between January 2002 and September 2006 from four sites [sites are blinded for this review process and will be included in the final manuscript]. We also approached eligible cases diagnosed at Children's Hospital of Philadelphia starting in January 2006. These children were assessed by the team in Chicago and included in the site's numbers. Staff at each site also recruited children without SSC who were frequency-matched to cases on the basis of child age, sex, and race/ethnicity, and family socioeconomic status (SES). Assessments were completed before surgery in cases and at a comparable age in controls (mean age = 7.4 months), and at 18 months, 36 months, and 7 years. The assessment at age 7 is the focus of this report. The study was approved by institutional review boards at each participating institution and all parents provided informed consent.

Cases—Infants with SSC were referred to the study at the time of diagnosis by a treating surgeon or pediatrician. Infants were eligible if they: (1) had SSC (isolated sagittal, metopic, unilateral, coronal, or unilateral lambdoid synostosis), confirmed by computed tomography scans when clinically indicated; (2) had not yet had cranial vault surgery; and (3) were ≤ 30 months of age at recruitment. Exclusion criteria included: (1) prematurity (<34 weeks gestation); (2) major medical conditions (e.g., cardiac defects, seizure disorders or significant health conditions requiring surgical correction); (3) presence of ≥ 3 extracranial minor malformations. We enrolled 270 cases (84% of those eligible), nine of whom were later found to be ineligible (for details regarding ascertainment; see Starr et al., 2012[2]). Three cases without surgery to correct the affected suture were also excluded. Among the 259 infant cases seen at baseline, 179 children (67%) participated in the school-age assessment.

For a subsample of 176 cases whose parents gave consent, we collected biospecimens and analyzed genetic data by array CGH and candidate and gene re-sequencing (for details see Cunningham et al. 2011[8]). Children with SSC who had a genetic variant (including a known or probable causal mutation for craniosynostosis) were eligible if they had no phenotypic features of a known syndrome and otherwise met all inclusion criteria.

Controls—Infants without SSC were recruited through pediatric practices, birthing centers, and announcements in publications of interest to parents of newborns. Infants were eligible as controls if they had no known craniofacial anomaly and met none of the exclusionary criteria for cases. Controls were frequency-matched to cases on factors potentially related to both neurodevelopment and craniosynostosis risk, including: (1) age at enrollment (within ± 3 weeks); (2) sex; (3) family SES within the same Hollingshead category[9]; and (4) race/ethnicity.

We enrolled 76% of all interested, eligible controls who were matched to enrolled cases (see Starr et al., 2012[2] for details). Among the 259 controls seen at baseline, 183 (71%) had a 7 year study visit.

Measures

NEPSY-II: A Developmental Neuropsychological Assessment (NEPSY-II). [10]—The Arrows subtest is a measure of visuospatial processing that gauges the ability of the child to judge line orientation.

Beery-Buktenica Test of Visual-Motor Integration (VMI)—The VMI[11] is a constructional test of design copying that quantifies developmental levels of visual-motor coordination. Supplementary tasks include a motor-free test of visual perception and a motor coordination task, which assess the relative contributions of visual perception and motor control to the overall VMI copying score.

Purdue Pegboard—The Purdue Pegboard test[12] uses a timed peg-placing task to measure the speed and accuracy of hand-eye coordination; i.e., dexterity. There are four subtests: three assessing performance using the preferred (dominant) hand, the non-preferred hand, or both hands, and a fourth subtest involving the assembly of pins, collars, and washers.

Caregiver-Family Information and Medical History Interview—Project staff completed a semi-structured interview with participants' primary caregivers to collect family information (socioeconomic status, race/ethnicity, family composition) and history of vision problems (e.g., strabismus or amblyopia). We also asked parents about treatments for vision problems (e.g., glasses, surgery, vision therapy), as well as about interventions for visuomotor impairments (e.g., occupational or physical therapy). This information was updated at each study visit.

Wonderlic Personnel Test (WPT)—The WPT is a timed, norm-referenced screening measure to assess general intelligence in adolescents and adults. The WPT has good reliability and correlates well with clinician administered measures (e.g., the Wechsler adult scale of intelligence.[13,14] The WPT was administered to mothers of participating children, allowing us to control for maternal IQ in analyses of differences between cases and control group participants.

Hand Preference Task (HPT)—The hand preference task[15] was used to assess handedness. Children were asked to perform 10 tasks, such as cutting with scissors, throwing a ball, and drinking from a cup. The test is repeated three times and dominance in the use of each hand is calculated across items. Children who used the same hand to perform 100% of the tasks were assigned as having left or right hand dominance; any other combination was defined as ambidextrous, and children were defined as having ambiguous dominance if they demonstrated within-item inconsistency on 3 or more tasks.

Procedures

Parents from the original cohort study were contacted by telephone six months prior to their child's 7th birthday and screened to determine their willingness to participate in a follow-up assessment. All testing sessions were administered by a trained psychometrist and video recorded and scored by a second psychometrist. Scoring errors were recorded and disagreements between psychometrists resolved by one of the psychologist investigators (Kapp-Simon, Collett or Speltz). Resolved scores were used for all analyses. Age-based standardized scores were used for all tests based on test norms. Parents who so desired were mailed a summary of their child's test results, which they were encouraged to share with their child's pediatrician or teacher.

Data Analysis

The distributions of demographic characteristics and visuomotor scores at elementary school age were calculated for cases and controls, as well as the number and percentage of children with a new or continuing vision problem. To assess for attrition bias, we also compared baseline demographic and neurodevelopmental characteristics of children who were lost to follow-up to those who participated in the age 7 study visit.

Linear regression with robust standard errors was used to estimate differences between cases and controls with corresponding 95% confidence intervals (CI). All analyses were adjusted for the child's age at assessment (in months, continuous), child sex, family SES (Hollingshead composite score, continuous), and maternal IQ (continuous, measured at baseline by the WPT).

In secondary analyses we used censored normal regression[16] to examine whether the receipt of interventions expected to improve children's visuomotor function (e.g., vision therapy, glasses, occupational therapy, physical therapy) may have influenced observed case versus control differences. This approach assumes that the scores of children who received intervention services would be at least as low as those observed in the absence of intervention, i.e. that they are "left-censored."

Using stratified linear regression, we examined whether case-control differences were modified by the presence of a parent-reported vision problem. Estimates were adjusted for age, sex, SES, and maternal IQ, and Wald tests were used to test for effect modification.

We also evaluated whether visuomotor scores differed by the site of the affected suture (sagittal, metopic, unicoronal, and lambdoid) using linear regression with robust standard errors and examined overall group differences using Wald tests. Controls were considered the referent category.

To examine the stability of our results, several sensitivity analyses were performed. Direct adjustment for additional confounders was not possible due to small sample sizes. To explore the potential impact of additional confounding, case-control differences were re-estimated by using propensity-score matching.[17] Propensity scores were estimated by using a logit model that predicted case status based on the four covariates included in the primary analyses plus race/ethnicity and study site. Analyses were also repeated after

excluding 19 children with SSC with known or probable causal mutations for craniosynostosis detected through array CGH and candidate gene re-sequencing.[8] Analyses were also repeated after excluding 14 cases and 5 controls who were born late-preterm, defined as between 34 and 36 weeks gestation. To explore the impact of selection bias from study attrition, we repeated the primary analyses using inverse probability weighting (IPW).[18] This method places greater weight on observations from subjects seen at age 7 who are similar in terms of baseline characteristics to those of children lost to follow-up, in essence resurrecting the full cohort through probability sampling. Weights were estimated based on factors observed at baseline in all subjects, including date of birth, sex, race/ethnicity, prematurity (< 38 weeks gestation, coded yes/no), case status, suture diagnosis, parents' marital status, maternal IQ, study site, and scores from the Bayley Scales of Infant Development-2 (Psychomotor Development Index [PDI]) and the Preschool Language Scale-3 (auditory comprehension [PLS-AC]) completed at baseline (see Starr et al., 2012[2]). Finally, prior reports indicate that girls outperform boys on visuomotor tasks.[19] We examined whether visuomotor scores differed by sex using linear regression and adjusting for all factors in the primary analysis plus case status. Girls were the referent category.

All analyses were performed using STATA version 12.

RESULTS

One hundred seventy-nine cases and 183 controls were seen at the school-age assessment. Mean age at the time of the assessment was 7.5 years for cases (range 6.9–9.5 years) and 7.4 years for controls (range 7.0–11.1 years). Over 90% of participants were in first or second grade. Both cases and controls were predominantly male, identified as white and non-Hispanic ethnicity, were of middle to upper SES (Hollingshead categories I-II), and were right-handed or ambidextrous (Table 1). Maternal IQ as measured by the WPT was lower in cases than controls. A similar proportion of cases (31%) and controls (29%) from the original cohort were lost to follow-up. Compared with children seen at age 7, children lost to attrition had lower average Bayley PDI and maternal IQ scores at study baseline and were of lower SES (44% Hollingshead categories III-V vs. 24% in participating subjects).

Case-control differences

Adjusted mean visuomotor scores were lower in children with SSC than controls for all tests and subtests; however, there was a range of case deficits across measures (Tables 2 and 3). The largest case-deficits were for the Purdue Pegboard, where cases scored on average −3.5 to −5.3 points lower than controls, equivalent to 0.2 to 0.4 SD (p-values ranged from 0.008 to 0.05). For the NEPSY-II Arrows and VMI total scores, average case deficits were modest, 0.1 SD or less (p-values ranged from 0.22 to 0.88). Estimates using propensity-score matching to account for additional confounding were similar in magnitude to most estimates from the primary analysis but were less precise (Supplemental Table 1). Estimates after excluding 19 cases with known or probable causal mutations for craniosynostosis and 19 subjects born late-preterm, as well as estimates using IPW to assess bias from attrition, yielded similar case-control differences (see Table, Supplemental Digital Content 1).

Adjusted visuomotor scores were higher in girls compared to boys for all measures of the Purdue Pegboard and VMI but lower for the NEPSY-II Arrows (see Table, Supplemental Digital Content 2).

Secondary analyses

Thirty-four percent of cases (61/179) and 19% of controls (34/182) received one or more interventions prior to the school age follow-up. Case deficits increased across all tests after accounting for the effects of intervention using censored normal regression, with adjusted differences ranging from 0.1 to 0.6 SD (p-values ranged from 0.41 to 0.003) (Table 4).

Twenty-nine percent of cases (53/179) and 17% of controls (31/183) had a parent-reported history of new or continuing vision problems. Among children without vision problems, visuomotor scores were consistently lower among cases than controls (see Table, Supplemental Digital Content 3). The estimated differences were similar in magnitude to case-control differences, and were greatest for the assembly component of the Purdue Pegboard. Differences from controls were slightly attenuated among children with vision problems (compared with children without vision problems). Confidence intervals for the case-control differences in the two groups overlapped greatly, and the p-values were high.

Differences by suture type

Compared with control group children, children with sagittal synostosis scored slightly higher on tests of visuomotor integration and visuospatial processing (e.g. NEPSY-II Arrows), but lower on all measures of the Purdue Pegboard (see Table, Supplemental Digital Content 4). Children with metopic, unicoronal, and lambdoid synostosis performed more poorly than controls on nearly all assessments. Estimates for differences within each of the affected suture types were imprecise, and most of the p-values were high. There was little evidence for differences in hand preference by suture location. Preference for the right hand ranged from 33.3% (metopic) to 58.3% (lambdoid) (see Table, Supplemental Digital Content 5).

DISCUSSION

To our knowledge this is the largest case-control study to date comparing the visuomotor skills of school-age children with and without SSC. Children with SSC performed slightly worse than unaffected controls on all measures of visuomotor ability, though the magnitude of most differences was small. The largest group differences were found on a measure of manual dexterity, where cases scored an average of approximately 0.2 to 0.4 SD lower than unaffected controls. This is also one of the first studies to compare children with different affected sutures to unaffected controls. Children with unicoronal and metopic synostosis, who have the highest probability of orbital deformity, scored lower than controls on all tests of visuomotor performance.

To our knowledge, only two previous studies have been conducted to compare visuomotor function in children with SSC to that of unaffected controls.[1,5] Chieffo et al. (2010) evaluated neurocognitive skills in 95 children with and without SSC aged 8 to 15 years, including visuospatial, fine-motor, and visual motor skills. Children with sagittal synostosis

demonstrated elevated rates of visuospatial and constructional ability defects as measured by the Rey Complex Figure Test, but no defects in visual motor integration. Children with unicoronal synostosis had elevated rates of defects in visual motor integration and visual perception, but no visuospatial defects. Virtanen et al. (1999) compared the fine-motor and visual-motor performance of 18 children, all with sagittal synostosis, with 18 age- and sex-matched control children between the ages of 8 and 16 years. No differences were observed in Beery VMI and Purdue Pegboard test scores between children with and without SSC. Two additional studies[20,21] compared VMI scores for children with sagittal SSC to standardized test norms. In both studies, children with sagittal SSC had lower average scores than test norms.

We observed few differences in hand preference between cases and controls, and only modest differences by suture location. This is inconsistent with findings by Rogers et al. (2015) [22] who observed large group differences in hand preference among children with unilateral coronal synostosis by surgical technique: In their study, about 30% of children treated with fronto-orbital advancement were left-handed, in contrast to 5% of children treated with endoscopic suturectomy and 11.5% of healthy unaffected controls. The authors speculated that endoscopic procedures, which are generally shorter, less invasive, and performed at younger ages than open procedures such as fronto-orbital advancement, may influence cerebral functional lateralization. All children with SSC in our study received open procedures so we were not able to compare handedness in open vs. endoscopic procedures. Nonetheless, the rate of left-handedness in our population, including those with unicoronal SSC, was markedly lower than that observed in the population of children who received open procedures in Rogers et al.

This variability in visuomotor findings across studies—including our own—is probably due to several factors, including the diagnostic makeup of the SSC samples involved in this research. Specifically, most studies of visuomotor performance have included only children with sagittal synostosis, and these patients appear to be the least impaired in several neuropsychological domains.[1–3] Variable study outcomes may be also related to individual differences in response to developmental interventions of varying quality and intensity, and variations across craniofacial centers in surgeries used to repair the fused suture in infancy (e.g., age, duration and type of surgery including endoscopic vs. “open” methods such as total cranial vault remodeling and frontal orbital advancement).[21] Variable and inconsistent neuropsychological findings related to SSC are not confined to visuomotor outcomes. In our own work, we have observed modest case deficits in IQ and math [3] as well as language and memory[4], but limited evidence of deficits in reading and spelling[3] and executive functions.[4] At present no discernible pattern of neuropsychological findings has emerged in relation to functional domain, including visuomotor abilities.

The mechanisms by which craniosynostosis leads to impaired neurodevelopment are also unclear, but elevated intracranial pressure and abnormal brain growth have been hypothesized to be casual factors.[23] Primary malformations of the central nervous system that cause both SSC and impaired neurodevelopment have also been proposed.[23] Visuomotor function may be uniquely altered because of the effects of intracranial pressure

and cranial deformities on the eye. Swelling of the optic discs can occur with increased intracranial pressure, leading to pressure on the optic nerves and optic neuropathy.[24] Due to the location of the fused suture, children with unicoronal synostosis are particularly susceptible to orbital deformity, extraocular muscle dysfunction, and vision impairment.[25] Although surgery can correct orbital asymmetry, the trochlea may remain asymmetric and ocular muscle dysfunction and subsequent vision abnormalities may persist.[26] Our findings, however, suggest that these potential vulnerabilities do not necessarily produce measurable impairments in visual perception at school age. We observed group differences primarily in fine motor ability and particularly in bimanual dexterity, with fewer meaningful differences in visual-motor integration or visual perception without a motor component (i.e. line orientation).

Fine motor ability has been shown to be one of the stronger predictors of academic skills in kindergarten and the early elementary school years, particularly math, but also reading.[27] By kindergarten, fine motor skills are a better predictor of academic performance than gross motor skills.[28] This is not surprising given that nearly half of the typical kindergarten day is devoted to fine motor activities (e.g., paper and pencil work, cutting and pasting letters, using manipulatives to learn number concepts).[29] Bimanual dexterity is one important component of fine motor ability and is believed to involve interhemispheric transfer, primarily through the corpus callosum.[30] At least one other study has observed an association between bimanual dexterity and the size of the corpus callosum.[31] The link between the corpus callosum and motor performance has not been evaluated in children with SSC, nor has there been detailed study of the morphology of the corpus callosum in children with SSC relative to unaffected children. Our findings suggest that this may be a potentially important area for future research.

There were several limitations to this study. Approximately one third of the original study cohort was lost to attrition, with children of lower SES more likely to drop out. Prior studies have observed that children at higher SES score higher on tests of visuomotor integration than children of lower SES.[32] However cases and controls experienced similar attrition frequencies and sensitivity analyses using IPW did not materially change the results. Second, as we did not have access to ophthalmologic records, we relied on parent reports to confirm the presence of a diagnosed vision problem. Bias due to selective under- or over-reporting of vision problems between cases and controls may have therefore influenced our results. In addition we did not have information about the severity of vision impairment or whether the vision problem had been adequately corrected. Children with vision problems may comprise a heterogeneous population of individuals with varying degrees of vision impairment and this may have contributed to the inconsistency in case-control differences among the children with vision problems.

Conclusion

School-age children with SSC performed slightly worse than matched controls on all measures of visuomotor functioning examined in this study. However, group differences were modest and mostly evident on tasks that emphasized manual dexterity, particularly bimanual dexterity, with greater similarity between the two groups on measures that relied

more heavily on visual processing, with or without a motor component. Replication of these findings in other samples of children with SSC is required, given the great variability in findings across this and previous studies of visuomotor performance in children with SSC. Nevertheless, our findings support current recommendations for vision screening in children with SSC as well as the early assessment of fine motor skills in school readiness evaluations of young children with this condition.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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Table 1

Demographic characteristics of children with and without single-suture craniosynostosis evaluated at age 7 years

	Controls N=183		Cases N=179	
Characteristic	**N**	**%**	**N**	**%**
Age (in years)				
<7.5	145	79.2	122	68.2
≥ 7.5	38	20.8	57	31.8
Grade[a]				
<1	3	1.6	3	1.7
1–2	171	93.4	164	91.6
≥ 3	9	4.9	9	5.0
Sex				
Female	68	37.2	66	36.9
Male	115	62.8	113	63.1
Race/ethnicity[b]				
Non-white	47	25.7	37	20.7
White	136	74.3	142	79.3
Socioeconomic status				
I (highest)	53	29.0	42	23.5
II	103	56.3	82	45.8
III	14	7.7	33	18.4
IV	10	5.5	20	11.2
V (lowest)	3	1.6	2	1.1
Maternal IQ				
Very Superior	6	3.3	5	2.8
Superior	40	21.9	23	12.8
High Average	53	29.0	52	29.1
Average	70	38.3	76	42.5
Low Average	12	6.6	17	9.5
Borderline	1	0.5	2	1.1
Extremely Low	0	0.0	2	1.1
Site				
Seattle	72	39.3	74	41.3
Chicago	76	41.5	63	35.2
St. Louis	9	4.9	18	10.1
Atlanta	26	14.2	24	13.4
Handedness of Child				
Right	82	44.8	76	42.5
Left	6	3.3	9	5.0
Ambidextrous	64	35.0	56	31.3

	Controls N=183		Cases N=179	
Characteristic	**N**	**%**	**N**	**%**
Ambiguous	31	16.9	35	19.6

[a] Includes Hispanic/Latino ethnicity, Asian/Pacific Islander, Black/African American, and mixed races or ethnicities

[b] Grade level missing for 5 controls and 4 cases

Table 2

Distribution of perceptual-motor skill scores for children with and without single-suture craniosynostosis at age 7 years

	Controls			All Cases			Sagittal			Metopic			Unicoronal			Lambdoid		
Neurodevelopmental test	**N**	**Mean**	**SD**	**N**	**Mean**	**SD**	**N**	**Mean**	**SD**	**N**	**Mean**	**SD**	**N**	**Mean**	**SD**	**N**	**Mean**	**SD**
NEPSY-II																		
Arrows Total Score	174	10.9	2.8	171	10.5	2.7	74	11.4	2.3	45	10.0	2.7	42	9.7	3.0	10	10.0	2.9
Purdue pegboard																		
Preferred hand	173	87.1	16.3	166	82.2	15.8	73	82.8	14.1	41	82.1	20.1	42	82.1	15.1	10	78.1	12.9
Non-preferred hand	173	89.0	16.5	167	84.2	16.6	73	84.6	14.0	42	80.1	21.0	42	89.1	14.8	10	78.6	16.5
Both hands	173	90.5	16.7	166	84.7	16.9	73	84.7	14.5	41	83.1	20.3	42	88.1	16.8	10	76.3	17.1
Assembly	170	103.2	17.2	164	97.2	18.3	72	96.2	16.2	40	98.8	18.3	42	96.6	22.3	10	99.5	16.2
VMI																		
Beery VMI	175	96.4	11.0	170	94.2	12.2	72	96.0	12.1	43	91.8	10.3	44	93.5	12.4	11	95.4	18.0
Visual perception	170	106.8	17.3	164	102.3	17.0	70	104.2	17.0	42	101.0	17.1	41	102.1	15.8	11	96.5	21.2
Motor coordination	170	92.0	10.6	164	91.1	11.9	70	93.1	10.1	42	89.9	13.6	41	89.1	12.1	11	90.2	14.3

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Table 3

Comparison of mean perceptual-motor skill scores for children with and without single-suture craniosynostosis at age 7 years

	Cases vs. Controls							
	Unadjusted				Adjusted[a]			
Neurodevelopmental test	Mean Difference	95% CI		p-value	Mean Difference	95% CI		p-value
NEPSY-II								
Arrows Total Score	−0.4	−0.9	0.2	0.24	−0.1	−0.6	0.5	0.78
Purdue pegboard								
Preferred hand	−4.9	−8.3	−1.5	0.01	−3.5	−6.9	−0.1	0.05
Non-preferred hand	−4.8	−8.3	−1.3	0.01	−3.6	−7.0	−0.3	0.03
Both hands	−5.8	−9.4	−2.2	0.002	−4.4	−7.8	−1.1	0.01
Assembly	−6.0	−9.8	−2.2	0.002	−5.3	−9.3	−1.4	0.008
VMI								
Beery VMI	−2.2	−4.7	0.2	0.08	−1.2	−3.5	1.2	0.34
Visual perception	−4.4	−8.1	−0.7	0.02	−2.3	−6.0	1.4	0.22
Motor coordination	−0.9	−3.3	1.5	0.46	−0.2	−2.5	2.2	0.88

[a] Adjusted for age (continuous), sex, SES (continuous), maternal IQ (continuous)

Table 4

Comparison of mean perceptual-motor skill scores for children with and without single-suture craniosynostosis

	Adjusted, with censored normal regression[a,b]			
Neurodevelopmental test	**Mean Difference**	**95% CI**		**p-value**
NEPSY-II				
Arrows Total Score	−0.4	−1.2	0.4	0.33
Purdue pegboard				
Preferred hand	−5.1	−9.8	−0.5	0.03
Non-preferred hand	−5.8	−10.2	−1.3	0.01
Both hands	−6.2	−10.8	−1.6	0.01
Assembly	−8.4	−13.8	−2.9	0.003
VMI				
Beery VMI	−2.5	−5.8	0.9	0.15
Visual perception	−4.3	−9.2	0.7	0.09
Motor coordination	−1.3	−4.4	1.8	0.41

[a] Adjusted for age (continuous), sex, SES (continuous), maternal IQ (continuous)

[b] 61/179 cases (34%) and 34/182 controls (19%) received one or more interventions (PT, OT, vision, developmental therapy, or 0–3 services); information on services missing for 2 cases and 1 control