

J. Stieh  
H. H. Kramer  
P. Harding  
G. Fischer

# Gross and Fine Motor Development is Impaired in Children with Cyanotic Congenital Heart Disease

Department of Pediatric Cardiology, Christian-Albrechts-University Kiel, Kiel, Germany

## Abstract

Many factors may intervene with the motor development of children with congenital heart disease (CHD). Children aged 5 to 14 years with various CHD were examined for disturbances of gross and fine motor development using motometric tests and compared with 30 healthy controls. The results of the Körperkoordinationstest für Kinder (KTK) (a body coordination test for children) for gross motor development were significantly lower in patients with uncorrected cyanotic CHD (motor quotient MQ  $74.8 \pm 11.7$ , mean  $\pm 1$  standard deviation,  $n = 16$ ) and after corrective surgery (MQ  $81.2 \pm 16.6$ ,  $n = 25$ ) than in controls (MQ  $102.8 \pm 11.8$ ,  $n = 30$ ). No relationship between these results and the cardiopulmonary exercise capacity was found. In patients with cyanotic CHD, significant deficits in fine motor development

were present before corrective surgery (e.g. Ziel-punktiertest [dotting] MQ  $87.7 \pm 9.9$  vs.  $106.5 \pm 10.8$ ), but already two years afterwards the results reached nearly normal values (MQ  $97.1 \pm 17.0$ ). In contrast, children with acyanotic CHD demonstrated normal gross and fine motor development. These results indicate that long-standing hypoxemia in infancy must be considered as an important cause of the pronounced motor disturbances. Early neurological evaluation of these children and a specialized motor physiotherapy are recommended.

**Key words:** Congenital heart disease – Cyanosis – Gross and fine motor development – Psychomotor development

## Abbreviations

ANOVA:	analysis of variance
ASD:	atrial septal defect
AVSD:	atrioventricular septal defect
bpm:	beats per minute
BW:	body weight
BWG:	Bland-White-Garland syndrome
$\chi^2$ -Test:	Chi-squared test
CoA:	coarctation of the aorta
commiss:	commissurotomy
F:	female
inf.:	infundibular
M:	male
MQ:	motoric quotient
MRI:	magnetic resonance imaging
MV:	mitral valve
PS:	pulmonary valve stenosis
SV:	single ventricle
TAP:	transannular patch
TCA:	tricuspid atresia
TGA:	complete transposition
L-TGA:	corrected transposition of the great arteries
TOF:	tetralogy of Fallot
VSD:	ventricular septal defect

## Introduction

Normally inherited movement patterns become differentiated through individual experiences with a wide variety of internal

and external influencing factors. Motorical competence is a major fundament for interactions of individuals with their environment giving sureness and self confidence (33, 44). Children with CHD may experience an impaired motor development due to limitations caused by the severity of the cardiac lesion (1, 35–37, 51) and externally imposed restrictions (33, 44). Also, an older age at operation (2, 48) or the effects of cardiopulmonary bypass (10, 12) have been reported to be of negative value. Only few studies about the nature and severity of motor disturbances in children with CHD are available (7, 15, 25, 26, 35, 36, 45).

The aim of the present study was to determine if a) children with hemodynamically significant acyanotic heart disease and b) children with cyanotic heart disease show disturbances of their gross and fine motor development before and after palliative or corrective cardiac surgery. In addition, a group of children with insignificant acyanotic heart disease was examined. All study groups were compared with an appropriate control group of healthy children.

## Patients and methods

### Study groups

102 children (53 M, 49 F), aged 5.0 to 14.11 years, with various forms of CHD (Table 1) underwent psychomotor tests and an evaluation of their physical working capacity. Children with other risk factors for an abnormal motor development were excluded (49); these were acquired cerebral lesions, auditory or visual problems, additional major malformations, syndromes and embryopathies. The patients were divided into 5 groups according to the results of cardiac examination and surgical status (Table 1); uncorrected cyanotic CHD (Group I) i.e. after palliative surgery; cyanotic CHD after corrective surgery (Group II); hemodynamically significant acyanotic CHD before (Group III) and after (Group IV) corrective surgery; hemodynamically insignif-

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Study group	Number of patients	Classification of CHD	Cardiac diagnosis	Type of surgery
Group I	(n = 16) (9 M, 7 F)	uncorrected cyanotic CHD (i. e. after palliative surgery)	TOF (n = 5) TCA (n = 5) D-TGA, PS (n = 1) TGA, SV (n = 5)	shunt shunt shunt shunt
Group II	(n = 25) (11 M, 14 F)	cyanotic CHD after corrective surgery	VSD, inf. PS (n = 1) TOF (n = 13) D-TGA (n = 5) D-TGA, VSD (n = 3) D-TGA, PS (n = 2) AVSD, PS (n = 1) VSD (n = 2)	patch closure, resection TAP atrial switch atrial switch atrial switch patch closure, commis. patch closure
Group III	(n = 4) (1 M, 3 F)	acyanotic CHD before corrective surgery	ASD II, VSD (n = 2)	patch closure
Group IV	(n = 32) (18 M, 14 F)	acyanotic CHD after corrective surgery	ASD I, II (n = 10) VSD (n = 6) VSD, CoA (n = 2) CoA (n = 7) PS, inf. PS (n = 4) AOS (n = 2) BWG (n = 1)	patch closure patch closure patch closure, resection resection commiss., resection artificial valve coronary transfer
Group V	(n = 25) (14 M, 11 F)	acyanotic CHD without hemodynamic significance	ASD I (n = 1) VSD (n = 18) PS (n = 4) AOS (n = 1) MV-prolaps (n = 1)	
Group VI	(n = 30) (14 M, 16 F)	innocent murmur		

**Table 1** Description of the study and control groups.

icant acyanotic CHD without requirements for corrective surgery (Group V). Children within the first two years after cardiac surgery were not included (31). The group of acyanotic children requiring surgery (n = 4) was too small and therefore excluded from statistics. All examinations were performed by the first author, who was unaware of all cardiological findings.

### Control group

Thirty children (14 M, 16 F) with an innocent heart murmur and without significant differences for age, sex or socioeconomic status were evaluated on the same psychomotor test battery.

### Psychomotor tests

All children had a fully standardized assessment of psychomotor performance. Previously published motometric tests specifically designed to assess gross and fine motor coordination were used (5, 38, 42, 43, 49, 50). According to the type of the applied test, the regularity of movements was scored by recording the number of correct movements per time, the number of mistakes in precision and the time needed for each task. These values were transformed into age- and sex-independent standard values (motor quotient, MQ) (38, 42, 49).

### Gross motor coordination

The gross motor coordination was examined with the Körperkoordinationstest für Kinder (KTK) for children aged 5.0 to 14.11 years which consists of two low-speed (balancing backwards [B], monopedal skipping [S]) and two high-speed components (sideward jumping [J], sideward moving on boards [M]) (40, 43).

### Fine motor coordination

Three tests of fine motor development were carried out for both, the dominant and non-dominant hand: Zielpunktieretest (dotting) (ZP) (38, 50) and Kamelnachfahrttest (camel drawing test) (KA)

(5, 38) examine speed and accuracy of optomotor and fine motor performance (Figs. 1a and b). Tapping (TA) (50) allows evaluation of speed and dysrhythmia of finger movements.

All these tests were previously standardized for children from 7.0 to 12.11 years of age (own results, not published). A moderate disturbance of coordination was defined as a MQ  $\leq 85$  and a severe impairment as a MQ  $\leq 70$  for each of the motometric tests employed.

### Physical working capacity

Cardiopulmonary fitness defined as the short-term maximal working and endurance capacity (13, 16, 19, 22, 30) was investigated by submaximal exercise testing  $W_{170}$  (physical working capacity) (8, 28). Children with  $W_{170}$  values ( $W_{170}/\text{kg BW}$ ) below two standard deviations of normal values (19) were excluded from the gross motor tests because their results might have been influenced by limited cardiopulmonary fitness.

### Hypotheses

The study intended to evaluate the following hypotheses by statistical analysis:

- 1) Children with uncorrected cyanotic CHD (Group I) and acyanotic CHD (Group III) will show more gross motor disturbances than children of the control group (Group VI).
- 2) Children with corrected cyanotic CHD (Group II) and corrected acyanotic CHD (Group IV) do not show more gross motor disturbances than children of the control group (Group VI).
- 3) Children with hemodynamically insignificant acyanotic CHD (Group V) do not show gross or fine motor disturbances.
- 4) Children with cyanotic and acyanotic CHD do not show fine motor disturbances, neither before nor after corrective surgery.

### Statistical analysis

The mean values of the four study groups were compared with those of the control group by one-way ANOVA. In case of

Name:                      geb.:                      Datum:



Fig. 1a

Anzahl:              Zeit:              Fehler:

Name:                      geb.:                      Datum:

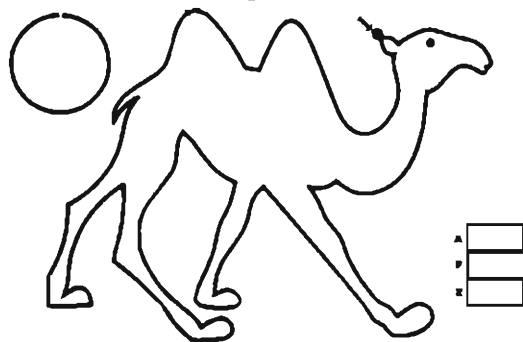


Fig. 1b

Fig. 1a and b Forms for fine motor tests: a) Zielpunktieretest (dotting), b) Kamelnachfahrtest (camel drawing test).

significance, *Student's t-test* for unpaired data was added. An  $\alpha$ -adjustment ( $\alpha^* = 0.0125$ ) was used for comparisons of the 4 study groups with the control group (21). For additional comparisons, the  $\alpha$ -value was not adjusted. Independence of the variables from each other (for example KTK from  $W_{170}$ ) was determined by the *Pearson's correlation coefficient* ( $r < 0.4$ ). Chi-square tests were performed to reveal differences of age, sex and socioeconomical status between study and control groups. Unless otherwise indicated, results are expressed as mean  $\pm$  standard deviation.

**Results**

The study groups showed no statistical differences to the control group with respect to age, sex or socioeconomical status ( $p >$

0.10) (Table 1). In the cyanotic CHD group, palliative and corrective surgery was carried out at an age of  $14 \pm 6$  weeks and  $22 \pm 7$  months, respectively. After palliative surgery, the arterial oxygen saturation was  $83 \pm 5\%$ . Children with acyanotic CHD underwent surgery with  $19 \pm 10$  months.

**Physical working capacity ( $W_{170}$ )**

The results of the physical working capacity are shown in Table 2. No significant differences were found between the study groups and the control group ( $2.15 \pm 0.47$  W/kg BW). However, the cyanotic children after palliative surgery (Group I) had the lowest values ( $1.8 \pm 0.48$  W/kg BW). After surgical repair, the children with cyanotic CHD (Group II) improved their physical working capacity ( $2.00 \pm 0.48$  W/kg BW), but their results remained below those of children with acyanotic CHD after corrective surgery (Group IV;  $2.21 \pm 0.57$  W/kg BW) or without need for surgery (Group V;  $2.27 \pm 0.53$  W/kg BW). The working capacity of three children from Group I and one child each from Groups II and VI was lower than 2 SD level of normal, so that they were excluded from gross motor tests.

**Gross motor coordination (KTK)**

**Cyanotic heart disease**

Patients with cyanotic CHD showed gross motor disturbances indicating a severe developmental delay after palliative (Group I,  $MQ = 74.8 \pm 11.7$ ) and also after corrective surgery (Group II,  $MQ = 81.2 \pm 16.6$ ). These results were significantly lower than the values of the control (Group VI) ( $MQ = 102.8 \pm 11.8$ ) and were not influenced by the physical working capacity (Group I  $r = 0.19$ ; Group II  $r = 0.08$ ) (Fig. 2). When the pre- and post-operative results were compared, only the subtest "sideward jumping" revealed a significantly better result ( $79.9 \pm 7.6$  vs  $91.3 \pm 17.7$ ;  $p < 0.01$ ), while the subtests "monopodal skipping" and "sideward moving on boards" showed little and the "balancing backwards" no improvement. However, the percentage of patients with cyanotic CHD with moderate ( $MQ \leq 85$ ) and severe ( $MQ \leq 70$ ) motor disturbances decreased after corrective surgery from 39 to 21%, and from 46 to 33%, respectively, while the percentage of children with no impairment increased from 15 to 46%. Children who were younger than 24 months at the time of corrective surgery had better values ( $MQ = 89.2 \pm 13.3$ ) than those who were operated later ( $MQ 75.6 \pm 15.3$ ) (Fig. 3).

**Acyanotic heart disease**

Gross motor development was slightly impaired in children with acyanotic CHD before corrective surgery (Group III,  $MQ = 91.3 \pm 12.9$ ), but normalized completely afterwards (Group IV,  $MQ = 101.1 \pm 16.5$ ). The motor skills of cardiac patients without need for a surgical intervention (Group V,  $MQ = 103.3 \pm 13.6$ ) did not differ from those of healthy children (Group VI) (Fig. 2).

**Fine motor coordination**

Children with uncorrected cyanotic heart disease (Group I) showed significant fine motor deficits compared with healthy children, pointed out by dotting (ZP- $MQ = 87.7 \pm 9.9$  vs.  $106.5 \pm 10.8$ ) and the camel drawing test (KA- $MQ = 91.9 \pm 23.3$  vs.  $117.4 \pm 20.4$ ). Tapping revealed distinct, but not significant differences between these groups (TA- $MQ = 91.3 \pm 13.3$  vs.  $101.5 \pm 14.9$ ). After corrective surgery (Group II), the fine

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motor development remained below the range of healthy children (ZP-MQ  $97.2 \pm 17.0$ , KA-MQ  $108.8 \pm 22.3$ , TA-MQ  $91.7 \pm 15.1$ , all differences not significant vs. the control group) because only the speed but not the accuracy of manual skills improved

(Table 2). Patients with acyanotic CHD had a normal fine motor development.

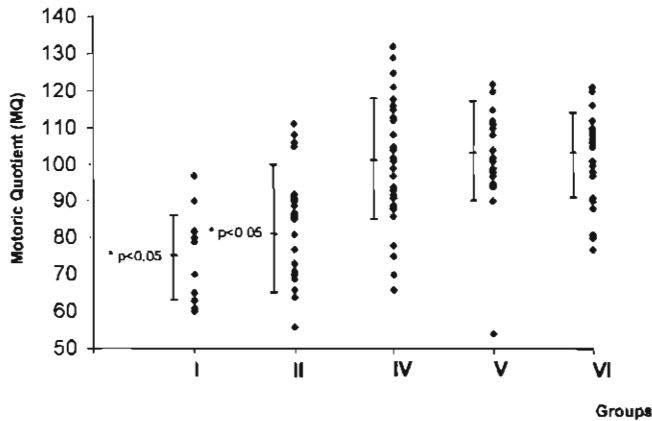
**Discussion**

We compared the gross and fine motor development of patients with congenital heart disease before and after corrective surgery with healthy children.

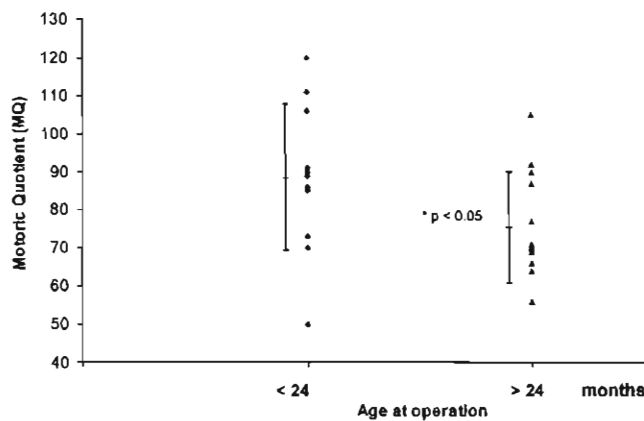
Gross and fine motor disabilities indicating a severe developmental delay were observed in children with cyanotic CHD before corrective surgery. Gross motor deficiencies remained overt even 2 years after corrective surgery. Children with hemodynamically significant acyanotic CHD seemed to have slight disturbances of their gross motor coordination before, but not after corrective surgery.

Several authors (3, 15, 25, 26, 36, 46) reported a significant delay of fine and gross motor development in cyanotic children before corrective surgery. However, in contrast to our results, they found no significant postoperative disturbances. Other authors reported either slightly worse (7), equally good (45) or even better (48) gross motor results for cyanotic patients in comparison to healthy children. These conflicting results on motor development are probably explained by different methods of evaluation, for example, no control group (3, 46), parent questionnaires instead of development tests (10, 45), no exclusion of children with syndromes or other neurological risk factors (7, 36) and also the choice of the applied test materials. While almost all previous studies employed general development tests including intellectual and motor test batteries, we used motospecific motometric tests which show a higher sensitivity and specificity for motor components (38, 40, 41, 42, 52) than the test batteries used in previous studies (7, 15, 25, 26, 36, 46, 48).

In the present study, children with obvious mental retardation and other risk factors than CHD for an abnormal neurological development (such as perinatally acquired cerebral lesions, auditory and visual problems etc.) were excluded from examination. However, unrecognized, non-overt mental and cognitive disabilities may have influenced their motor development. Previous psychomotor studies, especially those which were conducted before early cardiac surgery became available, reported a delay of cognitive, mental and motor development with special regard to cyanotic CHD (15, 25, 26, 46). As a result of an own previous



**Fig. 2** Results of the Körperkoordinationstest (KTK). Results of the t-test of the comparisons of Group I and II with the control group (Group VI) are statistically significant.



**Fig. 3** Relationship between age at operation and the Motoric Quotient (MQ) of children with cyanotic congenital heart disease. Result of the t-test of the comparison of children operated before and after 24 months of life is statistically significant.

**Table 2** Age, sex and socioeconomical status of the children in the control and study groups. Test results of physical working capacity ( $W_{170}$ ) and fine motor coordination.

	Group I	Group II	Group IV	Group V	Group VI (control)	Statistics
Age (years)	9.07 ± 1.08	9.07 ± 1.06	9.06 ± 1.08	10.02 ± 1.09	9.07 ± 1.06	p = 0.89 (ANOVA)
Sex	9 M, 7 F	11 M, 14 F	18 M, 14 F	14 M, 11 F	14 M, 16 F	p = 0.69 ( $\chi^2$ -test)
Social class*						
upper class (1-3)	3	6	4	3	1	p = 0.71 ( $\chi^2$ -Test)
middle class (4-7)	11	16	27	21	26	
lower class (8-10)	2	3	1	1	1	
$W_{170}/kg\ BW^{**}$	1.80 ± 0.48	2.00 ± 0.48	2.21 ± 0.57	2.27 ± 0.53	2.15 ± 0.47	p = 0.1 (ANOVA)
Dotting	87.7 ± 9.9	97.1 ± 17.0	102.6 ± 13.0	105.0 ± 12.0	106.5 ± 10.8	p < 0.01 (ANOVA) t-test I, II vs. VI
Camel drawing test	91.9 ± 23.3	108.8 ± 22.3	114.0 ± 25.9	118.6 ± 16.0	117.4 ± 20.4	p < 0.01 p = 0.03 (ANOVA) t-test I vs. VI, p > 0.01
Tapping	91.3 ± 13.3	91.7 ± 15.1	97.2 ± 16.0	94.8 ± 16.5	101.5 ± 14.9	p = 0.34 (ANOVA)

\* according to (15)

\*\*  $W_{170}/kg\ BW$  = Watt per kilogram bodyweight at a heart rate of 170 bpm

study (20), the intellectual development of the children with CHD, especially when physically fit, is not as much impaired that this may disturb their motor development. Although we have no data of intellectual development of the patients in the present study group, we are quite confident that mental retardation does not significantly contribute to the observed pathological findings.

The substantial fine and gross motor deficits of cyanotic children before corrective surgery are characterized by low speed as well as poor accuracy of performance. The speed improved after operation while the accuracy did not. Children of the small test group with acyanotic CHD requiring corrective surgery showed limitations of speed components, which were not present in the group tested after cardiac surgery.

The observation that motor performance was still abnormal even 2 years after corrective surgery suggests a significant functional damage of the nervous system. Since this impairment was only observed in cyanotic children, their chronic hypoxemia or the resulting polycythemia and increased blood viscosity seem to be responsible.

The duration of hypoxemia may be also important, as children with corrective surgery before two years of age showed a better development than patients who underwent surgery at an older age and did not improve their gross motor abilities. These observations are in accordance to those of O'Dougherty et al (35, 36) and Newburger et al (34) who studied the effects of prolonged hypoxemia in patients with complete transposition: they found a more impaired motor function and lower academic achievement when atrial rerouting had been carried out after the age of 2 years, as well as a progressive impairment of cognitive function when surgery was delayed from infancy up to more than 4 years. Aisenberg et al (3) also described a particular susceptibility of psychomotor development to chronic hypoxemia. Surprisingly, Stevenson et al (48) and Blackwood et al (7) found no significant delay of psychomotor development of children with severe, mostly cyanotic CHD before and after cardiac surgery.

Except for these previous examinations in children with cyanotic CHD, no reports about the sequelae of chronic hypoxemia in childhood have been published; in contrast, there are numerous studies about the neurodevelopmental outcome of acute hypoxic events especially in the perinatal period (24, 39, 47). Several investigators used neuroimaging techniques to determine the targeting regions of brain injury after acute hypoxia (4, 6). They report a special susceptibility of the primary sensory and fore-brain motor systems, e.g. the basal ganglia and the ventral thalamus, which function in sensorimotor integration and movement control. Neuronal damage of these regions has also been shown by experimental studies in neonatal piglets (23). Cerebral ultrasound studies (17) obtained early after cardiac surgery in infants with CHD showed a higher incidence of abnormalities (e.g. cerebral atrophy and linear echodensities in the basal ganglia and thalamus) than control infants (59% vs. 14%). Up to now, only two small cerebral magnetic resonance imaging (MRI) studies of infants and children with CHD have been published (27, 29). The first one compared the MRI results obtained before and immediately after cardiac surgery: preoperatively, 5 of 15 of the patients (all but one with cyanotic CHD) had cerebral atrophy with dilatation of the subarachnoid spaces and ventriculomegaly, while the postoperative images showed an increase especially in the bicaudate and third ventricular diameters. The second study reported pathological MRI findings in 17 of 23 patients at a mean age of 66 months after open heart surgery in infancy. Interestingly, 8 of 9 patients with diffuse changes of the grey and white

matter consistent with hypoxic-ischemic encephalopathy showed pathological neurodevelopmental findings (cerebral palsy and mental retardation), but only one of the six children with focal cortical infarctions.

The improved motor performance of patients with cyanotic as well as acyanotic CHD after corrective surgery is also explained by a better motor experience, which is in part related to the increase of the physical working capacity. The improved hemodynamic situation and the better tissue oxygen delivery after relief of cyanosis contribute to a better physical fitness and a diminished exhaustibility shown by the shifted anaerobic threshold (3, 9, 11, 14, 32). It is this better physical condition which facilitates training of coordinative skills and attainment of safety even for difficult motor requests.

An appropriate offer of perceptive and movement stimuli is essential for an adequate high-grade differentiation of movement patterns. However, parents may tend to overprotection because they fear overstrain of their sick children; in turn, this often reduces their activity level. Some physicians may advise a reduction of physical activity and, thereby further limit the field of possible experience. In addition, the increased feelings of anxiety found in cyanotic children (20) can impede the pleasure of physical experimentation. In this way, the children learn only simplified, goal-directed movement patterns, so-called motor adaptations (32, 43) and will fail in situations with rapidly changing motor requirements (e.g. ball games, climbing etc.). Many of these motor adaptations persist even after corrective surgery and may explain the observed lack of improvement in movement quality.

The experience of physical disability has important consequences for children with CHD. These are not only the exclusion from games with other children and a different recreational behavior, but also drawbacks from educational and other social factors which may negatively affect the development of personality and identity (20). Abnormalities of behavior and learning disorders may also be attributed to this field of psychomotor deficiencies (10, 25, 36).

Corrective surgery of severe congenital heart disease should be carried out at the earliest possible time. In addition, follow-up after surgery should include not only cardiological aspects but also examinations of neurological development. In order to detect motor disturbances, motoscopic and motometric tests will provide valuable clues. As motor development represents an important aspect of general development, patients even with severe CHD should be trained in motor skills.

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J. Stieh, M. D.

Department of Pediatric Cardiology  
Christian-Albrecht-University Kiel  
Schwanenweg 20  
D-24105 Kiel  
Germany