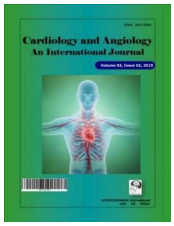

Cardiology and Angiology: An International Journal
4(3): 90-97, 2015, Article no.CA.2015.031
ISSN: 2347-520X, NLM ID: 101658392

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Kindergarten Children with Congenital Heart Disease Show Good Physical Activity but Reduced Motor Skills in Comparison with Healthy Children

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Authors' contributions

Authors AE, PBH and AH were responsible for conception and design of the study. Author AE sampled the data in the clinic and the kindergartens. Authors AE and AH were responsible for data monitoring, integrity, analysis and drafted the manuscript. Authors RO and PE gave important input for revising the manuscript. All authors have read and approved the final version of the manuscript.

Article Information

DOI: 10.9734/CA/2015/17997
Editor(s):
(1) Francesco Pelliccia, Department of Heart and Great Vessels University La Sapienza, Rome, Italy.
Reviewers:
(1) Anonymous, Medical University, Poland.
(2) Pietro Scicchitano, Cardiology Department, "San Giacomo" Hospital, Monopoli, Italy.
Complete Peer review History: http://sciencedomain.org/review-history/10292

Original Research Article

Received 31st March 2015
Accepted 8th July 2015
Published 24th July 2015

ABSTRACT

Objective: For the interaction of individuals with their environment, motor competence is of major importance. It is known that school children with congenital heart disease (CHD) have motoric limitations even without hemodynamic residuals. Data from kindergarten children is lacking. This study was to compare the motor competence of kindergarten children with congenital heart disease (4-6 years) with healthy children of the same age group.
Patients and Methods: A motor test "MOT 4-6" with 18 tasks in different groups of motor skills was performed in 62 children (19 female, 43 male) with various forms of CHD and compared to 39 healthy children (22 female, 17 male). In addition to the motor test all subjects answered the Kiddy-KINDL® quality of life questionnaire, and wore an accelerometer to capture daily physical activity for seven consecutive days.
Results: The median (quartile 1; quartile 3) motor quotient in the CHD group (104 [96;113]) was

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significantly lower than in the control group (111 [104;116]; Mann-Whitney-U test p=0.005). Quality of life did not differ significantly (p=0.774, parents' questionnaire p=0.066), nor the minutes in moderate and vigorous physical activity (p=0.093). No correlation between the motor quotient and the other variables could be shown.
Conclusion: Kindergarten children with CHD should be screened for a normal motor development. This delay seems to be independent from daily physical activity.

Keywords: Congenital heart disease; motor development; physical activity.

1. INTRODUCTION

Motor development is one of the essential aspects in young children [1]. The perceptual and motor experience of children influences not only physical and motor, but also emotional, psychosocial, and cognitive development [2]. Deficits in motor activities might affect the child's entire personal development [1,3]. Most children born with congenital heart disease (CHD), who were repaired successfully in infancy, are able to participate in all normal age-appropriate physical activities with their healthy peers [4]. However, in more detailed tests several children show neurodevelopmental deficits. The deficits in motor development are dependent of the severity of the cardiac lesion [5], and are also seen in children without hemodynamic residuals. These deficits might result from immobilisation periods after surgery or catheter intervention, but also because of an overprotective behaviour of the parents.

Few studies have focused on motor development of children with congenital heart disease [1-3, 5-7]. Most of them examined school children. Earlier data about the motor development of kindergarten children with CHD are rare. In a review about motor and cognitive outcomes after early surgery for CHD [8], only three studies focused on kindergarten children. Only one of these three studies investigated motor development in this age group. None of these studies have included physical activity data to investigate the deficits in relation to daily physical activity [8].

The aim of the present study was to examine kindergarten children with CHD, to compare their gross and fine motor skills with healthy peers of the same age and to relate these findings to daily physical activity.

2. PATIENTS AND METHODS

2.1 Study Design

From May to October 2010 and May to September 2011, we recruited all children at the age of 4-6 years, attending our outpatient department for routine follow-up of their CHD. We chose to recruit only in spring and summertime, when outdoor activity is performed regularly to avoid a bias in daily activity by seasonal effects. Children with syndromes, physical disabilities (severe neurodevelopmental retardation such as hydrocephalus, microcephaly or trisomy 21), who were expected to be unable to perform any task of the motoric test and children with cardiac intervention less than six month ago were excluded (Fig. 1). The sample of the tested children represented the complete spectrum of congenital cardiac disease. The detailed diagnoses corresponding to complex, moderate and mild lesions [9] are presented in Table 1. In parallel, healthy preschool children of three kindergartens were recruited. The age range of the included children was 4-6 years. All anthropometric data is depicted in Table 2.

All subjects performed the "motoric test for kindergarten children (MOT 4-6)", answered the "Kiddy-KINDL® questionnaire", and wore an accelerometer to capture daily activity for the next seven days after the test. In addition the parents were asked to rate their children's quality of life.

The study was designed as a prospective cross-sectional cohort study. The study protocol was in accordance with the declaration of Helsinki. It was approved by the local ethical board (project number 2782/10). All patients' parents gave written informed consent. All authors read the final version of the manuscript and agreed with its publication.

2.2 Motor Development Assessment

To assess the motor development, the "MOT 4-6" from R. Zimmer and M. Volkamer was used [10]. It was developed in 1984 to capture the motor development of 4-6 year old children. The test consists of 18 tasks in different groups of motor skills: gross motor skills of the whole body (5 tasks), fine motor skills (3 tasks), balance (5 tasks), reaction (2 tasks), bounce (2 tasks),

speed (3 tasks), and control of motion (2 tasks). The first task for the children is to acclimatize and get comfortable with the test situation. The other tasks are scored with zero (not able to perform), one (intermediate performance) or two points (correct performance). Precise instructions are given for the 18 tasks, as well as the definition of "intermediate" and "correct" performance. At the end all task scores are summed up to a raw value and transformed into a motor quotient, according to the age-dependent reference values [10]. A higher motor quotient represents a better motor ability with a mean of 100 and a standard deviation of 15 in a reference population.

The motoric test for children with CHD was performed in our hospital in a physio therapy room. The healthy children were tested in their kindergarten in a gym. All examinations were performed by one of the authors (A.E.).

2.3 Daily Activity Assessment

Daily physical activity was measured by the tri-axial accelerometer "RT3" (Stayhealthy, Monrovia, California, USA) for seven days after the motoric tests. The accelerometer was allowed to be taken off only during showering, swimming or at bedtime. Data sets accounting for less than 5 days were discarded.

The "RT3" is designed as a complete activity recording and measurement device for clinical and research applications. Worn on the waist, it continuously tracks activities throughout the day with the use of piezo-electric accelerometer technology. It measures motion in three dimensions and provides tri-axial vector data in activity units.

In our study, vector magnitudes were used to calculate the three dimensions with a sampling epoch of one minute. The daily minutes in moderate (3-6 metabolic equivalents) and vigorous activity (>6 metabolic equivalents) were calculated, using the published cut-off-points for moderate (> 970 count/min) and vigorous (> 2333 counts/min) activity [11]. Data were averaged for the sampling days. For statistics, the data from moderate to vigorous activity representing all activity >3 metabolic equivalents were pooled.

2.4 Quality of Life Assessment

The KINDL® questionnaire was applied to assess the health-related quality of life for children. It is used both for clinical assessment as well as for healthy children. It was developed for three different age groups (4-7 years, 8-12 years and 13-16 years) and additionally for parents to classify their children. The questionnaire was tested and evaluated in several studies with 3000 healthy children and children with chronic disease [12]. It has proven to be a flexible, modular, and accepted psychological method to evaluate the quality of life for children.

In our study we used the Kiddy-KINDL® for children aged 4-7 years and the Kiddy-KINDL® questionnaire for parents of children from the same age group. The questionnaire for children consists of 12 items about body, feelings, self-estimation, family, friends, and kindergarten (two items each). For the parents' questionnaire, the same items as in the children's questionnaire are used but supplemented by 22 general questions about their relation to the child.

Table 1. primary cardiac diagnosis in the CHD group

15	**Simple CHD**
7	Atrial septal defect
2	Ventricular septal defect
2	Persistant foramen ovale
1	Persistent arterial duct
3	Primary arrhythmia (supraventricular tachycardia/premature ventricular contraction)
22	**Moderate CHD**
4	Tetralogy of Fallot
5	Aortic stenosois
5	Coarctation of the aorta
2	Pulmonary stenosis
2	Totally anomalous pulmonary venous connection
2	Ebstein's malformation
1	Aortic regurgitation
1	Shone complex
25	**Complex CHD**
6	Transposition of the great arteries
5	Pulmonary atresia
4	Congenital corrected transposition of the great arteries
4	Double outlet right ventricle
3	Complex lesions/functionally univentricular heart
3	Tricuspid atresia

Diagnosis corresponding to complex, moderate and mild lesions [9] CHD (congenital heart disease)

All questions were read to the children by the investigator and each child had to choose one of

three given answers (1=never, 2=sometimes, 3=very often). None of the questions were explained to the child, if it was not able to answer a question, the question was ignored. From the answers a score was calculated from 0-100 with higher scores representing a better quality of life.

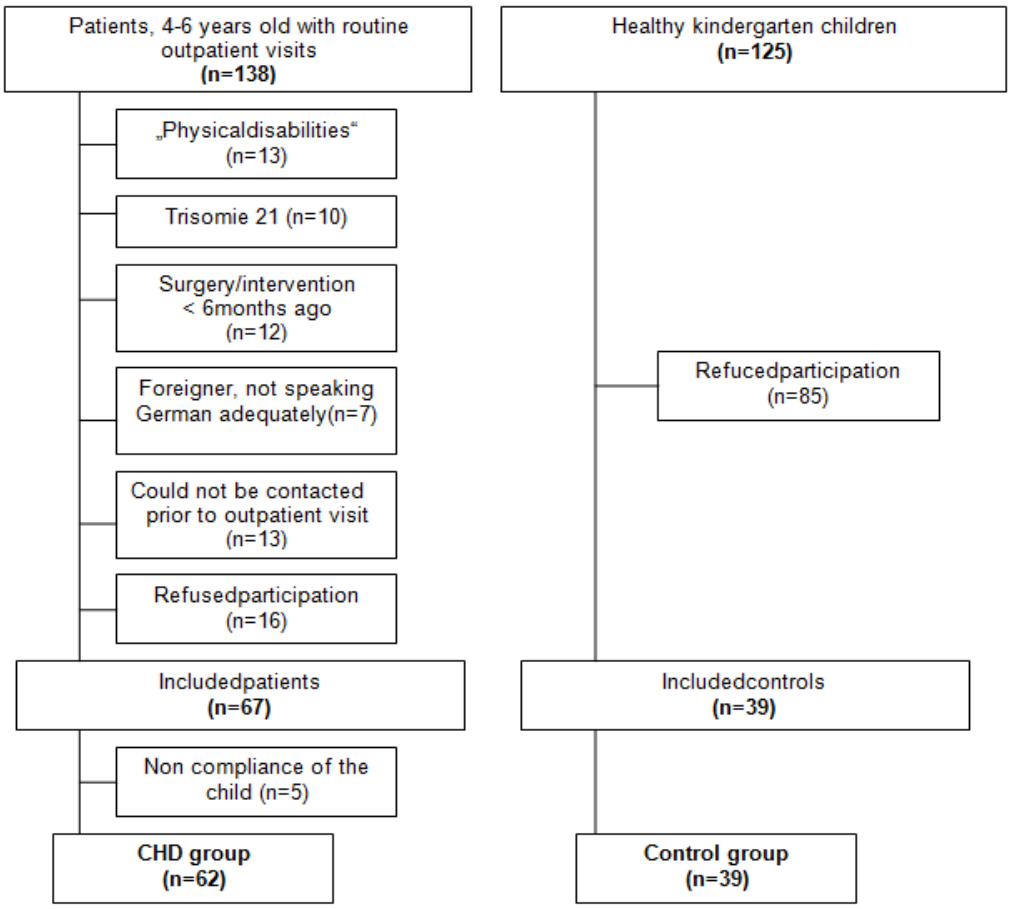

Fig. 1. Recruitment and participation of the children from our outpatient department and from three local kindergartens

CHD, congenital heart disease

Table 2. Anthropometric data and results of the tested parameters

	CHD	Control group	P*
	Median (Q1;Q3)	Median (Q1;Q3)	
Sex (male/female)	43/19	17/22	0.059
Age (years)	5.1 (4.5;5.9)	5.5 (4.8;6.2)	0.099
Body mass (kg)	18.0 (17.0;22.0)	21.0 (17.5;22.5)	0.129
Body length (cm)	110 (104;116)	118 (110;122)	**0.003**
Body mass index (kg/m²)	15.2 (14.2;16.7)	14.8 (13.9;15.5)	0.173
Motor quotient“MOT4-6”	104 (96;113)	111 (104;116)	**0.005**
Daily moderate activity (minutes per day)	123 (106;163)	109 (91;124)	0.060
Daily vigorous activity (minutes per day)	25 (16;42)	22 (16;32)	0.534
Daily moderate and vigorous activity	155 (124;190)	136 (112;163)	0.093
“Kiddy-KINDL®” questionnaire	75 (67;91)	79 (70;87)	0.744
“KINDL parents®” questionnaire	75 (70;80)	78 (73;82)	0.066

children with congenital heart disease (CHD) compared with the control group
(p values from a Wilcoxon rank sum test, except for sex: Chi² test was used)*

The parents (either mother or father) had to complete the parents' questionnaire without any help.

2.5 Statistical Analysis

All statistic calculations were performed with SPSS 19.0.0 (SPSS Inc., an IBM Company, Chicago, Illinois, USA). All investigated variables were skewed and normal distribution was rejected by a Shapiro Wilk test. Therefore, all measurements were depicted as median (1st quartile; 3rd quartile) and non-parametric tests were used. For the primary question, to describe the difference of the motor quotient for children with CHD in comparison with healthy children, the Wilcoxon rank sum test was performed. To compare activity and Kiddy-KINDL® questionnaire as secondary questions also Wilcoxon rank sum test was performed. With the Spearman rank correlation the relation of the motor quotient with sex, age, body mass index (BMI), activity and life quality was tested. Two-sided p-values <0.05 were considered significant.

3. RESULTS

Our study group did not differ significantly from the control group in respect to age, sex, body weight, and body mass index. Healthy children had a slightly higher body length (p=0.003, Table 2).

The motoric test could be performed in all children except in five patients who were all four years old and too much afraid of the test situation. These five children were excluded from the analysis. In the CHD group 27 children and in the control group 6 children refused to wear the accelerometer or had incomplete data sets. Six children with CHD were not able to answer the Kiddy-KINDL® questionnaire because of poor German or they didn't want to answer it. Among the parents only one was not able to answer the questions due to language barrier.

3.1 Motor Development

The median motor quotient of the CHD group was 104 (96;113) and significantly lower than in the control group with 111 (104;116) (p=0.005, Table 2). Only 6% of the CHD children showed motor quotient values below 85, but none of the healthy children.

This result was confirmed in an additional post-hoc stepwise multiple regression analysis. CHD was the most prominent factor for an deminished motor quotient, responsible for a loss of MQ of 5.6 (p=0.020). This was followed by the body mass index, responsible for a loss of 1,4 per every additional 1 kg/m² of BMI (p=0.029). After including those two factors, sex, age, body length, body mass did not reach significance in the model.

A post-hoc power analysis revealed, that a sample size of 32 subjects in each of the two groups would have been enough to detect the measured MQ difference of 6 with the measured standard deviations of 8.8 and 11.9 and a given p-value of 0.05 with a power of 90%.

3.2 Daily activity

Children with CHD trended to be slightly more active (155 [124; 190]) minutes of moderate and vigorous activity per day) than healthy children (136 [112; 163] minutes of moderate and vigorous activity per day, p=0.093). This was the result of a slightly increased time at moderate activity. However, both differences did not reach significance (Table 2).

3.3 Quality of Life

The assessment of life quality by the children showed no significant difference in children with CHD (75 [67; 92]) and healthy children (79 [71; 87] p=0.744). The parents estimated the life quality of their children similarly, (CHD group 75 [67; 92], control group 78 [74; 83] p=0.066).

3.4 Correlation to Motor Development

No correlation of the motor quotient to age, sex, daily activity or quality of life was found, neither in the CHD group nor in the control group (Table 3).

4. DISCUSSION

This study showed that kindergarten children with CHD have a slightly reduced motor ability in comparison with healthy children. These limitations could not be related to physical activity or quality of life.

During the last fifteen years, several studies have investigated the motor development of children with various congenital heart defects. Some of them analysed a mixed group of CHD [1,3,5], others investigated specific groups of CHD [6,7, 13-15]. Only two studies compared their results with healthy children of the same age group [1,5].

Table 3. Spearman correlation of the motor quotient with other tested parameters

	CHD	Controls
	r (p)	r (p)
Activity (min/day)	0.009 (0.959)	0.186 (0.308)
Body mass index (kg/m²)	-0.176 (0.175)	-0.084 (0.643)
"Kiddy-Kindl®" questionnaire	0.260 (0.053)	0.117 (0.479)
"KINDL parents®" questionnaire	0.067 (0.605)	-0.091 (0.580)

Furthermore, two studies examined very large age ranges between 5-14 years, whereas only one focussed on kindergarten children [8].

All studies found deficits in the motor development of children with congenital heart disease. Our findings confirm these results that there is a reduced motor ability in children with congenital heart diseases compared to a current control group. Nevertheless, in our group most of the children with congenital heart diseases had normal values in their motor development, when they are compared to the established reference values. This was also shown in our study on motor training, where most of the children with CHD also showed values within the reference range in their motor ability [16]. It was our control group that showed supranormal values. This might be a regional phenomenon. However, it cannot be ruled out that a selection bias of the control group was responsible for the supranormal values. In the patient group, far less subjects denied participation and such a bias is less likely.

To test the motor ability of the children some of the studies used the body coordination test for children (KTK) [1,3,5]. The "KTK" has only four gross motor skill tests and is recommended for children from 5 to 15 years. With its four tasks it is rather limited in the estimation of the complete motor development. In contrast we used the "MOT 4-6". It is a special motoric test for kindergarten children (4-6 years). With its 18 tasks it analyses many areas of motor development. The results of the motoric test were summed up to a single score. So we could not differentiate whether children had deficits in one area of motor development. But most children enjoyed the motoric test, maybe because it is so much diversified. Only five patients, all of them were four years old, were so afraid about the test situation that they denied participation.

Bjarnason-Wehrens [1] speculated that over-protection of the parents could be the main reason for motor deficits. They investigated school children and found motor limitations that were independent from the severity of the defect. These limitations could even be shown in patients with simple defects that were repaired with no or only mild residuals, which are very unlikely to cause any hemodynamic limitations. However, they did not investigate daily activity or parental education style itself to affirm their speculation. Our findings do not support this speculation. Our children with congenital heart disease were at least as active as healthy children. Children with CHD even seem to be more active than the healthy control group. This was surprising. McCrindle [17] as well as our group [18] could show in school children with Fontan circulation that they indeed have a reduced daily activity. However, in most previous studies on children with simpler congenital heart defects no daily activity was measured.

Honestly, some of the enhanced activity might also be explained by the fact that the CHD group had slightly more boys than the control group and boys are known to be physically more active than girls [18].

To our opinion the best explanation for the normal activity of children with congenital heart disease is a change in the attitude towards sport activities in these children during the last decade. Nowadays, the recommendation of the medical societies concerning leisure and even competitive sport participation became much more liberal [19]. For the last decade, in our institution physical activity has been promoted in patients with CHD, if no clear medical contraindications are present.

5. LIMITATIONS

Our control group showed supranormal values in the motor test. This might be a regional phenomenon. However, it cannot be ruled out that a selection bias of the control group was responsible for the supranormal values. But the small number of the control group, in contrast to the CHD-group could be a limitation. Unfortunately a substantial number of parents,

from the kindergarten children did not agree to the participation of their child. In the patient group, far less subjects denied participation and such a bias is less likely.

6. CONCLUSION

For the daily clinical work, it seems to be very important to focus more on the motor development during medical follow-up check-ups of the children. We agree with Hövels-Gürich [20] that at the age of 2 and 5 years a motoric test should be performed in all children with congenital heart disease. In addition, assessment of life quality should be implemented. At this age group such children showing limitations could be sent to a specialized motor training [16]. Once their condition improves, they can join regular sport classes at school. This avoids separation from their playmates, and helps to prevent consecutive social drawbacks. For those without deficits and no clear contraindications, physical activity of at least 60 minutes a day should be promoted, like for all children [19].

ETHICAL APPROVAL

The authors assert that all procedures contributing to this work comply with the ethical standards of the relevant national guidelines on human experimentation (Good clinical practice ISO 14155:2011) and with the Helsinki Declaration of 1975, as revised in 2013. The study has been approved by the institutional ethical board (ethical board of the medical faculty, Technische Universität München, project number 2782/10).

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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Peer-review history:
The peer review history for this paper can be accessed here:
http://sciencedomain.org/review-history/10292