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ORIGINAL RESEARCH

Mobility in preschool age children with and without Down syndrome: an exploratory cross-sectional study

Mobilidade em pré-escolares com e sem síndrome de Down: um estudo transversal exploratório

Movilidad en preescolares con y sin síndrome de Down: un estudio transversal exploratorio

Barbara Raiza Taranto Silva[1], Marina Almeida de Souza[2], Isabella Saraiva Christovão[3], Ana Cristina Resende Camargos[4]

ABSTRACT | Few studies have investigated the mobility of preschool age children with Down syndrome (DS). This study aimed to compare the mobility of preschool age children with and without DS and to verify if cognitive function and gait acquisition age may explain mobility outcomes. This was an exploratory cross-sectional study involving 38 children: 19 in the DS group and 19 in the typical development (TD) group. The 10-meter walk test and the modified Timed Up and Go (mTUG) test were used to evaluate mobility. The explanatory factors were the cognitive function screening test score and the age of gait acquisition. Stepwise multiple linear regression models were used. The children in the DS group had slower gait speed (p=0.0001) and took longer to complete the mTUG test (p=0.0001). The cognitive function screening test score and age of gait acquisition explained the variability in gait speed (R^2=0.52; p=0.0001) and the variability in the time to complete the mTUG test (R^2=0.68; p=0.0001). Children with DS showed a poorer mobility when compared to the children in the TD group. The outcomes of mobility in this age group were partially explained by the age of gait acquisition and the cognitive function screening test score.

Keywords | Down Syndrome; Walking Speed; Mobility Limitation; Cognition.

RESUMO | Poucos estudos investigaram a mobilidade de pré-escolares com síndrome de Down (SD). Dessa forma, os objetivos desta pesquisa foram comparar a mobilidade de pré-escolares com e sem SD, bem como verificar se a função cognitiva e a idade de aquisição da marcha podem explicar os desfechos de mobilidade. Estudo transversal exploratório com 38 crianças: 19 do grupo SD e 19 do grupo desenvolvimento típico (DT). O teste de caminhada de 10 metros e o *timed up and go* modificado (mTUG) foram utilizados para avaliar a mobilidade. Os fatores exploratórios foram: a pontuação da triagem da função cognitiva e a idade de aquisição da marcha. Foram usados modelos de regressão linear múltipla *stepwise*. As crianças do grupo SD apresentaram menor velocidade de marcha (p=0,0001) e necessitaram de mais tempo para completar o mTUG (p=0,0001). A pontuação da triagem da função cognitiva e a idade de aquisição da marcha explicaram a variabilidade na velocidade da marcha (R2=0,52; p=0,0001) e o tempo para completar o teste (R2=0,68; p=0,0001). Crianças com SD apresentaram pior capacidade de mobilidade quando comparadas às com DT. Os desfechos de mobilidade nessa faixa etária foram parcialmente explicados pela idade de aquisição da marcha e pelo escore da triagem da função cognitiva.

Descritores | Síndrome de Down; Velocidade de Caminhada; Limitação de Mobilidade; Cognição.

[1]Universidade Federal de Minas Gerais (UFMG), Departamento de Fisioterapia. Belo Horizonte (MG), Brazil. E-mail: brtaranto@gmail.com. ORCID-0009-0002-6863-335X
[2]Universidade Federal de Minas Gerais (UFMG), Departamento de Fisioterapia. Belo Horizonte (MG), Brazil. E-mail: marinafisioufmg@gmail.com. ORCID-0009-0007-7962-8771
[3]Universidade Federal de Minas Gerais (UFMG), Programa de Pós-Graduação em Ciências da Reabilitação. Belo Horizonte (MG), Brazil. E-mail: isabellasaraiva_@hotmail.com. ORCID-0000-0002-6599-4427
4Universidade Federal de Minas Gerais (UFMG), Departamento de Fisioterapia, Programa de Pós-Graduação em Ciências da Reabilitação. Belo Horizonte (MG), Brazil. E-mail: brtaranto@gmail.com. ORCID-0009-0002-6863-335X

Corresponding address: Ana Cristina Resende Camargos – EEFFTO – Escola de Educação Física, Fisioterapia e Terapia Ocupacional. Universidade Federal de Minas Gerais (UFMG) – Av. Pres. Antônio Carlos, 6627 Campus Pampulha – Belo Horizonte (MG), Brazil – Zip code: 31270 – E-mail: anacristinarcamargos@eeffto.ufmg.br – Financing source: nothing to declare – Presentation Apr. 21st, 2023 – Accepted for publication: Feb. 29th, 2024 – Approved by the Research Ethics Committee: CAAE 96264118.3.0000.5149.

RESUMEN | Son pocos estudios que han investigado la movilidad de preescolares con síndrome de Down (SD). En este contexto, los objetivos de este estudio fueron comparar la movilidad de los preescolares con y sin SD, así como comprobar si la función cognitiva y la edad de adquisición de la marcha pueden explicar los resultados de la movilidad. Se trata de un estudio transversal exploratorio con 38 niños: 19 del grupo con SD y 19 del grupo con desarrollo típico (DT). Para evaluar la movilidad se utilizaron la prueba de marcha de 10 metros y la prueba de levantarse y andar cronometrada modificada (mTUG). Los factores exploratorios fueron la puntuación del cribado de la función cognitiva y la edad de adquisición de la marcha. Se utilizaron modelos de regresión lineal múltiple por pasos. Los niños del grupo con SD tenían una velocidad de marcha inferior (p=0,0001) y necesitaban más tiempo para completar la mTUG (p=0,0001). La puntuación del cribado de la función cognitiva y la edad de adquisición de la marcha explicaron la variabilidad en la velocidad de la marcha (R2=0,52; p=0,0001) y el tiempo para completar la prueba (R2=0,68; p=0,0001). Los niños con SD tuvieron peor movilidad en comparación con los niños con DT. Los resultados de la movilidad en este grupo de edad se deben parcialmente a la edad de adquisición de la marcha y a la puntuación del cribado de la función cognitiva.

Palabras clave | Síndrome de Down; Velocidad al Caminar; Limitación de la Movilidad; Cognición.

INTRODUCTION

Down syndrome (DS) is a genetic condition characterized by an inappropriate distribution of chromosomes, which causes a full or partial extra copy of chromosome 21[1]. All individuals with DS present an alteration in cognitive function[2,] and this condition is also associated with a delayed development of gross and fine motor skills[3-5]. Children with DS need more time to acquire new skills and have a motor development repertoire that differs from that of children with typical development (TD)[6].

Regarding gross motor skills, children with DS have a noticeable delay in independent gait acquisition: for them, this process happens between 18 months and three years of age, whereas in children with TD, it occurs at approximately 13 months of age. Moreover, with increasing motor complexity, children with DS may need more time to learn movements such as running, jumping, and climbing stairs[6]. In clinical practice, physical therapy interventions in children with DS are considerably reduced or ceased after gait acquisition. However, the significance of gait acquisition extends beyond the acquisition of new motor abilities. The development of motor skills offers infants novel chances to explore and understand the environment[4]. Furthermore, there are developmental difficulties related to changes in static and dynamic balance[7,8]. Thus, children with DS have difficulty initiating and maintaining movements, as well as adapting them[6] to different task complexities and to changing environmental conditions[7,8], which can reduce their mobility[9] and restrict their participation in recreational activities with peers[10].

Mobility can be assessed with quick, simple and cost-effective tests such as the 10-meter walk test and the timed up and go (TUG) test. The 10-meter walk test have been used to assess the self-selected gait speed of healthy children[11] and children with neuromuscular disease[12], and it can easily be conducted in a clinical or home environment without sophisticated equipment. The timed up and go (TUG) test was developed to assess the mobility of older adults when carrying out activities that may increase the risk of falling, including activities performed on a daily basis, such as changing basic body positions, maintaining a body position, walking, and moving[13]. The original test evaluates the time (in seconds) it takes for an individual to rise from a standard armchair, walk three meters, turn around, walk back to the chair and sit down again[13]. However, for testing children and adolescents, a modified TUG (mTUG), which is also validated for typical preschool age children, is recommended[14]. This modification was employed in the validation study of the test for children with DS[9].

Few studies have investigated the mobility of preschool age children with DS[9,15], and no studies evaluating the factors that may explain mobility outcomes have been found in the literature. A delay in cognitive development can influence motivation and the ability to learn and practice motor skills, including walking[4,5]. Considering that motor and cognitive development are essential and closely intertwined[16],

it is important to verify whether cognitive functions and the age of gait acquisition can explain mobility outcomes in preschool age children.

In preschool age children with DS, mobility is an important prerequisite for independence and participation in social and recreational activities[10,17]. Thus, it is necessary to identify factors that may explain mobility capacity. Understanding these factors may assist in the planning of therapeutic interventions proposed by rehabilitation teams. Therefore, this study aimed to compare the mobility of preschool children with and without DS and to verify if cognitive function and the age of gait acquisition can explain mobility outcomes in children this age.

METHODOLOGY

Study design

An exploratory cross-sectional study was conducted from November 2018 to April 2019.

Population

Participants in this study, that is, children aged from three to five years old, were divided into two groups: the Down syndrome (DS) group and the typical development (TD) group.

Local

Children were recruited from associations, institutions and municipal preschools in Belo Horizonte, Minas Gerais, Brazil.

Selection criteria

To be included in the DS group, children had to have a clinical diagnosis of DS, be able to walk independently and be able to follow verbal instructions and commands to perform the tests, regardless of whether they were undertaking physical therapy treatment or not. Children who had significant visual or hearing impairments or whose parents refused to participate were excluded. For each child in the DS group, one child of the same sex and age was included in the TD group. In the TD group, children who were under medical supervision or had undergone physical therapy treatment were excluded.

Sample definition

The sample size calculation was based on Mancini et al.[18] for the mobility variable. Considering a 0.80 power and a 0.05 alpha, it was calculated that each group should include 19 children, totaling 38 children.

Data measures

To assess mobility outcomes, the 10-meter walk and the mTUG tests were used. The 10-meter walk test was used to document gait speed, reliable for children with neuromuscular disease (ICC=0.91)[12], and the time it took for each child to walk a distance of 10 meters was recorded using a stopwatch. The verbal command "Go" was used to start the test, and the child was encouraged to walk as fast as possible, without running. The test was performed in a 14-meter linear space that was measured with a tape measure and outlined with colored tape. The initial two meters and the final two meters were disregarded for speed calculation[19]. Gait speed was calculated using the equation speed = distance/time. Three repetitions were performed and the mean between them was used for the analysis.

The mTUG test was performed to measure the time it took for each child to get up from a bench, walk three meters, return and sit again. The mTUG test is a modified version of the TUG test, a reliable instrument to evaluate preschool age children without (ICC=0.95) and with DS (ICC=0.82)[9]. The test was performed by placing an armless bench three meters from a wall. Children sat with their feet flat on the floor so that their hips and knees remained flexed at a 90° angle. The task was to stand up, walk three meters, touch a child character figure on the wall, turn around, return to the bench and sit down again. The distance was measured with a tape measure, and the task was explained and demonstrated before the test. The verbal command "Ready, aim and go" was used for the child to start the test. Positive encouragement was provided as needed throughout the trials to promote better performance. The timer started when the child

got up from the bench and ended when they sat back down[15]. Three repetitions were performed, and the mean time was used for the analysis.

To assess cognitive function, the mini-mental state examination (MMSE), adapted for children, was applied. The MMSE is a test that evaluates and monitors five domains of cognitive function: orientation, attention/concentration, registration, recall, and language; and its adapted version can be used in children aged 3–14 years[20]. All children were given the same instructions during the test, and the examiner verbally asked them the questions. The MMSE score ranges from zero to 37 points, and a 24-point cut-off, established for typical children aged three to five years, was used to characterize the participants[20].

In addition, the children's parents and/or guardians individually responded to a questionnaire about the age of gait acquisition. Parents recall of motor milestones is considered accurate up to a 5-year follow-up[21]. Data on sex, date of birth, and participation in physical and/or sports activities and physical therapy interventions were also collected. The economic level of the families was assessed according to the Brazilian Criteria of Economic Classification[22]. The overall economic classification resulting from this criterion ranged from A to E.

Mobility measurements were administered by a single trained examiner and the MMSE was applied by another trained examiner. Each child was assessed individually in an appropriate place for the tests.

Data analysis

Data were analysed using SPSS version 20.0. Initially, descriptive statistics were used to characterize the sample. The Chi-squared test or Fishers exact test were performed to compare the proportions of the two groups. The Shapiro-Wilk test was used to verify data normality, and the Levene's test was used to verify homogeneity of variance. To compare the means between the two groups, the Student's t-test for independent samples was performed. A simple linear regression analysis was performed to verify the association between the cognitive function and age of gait acquisition and the outcomes (gait speed and mTUG). Finally, stepwise multiple linear regression models were used to verify the association between the explanatory factors and the outcomes. Since children's economic level and engagement in physical therapy or sports could be confounding factors, the analysis was adjusted for these variables. The residual analysis showed normal distribution and homogeneous variance in all regression models. The magnitude of the effect (d) was calculated.

Ethical aspects

After approval by the Research Ethics Committee, the associations, institutions and pre-schools that agreed to participate in the study signed a letter of consent authorizing the experiment to be conducted in their spaces. Children were included only after assent, and parents signed an informed consent form.

RESULTS

The children evaluated, 28 girls and 10 boys, were divided into two groups (19 in the DS group and 19 in the TD group). Table 1 shows the characteristics of each group. The groups were homogeneous regarding age, sex, economic level and sports practice ($p>0.05$). The groups differed in relation to the age of gait acquisition, which was greater for the children of the DS group, and the MMSE score, which was lower for the DS group (Table 1). Regarding the MMSE classification, all children with DS (100%) were below the cut-off point expected for their age. In the TD group, only seven children (36.8%) were within normal limits, and 12 children (63.2%) remained below the cut-off point expected for their age. All children in the DS group had already undergone physical therapy, and 12 children (63.2%) were currently undergoing treatment. In the DS group, two children (10.5%) practiced swimming, and one child (5.3%) practiced both swimming and ballet. In the TD group, two children (10.5%) performed sports initiation, and one child (5.3%) practiced artistic gymnastics.

When comparing the outcomes of the two groups, significant differences were observed in the gait speed and time required to perform the mTUG test. Children in the DS group had a slower gait speed ($p=0.0001$) and took longer to complete the mTUG test ($p=0.0001$) than children in the TD group, with a large effect size and statistical power (Table 2).

The age of gait acquisition and MMSE score were found to be significantly associated with gait speed ($R^2=0.38$, $p=0.0001$; $R^2=0.47$, $p=0.0001$) and the time for mTUG test ($R^2=0.64$, $p=0.0001$; $R^2=0.48$, $p=0.0001$), respectively.

Table 1. Down syndrome and typical development group characteristics. Belo Horizonte, Minas Gerais, Brazil, 2018-2019.

Characteristic	DS Group (n=19)	TD Group (n=19)	p value
Age (months)	51.32 (±9.24)	51.95 (±8.93)	0.83
Age of gait acquisition (months)	26.52 (±10.02)	12.53 (±2.29)	0.0001*
MMSE Score	6.31 (±5.58)	22.42 (±2.76)	0.0001*
Sex			1.00
Female	14 (73.7%)	14 (73.7%)	
Male	5 (26.3%)	5 (26.3%)	
Economic level			0.41
A	1 (5.3%)	3 (15.8%)	
B	9 (47.4%)	11 (57.9%)	
C	8 (42.1%)	5 (26.3%)	
D/E	1 (5.3%)	0 (0%)	
Sports practice			1.0
Yes	3 (15.8%)	3 (15.8%)	
No	16 (84.2%)	16 (84.2%)	

Caption: DS=Down syndrome; TD=typical development; MMSE=Mini Mental State Exam.

Table 2. Comparison of values obtained from the functional mobility test for the Down syndrome and typical development groups. Belo Horizonte, Minas Gerais, Brazil, 2018-2019.

Parameter	DS group	TD group	95% CI	d
Gait speed (m/s)	1.04 (±0.29)	1.46 (±0.29)	−0.61;−0.23	1.45
mTUG (s)	11.13 (±4.27)	6.19 (±1.10)	2.88;6.99	1.58

Caption: DS=Down syndrome; TD=typical development; CI=confidence interval; d=effect size; mTUG=modified Timed Up and Go.

Table 3 shows the results of the stepwise multiple linear regression analysis. The MMSE score alone explained 47% (p=0.0001; d=0.89) of the variability in gait speed. When the age of gait acquisition was included in the model, the explanation of gait speed variability increased to 52% (p=0.0001; d=1.08). When the predictors for the mTUG were analyzed, the age of gait acquisition alone could explain 64% (p=0.0001; d=1.78) of the variability in the time required to perform the mTUG test. When the MMSE score was included in the model, the explanation for the variation in the time for the mTUG test increased to 68% (p=0.0001; d=2.13). Lastly, economic level, physical therapy performance and sports practice were inserted in the regression model, but they did not affect the values of β and R^2.

Table 3. Stepwise multiple linear regression analysis between the factors and mobility outcomes (gait speed and modified timed up and go). Belo Horizonte, Minas Gerais, Brazil, 2018-2019.

Model	B (95% CI)	B	p	R^2	SEE	df
Gait speed (m/s)						
Model 1						
Constant	0.87 (0.71–1.03)	-	0.0001*	-	-	
MMSE score	0.03 (0.02–0.04)	0.69	0.0001*	0.47	0.26	1.36
Model 2						
Constant	1.20 (0.85–1.55)	-	0.0001*	-	-	
MMSE score	0.02 (0.007–0.03)	0.49	0.002*	-	-	
Age of gait acquisition (months)	−0.01 (−0.02–−0.001)	−0.32	0.04*	0.52	0.25	2.35
mTUG (s)						
Model 1						
Constant	2.04 (0.19–3.88)	-	0.03*	-	-	
Age of gait acquisition (months)	0.35 (0.26–0.44)	0.80	0.001*	0.64	2.38	1.35
Model 2						
Constant	5.59 (2.18–9.00)	-	0.002*	-	-	

(continues)

Tabela 4. Continuation

Model	B (95% CI)	B	p	R^2	SEE	df
Age of gait acquisition (months)	0.26 (0.15–0.37)	0.60	0.0001*	-	-	
MMSE score	−0.13 (-0.24–−0.02)	−0.31	0.02*	0.68	2.22	2.34

Caption: MMSE=mini mental state exam; mTUG=modified timed up and go; β=standardized regression coefficient; CI=confidence interval; B=regression coefficient; R^2=adjusted coefficient of determination; SEE=standard error of the estimate; df=degrees of freedom.
* Showed p-value< 0.05.

DISCUSSION

This study found that children with DS performed more poorly than their peers in mobility tests, presenting lower gait speed and taking longer to complete the mTUG test. Moreover, cognitive function and age of gait acquisition were considered significant factors that explained mobility outcomes. Notably, this is the first study to evaluate predictors that explain the variability of mobility outcomes in preschool age children with and without DS. The cognitive function screening test score was considered the main factor to explain gait speed variability, and age of gait acquisition was the main factor explaining the time taken to perform the mTUG test.

This study outcomes corroborate the findings in the literature that also indicate that children with DS need more time to perform the mTUG test.[9,15,23] Normative data from healthy children and adolescents demonstrated that the time it took for these groups to perform the mTUG test could be explained, at least partly, by age and body weight[9]. In typical preschool age (3–5 years) children, the normative value for the mTUG test time was 6.58 seconds[9]. In our study children in this age group took a mean of 6.19 (±1.10) seconds to complete the mTUG. A shorter time to perform the test is indicative of greater mobility[15]. In contrast, children with DS in the study performed the mTUG in a mean time of 11.13 (±4.27) seconds. In the study by Nicolini-Panisson and Donadio[9], the mTUG values of children with DS were more closely associated with their gross motor capacity, evaluated using the Gross Motor Function Measure (GMFM), than to variations in age and body weight. Therefore, higher mTUG values were associated with a lower capacity to walk, run, and jump.

Regarding the mTUG, age of gait acquisition was considered the main predictor and explained 64% of the variability in the time it took to perform the test. In this study, it was identified that the older the age of gait acquisition, the longer it took to perform the mTUG; an increase of one month in the age of gait acquisition led to an increase of six seconds in the time to perform the mTUG. According to Palisano et al.[6], children with DS started to walk independently from 18 months to three years of age. In the present study, it was found that children with DS had a significant delay in age of gait acquisition (26.52 months) when compared to their peers (12.53 months). Parents of infants with DS identify walking as one of the goals they value the most[24]. The ability to walk independently is important for infants to learn about the world, as it improves their active exploration of the environment, and can positively impact on cognitive, social, and emotional skills[25].

The cognitive impairment, evaluated using the MMSE score, was also significantly associated with the mTUG score. When the cognitive function scores were inserted into the model, the explanation of variability in mTUG time increased to 68%, demonstrating that the lower the MMSE score, the longer the time spent performing the mTUG. Other studies have also found an association between cognitive function and motor skills[3-5]. The MMSE can quickly assess cognitive function impairment in children, and cut-off scores two standard deviations below the mean for different ages can be used to identify early cognitive dysfunction and monitor the progression of the conidition[20]. In Brazil, this instrument has been used to screen cognitive impairment in children with cerebral palsy[26], however, no studies were found using the MMSE in DS children. In our study, the test score was used to verify whether higher scores were associated with better outcomes, and an increase of one point in the MMSE score was effectively associated with a decrease of approximately three seconds in the mTUG test.

Furthermore, children with DS were also found to have a lower gait speed than children with TD. Gait speed is one of the essential characteristics of human gait[12], and healthy preschool age children showed improvements in gait speed as they aged[11]. According to Cimolin et al.[27], children with DS show kinetic and kinematic compensatory strategies and walk with a longer stance duration, reduced anterior step length, lower speed of progression and lower propulsion capacity. In the present study, children in the DS group

had a mean gait speed of 1.04 m/s, while children in the TD group had a mean gait speed of 1.46 m/s. In the study by Pereira et al.[11], children with TD aged two to six years showed slower gait than those found in our study (mean 0.77 m/s). However, these authors calculated children's usual gait speed, not asking them to walk as fast as possible without running, differing from our methodology.

The MMSE score was considered the main predictor of gait speed and was explained 47% of gait speed variability. In the present study, children with lower MMSE scores had a lower gait speed. Other studies have also shown an association between cognitive function and walking ability[28,29]. Authors such as Amboni et al.[28] noted that walking ability requires not only motor skills, but also cognitive skills such as attention and assessment. In addition to the MMSE score, age of gait acquisition was also able to predict gait speed variability. When this variable was included in the model, the explanation of gait speed variability increased to 52%. Also, an increase of one point in the MMSE score was associated with a decrease of approximately three seconds in the duration of 10-meter walk test, and an increase of one month in the age of gait acquisition led to an increase of approximately five seconds in the duration of 10-meter walk test. The age of gait acquisition is related to the length of time of independent gait. In the study by Rodriguez et al.[30], increased gait speed was associated with independent walking practice time. Thus, the younger the age of gait acquisition, the longer the practice time and the faster the gait speed.

Other variables that could influence mobility outcomes are physical therapy and sports practice, which can positively interfere with motor performance[31]. Children's motor development can also be influenced by their economic level[32]. However, in this study, when these variables were inserted in the regression model, they did not interfere with the mobility outcomes.

Some aspects of this study may limit the interpretation of the results. The first aspect refers to the difficulty children in this age group have in understanding and obeying commands, especially children with DS. Although these tests have already been validated and are reliable for use in age group[9,11], several demonstrations had to be made before initiating data collection. The mTUG has already been validated for children with Down syndrome, but the 10-meter walk test has not yet been validated for this health condition. In the study by Pereira et al.[11], the 10-meter walk test was conducted with children with typical development, aged from two to 12 years old, and the variability of the data was similar among age groups, which indicates that this test can be used in children in this age group. The second limiting aspect is related to the MMSE score. In our study, cognitive function was assessed using the MMSE, a cognitive function screening test, adapted for children, and the cut-off point used was established for Indian children aged 3–5 years[20]. In our study, all children with DS had results below the expected cut-off point for their ages, and 12 children (63.2%) in the TD group also obtained scores below those expected for their age group. According to Peviani et al.[33], this test is a useful tool for monitoring cognitive development of children aged from 36 to 72 months, but normative data is only available for Brazilian children aged five years and over[26]. Since the MMSE scores range from 0 to 37 points, numerical variables (instead of categories) were used in the regression analysis. Lastly, the third limiting aspect refers to the fact that children's height, weight, and body mass index (BMI) were not measured, and these variables could also influence gait speed outcomes[34] and mTUG times[23].

This study aimed to evaluate the mobility of preschool age children using simple, quick and cost-free tests that can be easily used in clinical practice. Thus, standardized tests that require more time and effort to be evaluated were not used. This study outcomes may contribute to clinical practice, since children with DS had worse mobility outcomes, which need to be considered at preschool age. Neal et al.[10] demonstrated that the main goals set by parents of students with DS who received school-based physical therapy were related to mobility outcomes. Children with DS need to learn motor skills and perform them with greater independence, speed, ease, and safety, in order to use them effectively within a variety of meaningful contexts[17]. Mobility is essential to promote participation in recreational activities with peers[10]. Moreover, mobility outcomes in individuals with DS tend to decline with age, thus, it is important to carry out interventions that limit the progression of such impairment[35]. Adolescents and adults with DS engage less in physical activity than their peers[36], presenting a more sedentary lifestyle, which makes them more likely to gain weight and increases their risk of cardiovascular disease[3]. All of these factors can lead to activity limitations and participation restrictions[37], making it necessary to consider long-term health impacts. Future investigations into older children can

be carried out to verify the effects of cognition, age of gait acquisition and other factors on mobility outcomes, in order to ensure better implications in clinical practice.

CONCLUSIONS

Children with Down syndrome performed more poorly in mobility tests than children with typical development. Mobility outcomes were explained by the age of gait acquisition and MMSE score. The age of gait acquisition was considered the main predictor of the variability in the time spent on the mTUG, and cognitive function was the main predictor of gait speed variability. The identification of predictors that influence mobility outcomes may help rehabilitation teams plan individualized therapeutic interventions that meet children's real needs.

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