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Impaired growth outcomes in children with congenital colorectal diseases

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ABSTRACT

Background: Cloaca, Hirschsprung disease, and anorectal malformations (CHARM) are congenital anomalies of the hindgut. Small series have suggested that children suffering from one of these anomalies may be at risk for growth impairment. We sought to expand on these findings in a comprehensive cohort, hypothesizing that patients with Medicaid insurance or African-American (AA) race would be at higher risk for poor growth.

Methods: Following Institutional Review Board (IRB) approval, single-institution retrospective review of children with CHARM anomalies was performed (2009-2016). Body mass index (BMI) value Z-scores were obtained using the 2006 World Health Organization (age 0-24 mo) and 2000 Centers for Disease Control (CDC) (age >2 y) growth charts and calculators (statistical analysis system). Patient factors and BMI Z-scores were analyzed with descriptive statistics and Fisher's exact test.

Results: One hundred sixty-six patients (Cloaca $n = 16$, Hirschsprung disease [HD] $n = 71$, anorectal malformation [ARM] $n = 79$) were identified. The BMI Z-score distribution for the entire CHARM cohort was lower than controls ($P < 0.0001$). HD and ARM BMI Z-scores were also lower *versus* controls ($P < 0.0007$, $P < 0.0037$). Requiring more or less than the average number of surgeries did not impact BMI Z-score [P = non-significant (NS)]. Patients with Medicaid had lower Z-scores *versus* private or commercial insurance ($P < 0.0001$). AA race BMI Z-score distribution was lower than controls ($P < 0.0002$), but there was no statistical difference in BMI Z-scores when comparing AA *versus* non-AA CHARM patients (P = NS).

Conclusions: Patients born with CHARM anomalies are at risk for impaired growth. Furthermore study is warranted to identify modifiable risk factors contributing to this impairment. Longitudinal follow-up should include interventions to mitigate these risks.

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Introduction

Cloaca, Hirschsprung disease, and anorectal malformations (CHARMs) are a diverse group of complex congenital malformations involving the lower gastrointestinal tract. They constitute the most common hindgut anomalies in the newborn with an estimated cumulative incidence of approximately 40.3 per 100,000 live births.[1-3] There are no known differences in the incidences of these anomalies among different races or socioeconomic status. However, disparities in surgical treatment allocation, resource utilization, and long-term outcomes have been preliminarily identified as a function of primary payer status and patient race.[4] Demographic analyses have suggested that lower socioeconomic status and minority race may be associated with delayed recognition of pediatric surgical conditions, higher incidence of urgent and emergent procedures, increased length of hospital stay, as well as limited access to care and referral to specialized centers.[4,5]

Moreover, growth in the first 2 y of life has been shown to predict neurodevelopmental outcomes.[6,7] Children born with anomalies of the gastrointestinal tract exhibit suboptimal weight gain during the first year of life.[8] In addition, neonates who undergo surgery for both cardiac and noncardiac anomalies have cognitive and motor delay.[9,10] A number of factors may predispose children suffering from these anomalies to marked and persistent growth and developmental compromise. However, to date only a handful of studies have reported long-term growth outcomes in patients with individual CHARM anomalies.[1,2,11] Overall, these show trends toward impaired growth and raise concern for potential poor developmental consequences.

The aims of the present study were to (1) describe the long-term growth outcomes of a cohort of children with CHARM anomalies, and (2) investigate the impact of potentially modifiable risk factors for poor long-term growth outcomes. We hypothesized that children with CHARM anomalies suffer from long-term growth impairment compared to the general population. We further hypothesized that patients with Medicaid insurance, African-American (AA) race, and/or associated congenital anomalies are at greater risk for compromised growth.

Table 2 – Associated congenital anomalies.

System	Cloaca ($n = 16$)	Hirschsprung ($n = 71$)	ARM ($n = 78$)
Pulmonary	1	0	0
Skeletal	10	1	28
Urogenital	14	5	22
Cardiac	7	9	34
Gastrointestinal	9	2	9
Central nervous system	7	1	12
Total	16 (100%)	13 (18.3%)	53 (67.1%)

Methods

Patient identification and data extraction

This study was approved by The University of Tennessee Health Science Center Institutional Review Board. A single-institution retrospective review of pediatric patients with cloaca, Hirschsprung disease (HD), and anorectal malformation (ARM) (collectively CHARM) anomalies between

Table 1 – Demographics of the study population.

Characteristic	Cloaca ($n = 16$)	ARM ($n = 79$)	HD ($n = 71$)	P value
Gender (*n*, %)				<0.0001
Female	14 (87.5)	36 (45.6)	9 (12.7)	
Male	2 (12.5)	43 (54.4)	62 (87.3)	
Race (%)				0.1060
Hispanic/Latino	12.5	7.6	7	
Non-Hispanic white	31.25	45.6	33.8	
Non-Hispanic black/African-American	56.25	40.5	59.2	
Other	0	6.3	0	
Insurance (%)				0.7038
Commercial	0	11.4	12.7	
Government	87.5	83.5	78.9	
Private pay	0	1.3	1.4	
Unknown	12.5	3.8	7.0	
Congenital anomalies (%)	100	67	18	
Median # surgeries [interquartile range (IQR)]	4.5 (3-7.5)	2 (1-4)	3 (2-5)	
Median # inpatient days in the first year of life (IQR)	89.3 (35.4-162.2)	15.6 (10.8-28.8)	34.1 (24.4-40.8)	
Mean follow-up time (y)	7.2	4.6	6.6	