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Comparison of motor outcomes between preschool children with univentricular and biventricular critical heart disease not diagnosed with cerebral palsy or acquired brain injury

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Abstract

This comparison study of two groups within an inception cohort aimed to compare the frequency of motor impairment between preschool children with univentricular and biventricular critical congenital heart disease (CHD) not diagnosed with cerebral palsy/acquired brain injury, describe and compare their motor profiles and explore predictors of motor impairment in each group.

Children with an intellectual quotient <70 or cerebral palsy/acquired brain injury were excluded. Motor skills were assessed with the Movement Assessment Battery for Children-2. Total scores <5th percentile indicated motor impairment. Statistical analysis included $\chi 2$ test and multiple logistic regression analysis.

At a mean age of 55.4 (standard deviation 3.77) months, motor impairment was present in 11.8% of those with biventricular critical CHD, and 32.4% ($p < 0.001$) of those with univentricular critical CHD. The greatest difference between children with biventricular and univentricular CHD was seen in total test scores 8.73(2.9) versus 6.44(2.8) ($p < 0.01$) and in balance skills, 8.84 (2.8) versus 6.97 (2.5) ($p = 0.001$). Manual dexterity mean scores of children with univentricular CHD were significantly below the general population mean (>than one standard deviation). Independent odds ratio for motor impairment in children with biventricular critical CHD was presence of chromosomal abnormality, odds ratio 10.9 (CI 2.13–55.8) ($p = 0.004$); and in children with univentricular critical CHD odds ratio were: postoperative day 1–5 highest lactate (mmol/L), OR: 1.65 (C1.04–2.62) ($p = 0.034$), and dialysis requirement any time before the 4.5-year-old assessment, OR: 7.8 (CI 1.08–56.5) ($p = 0.042$).

Early assessment of motor skills, particularly balance and manual dexterity, allows for intervention and supports that can address challenges during the school years.

Children with critical congenital heart disease (CHD) are known to be at risk for neurodevelopmental delay, including motor delays/impairments.[1,2] More than a decade ago Majnemer et al[3] reported gross motor delays affected almost half of all school aged children who had survived open heart surgery. Holm et al found that children with critical CHD have a risk of severe motor impairment that is up to 11 times higher than those of same age healthy children;[4] and recently a systematic review showed all children with CHD are at high risk of motor impartment from birth to adolescence.[5]

Critical congenital heart disease can be characterised as biventricular or univentricular. In general, children with univentricular heart disease have been described to be at higher risk for neurodevelopmental impairments when compared with children with biventricular defects.[6,7]

While previous studies have compared the motor outcomes of children with univentricular critical CHD with those of healthy controls,[8–10] a comparison of the motor profile of preschool children with biventricular and univentricular critical CHD not diagnosed with cerebral palsy or acquired brain injury is yet to be completed.

This study aims to compare the frequency of motor impairment between preschool children with univentricular and biventricular critical CHD not diagnosed with cerebral palsy or acquired brain, to describe and compare the motor profiles of children with biventricular

and univentricular critical CHD, and to explore possible predictors of motor impairment in each group.

Methods

This comparison study of two groups within an inception cohort is part of the multiprovincial Western Canadian Complex Pediatrics Therapies Follow Up program. This study was conducted in five different developmental/rehabilitation sites across Western Canada: Vancouver, British Columbia; Edmonton, Alberta; Regina and Saskatoon, Saskatchewan and Winnipeg, Manitoba.[11] All children registered in this follow-up program are identified at the time of their first cardiac surgery and followed prospectively. At the time of their initial cardiac surgery their demographic, pre-, intra-, and post-surgical information is collected. All surviving children undergo neurodevelopmental testing at pre-specified intervals. Specific details on this project have been previously reported.[11] This study was approved by the health research ethics board at each site. The parents or legal guardians of all participating children signed informed consent.

Participants

All children with critical CHD registered with the Western Canadian Complex Pediatrics Therapies Follow Up program who underwent complex cardiac surgery at ≤6 weeks of age at Stollery Childrens' Hospital, Edmonton, Canada from 2009 to 2014, with the exception of those from the Calgary site, were eligible for inclusion. Children with an intellectual quotient <70 were excluded from the study, as lower scores in this group could represent difficulties with the ability to understand and follow the assessment's directions and tasks.

Those with a confirmed chronic neuromotor disability (including cerebral palsy/acquired brain injury)[12] were also excluded from this study, as were children who died before the 4.5-year-old assessment or were lost to follow-up.

For the purpose of this study participating children were divided into two groups, those with univentricular critical CHD and those with biventricular critical CHD. In this study univentricular hearts included those defects with an anatomical single ventricle, and those with "functionally univentricular hearts", representing essentially all lesions that require palliative surgery as they are not amenable for biventricular corrective surgical repair.[13,14] (Fig 1).

Childhood clinical assessments

As part of this follow-up program, all registered children underwent multidisciplinary assessment with standardised measures at approximately 4.5 years of age. At the time of this assessment, motor skills, the primary outcome of this study, were assessed by a physiotherapist or an occupational therapist with the Movement Assessment Battery for Children-second edition[15] a standardised measure of motor competence for children aged 3–16 years using eight tasks that are combined under three categories: Manual Dexterity, Aiming and Catching, and Balance. Results for each category are expressed in standard scores that have a mean of 10 and a standard deviation of 3; low standard scores indicate a poor performance. The test also provides a total score with a percentile equivalent; total scores <5th percentile indicate a motor function impairment.

In addition, all children were assessed by a developmental paediatrician who determined the presence of chronic neuromotor disability based on the medical history and findings from the physical examination and neuroimaging. Cognitive abilities were assessed by a psychologist using the Wechsler Preschool and Primary Scales of Intelligence – Third Edition;[16] adaptive abilities were determined through the parents completed Adaptive Behaviour Assessment System-2.[17]

Socio-demographic variables included maternal education in years, and the Blishen Index[18] an indicator of socioeconomic status based on employment, education, and prestige value of an occupation (population mean of 43 and SD of 13).

Acute care variables

Acute care information included birth gestation (weeks), birth weight (grams), gender, multiple birth, chromosomal abnormality, antenatal diagnosis, and first surgery preoperative ventilation days; preoperative and postoperative highest plasma lactate and inotrope score;[19] age (days), weight (kg), cardiopulmonary bypass time (minutes), X-clamp time (minutes), and use of deep hypothermic circulatory arrest at first cardiac surgery; the presence of pre- or postoperative sepsis, seizures, cardiopulmonary resuscitation, extra corporeal membrane oxygenation, dialysis; and the number of ventilated, intensive care unit, and hospital days. Overall events recorded were the number of cardiac surgeries with cardiopulmonary bypass and ventilation days, presence of sepsis, cardiopulmonary resuscitation, dialysis, extracorporeal membrane oxygenation, heart transplant, ventricular assist device support, and extracorporeal cardiopulmonary resuscitation for each child before the 4.5-year-assessment.

Statistical analysis

In this study categorical variables are presented as proportions and continuous variables are presented as means (standard deviation) or medians (inter quartile range). Frequency of motor impairment is given as percentage of assessed survivors. Student t-test and χ^2 test were used to compare groups. Multiple logistic regression analysis was conducted for each group individually and included demographic, operative and perioperative predictors of motor impairment having p value of <0.10 after screening for multicollinearity. Results are expressed as odds ratios with 95% confidence interval; significance considered at <0.05. Data analyses were performed using IBM SPSS Statistic Data Editor v 22 (IBM Corporation, Armonk, New York).

Results

At a mean age of 55.4 (standard deviation 3.77) months, 119 (72% of those eligible for inclusion) children (85 (71.4%) with biventricular critical CHD; 34 (28.6%) with univentricular critical CHD, 66.7% male) underwent testing with the Movement Assessment Battery for Children-second edition.

Table 1 describes the demographic, pre-, intra- and postoperative characteristics of children with biventricular and univentricular CHD. The growth, health and accompanying impairments of children with univentricular and biventricular critical CHD at time of testing are described in Table 2.

Overall, 10/85 (11.8%) of children with biventricular critical CHD, and 11/34 (32.4%) (p = 0.008) of those with univentricular critical CHD had total Movement Assessment Battery for Children-second edition scores <5th percentile, representing motor function impairment. On average, total Movement Assessment Battery for Children-second edition scores of children with univentricular heart disease were 2.3 points lower

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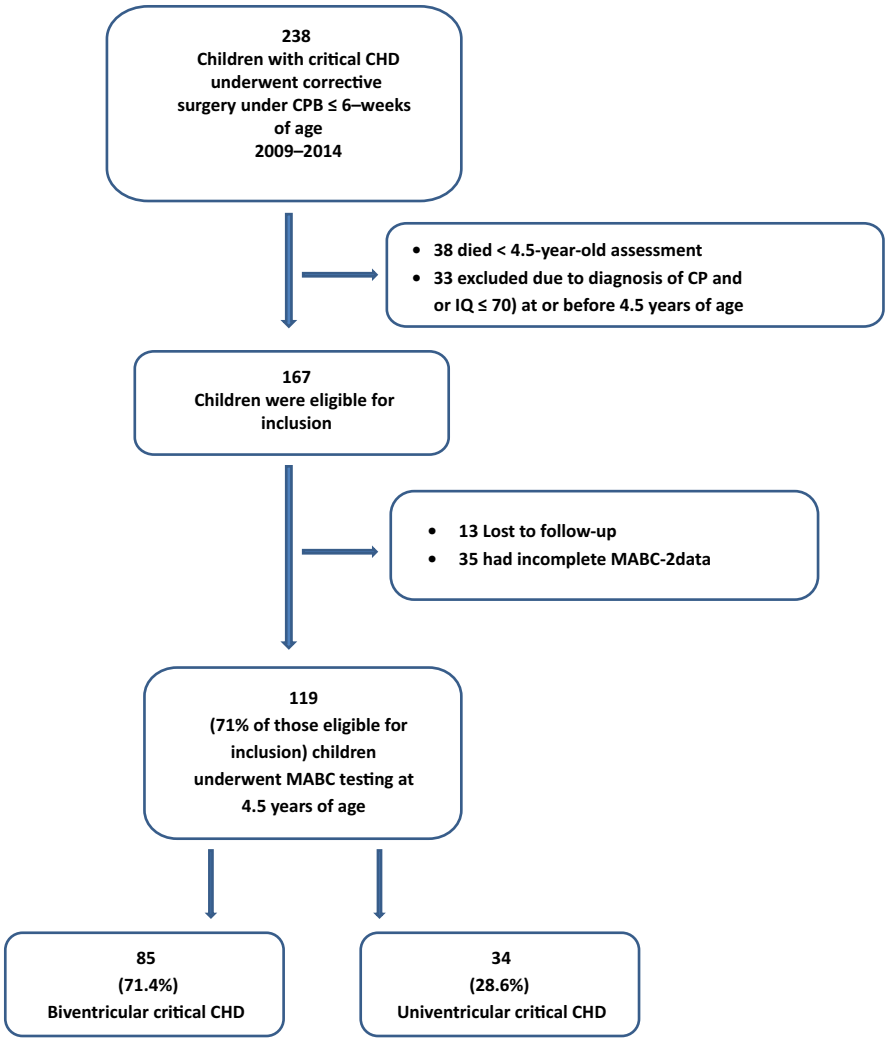

Figure 1. Flowchart of death, lost, excluded and assessed children after complex cardiac surgery at <6 weeks of age from the years 2009–2014.

than those of children with biventricular heart disease (6.44 (SD 2.8) versus 8.73 (2.9), p = <0.001). The comparison of the motor profiles (including Manual dexterity, Balance and Aiming, and catching scale scores) between children with univentricular and biventricular critical CHD is represented in Table 3.

Independent odds ratios for motor impairment in children with biventricular critical CHD was presence of chromosomal abnormality (OR: 10.9, 95% CI 2.13–55.8) (p = 0.004). In children with univentricular critical CHD independent odds ratios were: postoperative day 1–5 highest lactate (mmol/L) at first complex cardiac surgery, (OR 1.65, 95% CI 1.04–2.62) (p = 0.034), and dialysis requirement any time before the 4.5-year-old assessment, (OR: 7.8, 95% CI 1.08–56.5) (p = 0.042).

Discussion

Results of this study indicate that among preschool survivors of critical CHD without cerebral palsy, acquired brain injury, or intellectual impairment, children with univentricular critical CHD are at higher risk of motor impairment when compared to those with biventricular critical CHD, with one third meeting the diagnostic criteria for motor function impairment. Our findings are consistent with studies reporting higher rates of motor delays in children requiring multiple palliative surgeries that is, commonly those with univentricular heart disease, compared to those undergoing a corrective surgery.[20] Moreover, our group and others have previously identified children with univentricular CHD as being at higher risk of neurocognitive delays when compared to those with a biventricular defect.[7,21]

Prenatal abnormalities in brain volume,[22] in preoperative cerebral blood flow,[23] the greater number of required surgeries and postoperative hospitalisations, as well as higher incidence of feeding difficulties[24]often leading to a high need for gastrostomy tube feedings[25] among children with univentricular critical CHD compared to those with biventricular critical CHD may all play a role in explaining the differences in motor function impairment observed between the two groups. Moreover, both prenatally and for up to several years postnatally, the developing brain in children with univentricular critical CHD is subjected to prolonged periods of significantly decreased oxygenation; chronic hypoxemia has been shown to alter neuronal and glial protein expression in the fetal brain.[26]

In addition to physiologic factors, environmental factors such as parental anxiety and overprotective behaviors may also contribute to poorer motor outcomes in children with critical CHD, particularly for children with univentricular critical CHD who require repeated surgeries during infancy. Parental overprotectiveness may lead to reduced exposure to physical activity which, in turn, can affect motor performance.[27]

Results of this study demonstrate that children with univentricular critical CHD experience more challenges with balance tasks

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Table 1. Description of 4.5-Year-Old Children with biventricular and univentricular congenital heart disease n = 119: Mean (SD), Median (Interquartile Range), n (%)

	Total (n = 119) Mean (SD) Median (IQR)	Biventricular CHD N = 85 (71.4%)	Univentricular CHD N = 34 (28.6%)	p-value
Preoperative First CCS				
Family Socioeconomic Status	43.24 (11.4) 42 (35, 50)	42.8 (11.2)	44.32 (11.92)	0.469
Mother total schooling:	14.02 (2.23) 14 (12,16)	14.00 (2.5)	14.06 (9.72)	0.9
Birth Gestation: Weeks	38.35 (2.27) 39 (37,40)	38.18 (2.43)	38.79 (1.75)	0.181
Birth weight: Grams	3179.9 (684.2) 3290 (2791, 3610)	3166.18 (746.4)	3214.2 (504.2)	0.686
Sex Male	79 (66.4%)	54 (63.5%)	25 (73.5%)	0.297
Multiple birth	9 (7.6%)	5 (5.9%)	4 (11.8%)	0.273
Chromosomal abnormality	10 (8.4%)	8 (9.4%)	2 (5.9%)	0.531
Antenatal Diagnosis	71 (59.7%)	42 (49.4%)	29 (85.3%)	0.000
Preoperative 1st surgery ventilation days	2.9 (3.95) 1 (0,5)	2.95 (4.13)	2.76 (3.5)	0.816
Preoperative 1st surgery Inotrope	2.702 (5,24) 0 (0,5)	2.65 (5.3)	2.82 (5.22)	0.874
Preoperative 1st surgery Highest plasma lactate level: mmol/L	2.9 (2.44) 2.2 (1.7, 3.2)	2.97 (2.76)	2.74 (1.35)	0.625
	Total (n = 119) Mean (SD) Median (IQR)	**Biventricular CHD n = 85 (71.4%)**	**Univentricular CHD n = 34 (28.6%)**	**p-value**
Intra-operative First CCS				
Age at Surgery: Days	17.1 (24.4) 9 (7, 16)	20.42 (28.13)	8.82 (4.38)	<0.000
Weight: Kg	3.347 (.737) 3.300 (2.8,3.8)	3.380 (.81)	3.255 (.51)	0.299
CPB: min	113.8 (41.6) 104(81, 143)	111.39 (43.2)	119.82 (37.2)	0.32
X-clamp time: minute	61.47 (24.5) 60 (43,78)	62.99 (25.7)	57.68 (21.13)	0.288
DHCA: Yes	95 (79.8%)	62 (72.9%)	33 (97.1%)	0.002
C. Postoperative: First CCS				
Day 1–5 highest plasma lactate mmol/L	4.96 (2.41) 4.5 (3.1, 6.4)	4.42 (2.06)	6.3 (2.73)	0.000
Day 1–5 highest inotrope score:	9.42 (7.81) 8 (4, 13)	7.794 (6.62)	13.485 (9.09)	0.000
D. Overall 1st CCS				
Sepsis	8 (6.7%)	5 (5.9%)	3 (8.8%)	0.687
Seizures	5 (4.2%)	3 (3.5%)	2 (5.9%)	0.623

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Table 1. (*Continued*)

	Total (n = 119) Mean (SD) Median (IQR)	Biventricular n = 85 (71.4%)	Univentricular n = 34 (28.6%)	p-value
CPR	5 (4.2%)	1 (1.2%)	4 (11.8%)	0.02
ECMO	4 (3.4%)	1 (1.2%)	3 (8.8%)	0.07
Dialysis	21 (17.6%)	10 (11.8%)	11 (32.4%)	0.015
All ventilated days	9.79 (7.03)	8.34 (5.6)	13.41 (8.8)	0.003
	8 (5,12)			
All ICU days	12.66 (9.81)	10.56 (7.1)	17.88 (13.27)	<0.000
	10 (7,15)			
All hospital days	28.55 (24.4)	24.96 (19.89)	37.5 (31.7)	0.011
	21 (15,34)			
Overall prior to 4.5-year-assessment				
Sepsis	9 (7.6%)	5 (5.9%)	4 (11.8%)	0.274
CPR	5 (4.2%)	1 (1.2%)	4 (11.8%)	0.023
Dialysis	21 (17.6%)	10 (11.8%)	11 (32.4%)	0.015
ECMO	5 (4.2%)	1 (1.2%)	4 (11.8%)	0.023
Heart transplant	3 (2.5%)	0	3 (8.8%)	0.022
VAD	2 (1.7%)	1 (1.2%)	1 (2.9%)	0.492
E-CPR	0	0	0	
Number of interventions with CPB before 4.5 years	1.60 (.886)	1.08 (.277)	2.88 (.478)	<0.000
	1 (1,2)			
Total CPB time	162.34 (103.38)	120.26 (51.75)	267.53 (124.3)	<0.001
	142 (91, 201)			
+ days hospital <4.5years	34.97 (30.16)	24.94 (19.88)	60.03 (36.59)	<0.001
	25 (15,46)			
All ventilated days	9.99 (7.16)	8.36 (5.58)	14.06 (8.94)	0.001
	8 (5,13)			
>Than 1 CCS by 4.5	41 (34.5%)	7 (8.2%)	34 (100%)	<0.001

Abbreviations: CCS = complex cardiac surgery; CPB = cardiopulmonary bypass; CPR = cardiopulmonary resuscitation; DHCA = Deep hypothermic circulatory arrest; ECMO = extracorporeal membrane oxygenation; E-CPR = extracorporeal- cardiopulmonary resuscitation; ICU = intensive care unit; IQR = interquartile range; SD = standard deviation; VAD = ventricular assist device.

than those with biventricular critical CHD and overall display scores that are significantly below what is expected in the general population. Balance is a key component of motor proficiency. It provides the necessary base to support the movement of the head, torso and limbs; stabilising the body and keeping it in balance are "prerequisites for adaptive control of movement".[28] Difficulties with balance can not only limit the ability of a child to participate in sport related activities; studies suggest that balance skills are independently associated with spatial performance among 6-year-old children,[29] and with reading and mathematics academic achievement scores in elementary school children, potentially having far reaching impacts[30] Different movement education programmes targeting balance, including exergames and pedal less bikes have been proven to improve balance skills in children.[31,32]

Although the difference in manual dexterity scores between children with univentricular and biventricular critical CHD was not statistically significant, the manual dexterity score of those with univentricular critical CHD was substantially lower than that of the general preschool-age population (>1 SD below the norms). The co-existence of balance and manual dexterity challenges among children with univentricular critical CHD could be explained by the relationship between postural stability and manual control in children.[33]

Our study found that among children with univentricular critical CHD, higher lactate values in the first five postoperative days following the first cardiac surgery predicted motor impairments. Higher perioperative lactate values are known to be associated with a higher postoperative mortality and morbidity,[34,35] and have been identified to predict mental and motor delays as well as chronic neuromotor disability among children with critical CHD.[12,36] In addition, requirement of dialysis any time before the 4.5-year-old assessment was found to be a strong predictor of motor impairment. Children with cyanotic critical CHD have been identified to be at risk for acute renal failure leading to the requirement of dialysis;[37] and importantly an association between postoperative lactate and the need for dialysis was described by Maarslet and

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Table 2. Growth, health, and accompanying impairments at 4.5 years (n = 119): Mean (SD), Median (Interquartile Range), n (%).

	Total (n = 119) Mean (SD) Median (IQR)	Biventricular CHD n = 85	Univentricular CHD n = 34	p-value
Variables at 4.5 years				
Height: z-score	−0.55(1.3)	−0.286 (1.3)	−1.23 (1.08)	<0.001
	−0.50 (−1.3,0.3)			
Weight: z-score	−0.29 (1.18)	−0.89 (1.15)	−0.81 (1.1)	0.002
	−0.2 (−1, 0.55)			
Gastrostomy at any time after the first surgery	17 (14.3%)	8 (9.4%)	9 (26.5%)	0.022
Number of hospitalisations not related to Cardiac Treatment	1.16 (2.28)	0.89 (2.05)	1.82 (2.7)	0.045
	0 (0, 1)			
Number of hospitalisations related to cardiac treatment	1.47 (1.82)	0.86 (1.25)	3 ((2.12)	<0.001
	1 (0,3)			
Number of medical specialists in addition to pediatrician	1.97 (1.28)	1.85 (1.27)	2.29 (1.27)	0.087
	2 (1,2)			
Medication for chronic pulmonary disease: yes	13 (10.9%)	10 (11.8%)	3 (8.8%)	0.75
Medication for chronic cardiac disease: yes	48 (40.3%)	17 (20%)	31 (91%)	<0.001
Vision Impairment	1 (0.8%)	0	1 (2.9%)	.286
Permanent hearing impairment	2 (1.7%)	2 (2.4%)	0 (0)	1.000
Epilepsy	0	0	0	
Autism Spectrum Disorder	0	0	0	
Full-scale IQ	97 (11.57)	97.02 (11.5)	96.97 (9.9)	0.98
	98 (89, 104)			
Visual – motor Integration	96.8 (9.23)	98.01 (8.74)	93.65 (9.9)	0.02
	97 (90, 103)			
ABAS GAC:	93.8 (14.8)	94.65 (14.34)	91.56 (16.04)	0.307
	93 (85, 103)			

Abbreviations: ABAS = Adaptive behavior assessment system; GAC = general adaptive composite; IQ = intellectual quotient; IQR = interquartile range; SD = standard deviation.

Table 3. Comparison of motor profile as determined by the Movement Assessment battery for Children-second edition results in relation to Biventricular or Univentricular critical CHD, (mean) SD.

MABC-2	Biventricular CHD n = 85	Univentricular CHD n = 34	p-value
Manual Dexterity Scale score	8.37 (3.9)	6.85 (4.3)	0.6
Aiming and Catching Scale score	10.07 (2.9)	9.06 (3.8)	0.1
Balance Scale score	8.84 (2.8)	6.97 (2.5)	0.001
Total test scores	8.73 (2.9)	6.44 (2.8)	<0.001
MACB-2 total scores <5th percentile	10 (11.8%)	11 (32.4%)	0.008

collaborators.[35] According to a study by Warady et al,[38] gross motor delays are common among children receiving peritoneal dialysis for end-stage renal disease. While in this study children did not receive dialysis for chronic renal disease, the requirement of dialysis among our cohort may be an indicator of the level of severity of illness. Moreover, increased illness severity could also indicate decrease chances for physical activity in these children.

Among children with biventricular critical CHD, the presence of a chromosomal abnormality with an intellectual quotient >70, was predictive of significantly poorer motor outcomes. This finding is consistent with previous studies that showed children with deletion 22q11.2 had significantly worse mental and psychomotor developmental index scores compared to those without deletion 22q11.2.[39]

The importance of studying motor skills relies on the essential role these play on a child's emotional, psychosocial, and overall development. Motor development is known to have a broad and profound impact on all other developmental domains. Development of a child's mobility grants the child independence and self-assurance, thereby ensuring psycho-emotional stability. At preschool, development of cognitive and social skills such as sharing and cooperation are also highly dependent on a child's ability to be mobile and actively participate in games and activities with peers.[8] Motor proficiency and physical activity act synergistically to mutually reinforce one another. Obesity, although not identified in our study, is a common comorbidity in children with CHD and therefore, concurrently identifying motor impairments and promoting physical activity early are critical.[40]

Our study has several limitations. Data on 25 children who underwent testing with the Movement Assessment Battery for

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Children second edition was incomplete and as such could not be included in this study. Due to the observational nature of this study, we cannot account for the effect of other unmeasured confounders. Finally, the lack of routine brain imaging is a limitation for this study. Although we excluded all children with a confirmed diagnosis of cerebral palsy and/or acquired brain injury who represented those with abnormal findings on neuroimaging resulting in chronic motor difficulties, the lack of routine neuroimaging in all children may have resulted in missed evidence of brain injury in some of our subjects. The strengths of this study include the prospective design and the large number of children followed in this study.

Conclusions

Guidelines have been established regarding evaluation and management of neurodevelopmental outcomes in children with CHD.[41] In particular, children with univentricular critical CHD and those with biventricular critical CHD with a chromosomal anomaly merit early, close and active surveillance of their motor development. Early identification of motor impairments and particularly balance and manual dexterity impairments, through standardised testing allows for optimisation of interventions and supports that can ultimately improve children's motor skills and prevent future physical and psychosocial health problems.

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Conflict of interest. The authors have indicated they have no conflicts of interest to disclose.

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