A Comparison of Developmental Outcomes of Adolescent Neonatal Intensive Care Unit Survivors Born with a Congenital Heart Defect or Born Preterm

Kaitlyn Easson, BScH[1,2], Noémi Dahan-Oliel, PhD[3,4], Charles Rohlicek, MDCM, PhD[5], Sossy Sahakian, MSc[1], Marie Brossard-Racine, PhD[1,2,3,6], Barbara Mazer, PhD[3], Patricia Riley, MD[7], Désirée B. Maltais, PhD[8], Line Nadeau, PhD[8], Sean Hatzigeorgiou, BSc[9], Norbert Schmitz, PhD[10], and Annette Majnemer, PhD[1,3,6]

Objective To compare cognitive, motor, behavioral, and functional outcomes of adolescents born with a congenital heart defect (CHD) and adolescents born preterm.

Study design Adolescents (11-19 years old) born with a CHD requiring open-heart surgery during infancy (n = 80) or born preterm ≤29 weeks of gestational age (n = 128) between 1991 and 1999 underwent a cross-sectional evaluation of cognitive (Leiter International Performance Scale-Revised), motor (Movement Assessment Battery for Children-II), behavioral (Strengths and Difficulties Questionnaire), and functional (Vineland Adaptive Behavior Scale-II) outcomes. Independent samples *t* tests and Pearson χ^2 or Fisher exact tests were used to compare mean scores and proportions of impairment, respectively, between groups.

Results Adolescents born with a CHD and adolescents born preterm had similar cognitive, motor, behavioral, and functional outcomes. Cognitive deficits were detected in 14.3% of adolescents born with a CHD and 11.8% of adolescents born preterm. Motor difficulties were detected in 43.5% of adolescents born with a CHD and 50% of adolescents born preterm. Behavioral problems were found in 23.7% of adolescents in the CHD group and 22.9% in the preterm group. Functional limitations were detected in 12% of adolescents born with a CHD and 7.3% of adolescents born preterm.

Conclusions Adolescents born with a CHD or born preterm have similar profiles of developmental deficits. These findings highlight the importance of providing long-term surveillance to both populations and guide the provision of appropriate educational and rehabilitation services to better ameliorate long-term developmental difficulties. *(J Pediatr 2019;207:34-41).*

Infants born preterm or with a congenital heart defect (CHD) both experience critical illness in early life, and subsequently, are both at risk of experiencing long-term developmental impairments. These groups present with well-documented cognitive impairments, motor difficulties, and behavioral problems throughout childhood.[1-9] With advances in medical care over the past decades, more infants born preterm or with a CHD are now surviving long past infancy and childhood and into adolescence.[10-12]

Follow-up studies tracking the development of adolescents born preterm have convincingly uncovered pronounced deficits in executive functioning[13-18] and other cognitive impairments,[13,14,17-20] accompanied by reductions in academic achievement,[13,21-23] with a limited number of studies documenting the impaired motor skills[24] and behavioral issues, such as poor socialization skills,[25,26] also present in this group. Similarly, existing research on adolescents born with a CHD requiring early open heart surgery have predominantly focused on cognitive deficits,[27-32] with a paucity of studies reporting motor impairments[30,33] and behavioral difficulties.[27-29,32]

This secondary analysis of a cross-sectional study aims to describe and compare the developmental status of adolescents born with a CHD and adolescents born preterm in a regional sample. To do so, we performed a comprehensive evaluation

CHD	Congenital heart defect
Leiter-R	Leiter International Performance Scale-Revised
MABC-II	Movement Assessment Battery for Children-II
MCH	Montreal Children's Hospital
SDQ	Strengths and Difficulties Questionnaire
VABS-II	Vineland Adaptive Behavior Scale-II

From the [1]Research Institute of the McGill University Health Centre; [2]Department of Neurology and Neurosurgery, Faculty of Medicine; [3]School of Physical and Occupational Therapy, McGill University; [4]Shriners Hospitals for Children; [5]Department of Cardiology, Montreal Children's Hospital; [6]Department of Pediatrics, Division of Child Neurology, McGill University; [7]Department of Pediatrics, Montreal Children's Hospital, Montreal; [8]Department of Rehabilitation, Université Laval, Québec City; [9]Department of Physiology; and [10]Department of Psychiatry, McGill University, Montreal, Québec, Canada

Supported by an operating grant from the Canadian Institutes of Health Research (MOP-102720). N.D-O., the doctoral student working on this project, received 2 studentships to support her work from the Montreal Children's Hospital Research Institute—Foundation of Stars (2009-2010) and the Fonds de recherche du Québec—Santé (2010-2013). This work was also supported by infrastructure from the Montreal Children's Hospital—Research Institute and Centre for Interdisciplinary Research in Rehabilitation of Greater Montreal, both of which are funded by the Fonds de recherche du Québec—Santé. The authors declare no conflicts of interest.

https://doi.org10.1016/j.jpeds.2018.11.002

of cognitive, motor, behavioral, and functional outcomes to fully characterize the domains commonly affected by developmental disorders.

Methods

Adolescents (11-19 years old) born between 1991 and 1999 were recruited if they were born very or extremely preterm ≤29 weeks of gestational age, or born with a complex CHD requiring surgical correction (ie, open-heart surgery), as part of the Determinants of Activity Involvement in Leisure for Youth study. The primary aim of the Determinants of Activity Involvement in Leisure for Youth study was to identify determinants of participation in leisure activities among adolescents.[34-37] Participants were included if they and their parents could both speak and read English or French.

Preterm participants were recruited through the Neonatal Follow-up Program of the Montreal Children's Hospital (MCH), which includes all infants born ≤29 weeks of gestational age regardless of developmental status. A letter containing information about the research project was sent to families of potential participants, after which parents were contacted by a nurse from this program to determine their interest in the study. For the CHD group, adolescents born with a CHD and admitted to the MCH neonatal intensive care unit or wards as infants for a corrective open-heart surgery during the first 2 years of life were recruited. These patients were all followed closely and regularly by a cardiologist at the MCH. Eligible participants were contacted by a member of the pediatric cardiology department of the MCH to appraise their interest in the study. For both groups, a member of the research team contacted the interested participants to clarify study procedures and obtain written informed consent from the guardian, and written assent from the youth as appropriate based on cognitive status.

Following enrollment in the study, participants attended a single appointment at the MCH, during which various evaluations were performed by experienced evaluators who were blinded to participants' medical history and the findings of other evaluators. A psychologist administered the Leiter International Performance Scale-Revised (Leiter-R)[38] to evaluate cognition and the Vineland Adaptive Behavior Scale-II (VABS-II)[39] to evaluate functional status. The Leiter-R is a normalized nonverbal test of global intelligence designed for children aged between 2 and 20 years. On the basis of the Leiter-R brief IQ scores, each participant's cognitive status was categorized by impairment level, with brief IQ scores of <70 considered as cognitive deficit. The VABS-II provides normalized measures of subdomains of functional status as related to communication, daily living skills, and socialization and on the basis of these subdomain scores, computes a composite score, the adaptive behavior composite. Each participant's functional status was categorized by impairment level. Scores of <78 (1.5 SD below the mean) were considered to be abnormal.

Motor ability was assessed using the Movement Assessment Battery for Children-II (MABC-II),[40] administered by a licensed occupational therapist or physical therapist. The MABC-II evaluates motor ability as related to manual dexterity, aiming and catching, and balance, and provides a total test score indicating the child's overall motor competence. For each of these 4 scores, participants were categorized as having no movement difficulty (>15th percentile), at risk of movement difficulty (6th-15th percentile), or having significant movement difficulty (≤5th percentile).

Information regarding participants' behavioral problems was collected using the Strengths and Difficulties Questionnaire (SDQ),[41] which was completed by participants' parents. The SDQ evaluates behavioral status by measuring participants' prosocial behaviors, emotional and conduct problems, hyperactivity, and peer problems, and the impact of behavioral issues on their daily lives. Participants' behaviors were classified as normal, borderline, or abnormal for each subscale based on established cut-offs. Parents also completed a questionnaire outlining sociodemographic information related to parental education level and total family income and reported medical morbidities experienced by their children. Recruitment and data collection occurred between May 2010 and December 2014. This study was approved by the MCH Research Ethics Board.

Descriptive statistics were used to examine the sociodemographic characteristics, medical morbidities, and developmental outcomes of the sample. Means and proportions were derived for each of the outcome measures by group. Independent samples t tests were used to compare the means of continuous variables between groups. Pearson χ^2 or Fisher exact tests were used to compare categorical variables between groups. Missing data were excluded, and all statistical analyses were performed using a level of significance of $P < .05$. Statistical analyses were performed using IBM SPSS Statistics Software v 23 (IBM, Armonk, New York).

Results

Eighty adolescents born with a complex CHD (39 male, 41 female) and 128 adolescents born preterm (61 male, 67 female) participated in this study. Mean age at assessment was 15.7 years (SD = 1.7 years, range = 11.4-19.9 years) in the CHD group and 16.0 years (SD = 2.4 years, range = 12.0-19.9 years) in the preterm group. Of the participants with a CHD enrolled, 63 had a cyanotic CHD and 17 had an acyanotic CHD, and 13 had a single-ventricle physiology (eg, hypoplastic left heart syndrome, tricuspid atresia) and 67 had a biventricular physiology (eg, tetralogy of Fallot, transposition of the great arteries). Median age at first surgery was 1.1 months (range = 0.1-23.8 months). The majority of adolescents born with a CHD were born full-term; however, 5 participants were born preterm <37 weeks of gestational age, with 1 of these participants born ≤ 29 weeks. These 5 participants did not differ significantly from the rest of the group in terms of sex, age at assessment, height, weight at testing, sociodemographic characteristics, or medical morbidities. Mean gestational age at birth of the preterm group was 26.5 weeks (SD = 1.8 weeks, range = 22.4-29.6 weeks). The CHD and preterm groups did not significantly differ in terms of sex ($\chi^2 = 0.0236$, $P = .878$), age at assessment ($t = -0.771$,

Table I. Comparison of participants' sociodemographic variables

	Categories	CHD frequency (%) n = 80	Preterm frequency (%) n = 128	*P* value
Maternal education	High school incomplete	4 (5.3%)	11 (9.1%)	.816
	High school	25 (32.9%)	37 (30.6%)	
	CEGEP/college	22 (28.9%)	33 (27.3%)	
	University degree	19 (25.0%)	27 (22.3%)	
	Graduate degree	6 (7.9%)	13 (10.7%)	
	Missing	4	7	
Paternal education	High school incomplete	15 (20.8%)	12 (11.0%)	.204
	High school	22 (30.6%)	27 (24.8%)	
	CEGEP/college	15 (20.8%)	34 (31.2%)	
	University degree	12 (16.7%)	25 (22.9%)	
	Graduate degree	8 (11.8%)	11 (10.1%)	
	Missing	8	19	
Total household income (before taxes)	0-$19 999	5 (6.9%)	12 (10.7%)	.660
	$20 000-$39 999	8 (11.1%)	18 (16.1%)	
	$40 000-$59 999	13 (18.1%)	22 (19.6%)	
	$60 000-$79 999	11 (15.3%)	16 (14.3%)	
	$80 000+	35 (48.6%)	44 (39.3%)	
	Missing	8	16	
Primary language	English	22 (28.9%)	58 (47.2%)	<.001
	French	47 (61.8%)	31 (25.2%)	
	Other	4 (5.3%)	16 (13.0%)	
	Mixed	3 (3.9%)	18 (14.6%)	
	Missing	4	5	

CEGEP, College d'enseignement général et professionnel (College of general and vocational education).
Percentages (%) reported exclude the missing variables.

$P = .441$), height ($t = 1.24$, $P = .218$), or weight at testing ($t = 1.12$, $P = .264$).

The parent-reported sociodemographic information for the 2 groups is presented in Table I. The 2 groups did not differ significantly in terms of maternal and paternal education, or total household income. The groups differed significantly with respect to primary language spoken at home: although most preterm participants were primarily English-speaking, most CHD participants were primarily French-speaking. The parent-reported medical morbidities of the 2 groups are presented in Table II. Parent-reported medical morbidities did not differ significantly between the 2 groups with the exception of asthma, which was more prevalent in the preterm group, and genetic syndromes and chromosomal abnormalities, which were more prevalent in the CHD group. Adolescents born with a CHD were also more frequently taking daily medication compared with those born preterm.

The mean scores of adolescents from the 2 groups on the various assessments are presented in Table III. Proportions of impairment in each group for all outcome measures are presented in Table IV.

Adolescents born with a CHD and adolescents born preterm had comparable cognitive outcomes, with no significant difference in Leiter-R brief IQ. Proportion of impairment did not differ significantly between the 2 groups, with 14.3% of adolescents born with a CHD and 11.8% of adolescents born preterm presenting with mild to severe cognitive delay (brief IQ <70).

Adolescents born with a CHD and adolescents born preterm had comparable motor outcomes, with no significant differences in MABC-II manual dexterity, aiming and catching, or balance scores or MABC-II total test score. Proportion of overall motor impairment did not differ significantly between the 2 groups, with 43.5% of adolescents born with a CHD and 50.0% of adolescents born preterm classified as at risk or as having significant

Table II. Group comparison of participants' medical morbidities

Medical morbidities (parent report)	CHD frequency (%) n = 80	Preterm frequency (%) n = 128	*P* value
Allergies	1 (1.3%)	7 (5.7%)	.158
Asthma	1 (1.3%)	15 (12.2%)	.013
Currently taking daily medication	23 (30.3%)	22 (17.9%)	.043
History of epilepsy	6 (7.9%)	5 (4.1%)	.341
Visual impairment (without glasses)	4 (5.3%)	16 (13.0%)	.082
Wears corrective lenses (glasses or contacts)	26 (36.1%)	52 (50.0%)	.068
Hearing impairment	5 (6.6%)	6 (4.9%)	.751
Speech/language impairment	1 (1.3%)	6 (4.9%)	.255
Cerebral palsy	0 (0%)	6 (4.9%)	.084
ADHD	19 (26.8%)	22 (22.2%)	.495
ASD/developmental delay	1 (1.3%)	7 (5.7%)	.158
Depression/anxiety	20 (28.2%)	23 (23.5%)	.489
Learning disability	25 (35.2%)	34 (34.7%)	.944
Genetic syndrome/ chromosomal abnormality	7 (8.5%)	2 (1.7%)	.033
Parents did not complete questionnaire	4 (5.0%)	5 (3.9%)	.736

ADHD, attention deficit hyperactivity disorder; *ASD*, autism spectrum disorder.
Percentages (%) reported exclude the missing variables.

Table III. Mean outcome scores of adolescents born with a CHD or born preterm

Outcomes	CHD mean ± SD (range)	Preterm mean ± SD (range)	*P* value
Leiter-R	**n = 77**	**n = 127**	
Brief IQ	89.6 ± 21.8 (36-127)	91.9 ± 18.7 (36-131)	.415
MABC-II	**n = 76**	**n = 126**	
Manual dexterity	7.54 ± 3.42 (1-14)	6.82 ± 3.22 (1-18)	.133
Aiming and catching	8.13 ± 3.98 (1-16)	7.83 ± 3.48 (1-15)	.577
Balance	9.45 ± 4.00 (1-15)	9.29 ± 3.67 (1-15)	.781
Total test score	67.0 ± 20.9 (11-97)	64.6 ± 18.3 (12-100)	.395
SDQ	**n = 76**	**n = 118**	
Prosocial scale score	8.18 ± 2.04 (3-10)	8.50 ± 1.72 (3-10)	.249
Emotional symptoms scale score	2.74 ± 2.14 (0-8)	2.54 ± 2.40 (0-10)	.566
Conduct problems scale score	1.42 ± 1.40 (0-6)	1.36 ± 1.48 (0-6)	.760
Hyperactivity scale score	3.37 ± 2.39 (0-10)	3.13 ± 2.58 (0-10)	.516
Peer problems scale score	2.16 ± 2.17 (0-8)	2.29 ± 1.93 (0-8)	.662
Impact scale score	3.68 ± 4.01 (0-13)	2.93 ± 3.86 (0-15)	.194
Total score	9.68 ± 5.92 (0-22)	9.31 ± 5.85 (0-27)	.669
VABS-II	**n = 75**	**n = 123**	
Communication	87.0 ± 15.4 (43-124)	91.2 ± 15.5 (45-124)	.066
Daily living skills	94.5 ± 17.2 (51-130)	91.6 ± 17.7 (35-136)	.264
Socialization	97.6 ± 19.0 (38-150)	98.9 ± 17.1 (42-135)	.617
Adaptive behavior composite	91.9 ± 17.0 (46-135)	92.7 ± 16.2 (44-135)	.764

movement difficulty (total test score ≤ 67). Proportion of impairment did not differ significantly between the 2 groups with respect to manual dexterity, aiming and catching, or balance. For both groups, the proportion of impairment was lowest for balance and highest for manual dexterity.

Adolescents born with a CHD and adolescents born preterm had comparable behavioral outcomes, with no significant differences in SDQ total test score or prosocial behaviors, emotional symptoms, conduct problems, hyperactivity, peer problems, or impact of behavior. Proportion of behavioral impairment, across all domains, did not differ significantly between the 2 groups. In summary, 23.7% of adolescents born with a CHD and 22.9% of adolescents born preterm had overall behavioral problems as indicated by borderline or abnormal SDQ total test scores. In addition, 57.8% of adolescents born with a CHD and 49.1% of adolescents born preterm had borderline or abnormal impact scale scores, indicating that these behavioral problems caused these individuals considerable distress or interfered with their home lives, friendships, classroom learning, or leisure activities, as reported by participants' parents.

Adolescents born with a CHD and adolescents born preterm had comparable functional outcomes, with no significant differences in VABS-II communication, daily living skills, or socialization scores or the VABS-II adaptive behavior composite. Furthermore, proportions of overall functional impairment and impairment in communication and socialization did not differ significantly between the two groups. Overall, 12.0% of adolescents born with a CHD and 7.3% of adolescents born preterm presented with mild to moderate functional deficits, as described by the VABS-II adaptive behavior composite. In contrast, the 2 groups differed significantly in terms of level of functional impairment related to daily living skills. However, when participants' VABS-II daily living skills scores were dichotomized as normal (≥78) or abnormal (<78), there was no significant difference in the distribution of scores between the 2 groups ($\chi^2 = 0.752$, $P = .386$), with 14.7% of adolescents born with a CHD and 19.5% of adolescents born preterm classified as having abnormal daily living skills.

To further explore the developmental deficits observed in adolescents born with a CHD, follow-up subgroup analyses were performed to compare the developmental outcomes of adolescents born with a single-ventricle CHD to those born with a biventricular CHD. Adolescents born with a single-ventricle CHD presented with significantly lower MABC-II manual dexterity scores and VABS-II communication, socialization, and adaptive behavior composite scores compared with adolescents born with a biventricular CHD, with a trend toward lower Leiter-R brief IQ scores (**Table V**; available at www.jpeds.com). Furthermore, a greater proportion of adolescents born with a single-ventricle CHD presented with significant difficulty with manual dexterity on the MABC-II, abnormal conduct problem scores and borderline total test scores on the SDQ, and moderately low adaptive behavior composite scores on the VABS-II (**Table VI**; available at www.jpeds.com). However, when participants' VABS-II adaptive behavior composite scores were dichotomized as normal (≥78) or abnormal (<78), there was no significant difference in the distribution of scores between the 2 groups ($P = .151$), with 33.3% of adolescents born with a single-ventricle CHD and 13.6% of adolescents born with a biventricular CHD classified as having abnormal functional outcome.

Discussion

Cognitive impairments have been previously documented in adolescents born with a CHD and adolescents born preterm. Previous studies have consistently uncovered deficits in a broad range of executive function, including working memory and

Table IV. Categorized outcomes of adolescents born with a CHD or born preterm

Outcomes	Category	CHD frequency (%)	Preterm frequency (%)	*P* value
Leiter-R		**n = 77**	**n = 127**	
Brief IQ	Very high/gifted (130-170)	0 (0%)	1 (0.8%)	.559
	High (120-129)	7 (9.1%)	7 (5.5%)	
	Above average (110-119)	9 (11.7%)	15 (11.8%)	
	Average (90-109)	25 (32.5%)	46 (36.2%)	
	Below average (80-89)	13 (16.9%)	26 (20.5%)	
	Low (70-79)	12 (15.6%)	17 (13.4%)	
	Very low/mild delay (55-69)	6 (7.8%)	12 (9.4%)	
	Moderate delay (40-54)	2 (2.6%)	3 (2.4%)	
	Severe delay (30-39)	3 (3.9%)	0 (0%)	
MABC-II		**n = 76**	**n = 126**	
Manual dexterity	No movement difficulty (≥7)	49 (64.5%)	66 (52.4%)	.168
	At risk (6)	6 (7.9%)	19 (15.1%)	
	Significant movement difficulty (≤5)	21 (27.6%)	41 (32.5%)	
Aiming and catching	No movement difficulty (≥7)	52 (68.4%)	80 (63.5%)	.718
	At risk (6)	5 (6.6%)	8 (6.3%)	
	Significant movement difficulty (≤5)	19 (25.0%)	38 (30.2%)	
Balance	No movement difficulty (≥7)	62 (81.6%)	100 (79.4%)	.957
	At risk (6)	3 (3.9%)	6 (4.8%)	
	Significant movement difficulty (≤5)	11 (14.5%)	20 (15.9%)	
Total test score	No movement difficulty (>67)	43 (56.6%)	63 (50.0%)	.648
	At risk (57-67)	17 (22.4%)	31 (24.6%)	
	Significant movement difficulty (<57)	16 (21.1%)	32 (25.4%)	
SDQ		**n = 76**	**n = 118**	
Prosocial scale score	Normal (6-10)	65 (85.5%)	108 (91.5%)	.232
	Borderline (=5)	5 (6.6%)	7 (5.9%)	
	Abnormal (0-4)	6 (7.9%)	3 (2.5%)	
Emotional symptoms scale score	Normal (0-3)	51 (67.1%)	84 (71.2%)	.543
	Borderline (=4)	11 (14.5%)	11 (9.3%)	
	Abnormal (5-10)	14 (18.4%)	23 (19.5%)	
Conduct problems scale score	Normal (0-2)	60 (78.9%)	94 (79.7%)	.984
	Borderline (=3)	9 (11.8%)	13 (11.0%)	
	Abnormal (4-10)	7 (9.2%)	11 (9.3%)	
Hyperactivity scale score	Normal (0-5)	63 (82.9%)	95 (80.5%)	.596
	Borderline (=6)	6 (7.9%)	7 (5.9%)	
	Abnormal (7-10)	7 (9.2%)	16 (13.6%)	
Peer problems scale score	Normal (0-2)	47 (61.8%)	68 (57.6%)	.534
	Borderline (=3)	9 (11.8%)	21 (17.8%)	
	Abnormal (4-10)	20 (26.3%)	29 (24.6%)	
Impact scale score	Normal (=0)	32 (42.1%)	60 (50.8%)	.457
	Borderline (=1)	3 (3.9%)	3 (2.5%)	
	Abnormal (2-10)	41 (53.9%)	55 (46.6%)	
Total test score	Normal (0-13)	58 (76.3%)	91 (77.1%)	.083
	Borderline (14-16)	4 (5.3%)	15 (12.7%)	
	Abnormal (17-40)	14 (18.4%)	12 (10.2%)	
VABS-II		**n = 75**	**n = 123**	
Communication	Moderately high (115-129)	4 (5.3%)	8 (6.5%)	.538
	Adequate (86-114)	37 (49.3%)	72 (58.5%)	
	Moderately low (71-85)	23 (30.7%)	32 (26.0%)	
	Mild deficit (50-70)	9 (12.0%)	10 (8.1%)	
	Moderate deficit (35-50)	2 (2.7%)	1 (0.8%)	
Daily living skills	High (130-160)	2 (2.7%)	1 (0.8%)	.013
	Moderately high (115-129)	5 (6.7%)	10 (8.1%)	
	Adequate (86-114)	50 (66.7%)	62 (50.4%)	
	Moderately low (71-85)	9 (12.0%)	40 (32.5%)	
	Mild deficit (50-70)	7 (9.3%)	8 (6.5%)	
	Moderate deficit (35-50)	2 (2.7%)	1 (0.8%)	
	Severe deficit (20-35)	0 (0%)	1 (0.8%)	
Socialization	High (130-160)	1 (1.3%)	3 (2.4%)	.941
	Moderately high (115-129)	12 (16.0%)	18 (14.6%)	
	Adequate (86-114)	44 (58.7%)	76 (61.8%)	
	Moderately low (71-85)	12 (16.0%)	19 (15.4%)	
	Mild deficit (50-70)	4 (5.3%)	5 (4.1%)	
	Moderate deficit (35-50)	1 (1.3%)	2 (1.6%)	
	Severe deficit (20-35)	1 (1.3%)	0 (0%)	
Adaptive behavior composite	High (130-160)	1 (1.3%)	1 (0.8%)	.827
	Moderately high (115-129)	5 (6.7%)	8 (6.5%)	
	Adequate (86-114)	48 (64.0%)	79 (64.2%)	
	Moderately low (71-85)	12 (16.0%)	26 (21.1%)	
	Mild deficit (50-70)	7 (9.3%)	7 (5.7%)	
	Moderate deficit (35-50)	2 (2.7%)	2 (1.6%)	

attentional control,[13-18] and impairments in general intelligence, perceptual reasoning, and visual-spatial skills.[13,14,17-20,27-32] However, although existing studies have largely focused on cognitive impairments, the prevalence of these deficits is actually relatively low in these 2 populations. Previous studies have detected cognitive impairments, using a threshold IQ of 70, in 4.3%-20.2% of adolescent preterm survivors[14,18-20] and 11%-17% of adolescents born with a CHD.[27,28,30] Our findings fall within the range in the literature, as within our sample, 14.3% of adolescents born with a CHD and 11.8% of adolescents born preterm presented with Leiter-R brief IQ < 70. As the Leiter-R is a nonverbal test of intelligence, the cognitive deficits seen in our sample are independent of difficulties that may arise due to the language delays observed in these populations.[4,17,18]

Cerebral palsy is a common comorbidity of preterm birth,[42] and clinical motor dysfunction has been documented in adolescents born with a CHD.[30] In addition to these severe neuromotor deficits, more subtle motor difficulties may impede the everyday lives of adolescents. However, these have remained largely unexplored in adolescents born preterm or with a CHD. In our study, motor impairment was the most common developmental deficit, with at-risk or definitively impaired performance identified in 43.5% of adolescents born with a CHD and 50.0% of adolescents born preterm. Proportions of difficulties in manual dexterity, aiming and catching, and balance were comparable between the 2 groups, with the highest rate of impairment in manual dexterity and the lowest rate of impairment in balance. These findings are consistent with a previous study in which 51% of adolescents born preterm aged 12-13 years were at risk or had significant movement difficulty, with the most significant deficits in manual dexterity compared with a control group.[24] In contrast, in another study, only 14.8% and 5.6% of adolescents born preterm aged 15 years had gross and fine motor impairments, respectively.[42] However, this study only reported the prevalence of participants with definite motor impairments, and not those within the at-risk range, and excluded participants with cerebral palsy from their calculations, potentially explaining this lower reported rate of motor impairment.

Follow-up regarding the behavioral outcomes of adolescents born with a CHD or born preterm has largely focused on the prevalence of psychiatric disorders in these populations.[26,32] Within our sample, 23.7% of adolescents born with a CHD and 22.9% of adolescents born preterm presented with overall behavioral problems, with particular difficulties with emotional problems and peer problems. These results are consistent with previous findings of behavioral problems in 24%-25% of adolescents born preterm aged 14 years old[43] and 19% of adolescents born with a CHD.[44] These behavioral problems may be particularly important to address, as our findings suggest that behavioral difficulties interfere with daily life in about one-half of adolescents born with a CHD or born preterm.

In our sample, 12.0% of adolescents born with a CHD and 7.3% of adolescents born preterm presented with overall functional deficits, ranging from mild to moderate, and also presented with specific deficits in communication, daily living, and socialization. It is important to consider that these functional limitations may not simply be a result of an actual inability to perform certain tasks, but of a perceived inability resulting from high levels of attention from and dependence on parents and caregivers in daily life. This interpretation is consistent with a phenomenological study in which parents of young adults with developmental disabilities viewed themselves as having vital and complex ongoing roles in their children's lives, acting as important sources of daily support and advocacy and often filling in the gaps left by external service providers.[45]

We found poorer cognitive, motor, behavioral, and functional outcomes in adolescents born with a single-ventricle CHD compared with those born with a biventricular CHD. These results are consistent with previous reports of poorer developmental outcomes in infants and children born with a single-ventricle CHD compared with a biventricular CHD.[46,47] However, our findings are limited by the small sample size of adolescents born with a single-ventricle CHD in this study and should be interpreted with caution.

This study provides further support to a growing body of evidence that suggests very similar patterns of developmental deficits during childhood and adolescence in individuals born with a CHD and individuals born preterm. These 2 perinatal conditions may have similar neurologic effects, in terms of the patterns of brain injury, specifically white matter injury, seen in individuals born with these conditions.[48-51] This suggests that different types of critical illness in early life may have similar effects on the immature brain. Unfortunately, although many of the adolescents born preterm in our sample had undergone head ultrasound as a part of their clinical follow-up, few adolescents born preterm had undergone brain magnetic resonance imaging and few adolescents born with a CHD had undergone any neuroimaging at the time, preventing us from confirming this hypothesis in our sample. Future studies comparing brain development between these populations and examining the extent to which observed brain abnormalities may impact these individuals' developmental outcomes are required.

Our findings highlight the need for improvements in the targeted delivery of educational and rehabilitation services to these at-risk adolescent populations. With sufficient long-term follow-up and evaluation, adolescents identified as experiencing cognitive, motor, behavioral, or functional deficits can be provided with targeted and individualized delivery of educational and rehabilitation services to ameliorate their particular difficulties.

There were several limitations in our study. First, the 2 groups differed in terms of several sociodemographic and medical variables. There was a significant difference in the primary language spoken by our 2 groups, representing a possible selection bias. Although there are differences in language acquisition patterns between English- and French-speaking children,[52] these differences are likely to be minimal by adolescence. Each participant was evaluated in their preferred language, ensuring optimal understanding of evaluation instructions by both English- and French-speaking participants. Furthermore, administration of a nonverbal test of intelligence limited confounding effects of possible differences in language ability on

cognitive outcome. There was a higher prevalence of asthma in the preterm group compared with the CHD group, in keeping with the documented long-term impairments in respiratory function in individuals born preterm.[53-55] There was a higher prevalence of genetic syndromes and chromosomal abnormalities in the CHD group. Genetic testing was not routinely performed for all infants born preterm or with a CHD, so we may be under-reporting the true prevalence of genetic conditions in our sample. In addition, a greater proportion of adolescents born with a CHD were taking medication on a daily basis than adolescents born preterm, possibly reflecting ongoing pharmacological management of residual impairments in cardiac function. Secondly, 5 members of the CHD group were born preterm <37 weeks of gestational age, but these participants did not differ significantly from the rest of the group in terms of sociodemographic characteristics or medical morbidities. Only 1 of these participants met the threshold for inclusion in the preterm group. However, the developmental outcomes of this participant fell within the typical range of the CHD group, so the inclusion of this participant likely did not skew the overall group outcomes. Finally, sociodemographic and medical morbidity data were collected using parental questionnaires and therefore, biased answers could have been introduced because of parents' subjective interpretations of the questions or possibly inaccurate answers.

Our findings highlight the importance of providing high-caliber long-term follow-up and care to both of these populations of at-risk adolescents. These findings emphasize the importance of targeted delivery of educational and rehabilitation services to adolescents born with a CHD or preterm to help them overcome the developmental difficulties they experience and to empower them to navigate the difficult and demanding transition from childhood to adulthood. ■

We acknowledge the invaluable contributions of our research assistants Anna Radzioch, Joey Waknin, Marie-Linda Boghdady, and Christopher Saunders. We also thank Patricia Grier, RN for her assistance in the recruitment of the preterm group and Dr Keiko Shikako-Thomas, Shira Vasilevsky, Melissa Turner, Rena Birnbaum, Marie-Elaine Lafrance, Corinne Mercier, Rochelle Rein, Nathalie Bilodeau, Anna Radzioch, and Dr Catherine Zygmuntowicz for performing developmental evaluations. These aforementioned individuals were compensated for their work with funds from an operating grant from the Canadian Institutes of Health Research (MOP-102720). None of these individuals have any industry affiliations or conflicts of interest to disclose. Finally, we especially thank the adolescents and their families for their participation in the study.

Submitted for publication Jul 24, 2018; last revision received Oct 31, 2018; accepted Nov 1, 2018

Reprint requests: Annette Majnemer, PhD, 5252 boul. de Maisonneuve Ouest—3F.44, Montreal, QC H3G 2M, Canada. E-mail: annette.majnemer@mcgill.ca

References

1. Anderson P, Doyle LW, Victorian Infant Collaborative Study Group. Neurobehavioral outcomes of school-age children born extremely low birth weight or very preterm in the 1990s. JAMA 2003;289:3264-72.
2. Bellinger DC, Wypij D, duPlessis AJ, Rappaport LA, Jonas RA, Wernovsky G, et al. Neurodevelopmental status at eight years in children with dextro-transposition of the great arteries: the Boston Circulatory Arrest Trial. J Thorac Cardiovasc Surg 2003;126:1385-96.
3. Bellinger DC, Wypij D, Kuban KC, Rappaport LA, Hickey PR, Wernovsky G, et al. Developmental and neurological status of children at 4 years of age after heart surgery with hypothermic circulatory arrest or low-flow cardiopulmonary bypass. Circulation 1999;100:526-32.
4. Hovels-Gurich HH, Bauer SB, Schnitker R, Willmes-von Hinckeldey K, Messmer BJ, Seghaye MC, et al. Long-term outcome of speech and language in children after corrective surgery for cyanotic or acyanotic cardiac defects in infancy. Eur J Paediatr Neurol 2008;12:378-86.
5. Hovels-Gurich HH, Konrad K, Wiesner M, Minkenberg R, Herpertz-Dahlmann B, Messmer BJ, et al. Long term behavioural outcome after neonatal arterial switch operation for transposition of the great arteries. Arch Dis Child 2002;87:506-10.
6. Majnemer A, Limperopoulos C, Shevell MI, Rohlicek C, Rosenblatt B, Tchervenkov C. A new look at outcomes of infants with congenital heart disease. Pediatr Neurol 2009;40:197-204.
7. Vohr BR, Wright LL, Dusick AM, Mele L, Verter J, Steichen JJ, et al. Neurodevelopmental and functional outcomes of extremely low birth weight infants in the National Institute of Child Health and Human Development Neonatal Research Network, 1993-1994. Pediatrics 2000;105:1216-26.
8. Wolke D, Meyer R. Cognitive status, language attainment, and prereading skills of 6-year-old very preterm children and their peers: the Bavarian Longitudinal Study. Dev Med Child Neurol 1999;41:94-109.
9. Woodward LJ, Anderson PJ, Austin NC, Howard K, Inder TE. Neonatal MRI to predict neurodevelopmental outcomes in preterm infants. N Engl J Med 2006;355:685-94.
10. Greutmann M, Tobler D. Changing epidemiology and mortality in adult congenital heart disease: looking into the future. Future Cardiol 2012;8:171-7.
11. Khairy P, Ionescu-Ittu R, Mackie AS, Abrahamowicz M, Pilote L, Marelli AJ. Changing mortality in congenital heart disease. J Am Coll Cardiol 2010;56:1149-57.
12. Stoll BJ, Hansen NI, Bell EF, Walsh MC, Carlo WA, Shankaran S, et al. Trends in care practices, morbidity, and mortality of extremely preterm neonates, 1993-2012. JAMA 2015;314:1039-51.
13. Litt JS, Taylor HG, Margevicius S, Schluchter M, Andreias L, Hack M. Academic achievement of adolescents born with extremely low birth weight. Acta Paediatr 2012;101:1240-5.
14. Luu TM, Ment L, Allan W, Schneider K, Vohr BR. Executive and memory function in adolescents born very preterm. Pediatrics 2011;127:e639-46.
15. Rushe TM, Rifkin L, Stewart AL, Townsend JP, Roth SC, Wyatt JS, et al. Neuropsychological outcome at adolescence of very preterm birth and its relation to brain structure. Dev Med Child Neurol 2001;43:226-33.
16. Soria-Pastor S, Gimenez M, Narberhaus A, Falcon C, Botet F, Bargallo N, et al. Patterns of cerebral white matter damage and cognitive impairment in adolescents born very preterm. Int J Dev Neurosci 2008;26:647-54.
17. Taylor HG, Minich N, Bangert B, Filipek PA, Hack M. Long-term neuropsychological outcomes of very low birth weight: associations with early risks for periventricular brain insults. J Int Neuropsychol Soc 2004;10:987-1004.
18. Taylor HG, Minich NM, Klein N, Hack M. Longitudinal outcomes of very low birth weight: neuropsychological findings. J Int Neuropsychol Soc 2004;10:149-63.
19. Doyle LW, Casalaz D, Victorian Infant Collaborative Study Group. Outcome at 14 years of extremely low birthweight infants: a regional study. Arch Dis Child Fetal Neonatal Ed 2001;85:F159-64.
20. Hille ET, Weisglas-Kuperus N, van Goudoever JB, Jacobusse GW, Ens-Dokkum MH, de Groot L, et al. Functional outcomes and participation in young adulthood for very preterm and very low birth weight infants: the Dutch Project on Preterm and Small for Gestational Age Infants at 19 years of age. Pediatrics 2007;120:e587-95.

21. Lee ES, Yeatman JD, Luna B, Feldman HM. Specific language and reading skills in school-aged children and adolescents are associated with prematurity after controlling for IQ. Neuropsychologia 2011;49:906-13.
22. Nosarti C, Giouroukou E, Healy E, Rifkin L, Walshe M, Reichenberg A, et al. Grey and white matter distribution in very preterm adolescents mediates neurodevelopmental outcome. Brain 2008;131:205-17.
23. Saigal S, Hoult LA, Streiner DL, Stoskopf BL, Rosenbaum PL. School difficulties at adolescence in a regional cohort of children who were extremely low birth weight. Pediatrics 2000;105:325-31.
24. Powls A, Botting N, Cooke RW, Marlow N. Motor impairment in children 12 to 13 years old with a birthweight of less than 1250 g. Arch Dis Child Fetal Neonatal Ed 1995;73:F62-6.
25. Healy E, Reichenberg A, Nam KW, Allin MP, Walshe M, Rifkin L, et al. Preterm birth and adolescent social functioning-alterations in emotion-processing brain areas. J Pediatr 2013;163:1596-604.
26. Indredavik MS, Vik T, Heyerdahl S, Kulseng S, Brubakk AM. Psychiatric symptoms in low birth weight adolescents, assessed by screening questionnaires. Eur Child Adolesc Psychiatry 2005;14:226-36.
27. Bellinger DC, Rivkin MJ, DeMaso D, Robertson RL, Stopp C, Dunbar-Masterson C, et al. Adolescents with tetralogy of Fallot: neuropsychological assessment and structural brain imaging. Cardiol Young 2015;25:338-47.
28. Bellinger DC, Watson CG, Rivkin MJ, Robertson RL, Roberts AE, Stopp C, et al. Neuropsychological status and structural brain imaging in adolescents with single ventricle who underwent the fontan procedure. J Am Heart Assoc 2015;4:e002302.
29. Bellinger DC, Wypij D, Rivkin MJ, DeMaso DR, Robertson RL Jr, Dunbar-Masterson C, et al. Adolescents with d-transposition of the great arteries corrected with the arterial switch procedure: neuropsychological assessment and structural brain imaging. Circulation 2011;124:1361-9.
30. Heinrichs AK, Holschen A, Krings T, Messmer BJ, Schnitker R, Minkenberg R, et al. Neurologic and psycho-intellectual outcome related to structural brain imaging in adolescents and young adults after neonatal arterial switch operation for transposition of the great arteries. J Thorac Cardiovasc Surg 2014;148:2190-9.
31. Latal B, Patel P, Liamlahi R, Knirsch W, O'Gorman Tuura R, von Rhein M. Hippocampal volume reduction is associated with intellectual functions in adolescents with congenital heart disease. Pediatr Res 2016;80:531-7.
32. Pike NA, Roy B, Gupta R, Singh S, Woo MA, Halnon NJ, et al. Brain abnormalities in cognition, anxiety, and depression regulatory regions in adolescents with single ventricle heart disease. J Neurosci Res 2018;96:1104-18.
33. von Rhein M, Scheer I, Loenneker T, Huber R, Knirsch W, Latal B. Structural brain lesions in adolescents with congenital heart disease. J Pediatr 2011;158:984-9.
34. Dahan-Oliel N, Mazer B, Maltais DB, Riley P, Nadeau L, Majnemer A. Child and environmental factors associated with leisure participation in adolescents born extremely preterm. Early Hum Dev 2014;90:665-72.
35. Dahan-Oliel N, Mazer B, Maltais DB, Riley P, Nadeau L, Majnemer A. What do you like to do in your free time? Activity preferences of adolescents born extremely preterm. J Child Adolesc Behav 2014;3:186.
36. Dahan-Oliel N, Mazer B, Riley P, Maltais DB, Nadeau L, Majnemer A. Participation and enjoyment of leisure activities in adolescents born at < 29 week gestation. Early Hum Dev 2014;90:307-14.
37. Majnemer A, Rohlicek C, Dahan-Oliel N, Hatzigeorgiou S, Mazer B, Maltais DB, et al. A profile of participation in adolescents with congenital heart defects at high risk for restricted engagement. Dev Med Child Neurol 2015;57:17.
38. Roid GH, Miller LJ. Leiter international performance scale-Revised (Leiter-R). Madrid: Psymtec; 2011.
39. Sparrow S, Cicchetti D, Balla D. Vineland adaptive behavior scales: (Vineland II), survey interview form/caregiver rating form. Livonia (MN): Pearson Assessments; 2005.
40. Smits-Engelsman BC, Fiers MJ, Henderson SE, Henderson L. Interrater reliability of the Movement Assessment Battery for Children. Phys Ther 2008;88:286-94.
41. Goodman R. The Strengths and Difficulties Questionnaire: a research note. J Child Psychol Psychiatry 1997;38:581-6.
42. Skranes J, Evensen KI, Lohaugen GC, Martinussen M, Kulseng S, Myhr G, et al. Abnormal cerebral MRI findings and neuroimpairments in very low birth weight (VLBW) adolescents. Eur J Paediatr Neurol 2008;12:273-83.
43. Stevenson CJ, Blackburn P, Pharoah PO. Longitudinal study of behaviour disorders in low birthweight infants. Arch Dis Child Fetal Neonatal Ed 1999;81:F5-9.
44. Spurkland I, Bjornstad PG, Lindberg H, Seem E. Mental health and psychosocial functioning in adolescents with congenital heart disease. A comparison between adolescents born with severe heart defect and atrial septal defect. Acta Paediatr 1993;82:71-6.
45. Bianco M, Garrison-Wade DF, Tobin R, Lehmann JP. Parents' perceptions of postschool years for young adults with developmental disabilities. Intellect Dev Disabil 2009;47:186-96.
46. Forbess JM, Visconti KJ, Hancock-Friesen C, Howe RC, Bellinger DC, Jonas RA. Neurodevelopmental outcome after congenital heart surgery: results from an institutional registry. Circulation 2002;106:I95-102.
47. Ibuki K, Watanabe K, Yoshimura N, Kakimoto T, Matsui M, Yoshida T, et al. The improvement of hypoxia correlates with neuroanatomic and developmental outcomes: comparison of midterm outcomes in infants with transposition of the great arteries or single-ventricle physiology. J Thorac Cardiovasc Surg 2012;143:1077-85.
48. Back SA, Miller SP. Brain injury in premature neonates: a primary cerebral dysmaturation disorder? Ann Neurol 2014;75:469-86.
49. Beca J, Gunn JK, Coleman L, Hope A, Reed PW, Hunt RW, et al. New white matter brain injury after infant heart surgery is associated with diagnostic group and the use of circulatory arrest. Circulation 2013;127:971-9.
50. Brossard-Racine M, du Plessis A, Vezina G, Robertson R, Donofrio M, Tworetzky W, et al. Brain injury in neonates with complex congenital heart disease: what is the predictive value of MRI in the fetal period? AJNR Am J Neuroradiol 2016;37:1338-46.
51. Miller SP, McQuillen PS, Hamrick S, Xu D, Glidden DV, Charlton N, et al. Abnormal brain development in newborns with congenital heart disease. N Engl J Med 2007;357:1928-38.
52. Pierce AE. Language acquisition and syntactic theory: a comparative analysis of French and English child grammars. Dordrecht: Kluwer Academic Publishers; 1992.
53. Anand D, Stevenson CJ, West CR, Pharoah PO. Lung function and respiratory health in adolescents of very low birth weight. Arch Dis Child 2003;88:135-8.
54. Chan KN, Noble-Jamieson CM, Elliman A, Bryan EM, Silverman M. Lung function in children of low birth weight. Arch Dis Child 1989;64:1284-93.
55. Coates AL, Bergsteinsson H, Desmond K, Outerbridge EW, Beaudry PH. Long-term pulmonary sequelae of premature birth with and without idiopathic respiratory distress syndrome. J Pediatr 1977;90:611-6.

Table V. Mean outcome scores of adolescents born with a single-ventricle or biventricular CHD

Outcomes	Single-ventricle CHD mean ± SD (range)	Biventricular CHD mean ± SD (range)	*P* value
Leiter-R	**n = 12**	**n = 65**	
Brief IQ	78.4 ± 21.1 (38-113)	91.6 ± 21.5 (36-127)	.053
MABC-II	**n = 12**	**n = 64**	
Manual dexterity	5.17 ± 3.83 (1-14)	7.98 ± 3.18 (1-14)	.008
Aiming and catching	6.25 ± 3.98 (1-13)	8.48 ± 3.91 (1-16)	.074
Balance	9.33 ± 4.66 (1-14)	9.47 ± 3.91 (1-15)	.915
Total test score	58.1 ± 23.0 (21-91)	68.7 ± 20.3 (11-97)	.109
SDQ	**n = 11**	**n = 65**	
Prosocial scale score	8.09 ± 2.30 (4-10)	8.20 ± 2.02 (3-10)	.871
Emotional symptoms scale score	3.00 ± 2.14 (0-7)	2.69 ± 2.16 (0-8)	.663
Conduct problems scale score	2.18 ± 1.99 (0-6)	1.29 ± 1.25 (0-5)	.178
Hyperactivity scale score	3.91 ± 1.92 (2-8)	3.28 ± 2.47 (0-10)	.422
Peer problems scale score	2.73 ± 2.00 (0-6)	2.06 ± 2.19 (0-8)	.349
Impact scale score	6.36 ± 5.20 (0-13)	3.23 ± 3.63 (0-13)	.080
Total score	11.8 ± 6.29 (2-22)	9.32 ± 5.83 (0-22)	.198
VABS-II	**n = 9**	**n = 66**	
Communication	75.6 ± 9.5 (62-93)	88.6 ± 15.5 (43-124)	.017
Daily living skills	89.9 ± 16.5 (68-111)	95.1 ± 17.3 (51-130)	.400
Socialization	85.2 ± 13.7 (64-106)	99.3 ± 19.1 (38-150)	.036
Adaptive behavior composite	80.9 ± 11.3 (63-101)	93.5 ± 17.1 (46-135)	.036

Table VI. Categorized outcomes of adolescents born with a single-ventricle or biventricular CHD

Outcomes	Category	Single-ventricle CHD frequency (%)	Biventricular CHD frequency (%)	*P* value
Leiter-R		**n = 12**	**n = 65**	
Brief IQ	High (120-129)	0 (0%)	7 (10.8%)	.063
	Above average (110-119)	1 (8.3%)	8 (12.3%)	
	Average (90-109)	4 (33.3%)	21 (32.3%)	
	Below average (80-89)	1 (8.3%)	12 (18.5%)	
	Low (70-79)	1 (8.3%)	11 (16.9%)	
	Very low/mild delay (55-69)	4 (33.3%)	2 (3.1%)	
	Moderatedelay (40-54)	0 (0%)	2 (3.1%)	
	Severe delay (30-39)	1 (8.3%)	2 (3.1%)	
MABC-II		**n = 12**	**n = 64**	
Manual dexterity	No movement difficulty (≥7)	3 (25.0%)	46 (71.9%)	.003
	At risk (6)	1 (8.3%)	5 (7.8%)	
	Significant movement difficulty (≤5)	8 (66.7%)	13 (20.3%)	
Aiming and catching	No movement difficulty (≥7)	6 (50.0%)	46 (71.9%)	.096
	At risk (6)	0 (0%)	5 (7.8%)	
	Significant movement difficulty (≤5)	6 (50.0%)	13 (20.3%)	
Balance	No movement difficulty (≥7)	10 (83.3%)	52 (81.2%)	1.000
	At risk (6)	0 (0%)	3 (4.7%)	
	Significant movement difficulty (≤5)	2 (16.7%)	9 (14.1%)	
Total test score	No movement difficulty (>67)	6 (50.0%)	37 (57.8%)	.578
	At risk (57-67)	2 (16.7%)	15 (23.4%)	
	Significant movement difficulty (<57)	4 (33.3%)	12 (18.8%)	
SDQ		**n = 11**	**n = 65**	
Prosocial scale score	Normal (6-10)	8 (72.7%)	57 (87.7%)	.127
	Borderline (=5)	2 (18.2%)	3 (4.6%)	
	Abnormal (0-4)	1 (9.1%)	5 (7.7%)	
Emotional symptoms scale score	Normal (0-3)	8 (72.7%)	43 (66.2%)	1.000
	Borderline (=4)	1 (9.1%)	10 (15.4%)	
	Abnormal (5-10)	2 (18.2%)	12 (18.5%)	
Conduct problems scale score	Normal (0-2)	6 (54.5%)	54 (83.1%)	.042
	Borderline (=3)	2 (18.2%)	7 (10.8%)	
	Abnormal (4-10)	3 (27.3%)	4 (6.2%)	
Hyperactivity scale score	Normal (0-5)	9 (81.8%)	54 (81.1%)	1.000
	Borderline (=6)	1 (9.1%)	5 (7.7%)	
	Abnormal (7-10)	1 (9.1%)	6 (9.2%)	
Peer problems scale score	Normal (0-2)	6 (54.5%)	41 (63.1%)	.793
	Borderline (=3)	1 (9.1%)	8 (12.3%)	
	Abnormal (4-10)	4 (36.4%)	16 (24.6%)	
Impact scale score	Normal (=0)	3 (27.3%)	29 (44.6%)	.497
	Borderline (=1)	0 (0%)	3 (4.6%)	
	Abnormal (2-10)	8 (72.7%)	33 (50.8%)	
Total test score	Normal (0-13)	6 (54.5%)	52 (80.0%)	.010
	Borderline (14-16)	3 (27.3%)	1 (1.5%)	
	Abnormal (17-40)	2 (18.2%)	12 (18.5%)	
VABS-II		**n = 9**	**n = 66**	
Communication	Moderately high (115-129)	0 (0%)	4 (6.1%)	.141
	Adequate (86-114)	2 (22.2%)	35 (53.0%)	
	Moderately low (71-85)	4 (44.4)	19 (28.8%)	
	Mild deficit (50-70)	3 (33.3)	6 (9.1%)	
	Moderate deficit (35-50)	0 (0%)	2 (3.0%)	
Daily living skills	High (130-160)	0 (0%)	2 (3.0%)	.502
	Moderately high (115-129)	0 (0%)	5 (7.6%)	
	Adequate (86-114)	5 (55.6%)	45 (68.2%)	
	Moderately low (71-85)	2 (22.2%)	7 (10.6%)	
	Mild deficit (50-70)	2 (22.2%)	5 (7.6%)	
	Moderate deficit (35-50)	0 (0%)	2 (3.0%)	
Socialization	High (130-160)	0 (0%)	1 (1.5%)	.147
	Moderately high (115-129)	0 (0%)	12 (18.2%)	
	Adequate (86-114)	4 (44.4%)	40 (60.6%)	
	Moderately low (71-85)	4 (44.4%)	8 (12.1%)	
	Mild deficit (50-70)	1 (11.1%)	3 (4.5%)	
	Moderate deficit (35-50)	0 (0%)	1 (1.5%)	
	Severe deficit (20-35)	0 (0%)	1 (1.5%)	
Adaptive behavior composite	High (130-160)	0 (0%)	1 (1.5%)	.004
	Moderately high (115-129)	0 (0%)	5 (7.6%)	
	Adequate (86-114)	2 (22.2%)	46 (69.7%)	
	Moderately low (71-85)	6(66.7%)	6 (9.1%)	
	Mild deficit (50-70)	1 (11.1%)	6 (9.1%)	
	Moderate deficit (35-50)	0 (0%)	2 (3.0%)	