Developmental outcomes of a cohort of preschool children requiring craniosynostosis surgery in Manitoba

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INTRODUCTION

Children with craniosynostosis have been previously identified to be at **risk of developmental delays**. Historically **in Manitoba**, all preschool children with surgically managed craniosynostosis were referred to the **Child Development Clinic (CDC)** for neurodevelopmental evaluation. In spring of 2021, due to heightened volume pressures, there was a decision to limit the number of referrals for children with craniosynostosis. To date, there has not been a rigorous evaluation of the developmental outcomes of our own patient population.

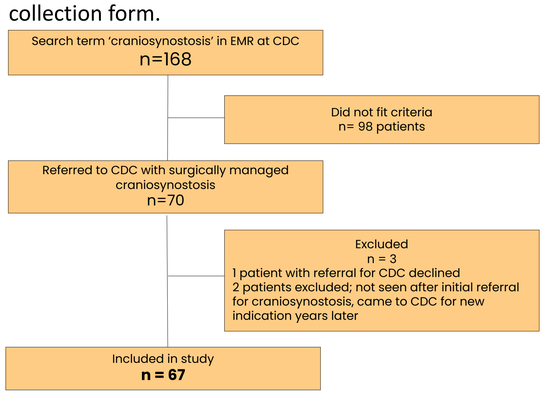
To address this knowledge gap, we aimed to:

1) Describe developmental outcomes of children with surgically managed craniosynostosis in Manitoba by:
a) Reporting the proportion of children with developmental delay
b) Identifying characteristics associated with delay

2) **Inform clinical follow-up strategy**

METHODS

We conducted **a historical cohort study** of all cases of surgically managed craniosynostosis referred to CDC for neurodevelopmental assessment between July 1st 2016 to December 1st 2021. Data was extracted from the electronic medical record using a standardized collection form.



Peri-operative developmental quotient (DQ) scores were collected per domain (motor, language, cognitive). For this study, patients were scored as:

i) **Normal** (with overall DQ 85-100, and no reported delays or a clinical evaluation stating age-appropriate development)

ii) **Mild Delay** (1 area of mild delay with DQ 75-80)

iii) **Developmental Delay** (≥2 areas of delay, and/or overall DQ <70 and/or clinical diagnosis of Global Developmental Delay (GDD)).

Data was collected on demographic information, timeline of surgery, follow up appointments, medical comorbidities and enrolment in Children's Disability Services. Descriptive statistics were used to describe and compare outcomes between groups.

RESULTS

Developmental Outcomes

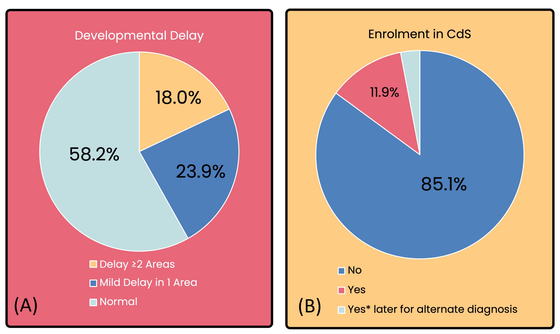


Figure 1. (A) Frequency of developmental delay; 58.2% (n=39) of children were normal, 23.9% (n=16) had a mild delay in one area, and 17.9% (n=12) had delay in ≥2 areas. (B) Frequency of enrolment in Children's Disability Services (CdS) at initial assessment at the CDC was 11.9%.

Characteristics

Table 1: Characteristics of preschool children who have undergone craniosynostosis surgery in Manitoba, by presence/absence of developmental delay.

Characteristics	Total (n=67)	Craniosynostosis with Developmental Delay in ≥2 Areas (n=12)	Craniosynostosis without Development Delay (n=55)	P value
Male, n (%)	45 (67.2)	8 (66.7)	37 (67.3)	1.000
Suture Involvement, n (%)				
Sagittal	29 (43.3)	2 (16.7)	27 (49.1)	**0.011***
Metopic	21 (31.3)	3 (25.0)	18 (32.7)	
Unicoronal	8 (11.9)	3 (25.0)	5 (9.1)	
Bicoronal	2 (3.0)	2 (16.7)	0 (0.0)	
Multisutural	6 (9.0)	2 (16.7)	4 (7.3)	
Unilambdoidal	1 (1.5)	0 (0.0)	1 (1.8)	
Syndrome associated with craniosynostosis, n (%)	8 (11.9)	6 (50.0)	2 (3.6)	**<0.001***
Prematurity, n (%)	15 (22.7)	3 (25.0)	12 (22.2)	1.000
Parental SES Score, median (IQR)	52.68 (17.53)	Median:45.55 Mean Rank: 17.05	Median:52.85 Mean Rank: 28.75	U=115.50 **p=0.028***

Table 2: Referral patterns and service utilization of preschool children who have undergone craniosynostosis surgery in Manitoba, by presence/absence of developmental delay

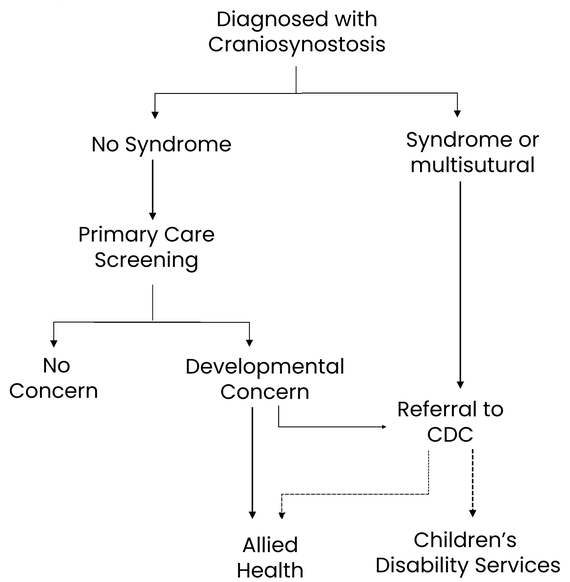
Characteristics	Total (n=67)	Craniosynostosis with Developmental Delay in ≥2 Areas (n=12)	Craniosynostosis without Development Delay (n=55)	P value
Age at craniosynostosis surgery (months), median (IQR)	7.25 (5.44)	9.00	6.75	U = 394.0 p=0.243
Age at referral to SSCY (months), median (IQR)	3.50 (3.00)	4.00	3.25	U=321.5 P=0.518
Age at first assessment at SSCY (months), median (IQR)	7.00 (5.50)	7.5	7.0	U=408.0 P=0.201
Surgery status at visit, n (%)				
Only Pre-op	17 (25.4)	2 (16.7)	15 (27.3)	0.915
Only Post-op	33 (49.3)	7 (58.3)	26 (47.3)	
Both	17 (25.4)	3 (25.0)	14 (25.5)	
Use of Allied Health, n (%)				
OT	20 (29.9)	9 (75.0)	11 (20.0)	<0.001*
PT	36 (53.7)	12 (100)	24 (43.6)	<0.001*
SLP	18 (26.9)	7 (58.3)	11 (20.0)	0.012*
Audiology	19 (28.4)	8 (66.7)	11 (20.0)	0.003*
Visual Impairment, n (%)	3 (4.5)	1 (8.3)	2 (3.6)	0.452
Hearing Impairment, n (%)	5 (7.5)	4 (33.3)	1 (1.8)	**0.003***
Number of appointments, n (%)				
1	40 (59.7)	6 (50.0)	34 (61.8)	0.524
≥2	27 (40.3)	6 (50.0)	21 (38.2)	

Suture type and syndromic involvement were significantly related (p<0.001). Binary Logistic Regression was performed for delay in ≥2 areas:
-Presence of Genetic Syndrome:
OR = 218.352 (95% CI 6.843, 6967.766).
- SES Score**:**
OR = 0.857 (95%CI 0.766, 0.959)

FINDINGS OF THIS STUDY

Developmental Delays in ≥2 areas are present in approximately 18% of children undergoing surgery for craniosynostosis; these children likely benefit most from CDC involvement.
Syndrome and suture type are factors that can be targeted to capture high risk patients.
Mild Delay in 1 area (23.9%); important role for identification and intervention.

Proposed follow up plan:



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