Long-term Behavioral Outcomes are Different Between School-age Children with Metopic and Sagittal Synostosis

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BACKGROUND

- Nonsyndromic craniosynostosis has been associated with neurocognitive and behavioral deficits
- Children with metopic synostosis have been described to have more behavioral difficulties, including ADHD and autism
- Evidence of differences between suture types has been mixed and due to the variability in sample size, age, and assessment metrics, conclusions have been limited

METHODS

- Parents of children ages 6-18 years old with surgically corrected metopic and sagittal synostosis were recruited
- Demographic factors: age at surgery, age at testing, type of surgery, sex, race, parental education attainment, and household income
- Behavioral assessment completed
  - Conners-3: ADHD
  - Social Responsiveness Scale-2: ASD
  - Behavior Rating Inventory of Executive Function-2: Executive Function
  - Child Behavior Checklist: Overall emotional and behavioral problems

METHODS (CONT.)

- Raw scores are converted to standardized T scores
- Whether borderline clinical levels were reached or exceeded were compared between children with metopic and sagittal synostosis
- ANCOVA was conducted to determine the difference in behavioral scores while controlling for sociodemographic risk, age at surgery, surgery type, and IQ

RESULTS

- There were 106 patients, 60 with metopic synostosis and 46 with sagittal synostosis
- Children with sagittal synostosis were younger (8.4 ± 2.1 vs 10.1 ± 3.6 years, p<0.001) at the time of assessment.
- There was no difference in the average age at surgery (SS: 6.8 ± 5.1 vs MS: 9.3 ± 7.8 months)
- More children with metopic synostosis had problems with executive function **(Table 1)**
  - Emotional regulation index: 33.3% vs 17.4%, p=0.03
  - Global executive composite: 31.7% vs 17.4%, p=0.05
- More children with metopic synostosis had features of ASD:
  - Restricted interests and repetitive behaviors: 30.0% vs 10.9%, p=0.01

RESULTS (CONT.)

Table 1. Behavioral assessment scores and proportion of children who reached or exceeded borderline clinical levels

Behavioral Assessment Subscale	Metopic n=60	Sagittal n=46	p-value
Conners-3 Parent Short			
Inattention	29 (48.3)	16 (34.9)	0.14
Hyperactive	27 (45.0)	19 (41.3)	0.65
Learning Problems	12 (20.0)	7 (15.2)	0.50
Executive Function	20 (33.3)	12 (26.1)	0.39
Defiance	9 (15.0)	8 (17.4)	0.77
Peer Relations	26 (43.3)	13 (28.3)	0.09
Social Responsiveness Scale-2			
Social Awareness	14 (23.3)	10 (21.7)	0.70
Social Cognition	13 (21.7)	8 (17.4)	0.47
Social Communication	15 (25.0)	7 (15.2)	0.16
Social Motivation	14 (23.3)	7 (15.2)	0.22
Restricted Interests and Repetitive Behavior	**18 (30.0)**	**5 (10.9)**	**0.01**
Total	13 (21.7)	7 (15.2)	0.31
Behavior Rating Inventory of Executive Function-2			
Behavior Regulation Index	14 (23.3)	6 (13.0)	0.12
Emotion Regulation Index	**20 (33.3)**	**8 (17.4)**	**0.03**
Cognitive Regulation Index	18 (30.0)	9 (19.6)	0.14
Global Executive Composite	**19 (31.7)**	**8 (17.4)**	**0.05**
Child's Behavior Check List 6-18			
Anxious	9 (15.0)	9 (19.6)	0.70
Withdrawn	5 (8.3)	5 (10.9)	0.78
Somatic	**2 (3.3)**	**7 (15.2)**	**0.04**
Social	6 (10.0)	6 (13.0)	0.76
Thought	12 (30.0)	9 (30.4)	0.75
Attention	12 (20.0)	7 (15.2)	0.37
Rule Breaking	2 (3.3)	2 (4.3)	0.87
Aggressive	4 (6.7)	4 (8.7)	0.81
Internalizing Problem	20 (33.3)	16 (34.8)	0.83
Externalizing Problem	7 (11.7)	7 (15.2)	0.74
Total Problems	17 (28.3)	9 (19.5)	0.18

Table 2. Differences in scores between children with metopic and sagittal synostosis

Behavioral Assessment Subscale	Metopic n=60	Sagittal n=46	p-value	F	Partial ETA
Conners-3 Parent Short					
Inattention	59.17 ± 1.98	57.93 ± 2.26	0.69	0.16	0.002
Hyperactive	59.39 ± 2.17	62.37 ± 2.48	0.39	0.76	0.008
Learning Problems	51.47 ± 1.39	52.58 ± 1.59	0.61	0.26	0.003
Executive Function	54.34 ± 1.72	56.39 ± 1.97	0.45	0.57	0.006
Defiance	52.16 ± 1.43	52.42 ± 1.63	0.57	0.32	0.003
Peer Relations	58.74 ± 2.30	56.22 ± 2.63	0.49	0.49	0.005
Social Responsiveness Scale-2					
Social Awareness	53.40 ± 1.43	53.17 ± 1.58	0.92	0.01	0.00
Social Cognition	51.41 ± 1.49	51.11 ± 1.65	0.90	0.02	0.00
Social Communication	51.70 ± 1.50	50.09 ± 1.66	0.49	0.49	0.005
Social Motivation	51.99 ± 1.53	50.93 ± 1.69	0.65	0.21	0.002
Restricted Interests and Repetitive Behavior	53.95 ± 1.74	51.44 ± 1.93	0.35	0.88	0.01
Total	52.82 ± 1.60	51.29 ± 1.77	0.54	0.39	0.004
Behavior Rating Inventory of Executive Function-2					
Behavior Regulation Index	50.72 ± 1.58	50.11 ± 1.75	0.80	0.06	0.001
Emotion Regulation Index	53.48 ± 1.62	52.09 ± 1.79	0.58	0.31	0.004
Cognitive Regulation Index	52.78 ± 1.58	51.75 ± 1.75	0.67	0.17	0.002
Global Executive Composite	53.51 ± 1.66	52.31 ± 1.85	0.64	0.22	0.003
Child's Behavior Check List 6-18					
Anxious	56.75 ± 1.23	58.83 ± 1.34	0.27	1.24	0.02
Withdrawn	55.74 ± 0.96	55.08 ± 1.04	0.65	0.21	0.002
Somatic	**54.89 ± 0.91**	**57.59 ± 0.99**	**0.05**	3.84	0.04
Social	55.32 ± 0.97	56.21 ± 1.06	0.55	0.36	0.004
Thought	57.27 ± 1.20	59.34 ± 1.30	0.26	1.29	0.02
Attention	56.43 ± 1.09	56.91 ± 1.19	0.77	0.08	0.001
Rule Breaking	**52.53 ± 0.74**	**55.33 ± 0.80**	**0.02**	6.23	0.07
Aggressive	53.65 ± 0.96	55.75 ± 1.04	0.16	2.06	0.02
Internalizing Problem	53.81 ± 1.54	55.51 ± 1.68	0.47	0.53	0.006
Externalizing Problem	**46.88 ± 1.75**	**52.32 ± 1.90**	**0.04**	4.19	0.05
Total Problems	52.20 ± 1.50	55.40 ± 1.63	0.17	1.97	0.02

RESULTS (CONT.)

- More children with sagittal synostosis had somatic complaints (15.2% vs 8.3%, p=0.04
- Higher scores among children with sagittal synostosis for **(Table 2):**
  - Somatic Complaints: 57.6 ± 1.0 vs 54.9 ± 0.9, p= 0.05
  - Rule Breaking: 55.3 ± 0.8 vs 52.5 ± 0.7, p=0.02
  - Externalizing Problems: 52.3 ± 1.9 vs 46.9 ± 1.8, p=0.04
- Multivariate linear regression showed sagittal synostosis associated with somatic complaints (p=0.05), rule breaking (p=0.01), and externalizing problems (p=0.05)

CONCLUSIONS

- Different behavioral difficulties are seen in nonsyndromic craniosynostosis based on suture subtype
- Children with metopic synostosis have more difficulties with executive function
- Children with sagittal synostosis have more difficulties with regulation emotions, such as rule breaking and externalizing behavior