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Full Length Research Paper

Reliability of balance tests in children with Duchenne muscular dystrophy

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The first aim of the study was to investigate the test-retest reliability of functional balance tests in children with Duchenne muscular dystrophy (DMD) and the second aim was to examine the relation between balance tests and motor functions in children with DMD. Timed up and go test (TUG) and functional reach test (FRT) were used for the evaluation of balance. Hammersmith motor ability scale (HMAS) was used to assess motor functions. Test-retest reliability was determined by using intra-class correlation coefficient (ICC). Spearman correlation coefficient was used to assess the relation between balance tests and HMAS. Both TUG and FRT had good test-retest reliability. ICC score for TUG was 0.86 (95%, CI=0.69 to 0.94) and 0.96 (95%, CI=0.92 to 0.98) for FRT. A significant negative correlation was found between TUG and HMAS with a correlation coefficient of r_{ho}= -0.69, p<0.01. A significant positive correlation was found between FRT and HMAS with a correlation coefficient of r_{ho}= 0.47, p<0.05. Our results provide evidence that, FRT and TUG are reliable measures and may be used to monitor change over time, particularly following interventions that aim to improve motor functions in children with DMD.

Key words: Duchenne muscular dystrophy, balance tests, reliability.

INTRODUCTION

Balance is the ability to maintain the body's center of gravity within the limits of stability as determined by the base of support (Horak, 1987). Children with many types of disabilities have been shown to have balance problems (Rose et al., 2002; Shum and Pang, 2009; Galli et al., 2008; Gagnon et al., 2004; Tsai et al., 2008). Duchenne muscular dystrophy (DMD) is a genetic disorder, characterized by the progressive weakness of skeletal muscles due to dystrophin protein deficiency in the muscle membrane (Uchikawa et al., 2004). Progressive muscle weakness and joint contractures lead to a poor standing balance (Vignos, 1968; Kelly et al., 1981), which is associated with an increased risk of fractures due to falls in DMD (McDonald et al., 2002; Yiu and Kornberg, 2008). Balance impairment can also restrict the mobility, independence and social participation due to a problem like fear of falling that further results in deconditioning, functional decline and poorer quality of life (Murphy et al., 2002; Franchignoni et al., 2005).

Despite the rapid advances in basic research on the etiology and pathophysiology of DMD, there is yet no cure (Liu et al., 2003). All therapeutic approaches like steroid therapy, strengthening and stretching exercises, and orthotic management aim to improve standing balance and prolong independent ambulation in order to delay confinement to a wheelchair along with its associated rapid progression of scoliosis (Parreira et al., 2007; Manzur and Muntoni, 2009). Standardized and reliable measures are needed in order to take appropriate and timely decisions about therapy plan, monitor the natural course of the disease and to evaluate the effect of different treatment modalities (Kembhavi et al., 2002) in DMD.

There are several tests that can be used for the evaluation of standing balance in children. We chose timed up and go test (TUG) and functional reach test

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Table 1. Descriptive characteristics of the children.

N = 23	Mean (SD)
Age (years)	7.17(1.26)
Height (cm)	120.3(11.19)
Weight (kg)	23.36(5.57)

SD: Standart deviation.

(FRT), because of their direct relevance to functional abilities, ease to use in a clinical setting in a short time without requiring any expensive equipment (Yim-Chiplis and Talbot, 2000). TUG was developed by Podsiadlo and Richardson (1991) for the assessment of basic functional ambulatory mobility, or dynamic balance. FRT was developed by Duncan et al. (1992) and it measures the margin of stability during a voluntary forward maximal reach. Both TUG and FRT were shown to provide reliable measurements in children with and without disabilities (Donahoe et al., 1994; Niznik et al., 1995; Williams et al., 2005), however the use and the reliabilty of these tests had not been examined in children with DMD. Therefore, the aim of our study was to investigate the test-retest reliabilty of TUG and FRT in children with DMD.

Balance is essential for the achievement of gross motor abilities (Kembhavi et al., 2002). Therefore, poor balance causes difficulties in activities of daily living involving simple or complex gross and fine motor tasks. In order to investigate how balance problems influence gross motor abilities we examined the relation between balance tests and motor functions in these children.

MATERIALS AND METHODS

The children who were diagnosed as DMD in Hacettepe University Pediatric Neurology Unit, Ankara, Turkey, and referred to Hacettepe University Faculty of Health Sciences, Physiotherapy and Rehabilitation Department for physiotherapy program were asked to join the study. The study was held between August-November 2007. 41 children met the inclusion criteria and 23 of them accepted to join the study. The children who were able to follow instructions, walk unassisted, rise from a chair independently (Vignos scale≤ 4) (Vignos et al., 1963) and achieve 90° shoulder flexion were included in the study. The study protocol was approved by the Ethical Committee of the Institute and informed consent was obtained.

Children's basic demographical data was recorded before the test procedure. Balance evaluations were carried out in a quiet room relatively free from disturbances that could affect the test results. For test-retest reliability, the balance tests were performed two times with a one week interval by the same physiotherapist with eight years of clinical experience in pediatric rehabilitation. Each test was performed three times and the average was used in statistical analysis. During the test procedure as needed, rest period was given for each participant.

For FRT, the child was positioned perpendicular to the wall with feet parallel in comfortable position. The shoulder of the dominant extremity flexed to 90° and the elbow extended without touching the wall. A tape measure was fixed to the wall parallel to the floor at the height of the child's acromion. The point corresponding to the tip of the third finger was marked, and then the child was asked to reach as forward as possible without taking a step or touching the wall. The point corresponding to the tip of the third finger was marked again. The distance between these two points was recorded in centimeters (cm) as a test result (Duncan et al., 1992).

For TUG, the child sat on a standard arm chair. On the word 'go', the child was asked to stand up, walk to a line on the floor 3 m away with their own comfortable speed, turn, walk back to the chair and sit down. A stopwatch was used to time the performance in seconds (s) (Podsiadlo and Richardson, 1991).

Gross motor functions were evaluated with Hammersmith motor ability scale (HMAS). HMAS was used to assess motor functions with an ability score based on 20 consecutive motor activities like get off a chair, standing and climbing stairs. It was graded on a three point (0-1-2) scale and the maximum score was 40 (Scott et al., 1982). The scale was designed by Scott et al., and is widely used for the evaluation of motor functions in DMD (Smith et al., 1991; Scott and Mawson, 2006; Mazzone et al., 2009).

SPSS statistical software was used for data analyses. Means and standard deviations of scores for each test were calculated. Test- retest reliability was determined by using intra-class correlation coefficient (ICC). The 95% confidence intervals for all ICCs were calculated. Spearman correlation coefficient was used to assess the relation between balance tests and HMAS. A p value of <0.05 was considered to indicate statistical significance.

RESULTS

The mean age of the children who were included in the study was 7.17 ±1.26 years, ranging from 5 to 10 years. 16 of the children were in stage one and 7 of them were in stage two according to Vignos scale. Table 1 shows the descriptive characteristics of the children.

Table 2 presents the means, standard deviations, ICCs and 95% confidence intervals of TUG and FRT. In our study, ICC score for TUG was 0.86 (95%, CI=0.69 to 0.94) and 0.96 (95%, CI=0.92 to 0.98) for FRT.

The mean score for HMAS was 32.65±3.88 points. A significant negative correlation was found between TUG and HMAS with a correlation coefficient of r_{ho}= -0.69 ($p<0.01$). A significant positive correlation was found between FRT and HMAS with a correlation coefficient of r_{ho}= 0.47 ($p<0.05$). Table 3 shows the relation between balance tests and HMAS.

DISCUSSION

In the follow-up of DMD, it is crucial to collect stable results from evaluation procedures. Test- retest reliability tests the stability of scores on two seperate occasions

Table 2. The means, standard deviations, ICCs and 95% confidence intervals of TUG and FRT.

N = 23	Measure 1:Mean(SD)	Measure 2: Mean(SD)	ICC	95% CI
TUG (s)	8.16(1.32)	8.35(1.55)	0.86	0.69–0.94
FRT (cm)	13.04(4.57)	12.39(4.90)	0.96	0.92–0.98

SD: Standart deviation, ICC: ıntraclass correlation coefficient, CI: confidence ınterval.

Table 3. Correlation between TUG and FRT with motor functions (HMAS) in children with DMD.

		TUG	FRT
HMAS	r_{ho}	-0.69	0.47
	p	<0.01	<0.05

r_{ho}:Spearman rank test.

when the score would not be expected to change (Tyson and DeSouza, 2004). The ICC is frequently chosen as a statistical method for assessing reliability (Bland and Altman, 1990). ICCs can vary from 0.00 to 1.00. Generally, ICC values less than 0.5 can be considered as indicating poor reliability, those between 0.5 and 0.75 as indicating moderate reliability, and those above 0.75 as indicating good reliability (Portney and Watkins, 1993).

Several authors have demonstrated the test-retest reliability of TUG and FRT in children with disabilities. In the study of Niznik et al. (1995), ICC value for FRT was 0.87 between sessions in children with lower extremity spasticity. Williams et al. (2005) showed the test-retest reliability of TUG with intraclass correlation coefficient of 0.83 in children with physical disabilities due to cerebral palsy and spina bifida. Gan et al. (2008) reported the test-retest reliability of TUG and FRT in children with cerebral palsy. In this study, the ICC values for TUG and FRT were 0.99 and 0.95 respectively. In our study, ICC values demonstrated good reliability for TUG and FRT in children with DMD. These results confirm that both TUG and FRT provide stable results and can be reliably used in the follow up of children with DMD.

Although these tests provide reliable results in DMD, both TUG and FRT have some limitations in children who are in the later stages of the disease. In our study we included the children who were in stages of 1 to 2 according to Vignos scale. This means that these children were able to walk unassisted and rise from a chair independently. The use of TUG is limited in children who can not rise from a chair. Similarly FRT can not be performed in children who can not achive 90° shoulder flexion because of the weakness of deltoid muscle. With disease progression, different evaluation procedures for balance must be employed in this population.

The connection between gross motor functions and balance evaluations were shown in previous studies. In a study of Williams et al. (2005), TUG scores showed moderate negative correlation with scores on the Standing and Walking dimensions of the Gross Motor Function Classification System ($r = 0.52$, $p = 0.01$) in children with disabilities. In another study, TUG and FRT showed significant correlation with GMFM-88 ($r = 0.86$ for FRT and $r = 0.89$ for TUG, $p<0.01$) in children with cerebral palsy (Gan et al., 2008). In our study we found a positive correlation between FRT and HMAS and a negative correlation between TUG and HMAS. Although, there was correlation between tests, the correlation coefficients were not high as previous studies. From our opinion, this result can be due to the small number of the study group, which can be a limitation of our study. These tests should be repeated on a larger sample in next studies.

Conclusions

Finally, our results provide evidence that FRT and TUG are reliable measures and may be used to monitor change over time, particularly following interventions that aim to improve gross motor functions in children with DMD.

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