

Original article

Motor and cognitive development of children with Down syndrome: The effect of acquisition of walking skills on their cognitive and language abilities

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Abstract

Objective: This study investigated the relationship between motor and cognitive/language development in children with Down syndrome (DS). We also tested the hypothesis that acquisition of walking skills facilitates later cognitive/language development.

Methods: Participants were 156 children with DS who were less than 48 months old and had undergone a health checkup by medical doctors and received rehabilitation treatment between April 2013 and March 2017 in Yokohama, Japan. To assess their development, the Kyoto Scale of Psychological Development (KSPD) 2001 was used, which measures development in three subdomains: Posture-Motor (P-M), Cognitive-Adaptive (C-A), and Language-Social (L-S). To investigate the relationship between motor and cognitive/language development, partial correlation analyses were conducted that controlled for participants' age. To test the effect of achieving walking skills, regression analyses were conducted using only data from participants who took the KSPD at least twice and could not walk at the initial test.

Results: P-M developmental age (DA) was significantly and positively correlated with both C-A DA and L-S DA in children 1–3 years old. The relationship strengthened with increased age. Acquisition of walking skills had a significant positive effect on both the C-A DA and L-S DA at the second test when controlling for the C-A DA and L-S DA at the first test and age at the second test.

Conclusion: Motor development was correlated with both cognitive and language development in young children with DS. Results also suggested that achievement of walking could facilitate later cognitive/language development in children with DS.

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Keywords: Down syndrome; Motor development; Cognitive development; Language development; Walking

1. Introduction

Down syndrome (DS) (21 trisomy abnormality) frequently causes developmental delay in several domains. The most common is motor delay, which is considered to be related to innate hypotonia [1]. Children with

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DS were shown to have lower motor performance than typically developing children from a very young age, even at 2–4 months of age [2]. On average, while typically developing children were found to sit without support by 7 months and start to walk independently by 12 months, children with DS sit by 15 months and walk by 30 months [3]. To address this marked delay in motor development, intervention programs suitable for children with DS have been developed [4].

Children with DS also show delay in cognitive development. In most cases, their IQs indicate moderate to severe mental retardation [5]. In one study, the IQ of DS children in early childhood averaged 44.7, ranging from 28 to 71 [6]. It was consistently shown that the IQ/DQ (Developmental Quotient) declined as DS children aged, indicating that the speed in cognitive development was slower than age-expected [7,8]. Moreover, language difficulties, specifically in linguistic production, have been commonly observed among children with DS [1]. For example, Ferreira-Vasques and Lamônica found that children with DS spoke their first words much later than typical children (mean age = 28.8 months vs. 12.7 months) [2]. Because of their low cognitive and language abilities, children with DS usually receive special education services at school although inclusion in mainstream education is promoted in some countries [9,10].

As the first step in interventions for children with DS, physical therapy has often been provided by clinicians because acquisition of basic locomotor skills and improvement in motor functioning are assumed to facilitate development in other areas, including cognition and language [11,12]. Despite such practices, only two studies [13,14] empirically investigated the relationship between motor development and cognitive/language development. One study [13] found that motor functioning assessed by Gross-Motor Function Measure 88 was associated with psychomotor function determined using the Brunet-Lezine scale for children aged 3 years or younger but was not associated with level of mental impairment identified by the Wechsler Intelligence Scale for Children for children aged 4 years or older. However, the relationship between motor and cognitive development for young children might have been unclear in this study because the Brunet-Lezine scale is not a pure measure of cognitive development. The results of the other study [14] indicated a positive but non-significant association between motor and cognitive development among children with DS, but the evidence was weak because motor development was measured using only two items, “head control” and “independent walking.” Taken together, despite frequent practice of prioritizing motor interventions for children with DS, their effect has not been strongly supported yet. Therefore, it is still worthwhile to investigate the relationship between motor and cognitive/language development in

these children and the effect of acquisition of basic locomotive skills on cognitive/language development.

Relationships between motor and cognitive/language development have been studied in other populations. In the case of typical children, results of a systematic review showed that for children aged 4–15 years, while overall motor skills and cognitive skills did not significantly correlate with each other, fine motor skills, bilateral body coordination, and timed performance in movements were found to be correlated with cognitive skills, specifically among pre-pubertal children [15]. For younger typical children, Iverson [16] pointed out that some language milestones are likely to be met just after acquisition of specific motor skills/actions; for example, reduplicated bubbles are likely to be observed soon after rhythmic arm movement emerged. This also indicated an interrelationship between motor and language development in typical children. Positive relationships between motor skills and cognitive functioning were also found among various specific populations, such as children with autistic spectrum disorders [17], children with spina bifida [18], and individuals with Parkinson’s disease [19]. Based on these findings, it is reasonable to expect a positive relationship between motor and cognitive/language functioning among children with DS.

In addition to confirming if there is a correlation between motor development and cognitive/language development, to justify prioritizing physical therapy as an intervention, it would be preferable to show a causal link between acquiring a specific motor skill and improving cognitive/language functioning. Because independent walking is often an important objective of an intervention, in this study, we tested the hypothesis that the acquisition of walking skills would positively impact cognitive/language development in children with DS. Following are the rationales supporting this hypothesis. In experiments with infants, Karasik et al. [20] found that in the transition from crawling to walking the range of environmental exploration by infants was expanded. As Piaget [21] proposed, active exploration of one’s environment is thought to facilitate further knowledge acquisition, which was supported by later experiments [22,23]. Thelen [24] maintained that the ability to move increases the amount of information that children can perceive from the surrounding world, which in turn facilitates their cognitive development. In addition, Bushnell and Boudreau [25] argued that the acquisition of certain motor skills precedes development of skills in other domains, such as haptic perception and depth perception, which is a component of spatial cognition. Moreover, the relationship between walking onset and cognitive development was empirically shown for pre-term children [26]. Taken together, it is expected that acquisition of independent walking skills would increase cognitive functioning of children with DS.

The link between the acquisition of walking and language development has been studied more directly in populations other than those with DS. For typical children, this relationship has been supported. Walle and Campos [27] reported that both receptive vocabulary and expressive vocabulary were found to increase immediately after children became able to walk. They also found that even after controlling for age, walking had a significant positive impact on language development. Transition from crawling to independent walking was also found to increase the frequency of prelinguistic behavior, including vocalizations, gestures, facial expressions directed toward adults, and the initiation of joint engagement [28–30], which are considered prerequisites for the development of expressive language. For children with autistic spectrum disorders, however, the findings are inconsistent; for such children, walking onset was significantly correlated with later language development in one study [31], but not in another study [32]. As the trajectory of language development in children with DS is somewhat different from that of typical children [1], it is an open question whether their acquisition of independent walking skills is associated with later language development, as with typical children.

In this study, we first investigated the relationship between motor skills and cognitive/language skills of children with DS by calculating the correlations among their domain scores in a standardized developmental test. The test, Kyoto Scale of Psychological Development 2001, is assumed to measure development in motor, cognitive, and language domains separately, with each domain consisting of multiple items. Then we tested the hypothesis that acquisition of walking skills would be associated with subsequent cognitive/language development by conducting regression analyses.

2. Methods

2.1. Participants

Data for this retrospective study were obtained from the medical records of children with DS who underwent a health checkup from pediatricians and began rehabilitation treatment between April 2013 and March 2017 at the Yokohama Rehabilitation Center, Japan, or at one of the 4 other local rehabilitation centers in the same city. For this study, we used data on 156 children (93 boys and 63 girls) who took a developmental test at least once before the age of 48 months during the period specified above. Because the data were collected retrospectively, at the time that we obtained the data some of the children were still very young and had recently taken the developmental test as an initial assessment while others had already used the service for several years and had been administered the developmental test several times. As a result, of the 156 participants, 83 were

tested once, 57 twice, and 16 three times. At the initial test, children ranged in age from 10 to 43 months (mean = 25.3 months). There was a significant difference ($t = -7.29, p < .001$) in the age at the initial test between children who were tested only once (mean = 28.49) and those who were tested multiple times (mean = 21.73). All the participants had received physical therapy from a certified physical therapist once to four times per month depending on need until they could walk without support.

For the first part of the study in which we investigated the relationship between motor skills and cognitive/language skills, we used the data on all 156 participants. For the second part of the study, we only analyzed data for 58 participants, who were those who took the developmental test twice or more and were unable to walk at the time of the first test.

2.2. Measures

Motor, cognitive, and language development. Kyoto Scale of Psychological Development 2001 (KSPD) [33] was used to measure motor, cognitive and language development of the participants. KSPD is a standardized developmental test that has been widely used in Japanese clinical settings because of its cultural suitability. The test provides the examinee's overall developmental age (DA) as well as the Total Developmental Quotient (DQ), which is calculated by the estimated DA divided by the child's chronological age. The test also yields DAs and DQs in three distinct developmental domains: Posture-Motor (P-M), Cognitive-Adaptive (C-A), and Language-Social (L-S). The P-M domain consists of items measuring gross motor skills (e.g., taking a few steps forward, climbing stairs using a handrail). The C-A domain contains items that assess non-verbal cognitive skills (e.g., stacking 4 blocks, pointing to correct shapes). The L-S domain includes items that assess verbal cognitive skills (e.g., recognizing specific words, repeating a sentence). According to data from the standardized sample ($N = 1589$ for age under 6) [33], the split-half reliabilities of each of the three domains were as follows: for P-M, 0.980 for age 0, and 0.888 for age 1–2; for C-A, 0.976 for age 0, 0.955 for age 1–2, and 0.931 for age 3–5; and for L-S, 0.922 for age 0, 0.938 for age 1–2, and 0.906 for age 3–5. With regard to the factorial validity of the KSPD, the scores on the three domains were moderately correlated (for children aged from 6 months to 3 years, 0.185–0.619 between P-M and C-A, 0.148–0.621 between P-M and L-S, 0.354–0.749 between C-A and L-S, depending on age). For this study, DAs were used for the analyses as the DA reflects the absolute developmental level that does not depend on the child's age.

Acquisition of walking skill. The medical records consulted for this study revealed the age at which a child

achieved unsupported walking. Unsupported walking refers to an act of moving forward by lifting and moving each foot forward in turn without any physical support while holding the head upright. The age at which a child acquired unsupported walking skill was defined as the time the physical therapist directly observed the child's unsupported walking. Because of the pattern of scheduling of physical therapy (see above), the age was recorded in units of months. In this study, the variable "acquisition of walking skill" was created using the information about the child's age at which a child acquired unsupported walking skill. The variable was coded as "0" if the child acquired the skills later than the month when he/she took the developmental test a second time, and coded as "1" if he/she acquired the skill at the same time or earlier than the month in which he/she took the developmental test.

2.3. Data collection procedures

Data were collected by reviewing medical records of the study participants obtained between April 2013 and March 2017 and stored in the Yokohama Rehabilitation Center, Japan or at one of the 4 other local rehabilitation centers in the same city. Medical records included the results of the KSPD, the written reports of physical therapists who trained the children, and the month when unsupported walking was achieved. The KSPD was administered to children with DS by a trained clinical psychologist.

This study was approved by the Institutional Review Board of the Yokohama Rehabilitation Center, Japan.

2.4. Statistical analyses

In the first part of the study, to investigate the relationship between motor development and cognitive/language development of the children with DS, the correlations among domain-level DAs (P-M DA, C-A DA, and L-S DA) were calculated. As many of the participants were tested more than once at different ages, we decided to calculate the correlations for each age group separately. Because the DA was expected to increase as the child aged for all three domains, to reduce the effect of age on the results, the age in months at which participants were tested was controlled for when correlation analyses were conducted.

In the second part of the study, we tested the hypothesis that acquisition of independent walking skill would facilitate cognitive/language development. Although a randomized controlled trial would be the best research design to test the effect of a child's achieving walking skills, in the real world we cannot control the timing of the acquisition of walking skills. Thus, we could not adopt such a study design. An alternative design would be conducting regression analyses on a longitudinal

dataset and examining whether acquisition of walking skill between two time points has a positive impact on cognitive/language functioning at the latter time. Therefore, for this study, regression analyses were conducted with acquisition of walking skill between the first and second test as the independent variable and C-A DA and L-S DA at the second test as dependent variables. In the analyses, we also controlled for age in months at the second test and C-A (L-S) DA at the initial test to more accurately examine the effect of achievement of walking.

3. Results

3.1. Cognitive/language and motor development of children with DS and the association between the two

Before the main correlational analyses, the means and standard deviations of the developmental age (DA) in motor, cognition, and language domains were calculated for participants in each age group. The results are shown in Table 1. As the children became older, their DA increased for all domains but their DQ decreased, which indicated that children with DS develop more slowly than typical children. Next, to examine the association between motor and cognitive/language development in children with DS, the correlations among P-M DA, C-A DA, and L-S DA were calculated as shown in Table 2. As can be seen in the table, P-M DA was significantly correlated with both C-A DA and L-S DA for all age groups even after controlling for the age at which the children were tested. The relationship between motor and cognitive/language development was found to strengthen with increases in age; for children aged 0–1, 2, and 3, the correlations between motor and cognitive development were 0.467, 0.531, and 0.583, respectively, and those between motor and language development were 0.315, 0.465, and 0.455, respectively.

3.2. Effect of acquisition of walking skill on later cognitive/language development

Before testing the hypothesis that acquisition of walking skills facilitates later cognitive development, we examined statistics on the age at which participants achieved unsupported walking. Of the 156 participants, 146 mastered independent walking by the time of data collection. The mean age of acquisition of walking skills was 28.71 months (SD = 8.21 months), which was consistent with previous studies. To test this hypothesis, we only analyzed data on participants who took the developmental test multiple times and could not walk at the first test. Among the 73 participants who were tested more than once, while 15 had already achieved unsupported walking by the first test (age of acquisition:

Table 1
Mean developmental age/developmental quotient for each domain at each age.

| Age (year) | Overall | | PM | | CA | | LS | |
|--------------|-----------------|-----------------|-----------------|------------------|-----------------|-------------------|-----------------|------------------|
| | DA (SD) | DQ (SD) | DA (SD) | DQ (SD) | DA (SD) | DQ (SD) | DA (SD) | DQ (SD) |
| 0–1 (n = 67) | 10.26 (2.29) | 53.39 (9.15) | 9.42 (2.95) | 48.31 (13.68) | 10.81 (2.59) | 55.31 (10.28) | 10.88 (3.14) | 55.06 (11.94) |
| 2 (n = 113) | 13.92 (3.08) | 47.83 (9.35) | 13.25 (3.70) | 45.04 (12.16) | 14.40 (3.44) | 49.40 (10.80) | 13.58 (3.41) | 46.74 (11.15) |
| 3 (n = 65) | 17.87 (3.35) | 45.08 (8.74) | 16.62 (4.27) | 41.34 (10.73) | 19.28 (4.80) | 47.26 (10.868) | 16.03 (5.51) | 39.23 (9.33) |

Note: PM = Posture-Motor, CA = Cognitive-Adaptive, LS = Language-Social, DA = Developmental age, DQ = Developmental quotient.

Table 2
Partial correlations of developmental age among each domain at different ages.

| Age (year) | PM-CA | PM-LS | CA-LS |
|--------------|----------|----------|----------|
| 0–1 (n = 67) | 0.467*** | 0.315* | 0.373** |
| 2 (n = 113) | 0.531*** | 0.465*** | 0.648*** |
| 3 (n = 65) | 0.583*** | 0.455*** | 0.765*** |

Note: PM = Posture-Motor, CA = Cognitive-Adaptive, LS = Language-Social.

Group Age 0 was combined with Group Age 1 because the former only included 2 children.

* $p < .05$.

** $p < .01$.

*** $p < .001$.

mean = 21.87 months, $SD = 4.63$ months), 58 could not walk at the time of the first test. Of those 58 participants, 38 children could walk by the second test (age of acquisition: mean = 29.50 months, $SD = 5.36$ months), but 20 were still unable to walk at that time. However, 16 of the 20 became able to walk by the time of data collection (age of acquisition: mean = 38.94 months, $SD = 6.69$ months).

The results of the regression analyses are shown in Tables 3 and 4. As expected, the achievement of walking between the first and second test had a significant positive effect on the C-A DA at the second test ($B = 2.544$, $SD = 1.250$, $p = .047$) after controlling for age at the second test and C-A DA at the first test. Similarly, when controlling for the age at the second test and L-S DA at the first test, achievement of walking was also found to

Table 3
Effect of achievement of walking on developmental age in the Cognitive-Adaptive domain at the second test.

| Variables | B | SD | β | t | p |
|------------------------|-------|-------|---------|-------|------|
| Achievement of walking | 2.544 | 1.250 | 0.240 | 2.035 | .047 |
| Age at 2nd test | 0.167 | 0.122 | 0.168 | 1.363 | .179 |
| CA at 1st test | 0.722 | 0.231 | 0.375 | 3.132 | .003 |

$R^2 = 0.305$ ***

Note: N = 58, CA = Cognitive-Adaptive.

*** $p < .001$.

have a significant positive effect on the L-S DA at the second test ($B = 3.292$, $SD = 1.442$, $p = .026$).

4. Discussion

This study investigated the relationship between motor and cognitive/language development among children with DS who received systematic and intensive rehabilitation in Yokohama city. The results showed that motor development was significantly correlated with both cognitive and language development at all time points between 1 and 3 years of age, which accords with the relationship observed among normative sample of the KSPD [33]. Our results were consistent with Kim et al.'s findings [14] that showed a non-significant but positive relationship between motor and cognitive development.

It is also interesting to point out that the relationship between motor and cognitive/language development was found to strengthen with increases in age, as can be seen in Table 2. This is inconsistent with the pattern observed among typical children that the relationship became weaker as the age of children increased [33]. Based on our clinical experiences, such a difference is considered to occur due to their difference in timing of achieving various developmental milestones. For example, around age one, abilities to walk and speak start to vary among typical children, while almost none of children with DS can walk or speak; in other words, variety of motor and cognitive/language development among children with DS is relatively small around this age. As they become age 2 or 3, they started to acquire and improve walking and speaking skills, which might have increased variety in their motor and cognitive/language abilities. In fact, children with DS in our data showed such an increase in variety of developmental level as they became older (see SDs in Table 1). The increase in variety among children is considered to eventually result in strengthening the correlation between motor and cognitive/language skills among children with DS.

With regard to the effect of achievement of walking skills on cognitive and language development, the results

Table 4
Effect of achievement of walking on developmental age in the Language-Social domain at the second test.

| Variables | B | SD | β | <i>t</i> | <i>p</i> |
|------------------------|--------|-------|---------|----------|----------|
| Achievement of walking | 3.292 | 1.442 | 0.262 | 2.282 | .026 |
| Age at 2nd test | −0.018 | 0.141 | −0.015 | −0.124 | .902 |
| LS at 1st test | 1.169 | 0.270 | 0.505 | 4.333 | ≤.001 |

$R^2 = 0.339^{***}$

Note: N = 58, LS = Language-Social.

*** $p < .001$.

supported our hypothesis that acquisition of walking skills facilitated cognitive and language development in children with DS, which was also shown in typical children and pre-term children [26–30]. As noted above, it has been considered that the achievement of walking enables children to explore their environment more broadly due to expansion of the range of movement, which in turn would increase the amount and variety of stimuli children are exposed to and facilitate cognitive and social development [20–25].

It is also possible that improvement in the various skills that are required to walk would facilitate cognitive and language development. According to studies of adults [34], appropriate gait requires not only motor skills, but also various other abilities, such as attention, executive function, and judgment of the environment and internal physical cues, which are regarded as bases of cognitive skills. In fact, based on neurological studies, some brain regions, such as the caudate nucleus and prefrontal cortex, which were found to be important for movement initiation and control, also appeared to be important for cognitive functioning [35]. This indicates the possibility that not acquisition of the ability to walk itself, but development of several other skills accompanying the mastery of walking may facilitate cognitive/language development.

The current study had several limitations. First, it is not clear whether the findings can be generalized to children with DS who do not receive the degree of intensive intervention as the participants in this study received. Children with DS who received early intervention were found to show better intellectual functioning and adaptive skills than those who did not, possibly due to increased sophisticated stimuli provided during the intervention [36]. Therefore, such experiences associated with the intervention might have moderated the effect of acquisition of walking on cognitive/language skills. Second, the age of the participants was restricted to 0–3 years because of the test battery used for the developmental evaluation. In the rehabilitation centers in Yokohama city, once children become around the age of 4, they are likely to be assessed using a different test (i.e., Tanaka-Binet intelligence test [37]) that does not measure motor development. For that reason, children 4 years or older were excluded from this study. There-

fore, it is unclear whether the relationship between motor and cognitive/language development is also found among older children with DS. Third, it is possible that KSPD measures somewhat different aspects of motor and cognitive/language abilities from those assessed in other developmental/intelligence tests (e.g., Tanaka-Binet Intelligence Test [37], Bailey Scales of Infant and Toddler Development [38]). To generalize the findings, it is necessary to replicate the findings of this study using other test batteries in future studies. Fourth, as we were not able to control the timing of the acquisition of walking skills, it is possible that the improvement in cognitive/language skills between the first and second tests cannot be explained by acquisition of walking, but by other factors that are associated with the acquisition of skills. Although in this study there were no relationships between the fact that children became able to walk between the first and second tests and several prenatal conditions, including low birth weight, preterm birth, and congenital heart disorders ($\chi^2(1) = 0.525, 0.345, 0.392$, respectively; $p > .10$ for all three variables), it is still possible that other factors that could not be ascertained from the data would explain the effect. Finally, we did not consider any moderating factors in this study; the relationship between motor and cognitive/language development might be different depending on other factors, such as children's temperament, physical complications, parents' educational background, and home environment.

Despite these limitations, this study provided evidence of a relationship between motor and the cognitive/language abilities in children with DS as well as a positive effect of acquisition of walking skills on cognitive/language development. Specifically, the findings emphasized the importance of acquisition of walking skills as a core facilitator of further development of children with DS. This information would be helpful for determining specific components of intervention programs for children, as well as the timing of their administration to each child. In future studies, it is desirable to replicate the findings of this study with older children and explore factors that would moderate and mediate the relationship between motor and cognitive/language development among children with DS. As an expansion of this study, it would be important to directly investi-

gate the effect of motor interventions on cognitive/language development among children with DS.

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