Impaired Motor Competence in School-aged Children With Complex Congenital Heart Disease

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Objective: To explore the extent and type of motor problems in children with complex congenital heart disease (CHD) compared with schoolchildren without any documented heart failure.

Design: Cohort study.

Setting: Biomechanical Laboratory, Rikshospitalet-Radiumhospitalet Medical Centre.

Participants: One hundred twenty children aged 7 to 12 years with complex CHD and 387 healthy schoolchildren in the same age range (control group).

Interventions: All children with CHD were surgically treated with multiple corrections within the first year of life.

Main Outcome Measures: Movement Assessment Battery for Children, grip strength, quadriceps muscle strength, and balance.

Results: Compared with the control group, children with CHD had a risk of having any degree of impaired motor competence of 5.8 (95% confidence interval, 3.8-8.8). The risk for having severe motor problems was 11.0 (95% confidence interval, 5.4-22.5). There were highly significant differences between the groups for manual dexterity, ball skills, grip strength, quadriceps muscle strength, and static and dynamic balance (P < .001).

Conclusions: Children with CHD have a risk of severe motor problems 11-fold that of schoolchildren without any known heart failure. This suggests that primary health care providers should screen the motor competence in children with CHD at an early age to initiate therapeutic actions for children who show incipient motor problems. Optimal rehabilitative, social, and environmental support may improve the children’s motor competence and prevent future health problems.

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Advances in cardiac surgery have improved the survival rate in children with complex congenital heart disease (CHD). In the United Kingdom, the survival rate for infants undergoing operation before 1 year of age was 90% in 2001.1 The morbidity, however, needs further elucidation.2 Several studies have shown that the survivors are at great risk for neurological, cognitive, and behavioral impairments.3–6 However, Mahle and Wernovsky7 claimed that, even if children with repaired CHD have an increased risk of neurocognitive deficits, most survivors perform within the normal range of most standardized measures and are comparable with survivors of other congenital lesions.

Previous surveys of children with CHD have mainly focused on neurological and behavioral problems and aerobic capacity.3,5,8–10 Less attention has been paid to motor competence, but there are some studies indicating that children with CHD have an increased prevalence of reduced motor skills, at least in early childhood. Limperopoulos et al8 found that 42% of children with CHD showed gross and/or fine motor delays 12 to 18 months after neonatal open heart surgery. Five years later, the gross and/or fine motor delays were present to the same extent.11 Whether and to what extent the motor problems still exist later in childhood is, however, unknown. Hövels-Gürich et al12 examined 77 children aged 3 to 9 years (mean age, 5.4 years) after the neonatal arterial switch operation. They found fine motor dysfunction in 22.1% and gross motor dysfunction in 23.4%. Karl et al13 evaluated neurodevelopmental outcome in 74 patients who had undergone the arterial switch operation after a minimum of 48 months. They used the Movement Assessment Battery for Children (Movement...
Because of the success of pediatric cardiology and heart surgery, the number of adults with CHD has increased. Motor competence below the reference range may influence the performance of daily life activities (eg, dressing and eating) and participation in children’s play, school activities, and sports. This may lead to a more inactive lifestyle and have potential implications for later health problems. In addition, motor competence may also be an important determinant of academic achievement, the child’s level of self-esteem and self-concept, and the child’s popularity and status within a peer group. It is unusual for motor problems in early childhood to simply disappear as the child gets older. In the absence of an intervention, the problems may still be present at the time of maturation.

Because of the success of pediatric cardiology and heart surgery, the number of adults with CHD has increased. Experts in cardiology suggest that the number of adults with CHD, regardless of repair, will approach the number of children with the disorder. Wren and O’Sullivan estimated that, in the United Kingdom alone, the number of patients with CHD needing follow-up beyond 16 years of age would be more than 1600 new cases each year or more than 200 cases per 100,000 live births. These estimates underline the importance of identifying potential developmental problems in early age, to give the children the best opportunities for optimal motor competence as adults. Having higher motor skills may increase options for participation in different work-related and physical activities as adults. The adolescents and adults with CHD will then benefit from the documented and positive effects of physical activities on musculoskeletal health, cardiovascular health, adiposity, and blood pressure.

The purpose of the present study was to explore the extent and type of motor problems in children with complex CHD compared with school-aged children with no known heart failure (control group). The findings will provide important information to all medical staff (cardiologists, general practitioners, physical therapists, etc) working with these children, as a basis for giving adequate advice concerning motor learning and physical activity to the children themselves, their parents, and personnel in preschools and primary schools.

METHODS

All children in Norway with severe CHD aged 7 to 12 years (183 eligible children) who had undergone a surgical repair with multiple and complex corrections within the first year of life and with no documented mental disturbances were invited by mail to participate in the study. They were traced from a database at the Rikshospitalet-Radiumhospitalet Medical Centre, which has the responsibility for all children in the country born with a serious heart disease. Patients with simple and moderate defects, such as single arterial septal defect, ventricular septal defect, coarctation of the aorta, aortic valve stenosis, and pulmonic stenosis, were excluded.

Schoolchildren in the Oslo area were recruited to participate in the control group. The schools were, for practical reasons, selected on the basis of accessibility. They represent the suburbs located no further than 4 to 5 km from the hospital (ie, an urban area) and may not be representative of the total population of Norwegian children. However, the main purpose was that they should match the children with CHD by age and sex. One of the study staff (P.M.F.) met with each class and told the children about the intention and design of the study. The children were given a take-home letter that included written information about the study and a request for consent from the parents to participate. Those who responded positively were contacted by e-mail and given information about where and when to meet for testing.

The testing was performed in the biomechanical laboratory at the hospital. The study staff consisted of 7 physical therapists who had extensive experience with the testing equipment. They were not blinded to group assignment.

Approval was obtained from the Regional Committee of Medical Research Ethics, the Data Inspectorate, and the City of Oslo Education Authority.

OUTCOME MEASURES

Anthropometric data were collected and included birth date, sex, ethnicity, class, school, weight, height, leg length, and chronic conditions other than CHD. The order in which the different tests were performed was unsystematic and depended on the number of children undergoing testing on the same day and on which device was free at the moment.

MOTOR IMPAIRMENTS

The Movement ABC was used to test the children’s motor skills. The Movement ABC is divided into 4 age bands: 4 to 6 years (age band 1), 7 to 8 years (age band 2), 9 to 10 years (age band 3), and 11 to 12 years (age band 4). The test consists of 8 tasks, grouped as the following 3 subscores: manual dexterity (3 tasks), ball skills (2 tasks), and static/dynamic balance (3 tasks). Each task is scored from 0 (best) to 5 (worst) and summed up in a total score ranging from 0 (best score) to 40 (worst score). Children whose total scores fall between the 5th and 15th percentiles of the population are defined as clumsy or at risk, and those who have a total score below the 5th percentile are defined as having a definite motor problem. According to a large sample of reference children given in the manual, the 5th and 15th percentiles represent 13.5 and 10.0 total score points, respectively. Depending on the age and capability of the individual child, the test took 25 to 40 minutes. Test-retest reliability of the Movement ABC, estimated using intraclass correlation coefficients, is high (range, 0.92-0.98) and the concurrent validity is moderate (Pearson r values, 0.60-0.90). The tests were guided by experienced physiotherapists (I.H., P.M.F., and M.A.F.).

MUSCLE FUNCTION

Knee extension and flexion were tested isokinetically using a dynamometer (Cybex 6000; Cybex-Lumex Inc, Ronkonkoma, New York). The test protocol consisted of 5 repetitions at an angular velocity of 60° per second (strength) followed by a 1-minute rest period and 30 repetitions at 240° per second (endurance). The variable used for analysis was total work. The isokinetic knee test is reliable in children, with intraclass correlation coefficients ranging from 0.78 to 0.90; however, to our knowledge, no validity studies have been performed in children.

Isometric grip strength of the dominant and nondominant hands was determined using a standard adjustable-handle hand-
grip dynamometer (Jamar, Bolingbrook, Illinois). The handle was adjusted to the child’s hand size, with setting 1 used for the younger children and setting 2 for the older children. Settings 3, 4, and 5 were never used. The subject was seated on a chair with the shoulder adducted and neutrally rotated. The elbow was flexed at 90° and the forearm was held in a neutral position. The children were told to make their best effort on the dynamometer, with 1 repetition for each hand. The reproducibility of the handgrip dynamometer is high (intraclass correlation coefficients, 0.91-0.93), and the validity is acceptable.28

**BALANCE**

Static balance was tested using a commercially available system (KAT 2000; OEM Medical, Carlsbad, California) consisting of a movable platform supported at its central point by a small pivot. A tilt sensor on the platform is connected to a computer, which registers the deviation of the platform from a reference position of 18.2 times each second. The distance from the central point to the reference position is measured at every registration, and from the summation of these distances, a score—the balance index—can be calculated (a low balance index indicates good ability to perform the balance task). Each child completed a 1-leg static balance test, with the dominant leg tested first, and 2 trials on each leg. The test is described in detail by Hansen et al.29 The balance platform system has not been tested for reliability and validity in children.

**STATISTICAL METHODS**

We used SPSS statistical software (version 13.0; SPSS Inc, Chicago, Illinois) for the statistical analyses. Differences in scores between the CHD and control groups were calculated and presented as means, standard deviations, and confidence intervals for group differences. Independent-samples t tests were used to compare the mean score of the 2 groups. The significance level was set to .01.

To estimate the risk for children with CHD to develop increased motor problems, we calculated risk ratios using a generalized linear model with a log link and binomial error distribution.

**RESULTS**

From October 1, 2003, through August 31, 2006, 387 school-aged children and 120 children with complex CHD agreed to participate in the present survey. Of the 183 children with CHD who received an invitation to the study, 120 (65.3%) responded positively. Their diagnoses included tetralogy of Fallot (24.2%), transposition of the great arteries (26.7%), hypoplastic right/left ventricle (15.0%), tricuspid atresia (2.5%), and others (31.6%). The main surgical procedures were the transannular patch (for tetralogy of Fallot), atrial switch procedure (Senning/Mustard atrial repair for transposition of the great arteries), and arterial switch. During surgery, 90 patients (75.0%) were connected to a heart-lung machine for a mean duration of 88 minutes. In the nonparticipating group (n=63), 41 patients (65%) did not answer our invitation letter and 22 patients (35%) answered that they did not want to participate or did not show up on the test day. The diagnoses for the nonparticipating group were the tetralogy of Fallot (16 [25%]), transposition of the great arteries (17 [27%]), hypoplastic right/left ventricle (13 [21%]), and others (17 [27%]). Two of the schoolchildren did not perform the Movement ABC test owing to lack of time.

The distribution of girls to boys was 38.2% to 61.8% and 51.4% to 48.6% for the CHD and control groups, respectively. For the CHD group, the mean (SD) age was 10.3 (1.7) years; height, 140.8 (12) cm; and weight, 35.2 (10.6) kg. The corresponding values for the control group were 10.2 (1.7) years, 142.3 (11.2) cm, and 36.3 (8.6) kg. For age, height, and weight, no statistical differences were found between the 2 groups. We found no sex difference for performance in the CHD or the control group.

Both parents were of Norwegian origin for 89.1% of the CHD group and 88.3% of the control group. The corresponding percentages for right-hand dominance were 89.4% and 88.3%. The CHD group showed significantly higher values than the control group for the total Movement ABC score and all 3 subscores (Table 1). The Figure shows that the distribution of the total Movement ABC score spans the entire range for the CHD group, in contrast to the much narrower distribution for the control group. When applying the common criteria for clumsiness and definite motor problems, we found that 20 of 120 children in the CHD group (16.7%) showed clumsiness compared with 19 of the 385 children in the control group (4.9%). Thirty-one of the 120 children in the CHD group (25.8%) had severe motor problems compared with 9 (2.3%) in the control group. Compared with the control group, the children with CHD had a risk of having any degree of impaired motor competence of 3.8

### Table 1. Mean Outcome Values and Differences Between Groups for the Movement ABC Total Score and Subscores

<table>
<thead>
<tr>
<th>Assessment (Range of Scores)</th>
<th>Children With CHD (n=120)</th>
<th>Healthy Children (n=385)</th>
<th>Mean Differences Between Groups (95% CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Total Movement ABC score (0-40)</td>
<td>10.0 (9.7-10.3)</td>
<td>4.7 (9.6-10.0)</td>
<td>5.3 (5.0-5.6)</td>
</tr>
<tr>
<td>Manual dexterity (0-15)</td>
<td>4.3 (4.0-4.6)</td>
<td>2.1 (1.6-2.6)</td>
<td>2.2 (1.9-2.5)</td>
</tr>
<tr>
<td>Ball skills (0-10)</td>
<td>2.4 (2.4-2.6)</td>
<td>0.9 (1.1-1.9)</td>
<td>1.5 (1.2-1.8)</td>
</tr>
<tr>
<td>Static/dynamic balance (0-15)</td>
<td>3.3 (3.3-3.5)</td>
<td>1.0 (1.0-1.2)</td>
<td>2.2 (1.9-2.5)</td>
</tr>
</tbody>
</table>

Abbreviations: CHD, congenital heart disease; CI, confidence interval; Movement ABC, Movement Assessment Battery for Children.

a The mean differences between groups were statistically significant for all assessments (P<.001 for all comparisons).
(95% confidence interval, 3.8-8.8). The risk for having severe motor problems was 11.0 (95% confidence interval, 5.4-22.5).

There were highly significant differences between the groups for grip strength, quadriceps muscle strength, and balance (Table 2). The CHD group showed a 21.4% lower quadriceps strength and 17.7% lower hand grip strength. The balance index was 27.5% higher (worse) for the CHD group. For the CHD group, the correlations between motor skills and muscle strength and between motor skills and balance were high (r = 0.87 and r = 0.65, respectively), indicating that the children with high Movement ABC scores also showed the lowest muscle strength and poorest balance.

In the present study, we found an increased amount of motor problems in children with CHD (42.5%) compared with healthy children (7.2%). The results indicate that children with CHD have a risk of any degree of impaired motor competence or severe motor problems that is almost 6 and 11 times, respectively, that of healthy age- and sex-matched children. The children with CHD also showed significantly reduced muscle strength and impaired balance performance. Muscular strength and balance are essential components of several motor skills in that a certain level of muscular strength and balance is necessary to perform specific tasks. On the other hand, the ability to perform several motor skill tasks is used as an indicator of specific aspects of strength and balance. The present study showed highly significant differences between the 2 study groups for all the different tasks executed. These findings indicate that the reduced performance and impaired motor competence found in the children with CHD may be a general problem that is not simply associated with particular tasks or qualities but concerns muscle strength, balance, and fine and gross motor skills.

Almost 60% of the eligible children with CHD agreed to participate in the study. The children who did not want to participate were not asked to explain their decision. However, we have no reason to assume that only the children with no or minor motor problems failed to participate, thereby resulting in a relative risk for impaired motor skills that is too high (57% of the test results were categorized as within the reference range). The prevalence of motor problems in the control group (7.2%) is identical to those found in previously published studies (6%-10%) and should therefore be a representative sample for comparison. The evaluations were not performed in a blinded fashion, which may have influenced the results. The examiners were aware of the group assignment but not the medical history. Most of them, however, had not participated in the hypothesis generation or in the planning of the study; they were hired only to administer the tests and therefore were minimally biased.

Cognitive and neurological outcomes have been the primary areas described in studies of children with CHD. Some studies, however, have focused on motor competence. Majnemer et al. found that more than 40% of the children with CHD undergoing neonatal open heart surgery had gross and/or fine motor delays 5 years after the surgery. Compared with the results from the present study, the percentage of children with CHD who showed motor problems is almost identical, indicating that the incidence of motor problems is quite high and remains stable as the child grows older. Chen et al. compared growth and development of preschool children with CHD and

### Table 2. Mean Outcome Values and Differences Between Groups for Strength and Balance

<table>
<thead>
<tr>
<th>Assessments</th>
<th>Children With CHD (n=120)</th>
<th>Age- and Sex-Matched Healthy Children (n=387)</th>
<th>Mean Differences Between Groups (95% CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Quadriceps strength, Nm</td>
<td>219 (106.3) [52 to 514]</td>
<td>278.8 (112.6) [76-706]</td>
<td>-59.7 (-83 to -36)</td>
</tr>
<tr>
<td>Grip strength, N</td>
<td>145.3 (47.4) [39.2 to 274.7]</td>
<td>176.6 (51.2) [58.9 to 372.8]</td>
<td>-31.3 (-44 to -21)</td>
</tr>
<tr>
<td>Static balance index</td>
<td>592 (302) [245 to 1803]</td>
<td>464 (193) [170 to 1363]</td>
<td>127.5 (81 to 174)</td>
</tr>
</tbody>
</table>

Abbreviations: CHD, congenital heart disease; CI, confidence interval; Nm, Newton meters.

- The mean differences between groups were statistically significant for all assessments (P < .001 for all comparisons).

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healthy preschool children and found significant differences in growth, language development, and gross motor development. The children with CHD weighed significantly less and were slightly shorter than the healthy preschool children. The present study did not show the same differences between the 2 groups concerning physical growth because there were no significant differences for height or body weight. This suggests that the delayed growth shown in the study by Chen et al may not be an explanatory factor for impaired motor competence in the CHD group.

Several studies have tried to identify factors that may increase the risk for persistent developmental deficits in children with CHD. The etiology of neurological deficits in this population appears to be multifactorial, with preoperative (eg, structural brain abnormalities), operative (eg, circulatory arrest), and postoperative factors (eg, low cardiac output). The same factors may influence motor development. However, Chen et al claimed that, in addition, children with CHD have a more sheltered upbringing and are often overprotected by their parents. This may limit their opportunities for participating in activities or interacting with peers and restrict their movement experience significantly. Adequate and repeated instruction about the positive effects of physical activity may, to a certain degree, eliminate this negative factor.

The high prevalence of motor problems shown among children with CHD in this study indicates that preventive or rehabilitative programs could be of great importance. It is crucial to give children with motor problems adequate physical challenges, a well-rounded and many-sided movement experience, and the opportunity to participate socially at home and in school. This active approach should be the responsibility of parents, teachers, and primary health care providers.

Today, most children born with complex CHD will survive and reach adolescence and adulthood. Regular physical activity is associated with substantial health benefits for children and adolescents. In addition, active youths are more likely to become active adults, contributing to health across the life span.

We found an increased amount of definite motor problems in children with CHD compared with age- and sex-matched children with no documented heart disease. There were highly significant differences between the 2 groups for manual dexterity, ball skills, muscle strength, and balance. This suggests that health care providers need to perform a routine screening of motor competence in all children with CHD at an early age to identify those who have a developmental delay.

CONCLUSIONS

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Author Contributions: All authors had full access to all data in the study. Dr Holm and Ms Olstad take responsibility for the integrity of the data and the accuracy of the data analysis. Study concept and design: Holm and Fredriksen. Acquisition of data: Holm, Fredriksen, and Fosdahl. Analysis and interpretation of data: Holm, Fosdahl, Olstad, and Vollestad. Drafting of the manuscript: Holm. Critical revision of the manuscript for important intellectual content: Fredriksen, Fosdahl, Olstad, and Vollestad. Statistical analysis: Holm, Fosdahl, Olstad, and Vollestad. Obtained funding: Holm and Vollestad. Administrative, technical and material support: Holm, Fredriksen, Fosdahl, and Vollestad.

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Trial Registration Required

In concert with the International Committee of Medical Journal Editors (ICMJE), Archives of Pediatrics and Adolescent Medicine will require, as a condition of consideration for publication, registration of all trials in a public trials registry (such as http://ClinicalTrials.gov). Trials must be registered at or before the onset of patient enrollment. This policy applies to any clinical trial starting enrollment after July 1, 2005. For trials that began enrollment before this date, registration will be required by September 13, 2005, before considering the trial for publication. The trial registration number should be supplied at the time of submission.

For details about this new policy, and for information on how the ICMJE defines a clinical trial, see the editorials by DeAngelis et al in the September 8, 2004 (2004;292:1363-1364) and June 15, 2005 (2005;293:2927-2929) issues of JAMA. Also see the Instructions to Authors on our Web site: www.archpediatrics.com.