Title: Activity participation intensity is associated with skeletal development in pre-pubertal children with developmental coordination disorder

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Abstract

Purpose: This study aimed (1) to compare the skeletal maturity and activity participation pattern between children with and without developmental coordination disorder (DCD); and (2) to determine whether activity participation pattern was associated with the skeletal development among children with DCD. **Material and methods:** Thirty-three children with DCD (mean age: 7.76 years) and 30 typically-developing children (mean age: 7.60 years) were recruited. Skeletal maturity was assessed with the Sunlight BonAge system. Motor ability was evaluated by the Movement assessment battery for Children-2 (MABC-2). Participation patterns were evaluated using the Children Assessment of Participation and Enjoyment assessment. Analysis of variance was used to compare the outcome variables between the two groups. Multiple regression analysis was performed to examine the relationship between skeletal development, motor performance and activity participation intensity in children with DCD. **Results:** The DCD group had significantly delayed skeletal development, lower MABC-2 derived scores, and participated less intensely in various types of physical activities than their typically-developing peers. After accounting for the effects of age and sex, activity participation intensity score remained significantly associated with delay in skeletal development, explaining 28.0% of the variance ($F_{change1,29}=11.341$, $p=0.002$). **Conclusion:** Skeletal development is delayed in pre-pubertal children with DCD. Limited activity participation intensity appears to be one of the contributing factors.

Keywords Clumsy children; activity; motor proficiency; balance; bone age

INTRODUCTION

Developmental coordination disorder (DCD) is a well-known motor-based problem that affects approximately 6% of children at primary school age (American Psychiatric Association, 2000). Due to their poor motor proficiency, DCD-affected children participate in fewer activities and less intensely than their typically-developing peers (Fong et al., 2011a; Fong et al., 2011b; Jarus et al., 2011). It is well known that participation in activities, particularly weight-bearing activities (e.g. soccer training), during pre-pubertal and pubertal periods is very important because external forces acting on the bones during different activities can facilitate bone growth and development (Rogol et al., 2000; Vicente-Rodriguez, 2006). Sedentary life style in children with DCD may have a negative impact on skeletal development.

Apart from bone strength, bone age is another useful parameter to indicate skeletal maturity. Traditionally, bone age is determined by the appearance of centres of ossification, fusion or non-fusion of epiphyses, and changes in size and shape of the wrist and hand bones (Cardoso, 2007). It is considered as the most valid biological measure of maturity (De Luca & Baron, 1999). Measurement of bone age is commonly used in clinical evaluation of growth and maturity status and is an important part of the diagnosis and management of pediatric growth disorders (Baxter-Jones et al., 2002). Previous studies have shown that bone age is positively associated with bone mass in preadolescent females (aged 8 to 13 years) (Ilich et al, 1996) and bone mineral density in children (aged 9 to 16 years) (Jones et al., 2005).

To date, no study has reported the skeletal maturity status and its relationship with activity participation in the DCD population. Therefore, the objectives of this study were (1) to compare the bone age, and activity participation pattern between children with and without

DCD; and (2) to determine whether activity participation pattern is associated with the skeletal development among children with DCD.

MATERIALS AND METHODS

All sample size calculations were based on a statistical power of 0.80 and an alpha level of 0.05 (two-tailed). A previous study (Fong et al., 2011a) showed that the physical activity intensity score, as assessed by the Children's Assessment of Participation and Enjoyment (CAPE), was 108.37 (28.67) and 133.76 (26.61) for the DCD group (n=81) and control group (n=67) respectively, which translates into a large effect size of 0.92. Therefore, the minimum sample size needed to detect a significant between-group difference in the activity participation outcomes would be 20 for each group (alpha = 0.05, power =0.80) (objective 1). Regarding the association between bone development and activity participation (objective 2), a previous study by Lehtonen-Veromaa et al. (2000) reported that in their multiple regression analysis, physical activity and other relevant variables combined to account for 54.7% and 63.4% of the variance in BMD of the femoral neck and lumbar spine, respectively, among peripubertal girls. Therefore, a large effect size ($R^2 = 0.4$, translating into $F^2 = 0.67$) was estimated for this study. A minimum sample size of 24 children with DCD was required to detect a significant association of activity participation intensity with skeletal development, after accounting for age, sex, and motor function (alpha = 0.05, power = 0.8, total number of predictors = 4).

Children with DCD were recruited from local Child Assessment Centres by convenience sampling. Inclusion criteria were (1) a formal diagnosis of DCD that was made by an interdisciplinary team (pediatrician, clinical psychologist, physiotherapist and occupational therapist) at the Child Assessment Centre according to criteria of the Diagnostic

and Statistical Manual of Mental Disorders (DSM-IV-TR) (American Psychiatric Association, 2000); (2) six to ten years old; (3) study in a regular education framework; (4) no intellectual disability; and (5) of Chinese ethnicity. Exclusion criteria were (1) diagnosis of emotional, endocrine, neurological, or other movement disorders; or (2) significant musculoskeletal, cardiopulmonary or medical conditions that may influence motor performance or skeletal development. Chinese children with normal development were recruited from the community on a volunteer basis as controls. The inclusion and exclusion criteria for the control group were same as the DCD group except that they did not have any history of DCD.

This study was approved by the human subjects ethics review subcommittee of the Hong Kong Polytechnic University. After explaining the study to each participant and their parents, written informed consent was obtained. Data were collected by an experienced pediatric physiotherapist in the Balance and Neural Control Laboratory of the Hong Kong Polytechnic University. All procedures were conducted in accordance with the Declaration of Helsinki.

Basic demographic information including sexual maturity (as indicated by the presence of pubic hair, onset of breast development and testicle volume in cubic centimeter) was obtained by interviewing the children and their parents. If the participants were not sure about the testicle volume, parents were invited to measure it with their boys in a closed room. The volume of water displaced by the testicles was documented. Skeletal development was determined ultrasonically with the 'Sunlight BonAge system' (Sunlight Medical Ltd., Tel Aviv, Israel). This device provides an accurate (intra-operator precision: 0.24 years for males and 0.25 years for females), radiation free assessment of skeletal age (chronological age: 5 to 18 years old) and the results are highly correlated with the conventional Greulich and Pyle method (Mentzel et al., 2005; Sunlight Medical Ltd., 2005). Each participant was asked to

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rest the left forearm on the BoneAge measurement table and position the distal tip of the left ulna styloid process between the two ultrasound transducers. Ultrasonic waves with a frequency of 750kHz were transmitted through the left wrist. Five to eleven cycles of measurement were performed to ensure high precision. The BonAge device calculated the speed of sound (velocity of the ultrasound wave increases when ultrasound is transmitted through ossified epiphyses in relatively matured radius and ulna) and used the distance between the two transducers under known and controlled pressure conditions, and a proprietary sex- and ethnicity-based algorithm to provide a numeric result of 'bone age' that was used for subsequent analysis (Mentzel et al., 2005; Sunlight Medical Ltd., 2005). In addition, 'delay in skeletal development' was calculated from the equation: chronological age – bone age.

The Movement assessment battery for Children-2 (MABC-2) was used to measure the fine and gross motor performances in all participants. It is a standardized tool for measuring motor performance in 3- to 16-year-old children and consists of eight tasks for each of the three age ranges. The eight tasks are categorized into three domains: manual dexterity, aiming and catching, and balance. Test items in the three domains are described in Henderson et al. (Henderson et al, 2007). The raw score of each item was converted into the item standard score, and the component score, standard score, and percentile of each domain were derived from the item standard scores. Additionally, the total test score, standard score, and percentile rank were derived. The total test score (reflects the general motor proficiency), manual dexterity component score, aiming and catching component score, and the balance component score were used for analysis. MABC-2 has demonstrated good test-retest reliability, inter-rater reliability, and criterion-related validity in children with and without motor difficulties (Henderson et al, 2007).

The Children's Assessment of Participation and Enjoyment (CAPE) was used to assess participation pattern in out-of-school time activities. It is a reliable and valid self-report measure of activity participation for children and youth (6 to 21 years old) (Imms, 2008; King et al., 2004). This questionnaire includes both formal (more structured activities) and informal domains (less structured activities that require less planning), and five activity types, namely recreational, physical, social, skill-based, and self-improvement activities (King et al., 2004). The specific activities assessed with CAPE were described in Fong et al. (2011a). Face-to-face interviews were conducted with participants and their parents to complete the CAPE questionnaire. The total activity diversity score (count of activities in which the child has participated over the previous four months), total activity intensity score (participation frequency for a set of activities), and the intensity scores of different activity types were used for analysis (King et al., 2004).

Statistical analyses were performed using SPSS version 17.0 (SPSS, Chicago, IL). Level of significance was set at 0.05 (two-tailed) for all statistical tests. Kolmogorov-Smirnov tests were used to check the normality of data. Independent t-tests were used to compare the age, height, weight, body mass index (BMI), MABC-2 derived scores and bone age related data between DCD and control groups while chi-square test was used to compare sex between the two groups. To compare the CAPE-derived participation scores between groups, multivariate analysis of variance (MANOVA) with Bonferroni adjustment was performed to reduce the risk of type I error due to multiple comparisons.

Pearson's correlation coefficients were used to determine the bivariate relationships between 'delay in skeletal development' and other demographic data, CAPE and MABC-2 derived variables among children with DCD. The relationship between 'delay in skeletal development' and sex was examined by Spearman's rho. Next, multiple regression analyses

were performed to identify which physical and activity participation parameters were strongest predictors of 'delay in skeletal development'. Selection of predictors for regression analysis was based on both biological relevance and results of the correlation analysis. Age and sex were first entered into the regression model followed by MABC-derived scores, and CAPE total activity intensity score. The degree of association among the potential independent variables was also checked to avoid multicollinearity.

RESULTS

Demographic characteristics, motor abilities, and 'delay in skeletal development' of the DCD group (n=33) and control group (n=30) are outlined in Table 1. All participants were at Tanner stage one (pre-puberty). The basic demographic variables and bone age of the two groups did not differ (p>0.05). As motor ability was one of the major criteria for diagnosing DCD, it was only natural to find significant between-group differences in MABC-2 total test score, balance component score, manual dexterity component score, aiming and catching component score (p≤0.05). There was also a significant between-group difference in 'delay in skeletal development' (calculated by chronological age - bone age) (p≤0.05) (Table 1). Intensity of activity participation also differed between groups (p≤0.01). Children with DCD participated less frequently in informal, recreational, physical and self improvement activities (p≤0.05) (Table 2).

Fair correlations were found between 'delay in skeletal development' and CAPE intensity scores (r=-0.339 to -0.532, p≤0.05) among children with DCD. Among the CAPE sub-category intensity scores, recreational activity intensity score demonstrated strongest correlation with 'delay in skeletal development' (r=-0.441, p≤0.05) (Table 3). MABC-2 balance component score was also negatively correlated with 'delay in skeletal development'

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(r=-0.360, p≤0.05). No correlation was found between the 'delay in skeletal development' and MABC-2 total test score, manual dexterity component score, or aiming and catching component score (Table 3). The results of multiple regression analysis showed that, after accounting for age and sex, adding CAPE total activity intensity score to the regression model accounted for an additional 28.0% of the variance in the 'delay in skeletal development' ($F_{change1,29}$=11.341, p=0.007). MABC-2 balance component score was not a significant predictor of the 'delay in skeletal development' (p=0.132) (Table 4).

DISCUSSION

Skeletal development in children with DCD

This is the first study to identify a significant skeletal growth delay in children with DCD when compared with typically-developing children. Although the mean body height (a measure of linear growth) of both groups was similar (Table 1), skeletal age (a measure of bone growth and development) lagged behind the chronological age by 1.09 years in the DCD group. Our present finding agrees with previous studies that suggest chronic diseases or disorders such as cerebral palsy (CP) in children may alter the skeletal maturation (Asworth & Millward, 1986; De Luca & Baron, 1999; Roberts et al., 1994). Similar to children with mild CP, many children with DCD experience minimal brain damage (e.g. disrupted cerebello-cerebral network and posterior parietal cortex dysfunction) since birth and the resulted movement dysfunctions are chronic in nature (Cermak & Larkin, 2002; Kashiwagi et al., 2009; Marien et al., 2010). Therefore, we postulated that DCD-affected children might also require more time to complete their skeletal growth process and might have delayed attainment of skeletal maturation or adult stature. The exact reasons for the delayed skeletal maturation in DCD-affected children are still unclear. This may be related to poor nutritional

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status and endocrine dysfunction as observed in children with CP (Henderson et al., 2005), or decreased activity participation as found in the present study (will be discussed in the next section). Further study should explore other potential factors affecting bone maturation and whether catch-up growth (i.e. acceleration in growth that occurs when a period of growth retardation ends) (Asworth & Millward, 1986) could happen in children with DCD if favourable conditions (e.g., good nutrition, movement therapy) were provided.

Activity participation pattern in children with and without DCD

Consistent with the results reported by Jarus et al. (2011), this study showed that children with DCD participated less intensely (frequently) in all activities generally. Specifically, they participated less frequently in informal, recreational, physical and self improvement activities. Participating in physical activities with sufficient intensity (frequency) is essential for inducing a positive effect on health and functional outcomes (American College of Sports Medicine, 2006). For example, it has been shown that if DCD-affected children could participate in Taekwondo activity one-hour daily for three months consecutively, their balance ability and vestibular function could be enhanced (Fong et al., 2012). On the contrary, children with DCD might not be able to benefit from exercises if the intensity is too low. Therefore, parents should encourage their children to participate in activities regularly and cultivate an exercise habit.

This study found that children with DCD participated in informal activities less intensely. Informal activities refer to those activities that are less structured (e.g. doing water sports, going to a party, doing crafts, drawing or colouring) and so may be less predictable (King et al., 2004). Participating in less structured and unpredictable activities may expose

the children's gross and/or fine motor weakness (Engel-Yeger & Kasis, 2010). Children with DCD may thus be less inclined to participate in these activities.

DCD-affected children also participated less intensely in recreational, physical and self improvement activities. Recreational activities (e.g. doing puzzles, going for a walk or hike) that are addressed by CAPE require certain level of gross or fine motor skills. Self improvement activities are also rather physical in nature (e.g. shopping, doing a chore) (King et al., 2004). Children with DCD have less efficient movement patterns and gross/fine motor functions, as reflected by their significantly lower MABC-2 scores. They may expend more energy and therefore fatigue faster during different types of activities (Wrotniak et al., 2006). This could negatively affect their activity participation intensity.

For the activity participation diversity, the present study showed no statistically significant difference between the DCD and control groups. This finding is quite different from Jarus et al. (2011) who reported that children with DCD participated in fewer activities than their typically-developing peers. The discrepancy in results could be attributed to the use of different age groups between studies. Our participants were relatively older (6 to 10 years old) than those participated in Jarus et al.'s study (5 to 7 years old) (Jarus et al., 2011). Increasing age is known to be related to participation in fewer activities in children with physical disabilities, perhaps because older children are less controlled by their parents and opt to be more inactive (King et al., 2009).

Determinants of 'delay in skeletal development' in children with DCD

In our multiple regression model, activity participation intensity was the only significant independent variable for predicting the delayed skeletal development in children with DCD after adjusting for age and sex. On the other hand, MABC-2 balance score, which

was used to classify DCD- and non-DCD-affected children, was not directly associated with the bone age lag as demonstrated in the present study.

This study showed that less intense activity participation is significantly associated with delay in skeletal development among children with DCD, accounting for 28.0% of its variance. It is well known that physical exercises with sufficient intensity can increase growth hormone secretion and thus promote skeletal growth (McArdle et al., 2001). Numerous studies have reported that participation in weight-bearing or muscle loading activities has an osteogenic effect (i.e. increase in bone mineral content and density) during growth (Baxter-Jones et al., 2008; Cech & Martin, 2002; Lehtonen-Veromaa et al., 2000; Schoenau, 2005; Vicente-Rodriguez, 2006). Our results showed that the lower level of participation in physical activity among children with DCD may have a negative impact on skeletal maturation.

Our correlation analysis further showed that participating in recreational activities frequently was mostly strongly related to skeletal development, followed by participating in informal and physical activities, while participating frequently in self improvement activities was not significantly related to skeletal maturation in children with DCD. This may be because the recreational activities, as measured by the CAPE, include many weight-bearing activities (e.g. going for a walk or a hike) and require more physical strength (King et al., 2004). In contrast, the sedentary self improvement activities are relatively static (e.g. reading, doing homework, writing letters or a story). Loading of the bones is thus more likely to occur during the recreational activities compared with self-improvement activities.

Clinical implication

Delay in skeletal maturation in children with DCD may have implications for factures and injuries, particularly during sports activities. For example, late maturers are associated

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with having lower bone mass (Ilich et al., 1996), higher risk of upper limb fracture (Jones & Ma, 2005) and are reported to have more injuries during sports (soccer) training (Backous et al., 1988). Physical injuries might further hinder their participation in activities. Thus a vicious cycle of activity avoidance, poor skeletal development, physical injuries, and decreased participation intensity in all activities may ensue. Interventions should aim to prevent this vicious cycle by encouraging children with DCD to participate in appropriate weight-bearing activities and increase the exercise intensity progressively (Rogol et al., 2000; Vicente-Rodriguez, 2006).

Limitations and consideration for future studies

DCD is heterogeneous in nature (Cermak & Larkin, 2002) and our DCD group also included children with co-morbidities such as attention deficit hyperactivity disorder (ADHD), autism spectrum disorders (ASD), asperger syndrome and dyslexia (Table 1). Mills et al. (2007) suggested that children with autism or ASD do not have an advanced bone age when compared to the norm. Gustafsson et al. (2008) also reported that there was no correlation between ADHD-symptoms and bone age. No previous study has examined the relation between dyslexia and bone age. Since only four children out of 33 DCD-affected children in our study had dyslexia, we assumed that the overall effect of co-morbidities on the skeletal development in the DCD group was minimal. However, the effect of this confounding factor could not be totally eliminated and therefore the results should be interpreted with caution. Furthermore, how the presence of co-morbidities affects activity participation is also unknown. Previous studies have shown that the activity participation patterns in children with ASD are quite different from typically developing children (Hilton

et al., 2008; Solish et al., 2010). Further study should include a DCD group without co-morbidities in order to delineate the effects of DCD on bone age and activity participation."

This study was cross-sectional in design. Therefore, natural variations of individuals in their rates of skeletal development may contribute to the discrepancy between bone age and chronological age (Cole et al., 1988). Further longitudinal studies with serial measurements could reveal greater accuracy the growth rate and patterns of skeletal development in children with DCD.

Moreover, our regression analysis only accounts for less than 35% of the variance in delay in skeletal development. Potentially important factors that may be associated with skeletal development such as nutrition, endocrine and paracrine functions (De Luca & Baron, 1999), and socio-economic status (Schmeling et al., 2000) should also be considered in future studies.

Future studies should also consider the factors that are associated with children's activity participation intensity (e.g. children's leisure interests and preferences, socioeconomic backgrounds, parental education and environment setting), so that children at risk for physical inactivity and hence delayed skeletal development can be more effectively identified (Jarus et al., 2011; King et al., 2006). While activity participation intensity is significantly associated with skeletal maturity in children with DCD, whether increasing the participation intensity in various activities (e.g. physical activities) can actually enhance skeletal development is still uncertain. Ample evidence shows that physical activity programs can enhance bone mass in typically-developing children at pre-pubertal and pubertal ages (Vicente-Rodriguez, 2006). A number of studies have demonstrated that exercise training can lead to significant improvement in motor performance and neuromuscular coordination as well in children with DCD (Fong et al., 2012; Tsai, 2009). However, no study has examined

the effects of different physical activity programs on skeletal development in children with DCD. Research is much needed in this important area.

Conclusions

Skeletal development is delayed in pre-pubertal children with DCD. Limited activity participation intensity appears to be one of the contributing factors.

ACKNOWLEDGEMENTS

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Table 1. Demographic characteristics, motor abilities and skeletal maturity of the participants

	DCD group Mean±SD (n=33)	Control group Mean±SD (n=30)	p value
Age, year	7.76 ±1.41	7.60 ±1.10	0.623
Sex (Boys/Girls), n	27/6	24/6	0.854
Height, m	127.70±10.65	127.53±8.65	0.947
Weight, kg	30.05±10.73	29.60±7.86	0.848
BMI, kg/m^2	17.86±3.53	17.93±3.04	0.936
Co-morbidity:			
Attention deficit hyperactivity disorder, n	3	0	
Attention deficit disorder, n	3	0	
Dyslexia, n	4	0	
Asperger syndrome, n	2	0	
Autism spectrum disorders, n	1	0	
MABC-2:			
Total test score	62.36±15.50	68.47±5.00	0.038*
Balance component score	28.21±7.07	31.80±3.40	0.012*
Manual dexterity component score	21.55±8.23	26.23±5.04	0.008**
Aiming and catching component score	12.61±4.73	18.90±5.65	<0.001***
Skeletal maturity:			
Bone age, year	6.67±2.00	7.46±1.33	0.066
Delay in skeletal development (Chronological age – Bone age), year	1.09±1.32	0.14±0.64	0.001***

*Significant difference at $p \leq 0.05$

**Significant difference at $p \leq 0.01$

***Significant difference at $p \leq 0.001$

Table 2. Comparison of activity participation patterns between children with DCD and their typically-developing peers

Outcome	DCD group (n=33) Mean±SD	Control group (n=30) Mean±SD	p value
CAPE total activities:			
Total diversity score	25.42±7.91	27.60±5.32	0.210
Total intensity score	110.06±32.06	130.97±29.12	0.009**
Intensity score of different types of activities:			
Informal activities	2.12±0.60	2.57±0.58	0.004**
Formal activities	1.63±0.80	1.88±0.65	0.177
Recreational activities	3.03±0.84	3.54±0.93	0.025*
Physical activities	1.21±0.74	1.63±0.78	0.030*
Social activities	1.67±1.00	1.99±0.76	0.165
Skill-based activities	1.39±0.77	1.65±0.85	0.217
Self improvement activities	2.64±0.85	3.08±0.79	0.038*

*Significant difference ($p \leq 0.05$) between two groups

**Significant difference ($p \leq 0.01$) between two groups

Table 3. Correlations with skeletal maturity in children with DCD

	Delay in skeletal development (Age – Bone age)
Age	-0.063
Sex	-0.012
Height	-0.323
Weight	-0.307
BMI	-0.290
MABC-2 Total test score	-0.306
MABC-2 Balance component score	-0.360*
MABC-2 Manual dexterity component score	-0.114
MABC-2 Aiming and catching component score	-0.267
CAPE Total activity intensity score	-0.532***
CAPE Physical activity intensity score	-0.339*
CAPE Recreational activity intensity score	-0.441*
CAPE Informal activity intensity score	-0.410*
CAPE Self improvement activity intensity score	-0.173

*Significant difference at $p \leq 0.05$

**Significant difference at $p \leq 0.01$

***Significant difference at $p \leq 0.001$

Table 4. Multiple regression analysis: Prediction of 'delay in skeletal development'

Independent variables	R^2 change	Unstandardized Regression Coefficient (B)	95% Confidence interval	Standardized Regression Coefficient (β)	p value
Age (year)	<0.001	-0.016	-0.314, 0.282	-0.017	0.913
Sex (boys=1, girls=2)	<0.001	0.184	-0.945, 1.313	0.054	0.741
CAPE Total activity intensity score	0.280	-0.019	-0.033, -0.006	-0.466	0.007*
MABC-2 Balance component score	0.057	-0.048	-0.112, 0.015	-0.259	0.132

*Significant difference at $p \leq 0.01$