Motor Performance in Children with Generalized Hypermobility: The Influence of Muscle Strength and Exercise Capacity

Yvonne B. Hanewinkel-van Kleef, PT, MSc, PCS, Paul J.M. Helders, PT, MSc, PhD, PCS, Tim Takken, MSc, PhD, and Raoul H. Engelbert, PT, PhD, PCS

Practice of Pediatric Physical Therapy (Y.B.H.-v.K.), Drachten, the Netherlands; Department of Pediatric Physical Therapy and Exercise Physiology (P.J.M.H., T.T.), Wilhelmina Children's Hospital, University Medical Center, Utrecht, the Netherlands; and University of Applied Sciences (R.H.E.), Amsterdam School of Health Professions, Amsterdam, the Netherlands

Purpose: The aim of this study was to investigate whether muscle strength and functional exercise capacity (FEC) influence motor performance in children with generalized joint hypermobility. **Methods:** Forty-one children (mean age: 8.1 years) with symptomatic generalized hypermobility were included. Motor performance was assessed using the Körperkoordinationstest für Kinder (KTK) and the Movement Assessment Battery for Children. Muscle strength and FEC were measured with a handheld dynamometer and the 6-minute walk test. **Results:** Only muscle strength was significantly positively associated with motor performance on the KTK. FEC was significantly decreased. Children's scores on the KTK were significantly lower ($p <$ 0.001) compared with scores on the Movement Assessment Battery for Children. **Conclusions:** The KTK is a more sensitive tool for detecting motor problems in children with generalized joint hypermobility, but is not associated with FEC. Along with the KTK, the 6-minute walk test can be used to independently assess and evaluate FEC. **(Pediatr Phys Ther 2009;21:194–200)** *Key words: child, joint instability/diagnosis, motor performance, movement disorders/diagnosis, muscle strength, physical exertion/physiology, predictive value of tests, range of motion/articular physiology, validity*

INTRODUCTION

Joint hypermobility or joint laxity is the result of ligamentous laxity and may be a feature of a heritable disorder of connective tissue. Hypermobile joints are those displaying a range of motion that is considered excessive, taking into consideration age, gender and ethnic background. If several joints are involved, it is regarded as generalized hypermobility, otherwise as pauciarticular hypermobility (1 to 4 joints).[1–3] Joint hypermobility declines with age, is greater in females than in males, and varies between ethnic groups.[1,2] In the white population, the prevalence is 10%, and in African and Asian populations, it is 25%.[4,5] In Dutch children aged 4 to 17 years, 14% generalized joint hypermobility is reported.[6] Generalized hypermobility can be measured using several diagnostic criteria. Carter and Wilkinson[7] proposed the first rating scale, which was modified by Beighton et al.[4] The Beighton scoring system is the most commonly used today. An alternative scale is the Bulbena scoring system. This scale includes a different cutoff for hypermobility for males and females and assesses more joints, especially in the lower extremities.[8] Although the Beighton score and the Bulbena score have only been validated for adults, in general practice it is used for children beginning at the age of 6 years[9] (Table 1).

If hypermobility is combined with musculoskeletal complaints, such as arthralgia in more than 2 joints for a period exceeding 12 weeks or exercise-induced pain and

0898-5669/109/2102-0194
Pediatric Physical Therapy

Address correspondence to: *Yvonne B. Hanewinkel-van Kleef, PT, MSc, PCS, Morra 83, 9204 KV Drachten, the Netherlands. E-mail: yhanewinkel@chello.nl*
This study was in partial fulfillment of Yvonne B. Hanewinkel-van Kleef's MSc degree in Physical therapy at Utrecht University, the Netherlands.

DOI: 10.1097/PEP.0b013e3181a3ac5f

TABLE 1
Beigthon and Bulbena Criteria for the Clinical Assessment of Generalized Joint Hypermobility[4,8]

Criteria	Beighton Score	Bulbena Score
Upper extremities		
Passive exorotation of the shoulder >85°		1
Passive hyperextension of the elbow ≥10°	1*	1
Passive apposition of the thumb to the flexor aspect of the forearm	1*	1 at <21 mm
Passive dorsiflexion of the fifth metacarpophalangeal joint ≥90°	1*	1
Lower extremities		
Passive hip abduction both legs >85°		1
Knee flexion allows the heel to make contact with the buttock		1
Passive hyperextension of the knee ≥10°	1*	
Passive shift of the patella to the lateral side of the tibia		1
Passive dorsiflexion of the ankle >20°		1
Passive dorsiflexion of the first metatarsophalangeal joint >10°		1
Spine		
Active lumbar flexion, putting both hands flat on the floor without bending the knees	1	
Skin		
Appearance of ecchymoses after hardly noticed, minimal trauma		1
Total score	**9**	**10**

*Each side one point.
Cutoff according to: Beighton criteria, ≥5; Bulbena criteria, male ≥4, female ≥5.

intolerance, it is defined as benign joint hypermobility syndrome. It is called benign to make a distinction from connective tissue disorders with life-threatening complications such as Marfan syndrome, Ehlers-Danlos syndrome, or Osteogenese Imperfecta. Not all persons with joint hypermobility experience complaints, some even benefit from their increased range of motion like ballet dancers, gymnasts, and musicians (violinists and pianists).[10,11]

In case of symptoms, they may occur at any age. As early as birth, congenital (benign) hypotonia and muscle weakness are associated with joint hypermobility.[7,12] This is sometimes followed by delayed motor development, which may be temporary for a number of children.[13–15] Decreased muscle strength and functional exercise capacity are often reported to be persistent.[7,9,12,16,17] From the age of 3 years, children may suffer from benign nocturnal leg pain, so-called growing pains (until 6 years), and musculoskeletal pain in the legs, ankles, and feet. Recurrent knee pain and ankle distorsion frequently appear at school age, followed by pain in the hips and back at approximately the age of 10 years.[9,12,18] Decreased muscle strength, functional exercise capacity, and delayed motor development limits the child in daily physical activities. Mostly motor skills such as balancing, walking, running, hopping, jumping, fast turning of direction, and sport activities are impaired.[9,15,19]

Several motor tests are available to assess children's motor performance. Because disease-specific instruments for the measurement of motor performance are lacking, pediatric physiotherapists have to use generic tests. It is not clear if those generic tests can measure the specific motor problems of children with generalized joint hypermobility and if they do so, which test is the most sensitive for this group of children. It is important for the quality of pediatric physical treatment to use the correct motor test to assess motor problems and to evaluate the results of treatment.

The first aim of this study was to investigate whether muscle strength and functional exercise capacity are predictors of motor performance as assessed and objectified by the Körperkoordinationstest für Kinder (KTK) and the Movement Assessment Battery for Children (M-ABC) in children with generalized joint hypermobility. The second aim was to compare the KTK and M-ABC in their sensitivity for detecting motor problems in children with generalized joint hypermobility.

The first hypothesis was that in children with symptomatic generalized joint hypermobility, muscle strength and functional exercise capacity are predictors of motor performance according to the KTK and the M-ABC. The second hypothesis was that the KTK and M-ABC do differ in the measurement of motor performance in children with symptomatic generalized joint hypermobility.

METHODS

Design

The study was performed in 2 practices of pediatric physical therapy by 2 experienced pediatric physical therapists (Y.B.M.H. and C.J.M.H.). The children were tested during February and March 2007. The parents were provided an informational letter about the purpose of the study. They indicated their consent for the use of the data in this study. Because patients were referred and treated according to "care as usual," no approval of the Medical Ethics Committee was needed.

Subjects

Forty-one children with symptomatic generalized hypermobility with a mean age of 8.1 years (range, 5–12 years) were included. The children were included in the study when generalized hypermobility was present according to the criteria of Bulbena (Table 1) and when they reported problems with motor performance. Children referred for the first time and children who came at long intervals for advice or a check-up were included in this study. Children with known neuromuscular diseases, cerebral palsy, a recent fracture or joint sprain in the lower extremity, and known collagen disorders were excluded.

Measurements

A large body mass index (BMI) will possibly interfere with gross motor performance. Therefore, it was determined to use BMI in the statistical analysis as a possible confounding variable. Body height and weight were determined using a wall assembled stadiometer and an electronic scale and rounded to the nearest centimeter and

100 g, respectively. During measurement, the children wore sport clothing and no shoes. BMI was calculated as body mass/height.[2]

Generalized joint hypermobility was diagnosed according to the criteria of Bulbena et al[8] (Table 1). Passive movements of 9 joints were examined. The presence and location of hypermobility was scored and the presence of ecchymoses (bruising after minimal trauma) was recorded. Generalized hypermobility of joints was considered to be present if the score was ≥5 in girls and ≥4 in boys. The Bulbena score has high concurrent validity (>0.85) with the Beighton score and high test-retest reliability (κappa >0.8).[8] The internal consistency of the items of the Bulbena score is more homogeneous (Cronbach's $\alpha = 0.933$) compared with the items of the Beighton score (Cronbach's $\alpha = 0.836$).[8] An interrater reliability study between the 2 testers was performed comparing a group of 25 children. The children were tested by both testers on the same day. The testers were blinded to each others test results. The interrater reliability was found to be high (κappa = 0.91).

Muscle strength of the shoulder abductors, the 3-point grip of the hand and the hip flexors was measured with a handheld dynamometer, type CT 3001 (C.I.T. Technics BV Haren/Groningen, the Netherlands). Measurements were performed 3 times consecutively and the highest value for each muscle group was registered. Those values were converted into z scores (standardized score; normal between −2 and 2)[20] according to age- and gender-related reference values of Beenakker et al.[21] The total z score of muscle strength was the summation of the 3 separate z scores. An interrater reliability study between the 2 testers was performed, comparing a group of 22 children on the same day. The testers were blinded to each other's test results. The highest score of the 3 attempts of each muscle was recorded to determine the interrater reliability. According to the limits of agreement (LOA), the interrater reliability was found to be acceptable (shoulder abductors 12% LOA, 3-point grip 12.5% LOA, and hip flexors 10% LOA).[20]

Functional exercise capacity was measured using the 6-minute walk test (6MWT). This is a self-paced endurance test. Because children pace themselves during their daily physical activities, it may be a better representation of the patterns of physical activity in children than traditional maximal exercise tests, as children rarely engage in sustained, heavy exercise.[22] The 6MWT is a well standardized, simple, safe test, and easy to perform. The test has a physiologic response at a submaximal, intense level of exercise.[23–26] The test was performed on an 8-m track.[27] Patients were instructed to cover the largest possible distance in 6 minutes at a self-chosen walking speed. Turns were made on both ends (marked with a line) of the 8-m track. Each 8-m distance walked was recorded. At the end of the test, the child was asked to stop and stand still. The distance covered in the final lap was measured with a measuring tape. The child was encouraged in a neutral way, by telling him when he had walked 10 laps and when a minute had passed. According to the age- and gender-related reference values of Geiger et al,[25] the distance walked was converted into a z score.

Motor performance was measured with the M-ABC and with the KTK. These tests were performed on different days with a maximum interval of 14 days. The order of testing was counterbalanced within the group.

The M-ABC is a norm-referenced motor test for children aged 4 to 12. It is a generic instrument used to identify children with impaired motor ability in daily life and contains 3 items for manual dexterity, 2 for ball skills, and 3 for static and dynamic balance.[28,29] (Table 2). The total score of the items is converted into a percentile score. A score less than the fifth percentile (P5) score indicates a definite motor delay; a score less than the fifteenth percentile (P15) indicates a motor performance that is at risk. The interrater reliability is good (0.70 to 0.90).[28,29]

The KTK was designed to identify and diagnose problems of motor development in children.[30] It differs from the M-ABC in that it focuses exclusively on gross motor coordination,[31] with more emphasis on muscle strength and endurance. The KTK is a norm-referenced motor test for children 5 to 14 years old. This test consists of 4 items that have to be performed 2 or 3 times. Item 2 and 3 have different raw scores for boys and girls (Table 3). All attempts of each item are recorded and added up to a total item score. This is converted into an item motor quotient. By adding up the 4 item motor quotients, the total motor quotient is calculated (mean 100, SD 15) and can be converted into a percentile score. A child having a percentile score less than the third percentile (P3) is considered to have a motor delay, whereas a motor performance with a

TABLE 2

Movement Assessment Battery for Children[28]

Manual dexterity
Speed task for both hands separately
Bimanual coordination
Eye-hand coordination with preferred hand (drawing)
Ball skills
Catching moving object
Aiming at goal
Balance
Static balance (stance)
Dynamic balance while moving fast (jumping, hopping)*†
Dynamic balance while moving slowly*

*Third attempt permitted if the norm score is not reached at the second attempt.

†Jumping or hopping depends on age.

TABLE 3

Körperkoordinationstest für Kinder[29]

	Number of Times
Balancing backwards along 3 balance beams (6, 4.5, 3 cm)	3 times on every beam
Hopping over a pile of foam (increasing height)*	Till maximum is reached
Jumping sideways*	2 times
Moving sideways on boxes	2 times

*Different norms for boys and girls.

TABLE 4
Clinical Characteristics of the Study Population (n = 41)

Clinical Features	Number (%)	Mean (SD)	Range
Girls	19 (46)		
Boys	22 (54)		
Bulbena score P50 (P25–P75)	6 (6–7)		
Bulbena score LE P50 (P25–P75)	3 (3–4)		
Age (yrs)		8.1 (1.9)	5 to 11.7
Length (m)		1.36 (0.15)	1.07 to 1.73
Weight (kg)		30.5 (7.6)	18.1 to 52.8
BMI (kg/m^2)		16.4 (1.8)	13.4 to 21.2
z score total muscle strength P50 (P25–P75)		−0.57 (−0.24 to −1.37)	
z score total muscle strength		−0.66 (0.80)	−2.18 to 1.23
z score 6MWT P50 (P25–P75)		−2.39 (−1.61 to −3.15)	
z score 6MWT		−2.47 (0.92)	−1.12 to −4.69
Total percentile score M-ABC		40.5 (29.8)	3 to 96
Total percentile score KTK		9.2 (12.9)	0 to 60

SD indicates standard deviation; P50, median; P25, upper quartile; P75, lower quartile; P, percentile; LE, lower extremities; 6MWT, 6-minute walk test; M-ABC, Movement Assessment Battery for Children; KTK, Körper koordinationstest fur̈ Kinder.

TABLE 5
Associations with Corresponding 95% Confidence Interval (CI) of the M-ABC, KTK, Muscle Strength, BMI, and Functional Exercise Capacity

Dependent Variable	Independent Variable	Beta	95% CI
M-ABC	*z* score total muscle strength	−0.19	−0.71 to 0.22
	BMI	0.24	−0.06 to 0.35
	z score 6-minute walk test	−0.27	−0.42 to 0.36
KTK	*z* score total muscle strength	0.43*	0.09 to 0.69
	BMI	−0.33*	−0.27 to −0.01
	z score 6-minute walk test	−0.07	−0.31 to 0.19

*Statistically significant associations were considered reached when 95% CIs did not include the null value.

M-ABC indicates Movement Assessment Battery for Children; KTK, Körper koordinationstest fur̈ Kinder.

score less than the sixteenth percentile (P16) is regarded as being at risk. The test-retest reliability of the different items and the total score is high: balancing backwards 0.80, hopping 0.96, jumping sideways 0.95, moving sideways on boxes 0.94, and for the total score of the KTK 0.97.[30] The relationship between the 3 subsections of the M-ABC and the 4 component items of the KTK is $r = 0.62$ ($p < 0.001$).[31]

Statistical Analysis

Frequencies, means and standard deviations (SD) were calculated of all variables. The data for the Bulbena score, the *z* scores of the muscle strength and 6MWT, the percentile score of the M-ABC and the KTK were presented as medians (50th percentile [P50]) and interquartile ranges (P25–P75). Normal distribution of the data was explored by the Komolgorov Smirnov test. The percentile scores of both the M-ABC and KTK were not normally distributed. Therefore, a nonparametric Wilcoxon's sign rank test was used for comparing both percentile scores. Chi square tests were used to compare the cutoff of both motor tests for normal, at risk, and delayed motor development.

Multiple regression analysis was used to study the association between motor performance as a dependent variable and total muscle strength, BMI and functional exercise capacity as predicting variables. This was done for the performance on each motor test. To reach a normal distribution of the percentile score on both motor tests, the data were transformed (squared root). Associations were expressed as linear regression coefficients with their corresponding 95% confidence interval (95% CI). Data were analyzed using SPSS, version 15.0 (SPSS Inc., Chicago, Ill).

To attend to the power of this study, the study population (n = 41) was based on the principle of a minimum of 10 children per independent variable, as recommended for multiple regression analysis.[32]

RESULTS

Descriptive data of all children are presented in Table 4. The Bulbena scores varied from 5 to 9, with a median score of 6 and interquartile scores of 6 (P25) and 7 (P75). The mean *z* score of total muscle strength was −0.66 (SD 0.80). The mean *z* score of the 6MWT was −2.47 (SD 0.92). Assessment of motor performance with the M-ABC showed a mean score above P15 (40.5; SD 29.8), whereas the mean score on the KTK was below P15 (9.2; SD 12.9). The children scored significantly lower on the KTK (Wilcoxon's $z = -4.99$; $p = 0.00$). The clinical relevance of the difference between both scores had a high effect size ($r = 0.78$).

No significant association was found between the M-ABC and KTK for the cutoff at P15 ($\chi^2 = 3.24$; $p = 0.072$) and at P5 ($\chi^2 = 0.211$; $p = 0.65$). According to the M-ABC, 32 children (78%) had normal motor performance, 3 children (7.4%) had motor problems being at risk (<P15), and 6 children (14.6%) had definite motor delay. This was in direct contrast to the findings on the KTK: 9 children (22%) had normal motor performance, 15 children (36.5%) were at risk for motor problems, and 17 children (41.5%) had definite motor delay.

The statistical associations are presented in Table 5. Multiple regression analysis showed that the percentile score of the M-ABC was not predicted by muscle strength ($\beta = -0.19$; CI −0.71 to 0.22), BMI ($\beta = 0.24$; CI −0.06 to 0.35) and the 6MWT score ($\beta = -0.27$; CI −0.42 to 0.36). On the other hand, significant associations were found between muscle strength ($\beta = -0.43$; CI 0.09 to 0.69) and BMI ($\beta = -0.33$; CI −0.27 to −0.004) with the percentile score of the KTK. No significant association was found for the 6MWT ($\beta = -0.07$; CI −0.31 to 0.19). The regression model was only significant for the KTK score ($p = 0.046$), with a prediction of the KTK score of $R^2 = 0.19$. For the M-ABC, the model was not significant ($p = 0.44$) with a prediction of $R^2 = 0.07$.

DISCUSSION

The first aim of this study was to investigate in children with generalized joint hypermobility whether muscle strength and functional exercise capacity are predictors of motor performance as objectified by the KTK and the M-ABC. The second aim was to compare the KTK and M-ABC in their sensitivity for detecting motor problems in children with generalized joint hypermobility. The hypothesis of the first aim was partly confirmed for the KTK motor score: muscle strength and BMI did predict the KTK motor score. For the motor score of the M-ABC, it was completely rejected. The hypothesis of the second aim was confirmed.

Children with generalized hypermobility are known to have less muscle strength and a lower functional exercise capacity and this may lead to problems in the participation of sports and physical activities like running and walking.[9,12,14,16,17] Decreased muscle strength was also observed in this study. Although within the normal range of −2 and + 2, the median z score was −0.57 and the P25–P75 was below 0. Functional exercise capacity, as measured by the 6MWT, was significantly decreased with a median z score of −2.47. This result, therefore, confirms mentioned problems with walking and is in concert with the results of Engelbert et al[17] in their study on functional exercise capacity. They stated that this was the result of deconditioning. The children they studied participated less in sports activities and had a higher mean BMI. Children in our study were younger: 8.1 years compared with 14.5 years in the study by Engelbert et al[17] and had a normal mean BMI of 16.4. The young children in our study are physically more active than Engelbert's older group, yet their exercise is decreased compared with their peers.

Only the motor performance as measured by the KTK was positively predicted by muscle strength and negatively by BMI. An explanation for this result can be found in the content of both motor tests: the KTK represents more sports-like elements as strength and speed than the M-ABC does. A negative correlation between BMI and gross motor performance has been found before.[33,34] Functional exercise capacity was not a predictor of performance on both motor tests. This is not a real surprise, because both tests were designed to objectify motor coordination.[29,30] The performance of the items does not take enough time to approximate a level of muscle endurance. Because the ability to walk for a certain distance is important to quality of life, especially for children participating in outdoor leisure activities and sports, one could consider the 6MWT as a tool to objectify this ability to participate. As mentioned before, children with generalized hypermobility often have problems in those daily, physical demanding activities.

A substantial difference was found in the comparison of the KTK and the M-ABC with the regard to the sensitivity in detecting motor problems in hypermobile children. The KTK found a motor delay in 78% of the children, whereas the M-ABC in only 22%. One explanation is that only muscle strength predicted the motor score of the KTK. Another explanation could be the differences in the constructs of both motor tests. Smits et al[31] studied a population consisting of children with and without motor problems and compared the construct validity of the M-ABC with the KTK as the "gold standard." A correlation between the total scores of 0.62 was found, and a concordance of 78% for the cutoff for a motor delay (this is exactly the opposite of the cutoff that was found in present study). Smits et al stated that the KTK is "oversensitive" for detecting motor problems. Since the KTK is a test from 1974, the norms may not be up to date anymore. The leisure habits of children have changed over time and consist of less physically demanding activities, such as watching TV and playing computer games,[35] so norms could be too stringent nowadays. This assumption is confirmed by several studies in Germany. The mean normal motor quotient tended to be around the 93, with more children having a standard score below the −1 standard deviations or more.[36] On the other hand, some remarks can be made on the sensitivity of the M-ABC; ceiling effects of some gross motor items are reported.[37,38] Therefore, too stringent norms of the KTK and the lax norms of the M-ABC may also have counted for the difference in our motor scores.

It can be concluded that the KTK is a more sensitive tool for detecting motor problems in children with joint hypermobility. The M-ABC score does not have a significant relationship with muscle strength and FEC and, therefore, is not able to measure the specific motor problems in this group of children. The KTK is not associated with functional exercise capacity. Because FEC was significantly decreased and the decreased ability to walk can interfere in the daily activities of children, the 6MWT can be recommended to be used along with the KTK. Both tests are needed in the assessment of motor performance and physical endurance in children with generalized joint hypermobility.

Comparing the results of motor delay with other studies, Engelbert et al[13] found a motor delay (<P15) in 46% of the children of 4 to 12 years. In our present study, a motor delay of 22% (<P15, M-ABC) was found. The children studied by Engelbert et al were visiting a hospital for their complaints due to generalized hypermobility. Our study was performed in primary healthcare settings. Possibly, children with more profound motor problems are referred to a hospital rather than to a general practitioner.

A set of criteria for the measurement of the amount and degree of hypermobility in children is not yet available.

Until now, the use of the adult criteria is advised after the age of 6 years.[9] This may lead to a false-positive diagnosis, because on average, children inherently have a greater range of motion of their joints than adults.[1,4,12,39] The available criteria are not only inadequate for the pediatric population, but also seem to lack validity for the adult population. Remvig et al[40,41] stated that for any of the available criteria, concurrent validity had not been investigated, as there is no golden standard that describes the normal range of motion for different races, ages, and gender while specifying which joints most discriminate ligamentous laxity. The lack of valid criteria may have led to an overestimation of the prevalence of joint hypermobility. Results of previous studies concerning complaints due to joint hypermobility should, therefore, be considered and interpreted with caution.[19,39–41] The first step should be the development of a golden standard. As soon as this has been established, the validity and cutoff of the existing screening criteria for hypermobility can be investigated. For the pediatric population, one should take in account the natural laxity and the diminishing of joint hypermobility with age. Children up to 12 years have inherently a greater range of joint motion. To detect collagen laxity, it might be necessary to look also for other symptoms of collagen laxity, like skin extensibility rather than just joint mobility.[16,40,41]

For pediatric physiotherapists, a valid score of joint hypermobility is essential: one needs to know if joint hypermobility is the main reason for existing problems or if the child is "just clumsy," with some inherent laxity. The intervention for the first group has a goal of stabilizing the affected joints and increasing muscle strength and endurance, captured in functional exercises. Similarly, education of the parents, teachers, and sport coaches about the nature and management of joint hypermobility is also a goal.[42] Until now there is no evidence of a long term treatment effect. This also needs to be the subject of future research.

To our knowledge, there are no previous studies into the influence of muscle strength and FEC on motor performance in children with generalized joint hypermobility. The use of a specific motor assessment tool for this group of children does not only yield better insight in motor problems, but it also provides a better opportunity to evaluate treatment results. A possible strength of our study is that it was performed in a primary healthcare setting. The study population corresponds with the average patient population seen in the community practice of a pediatric physiotherapist. A possible weakness of this study concerns the study population, because only children of the northern part of the Netherlands were involved. This could represent a selection bias. As mentioned before, the lack of a set of validated criteria for the measurement of the amount and degree of joint hypermobility in children is also a weakness. However, the use of adult criteria has been used before in several other studies.[9,13,16,17]

CONCLUSION

Muscle strength was moderately decreased while FEC was substantially decreased compared with reference values. Only muscle strength predicted motor performance when measured with the KTK. Functional exercise capacity as measured by the 6MWT did not predict motor performance on both the KTK and the M-ABC. For pediatric physiotherapists, the KTK can be recommended as an appropriate tool in the assessment of motor performance and evaluation of treatment results in children with generalized joint hypermobility. Along with the KTK, the 6MWT can be used to assess and evaluate the functional exercise capacity.

ACKNOWLEDGMENTS

The authors thank C.M.J. Hoffmann for her help in testing the children.

REFERENCES

1. Grahame R. The hypermobility syndrome. *Ann Rheum Dis.* 1990;49: 199–200.
2. Grahame R. Joint hypermobility and genetic collagen disorders: are they related? *Arch Dis Child.* 1999;80:188–191.
3. Kirk JA, Ansell BM, Bywaters EGL. The hypermobility syndrome: musculoskeletal complaints associated with generalized joint hypermobility. *Ann Rheum Dis.* 1967;26:419–425.
4. Beighton P, Solomon L, Soskolne CL. Articular mobility in an African population. *Ann Rheum Dis.* 1973;32:413–418.
5. Birrell FN, Adebajo AO, Hazleman BL, et al. High prevalence of joint laxity in West Africans. *Br J Rheumatol.* 1994;33:56–59.
6. Rikken-Bultman DG, Wellink L, van Dongen PW. Hypermobility in two Dutch school populations. *Eur J Obstet Gynecol Reprod Biol.* 1997; 73:189–192.
7. Carter C, Wilkinson J. Persistent joint laxity and congenital dislocation of the hip. *J Bone Joint Surg Br.* 1964;46:40–45.
8. Bulbena A, Duro JC, Porta M, et al. Clinical assessment of hypermobility of joints: assembling criteria. *J Rheumatol.* 1992;19:115–122.
9. Adib N, Davies K, Grahame R, et al. Joint hypermobility syndrome in childhood. A not so benign multisystem disorder? *Rheumatology (Oxford).* 2005;44:744–750.
10. Klemp P, Stevens JE, Isaacs S. A hypermobility study in ballet dancers. *J Rheumatol.* 1984;11:692–696.
11. Larsson LG, Baum J, Mudholkar GS, et al. Benefits and disadvantages of joint hypermobility among musicians. *N Engl J Med.* 1993;329: 1079–1082.
12. Murray KJ, Woo P. Benign joint hypermobility in childhood. *Rheumatology (Oxford).* 2001;40:489–491.
13. Engelbert RH, Kooijmans FT, van Riet AM, et al. The relationship between generalized joint hypermobility and motor development. *Pediatr Phys Ther.* 2005;17:258–263.
14. Jaffe M, Tirosh E, Cohen A, et al. Joint mobility and motor development. *Arch Dis Child.* 1988;63:159–161.
15. Tirosh E, Jaffe M, Marmur R, et al. Prognosis of motor development and joint hypermobility. *Arch Dis Child.* 1991;66:931–933.
16. Engelbert RH, Bank RA, Sakkers RJ, et al. Pediatric generalized joint hypermobility with and without musculoskeletal complaints: a localized or systemic disorder? *Pediatrics.* 2003;111:e248–e254.
17. Engelbert RH, van Bergen M, Henneken T, et al. Exercise tolerance in children and adolescents with musculoskeletal pain in joint hypermobility and joint hypomobility syndrome. *Pediatrics.* 2006;118: e690–e696.
18. Biro F, Gewanter HL, Baum J. The hypermobility syndrome. *Pediatrics.* 1983;72:701–706.
19. Murray KJ. Hypermobility disorders in children and adolescents. *Best Pract Res Clin Rheumatol.* 2006;20:329–351.
20. Portney LG, Watkins M. *Foundations of Clinical Research. Application to Practice.* 2nd ed. New Jersey: Prentice Hall, Inc.; 2000.
21. Beenakker EA, van der Hoeven JH, Fock JM, et al. Reference values of maximum isometric muscle force obtained in 270 children aged

4–16 years by hand-held dynamometry. *Neuromuscul Disord.* 2001;11:441–446.
22. Cooper DM. Rethinking exercise testing in children: a challenge. *Am J Respir Crit Care Med.* 1995;152(4 Pt 1):1154–1157.
23. Paap E, van der Net J, Helders PJ, et al. Physiologic response of the six-minute walk test in children with juvenile idiopathic arthritis. *Arthritis Rheum.* 2005;53:351–356.
24. Takken T, van der Net J, Helders PJ. Anaerobic exercise capacity in patients with juvenile-onset idiopathic inflammatory myopathies. *Arthritis Rheum.* 2005;53:173–177.
25. Geiger R, Strasak A, Treml B, et al. Six-minute walk test in children and adolescents. *J Pediatr.* 2007;150:395–399.
26. Li AM, Yin J, Au JT, et al. Standard reference for the six-minute-walk test in healthy children aged 7 to 16 years. *Am J Respir Crit Care Med.* 2007;176:174–180.
27. Gulmans VA, van Veldhoven NH, de Meer K, et al. The six-minute walking test in children with cystic fibrosis: reliability and validity. *Pediatr Pulmonol.* 1996;22:85–89.
28. Henderson SE, Hall D. Concomitants of clumsiness in young schoolchildren. *Dev Med Child Neurol.* 1982;24:448–460.
29. Henderson SE, Sugden DA. *Movement Assessment Battery for Children: Manual.* Kent, UK: Psychological Corporation; 1992.
30. Kiphard EJ, Schilling F. *Korperkoordinationstest für Kinder.* Weinheim: Beltz Test GmbH; 1974.
31. Smits-Engelsman BCM, Henderson SE, Michels CGJ. The assessment of children with developmental coordination disorders in the Netherlands: the relationship between the movement assessment battery for children and the Körperkoordinations Test für Kinder. *Hum Mov Sci.* 1998;(17): 699–709.
32. Dawson-Saunders B, Trapp RG. *Basics and Clinical Biostatics.* Norwalk CT: Appleton & Lange; 1998.
33. Graf C, Koch B, Dordel S, et al. Physical activity, leisure habits and obesity in first-grade children. *Eur J Cardiovasc Prev Rehabil.* 2004; 11:284–290.
34. Graf C, Koch B, Kretschmann-Kandel E, et al. Correlation between BMI, leisure habits and motor abilities in childhood (CHILT-project). *Int J Obes Relat Metab Disord.* 2004;28:22–26.
35. Hofferth SL, Sandberg JF. Changes in American children's use of time 1981–1997. In: Owens T, Hofferth SL, eds. *Children at the Millennium.* Amsterdam, Netherlands: Elsevier Publishers; 2001:193–229.
36. Kiphard EJ, Schilling F. *Körperkoordinationstest für Kinder. Manual.* 2nd ed. Göttingen: Beltz Test GmbH; 2007.
37. Miyahara M, Tsujii M, Hanai T, et al. The Movement Assessment Battery for Children: A preliminary investigation of its usefulness in Japan. *Hum Mov Sci.* 1998;17:679–697.
38. van Waelvelde H, De Weerdt W, De Cock P, et al. Aspects of the validity of de Movement Assessment Battery for Children. *Hum Mov Sci.* 2004;23:49–60.
39. Jansson A, Saartok T, Werner S, et al. General joint laxity in 1845 Swedish school children of different ages: age- and gender-specific distributions. *Acta Paediatr.* 2004;93:1202–1206.
40. Remvig L, Jensen DV, Ward RC. Epidemiology of general joint hypermobility and basis for the proposed criteria for benign joint hypermobility syndrome: review of the literature. *J Rheumatol.* 2007;34:804–809.
41. Remvig L, Jensen DV, Ward RC. Are diagnostic criteria for general joint hypermobility and benign joint hypermobility syndrome based on reproducible and valid tests? A review of the literature. *J Rheumatol.* 2007; 34:798–803.
42. Grahame R, Keer R. Hypermobility Syndrome, Recognition and Management for Physiotherapists. Edinburgh: Butterworth /Heinemann; 2003.