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ORIGINAL ARTICLE

Motor learning through a non-immersive virtual task in people with limb-girdle muscular dystrophies

Marcelo Prumes[1], Talita Dias da Silva[2,3,4], Camila Aparecida de Oliveira Alberissi[5], Camila Miliani Capelini[4], Lilian Del Ciello de Menezes[2], João Batista Francalino da Rocha[1], Francis Meire Favero[6], Carlos Bandeira de Mello Monteiro[1,4,5]

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[1]Laboratório de Delineamento de Estudos e Escrita Científica. Centro Universitário Saúde ABC (FMABC), Santo André, SP, Brazil.

[2]Departamento de Cardiologia, Escola Paulista de Medicina, Universidade Federal de São Paulo (UNIFESP), São Paulo, SP, Brazil.

[3]Faculdade de Medicina, Universidade Cidade de São Paulo (UNICID), São Paulo, SP, Brazil.

[4]Programa de Pós-Graduação em Ciências da Reabilitação, Faculdade de Medicina da Universidade de São Paulo, São Paulo, SP, Brazil.

[5]Escola de Artes, Ciências e Humanidades (EACH), Universidade de São Paulo (USP), São Paulo, SP, Brazil.

[6]Setor de Investigação nas Doenças Neuromusculares, Departamento de Neurologia e Neurocirurgia, Universidade Federal de São Paulo (UNIFESP), São Paulo, SP, Brazil.

Corresponding author
carlosmonteiro@usp.br

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Abstract

Introduction: Limb-girdle muscular dystrophies (LGMDs) are neuromuscular and genetic disorders that progress with weakness and damage of the proximal muscles, developing with loss of functionality. Virtual reality environments are suggested as an effective alternative for performance of daily life activities. However, there is no evidence in the literature on the use of virtual reality in this population.

Objective: Assess motor performance through a motor learning protocol in a coincident timing task.

Methods: 10 participants with LGMD and 10 healthy individuals were selected and included in the study to perform a non-immersive virtual reality task divided into three phases: acquisition (20 attempts), retention (5 attempts), and transfer (5 attempts, with speed increase).

Results: It is observed that the accuracy of movement improves from the beginning to the end of the acquisition ($p = 0.01$); however, there is a marginal difference between the groups in block A1 ($p = 0.089$). Regarding the variability of touches, observed by the variable error, both groups improved performance in all phases.

Conclusion: Even with lower performance than the control group at the beginning of the practice, individuals with LGMD showed the potential to optimize motor function during the practice of a non-immersive virtual reality activity and were able to match their performance with the control group after a few attempts.

Keywords: Muscular Dystrophies. Virtual Reality. Motor Learning. Limb-Girdle Muscular Dystrophies.

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Authors summary

Why was this study done?
Limb-girdle muscular dystrophies (LGMDs) is a peculiar type of muscular dystrophy characterized by weakness and wasting of the proximal upper and lower limb muscles, with progressive loss functionality. However, there is no cure yet. It is known that the management of this disease consists, among others, of rehabilitation services. Programs that use virtual reality in rehabilitation can be effective for the performance of activities of daily living in this population, since there are studies of virtual reality in other types of dystrophies, but not with LGMD.

What did the researchers do and find?
A motor learning protocol performed through a non-immersive virtual reality task compared participants with LGMD with healthy individuals. Thus, it was observed that despite the differences between the groups, both improved performance in all phases.

What do these findings mean?
People with LGMD have an inferior performance in a virtual reality task due to motor difficulties. However, they can improve motor function through practice, helping in the rehabilitation of these individuals.

INTRODUCTION

Muscular dystrophies (MDs) are a heterogeneous group of inherited disorders that share similar clinical features and dystrophic changes in muscle biopsy[1] associated with progressive skeletal muscle weakness and wasting[2]. The MD can be transmitted as autosomal dominant, autosomal recessive, or X-linked traits; sporadic cases may also arise as a result of the new mutation[3] and are characterized clinically by intense variability in severity and phenotype, and by anatomical changes of non-specific dystrophic pattern in muscle fiber[4]. Muscle weakness and atrophy occur due to degeneration of muscle tissue during this process[5-8], and destruction and recovery of muscle fibers occur, which are replaced by fibrous and adipose tissue[5,9].

Approximately 40 types of MDs, which differ from one another by genetic mechanism of inheritance, age of onset of first symptoms, involvement of affected muscle groups, and speed of disease progression, are identified[10-12]. A peculiar type of muscular dystrophy is limb-girdle muscular dystrophies (LGMDs), which are a diverse group of genetic neuromuscular conditions that usually manifest in the proximal muscles of the hip and shoulder girdles and have a typical age of onset between 10 and 30 years of age[13-15].

LGMDs are characterized by weakness and wasting of the proximal upper and lower limb muscles, with progressive loss of functionality. Distal and axial weakness is recognized in some forms, but the facial and bulbar muscles are usually spared[16]. The evolution of muscle weakness is usually symmetrical, but it varies in the severity of muscle fiber atrophy and speed of progression between men and women and between individuals[17,18].

Currently, there is no cure for MDs. However, treatment and management can consist of medication, surgery, and/or rehabilitation services, including strength training, training of aerobic capacity, or the use of aids and adaptations, such as arm supports to enable the performance of daily activities[19,20].

Some studies have been conducted with LGMDs directed mainly to cardiology[21-23], genetics[24-26], molecular biology[27,28], and differential diagnosis[29]. However, with recent advances in the development of computer-assisted technology, programs increasingly facilitate the use of virtual reality environments in rehabilitation, and those benefits of new technology can provide some improvement for LGMD individuals. Virtual reality allows an individual with LGMD to be exposed to a virtual interface and facilitates the capacity to interact with elements and targets. Although some studies with the use of virtual reality in individuals with different dystrophies have been conducted and presented positive results, Freitas *et al.*[30] analysed different device interactions in a virtual reality task in individuals with Duchenne Muscular Dystrophy (DMD), Heutinck[31] used virtual reality computer gaming with dynamic arm support in boys with Duchenne muscular dystrophy, and Massetti[32] compared between virtual and real tasks in DMD. We did not find any research using virtual reality in individuals with LGMD.

Moreover, with the development of computer-assisted technology, programs often include virtual reality environments as possibilities to be used as rehabilitation tasks providing movement, repeated muscle contraction, and possible motor learning[33]. Different studies have used the knowledge of virtual reality and motor learning to verify performance improvement with muscular dystrophy[30,32-36], and other motor diseases, such as cerebral palsy[37-41], Down syndrome[42,43], and autism spectrum disorder[44-46].

Motor learning is a phenomenon that refers to the relatively permanent internal changes involved in the ability to perform motor skills[47]. These changes take place in order to guarantee the achievement of the goal and come from the experience and practice that result in the acquisition and transfer of motor skills[48,49]. The motor learning phenomenon, although not directly observable, allows one to clearly discern changes in the interaction with objects and with other human beings. Thus, motor learning can be inferred through performance, while its improvement can be observed by increasing consistency, fluency in movement, reduction of execution error, and reduction in the total time of movement to perform the task[50].

Thus, in order to verify that motor learning has been solid, rather than comparing performance in the initial phase in relation to the final phase of acquisition, it is necessary to resort to performance in learning tests that is, to enable a transfer test that consists of the ability to adapt a motor behavior practiced in a different context through a small change in the motor task[51-53].

Due to the lack of studies using virtual reality in LGMD and according to the above deliberations,

we organized a motor learning protocol using a virtual reality timing coincident task, as used by Monteiro *et al.*[37], Monteiro *et al.*[43], Martins *et al.*[41] and Moraes *et al.*[46]. The results of the current study will provide evidence to determine if there is improvement in performance during acquisition practice with task retention and transfer in individuals with LGMD in a virtual reality task. Therefore, we aim to assess the motor performance in individuals with LGMD through a motor learning protocol in a virtual coincident timing task.

METHODS

Participants

Twenty individuals with a mean age of 27.9 ± 8.16 divided into two groups participated in a cross-sectional study[54]: an experimental group (EG), formed by 10 individuals with a genetic diagnosis of LGMDs, and a control group (CG), composed of 10 individuals without changes in posture and movement, matched for age and sex with the EG (Figure 1).

The participants signed a free and informed consent form before the tests started. For this protocol, we considered eligible all patients undergoing clinical follow-up at the Brazilian Association of Muscular Dystrophy who had the authorization of their parents or guardians to participate in the study as well as a diagnosis of muscular dystrophy of the girdle type confirmed by molecular method and/or protein expression skeletal muscle.

The exclusion criteria were individuals with cognitive impairment that prevents understanding of the task, malformations in the central nervous system, and/or neurological syndrome. This study was approved by the ethics committee on research of ABC Medical School (no. CAEE: 39396814.9.1001.0082 - approved on 20/01/2015 by no. 980.629). It was also approved by the ethics committee on research of the University of São Paulo (USP) no. CAEE: 02949412.2.0000.5390).

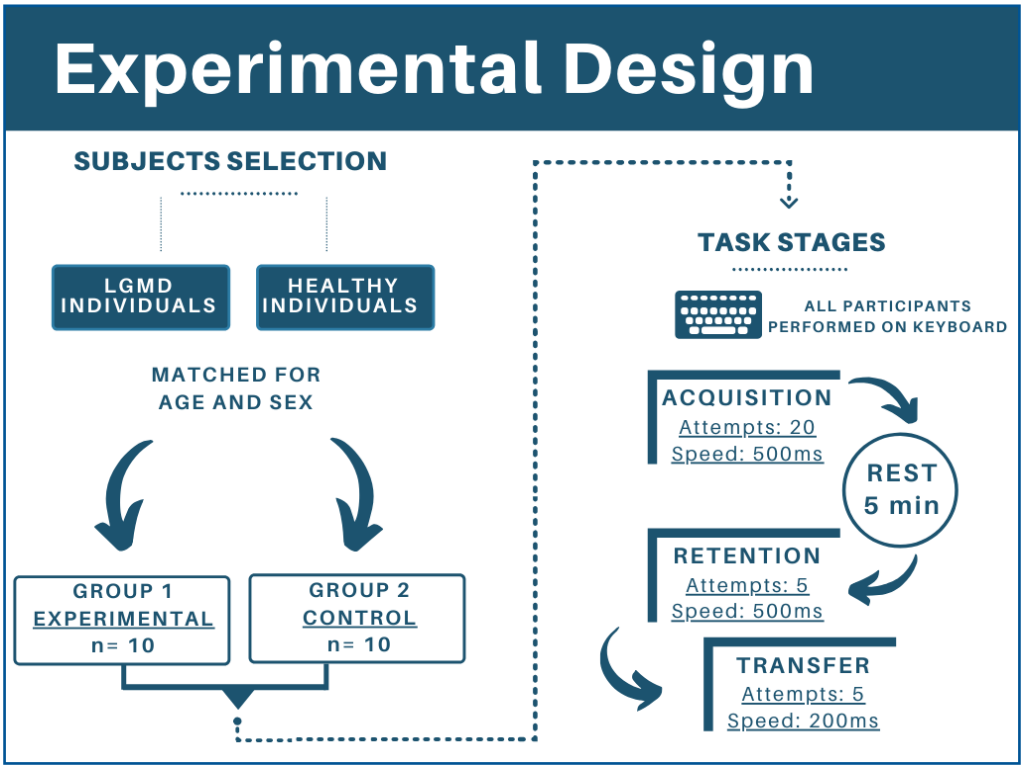

Figure 1: Experimental design: subject selection and task stages

Instrument: COINCIDENT TIMING

A coincident timing task was used to understand the human visuomotor system, including how motor control processes information involved in intercepting a moving object. In this study, the coincident timing software used three-dimensional images and enabled the task to be performed by pressing the space bar on a keyboard. The coincident timing task can provide results that show the possibility of motor learning based on the performance obtained by decreasing errors or the variability of errors[41].

For the coincident timing software, 10 spheres that turned on a red light were displayed on the computer screen in sequence until the last sphere was reached, which was considered the target. The participants received immediate feedback on the correctness or error of the task performance, either through different sounds (auditory interaction-differentiated sounds for correctness or error) or through images that changed color (visual interaction – green for correctness and red for an error). The individuals were instructed to place their hand next to the computer keyboard's space bar and were told that when the first/top sphere was activated, they could move their hand when they felt it was appropriate to touch the space bar exactly at the moment the last sphere (target sphere) was activated (lit)[41] (Figure 2).

Procedure

All participants were positioned comfortably in a chair that was adequate to enable task execution. Each participant was properly instructed on what should be done at each stage. Twenty attempts of the task were performed with the dominant upper limb at moderate speed that is, 500 ms between the lights of each circle for the acquisition phase. After the acquisition, the participants rested for 5 minutes, where they had no contact with the task, and then made five attempts in the retention phase. For the transfer phase, five more attempts were made with an increase of speed (200 ms) (Figure 1).

Figure 2: Illustration of the coincident timing task performed on a computer as proposed by Silva *et al.*[55]

Notes: (A) Demonstration of error performed by the participant (red light – unsuccessful in that attempt); (B) demonstration of hit performed by the participant (green light – successful in that attempt); (C) an example of a participant performing the task using the keyboard space button.

Statistical analysis

As performance measures (dependent variables), we used absolute (AE), variable (VE), and constant errors (CE), which represent precision, variability, and directional tendency of performance, respectively. The data were analyzed in blocks of attempts of five trials each, with four blocks of acquisition (A1 to A4), one block of retention (R), and one block of transfer (T). The dependent variables were analyzed using MANOVA with factor 2 (group: limb-girdle muscular dystrophy - "LGMD", healthy control - "Control") by 4 (blocks: first to fourth blocks of acquisition – A1 versus A2, A1 versus A3, and A1 versus A4). For retention and transfer phase comparisons, we used factor 2 (A4 versus R and A4 versus T, respectively). For the factor blocks, we used repeated measures. Partial eta-squared (ŋp2) was reported to measure effect size and was interpreted as small (effect size >0.01), medium (effect size >0.06), or large (effect size >0.14)[56]. Post-hoc comparisons were performed using Tukey's least significant difference (LSD) test ($p < 0.05$).

RESULTS

The MANOVA found a main effect for blocks (Wilks' lambda= 0.717, $F_{9, 126}$= 2.06, p= 0.038, η_p^2= 0.11). No further effects or interactions were found.

For analysis, the error measures in milliseconds of constant (CE), absolute (AE), and variable (VE) errors were considered. The constant error refers to the lack of anticipation and delay of the movement; that is, through this variable, the directional trend of the movement is evaluated. The absolute error represents the absolute difference between the time the ball arrives at the target and the response time; therefore, it demonstrates accuracy of movement and whether the individual is able to hit the target. The variable error is a standard deviation of the CE, indicating variability in the participant's touches/responses; in other words, it identifies whether there was precision in the movement[57]. The phases of motor learning will be described through the errors in the following sections.

Absolute error

Acquisition

There was a significant effect for block ($F_{3, 54}$= 5.11; p=0.009; η_p^2= 0.22). The post-hoc tests showed no significant improvement from A1 to A2 (p=0.825), but from A1 to A3 and from A1 to A4, the improvement was significant (p= 0.009 and 0.001, respectively). There was no main effect for groups; however, the post-hoc test showed that there was a trend of significant difference between groups in the first block of acquisition (p=0.089) (Figure 3).

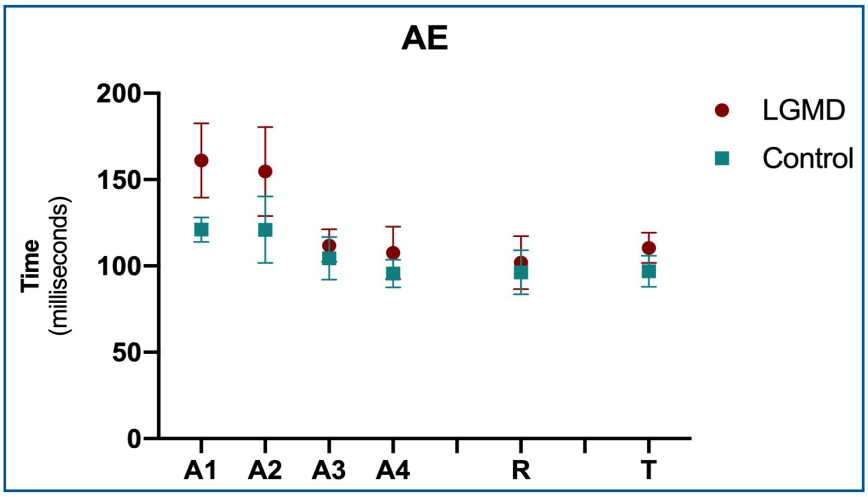

Figure 3: Mean and standard error of the blocks of attempts in all protocol phases in the absolute error

A1-A4: blocks of acquisition; R: retention block; T: transfer block; LGMD: limb-girdle muscular dystrophy

Retention

There were no significant effects or interactions for blocks or groups.

Transfer

Similarly to retention, there were no significant effects or interactions for blocks or groups.

Variable error

Acquisition

There was a significant effect for block ($F_{3, 54}$= 3.17; p=0.050; η_p^2= 0.15), in which both groups decreased the VE during practice. Similarly to absolute error, there was no significant improvement from A1 to A2 (p= 0.479), but between A1 and A3 and between A1 and A4, the improvement was significant (p= 0.038 and 0.016, respectively) (Figure 4). No further effects were found for groups.

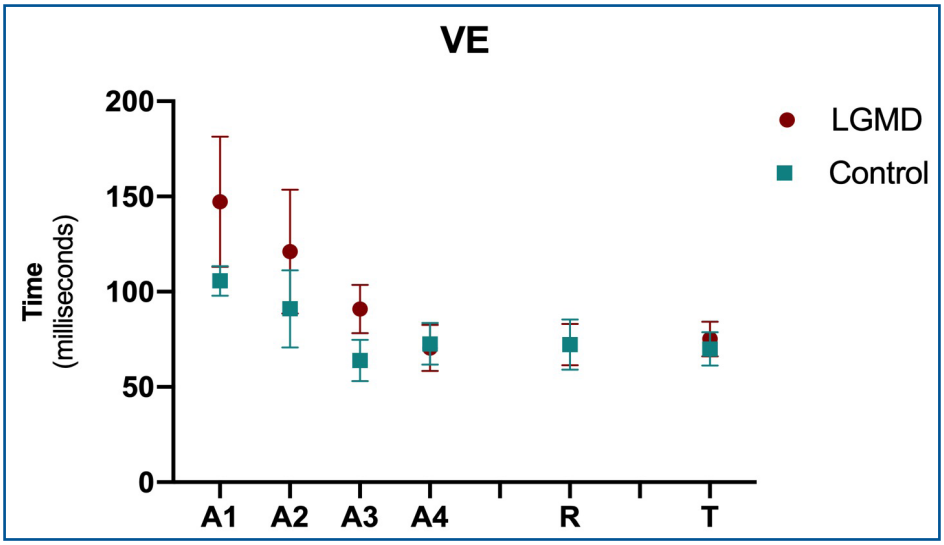

Figure 4: Mean and standard error of the blocks of attempts in all protocol phases in the variable error

A1-A4: blocks of acquisition; R: retention block; T: transfer block; LGMD: limb-girdle muscular dystrophy

Retention

There were no significant effects or interactions for blocks or groups.

Transfer

Similarly to retention, there were no significant effects or interactions for blocks or groups.

Constant error

The directional trend is observed in Figure 5. The EG presented a greater tendency to advance the touch and the CG to delay in most of the evaluated blocks. In the retention phase, the directional trend was towards advancement, and in the transfer test, it was delayed in both groups.

A1-A4: blocks of acquisition; R: retention block; T: transfer block; LGMD: limb-girdle muscular dystrophy

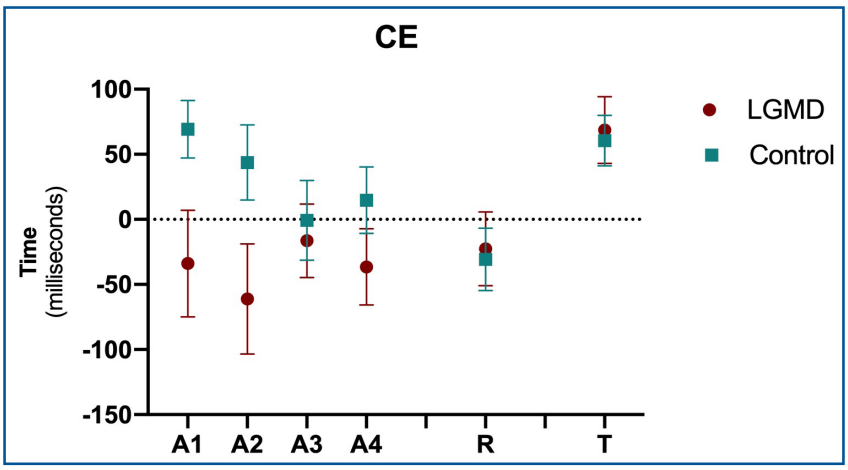

Figure 5: Mean and standard error of the blocks of attempts in all protocol phases in the constant error

DISCUSSION

Due to the motor difficulties presented by individuals with LGMD, this study used the knowledge from motor learning to verify the improvement of performance through the use of a coincide timing task in a non-immersive virtual environment.

Our results, as hypothesized, showed improvement in performance in the acquisition phase, considering the absolute and variable error (Figures 3 and 4) for both groups (EG and CG), which may suggest good adaptation to the task. Both groups showed performance stabilization, since the improvement in performance observed during the acquisition of the task was maintained during retention (no statistical difference in relation to the last block of the acquisition phase and retention) and transfer test (no statistical difference in relation to the retention test and transfer).

Despite this improvement, the participants in the CG performed better than individuals in the EG, but only in the first acquisition block. No statistically significant difference was found between the groups, considering the absolute and variable errors during the following blocks. These data contribute to justify that patients with LGMD, despite the proximal difficulties in tasks that require hand ability, were able to maintain adequate function for the task proposed in this study.

According to Mahjneh *et al.*[58], LGMD individuals presented mild weakness in the upper limb, and their proximal muscles are weaker than distal muscles, which can allow better distal performance[16]. We can speculate that a simple movement task and the necessity to use more hand and finger movement (distal movement) during our task were responsible for the improvement of performance in the LGMD group and match value with the CG during most protocol blocks.

These results are confirmed by the studies by Faulkner *et al.*[59] and Mahmood *et al.*[60], which showed that muscle contractions in LGMD may determine mild difficulty and no significant damage of muscle fibers during common daily activities, and they could provide benefits to improve performance in a simple task. Otherwise, exercise tolerance is affected in patients with LGMD, as a direct consequence of loss of muscle fibers, but also secondary to the sedentary lifestyle due to motor impairment[61].

On the other hand, McDonald *et al.*[62] and Stübgen *et al.*[63] found a progressive decline in muscle strength and ability in LGMD individuals, but it happened slowly per decade of disease duration. Thus, despite muscular weakness, damage to muscle fibers, and movement difficulty, characterizing LGMD, the individuals in this study still have the capacity to improve performance, even to match the value with the EG. Corroborating with these findings, Hunter *et al.*[64] cited that the patient responses highlight symptoms that may be responsive to intervention, and thus the muscle weakness in LGMD may be due to disuse in addition to degeneration from the disease itself; however, difficulties to perform specific activities may be amendable through appropriate practice and intervention using assistive devices[65].

Considering the difficulty for individuals with LGMD in quality of life care, physical and psychological aspects[66], and low self-esteem and feelings of sadness[67], new technology such as virtual reality can provide better improvement in individuals with disabilities and can be used as a resource to help gain motor function[68-70] and stimulate and enable interaction and involvement with the activity independently[71]. The ability of virtual reality to create opportunities for active repetitive motor/sensory practice adds to its potential for neuroplasticity and learning in individuals with neurologic disorders[72] and can represent the future for LGMD rehabilitation. Corroborating with Jensen *et al.*[73] hypothesis, our study agrees with their idea that people with LGMD have the potential to optimize motor function through motor learning when the training is performed in a safe environment, as is done during a virtual reality task.

Despite the improvement in performance found in this study, we can point out some limitations: (1) The small number of participants can diminish the sensitivity of the tests. Future studies should be conducted with more participants. (2) We did not analyze the motor function for the EG, and the results showed good performance probably because the participants in our study presented good function. Clinical assessments for future studies are necessary to better characterize the sample and understand the results. (3) We used a computer program that limited the outcome parameters to non-immersive virtual reality; future studies should use more immersive and non-contact tasks, since a different environment could provide

valuable information. (4) We used a protocol with only one day of practice; future studies should be conducted with a long-term protocol to perform a longer period of practice. Finally, (5) this is a simple computer task, and the results could not be extrapolated for other tasks; future studies using real and virtual tasks could provide better insight into our findings.

CONCLUSION

The LGMD group had lower performance than the CG at the beginning of the task. However, the results demonstrate that all participants, from both groups, improved their performance during the practice. Therefore, individuals with LGMD showed the potential to optimize motor function during the practice of a non-immersive virtual reality activity and were still able to match their performance with the CG after a few attempts.

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Resumo

Introdução: As distrofias musculares de cinturas (DMC) são distúrbios neuromusculares e genéticos que progridem com fraqueza e dano dos músculos proximais, desenvolvendo-se com perda de funcionalidade. Sugere-se ambientes de realidade virtual como uma alternativa eficaz para o desempenho das atividades da vida diária. No entanto, não há evidências na literatura sobre o uso da realidade virtual nessa população.

Objetivo: Avaliar o desempenho motor através de um protocolo de aprendizagem motora em uma tarefa de timing coincidente.

Método: 10 participantes com DMC e 10 indivíduos saudáveis foram selecionados e incluídos no estudo para realizar uma tarefa de realidade virtual não imersiva dividida em três fases: aquisição (20 tentativas), retenção (5 tentativas) e transferência (5 tentativas, com aumento de velocidade).

Resultados: Observou-se que a acurácia do movimento melhorou do início ao final da aquisição ($p = 0,01$); no entanto, existe uma diferença marginal entre os grupos no bloco A1 ($p = 0,089$). Em relação à variabilidade de toques, observada pelo erro variável, ambos os grupos melhoraram o desempenho em todas as fases.

Conclusão: Mesmo com desempenho inferior ao grupo controle no início da prática, os indivíduos com DMC mostraram potencial para otimizar a função motora durante a prática de uma atividade de realidade virtual não imersiva e foram capazes de corresponder seu desempenho com o grupo controle após poucas tentativas.

Palavras-chave: distrofias musculares, realidade virtual, aprendizagem motora, distrofias musculares de cinturas.