Adaptive VR-based rehabilitation to prevent deterioration in adults with cerebral palsy

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Abstract—Cerebral palsy (CP) is a disabling life-long condition progressively impeding a patient's independence. Although incident rates are high, a clear understanding of the disease is missing. CP is characterized by several motor disorders and sensory or perceptive comorbidities. This multifaceted nature complicates proper diagnosis and hampers the search for possible treatments. During adolescence and adulthood, individuals with CP experience a drastic deterioration in gross motor control, independence, and quality of life. There is poor evidence that physical therapy promotes the retention of function through aging, and no clinical studies exist that explore the potential of VR-based training to prevent deterioration. In this pilot randomized controlled trial, we expose 14 adults with CP to the Rehabilitation Gaming System (RGS) and examine its usability, effectiveness, and acceptability. Our results show that the RGS difficulty adaptation algorithm automatically matches the patients’ impairment level as captured by clinical scales (Barthel and Box & Blocks). The clinical effectiveness and acceptability of the RGS and conventional therapy were comparable. We conclude that VR-based physical therapy as an adjunct to usual treatment may be a promising approach for the prevention of deterioration in adolescents and adults with CP.

Keywords—cerebral palsy, virtual reality, motor function, physical therapy, rehabilitation

I. INTRODUCTION

Cerebral Palsy (CP) is a disorder identified by impaired muscle coordination (spastic paralysis) and/or other disabilities, typically caused by damage to the brain before or at birth. Among all limitations CP is the most frequent in affecting children during development (2 x 1000 live births) and given that life expectancy is rather high (30-70 years), there is an almost equivalent number of adults that are affected by this severe brain disease (Cans, C. 2000).

There is a lack of consensus on the definition of CP (Korzeniewski, et al. 2018). However at least four recurring elements can be observed: (1) brain lesion, (2) onset at birth or early childhood years, (3) impaired motor control, (4) non-progressive state of the disease. The motor disorders in patients with CP are complex. Namely, deficits include muscle tone abnormalities, impairment of balance and coordination, decreased strength and loss of selective motor control. Deficits are also often accompanied by comorbidities such as...
disturbances of sensation, cognition, communication, perception and/or behaviour (Rosenbaum, 2007). The interrelated nature of CP symptomatology hinders the diagnosis and treatment of CP (Zarrinkalam, et al. 2010).

<table>
<thead>
<tr>
<th>TABLE 1. PATIENT’S CHARACTERISTICS AT BASELINE</th>
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<td>N (%)</td>
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<tr>
<td>Subjects</td>
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<td>Male</td>
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<td>Laterality</td>
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<td>Affected Arm (Left/Right/Bilateral)</td>
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<td>Dominant Arm (Left/Right)</td>
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<tr>
<td>Type (tetraplegia/triplegia/diplegia/hemiplegia)</td>
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<tr>
<td>Clinical scales</td>
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<td>Age(years)</td>
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<td>GMFCS (count) (1/2/3/4/5)</td>
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<td>MACS (count) (1/2/3/4/5)</td>
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<tr>
<td>MAS (biceps brachii)</td>
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<td>MAS (pectoralis major)</td>
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<tr>
<td>Star Cancellation</td>
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<td>Box &amp; Blocks (dominant arm)</td>
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<td>Box &amp; Blocks (non-dominant arm)</td>
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<td>Barthel</td>
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Although CP is a permanent life-long condition of non-progressive nature, since adolescence individuals with CP experience a drastic deterioration in gross motor control, subsequently affecting the patient’s independence (Tarsuslu, et al. 2010, Strauss, et al. 2004). However, given that CP does not have a clear aetiology neither is a single pathology, no specific diagnostic and therapy protocol exists, therefore leaving this population unattended. Indeed, estimates from a recent study indicate that from 20 to 35 years old, 62% patients do not experience further improvements; 26% deteriorate; and 4% die (Strauss, et al. 2004). There is a need for knowledge and tools to facilitate a reliable and accurate prognosis of CP outcome as well as its treatment with affordable rehabilitation programs that prevent the degradation of function and that are suitable for sustained use across the patient’s lifespan.

So far conventional rehabilitation approaches for adolescents and adults with CP do not meet these criteria and instead combine supervised occupational therapy with recreational therapy. There is a consensus on the importance of encouraging patients with CP to be physically active and frequent exercise testing is recommended (Verschuren, et al. 2016). However, time constraints, financial and psychological burdens may hamper the individual’s ability to adhere to such intense recommendations. In light of these limitations, there is a need for an alternative approach that empowers the individual and provides multimodal sensorimotor training that is contextualized in goal-oriented actions to stimulate brain plasticity while targeting cognitive and motor functions in conjunction. This is in line with our current understanding of clinical and neuroscientific research (Verschure, 2012). Virtual Reality (VR) systems offer a set of advantages for the implementation of training protocols that exploit these general principles of neurorehabilitation, therefore providing embodied, intense, goal-oriented practice that is structured in time. For instance, VR-based systems allow to 1) provide concurrent implicit and explicit feedback, 2) monitor kinetic and kinematic descriptors of the patient’s full body movements, and 3) automatically adjust the training intensity to the specific capabilities of the patient. Training paradigms based on such principles have already shown significant efficacy in stroke patients (Cameirão, et al. 2010, 2012). The effectivity of therapies for individuals with CP ground on common general principles of motor learning and plasticity (Gordon, et al. 2007; Schmidt, et al. 2018; Nudo, et al. 2006), including goal-oriented training, intense patient-tailored training, massed practice, task-specificity and provision of feedback.

In this preliminary analysis study, we test the applicability of the Rehabilitation Gaming System (RGS) training paradigm to adolescents and adults living with CP. The RGS is a VR-based training system that provides adaptive motor and cognitive training after brain damage. RGS has already been clinically validated (Cameirão, et al. 2010, Ballester, et al. 2016) and has shown to facilitate the recovery of motor (Cameirão, et al. 2011, Ballester, et al. 2015, 2016, 2017) and cognitive functions (Maier, et al. 2015, Grechuta, et al. 2019). On these bases, we hypothesize the RGS functionalities, including the system’s tracking system, rehabilitation scenarios, and adaptive difficulty algorithms are suitable for individuals with CP.
II. METHODS

A. Participants

A total of 14 individuals with CP were recruited and assigned to the experimental (RGS-based training, EG) or the control group (conventional therapy, CG). Inclusion criteria were as follows:

- Adolescent and adult patients between 12-50 years old diagnosed with CP according to the Gross Motor Function Classification System (GMFCS, McCormick, et al. 2007). The GMFCS is a 5-level clinical classification system that describes the gross motor function of people with CP based on self-initiated movement abilities.
- Ashworth modified clinical scale < 3 (Bohannon, et al. 1987) for biceps brachii and pectoralis major.

Patients with severe visual or cognitive impairments such as mental retardation were excluded. Table 1 summarizes the demographical and clinical profile of each group at baseline. All the patients signed an informed consent form prior to participation. The study was approved by the local Ethical Committee of Adacen and the written consent to participate in the experiment was obtained from all patients recruited through the administrative staff of Adacen. This trial was registered in ClinicalTrials.gov before the onset of participants’ enrolment (NCT02938754).

B. Protocol

Clinical scores at baseline, and sociodemographic data including gender and age were collected immediately after recruitment. Furthermore, therapists did gather information about the frequency of occupational therapy and other forms of rehabilitation attended by the patients.

In addition to ‘treatment as usual’, we administered a supplementary intervention that consisted of 3 sessions of RGS training (experimental group) or conventional therapy (control group) per week during four consecutive weeks. Each session had a total duration of 30 minutes. During the first sessions with RGS, the experimenter explained the setup and the task to the subject. RGS scenarios combine movement execution with the observation of a correlated action by virtual arms displayed in a first-person perspective on a computer screen (Cameirão, et al. 2010) (Figure 1).

During each session, the experimental group was exposed to 3 different training protocols dedicated to the rehabilitation of the upper limbs, with a duration of 10 minutes each (Figure 2). The interaction design of these scenarios requires performing tasks of reaching and hitting spheres, or hockey pucks, spaced at different times, and varying in physics and appearance. The VR scenarios are divided into two workspaces, thus encouraging bi-manual training and preventing over-compensation. Each of the scenarios is defined by several difficulty parameters associated to performance descriptors. These parameters determine the complexity and intensity of the activity. For instance, in the Spheroids scenario, we identify 3 pairs of difficulty parameters and performance descriptors: 1) different trajectories of the spheres require different ranges of joint motion for elbow and shoulder, 2) the size of the spheres require different hand and grasp precision and perceptual abilities, 3) the velocity of the spheres require different movement speeds and timing. All these parameters are dynamically modulated by the RGS Adaptive Difficulty Controller to maintain the performance ratio (i.e. successful trials/total trials) above 0.6 and below 0.8, optimizing effort and reinforcement during training (Csikszentmihalyi, 1992). y.

These difficulty parameters in each scenario are modulated independently for the left and right workspaces, allowing lateralized control of the challenge in those cases in which the user presents hemiparesis or hemiplegia. The three rehabilitation scenarios are:
Spheroids: the user must perform planar horizontal reaching movements to intercept the maximum number of spheres approaching his location.

Air Hockey: introduces physics into the target’s behaviour, through a moving puck that bounces off from walls at a constant speed. In this task the patient is instructed to push the puck towards a goal area, therefore promoting movement planning in an environment that includes physical constraints.

Pinball: extends the physical constraints introduced in the Air Hockey scenario. This task consists of intercepting falling spheres in an environment in which moving cylindrical colliders may consecutively alter the trajectory of the targets. This training scenario demands high levels of attention for an accurate prediction of the objects’ behaviour.

C. Measurements and Data Analysis

We defined as the primary outcome the change in motor function of the affected extremity as measured by the Box and Blocks Test. As secondary outcomes we used three standardized clinical scales: 1) the Wechsler Adult Intelligence Scale (WAIS-III, Wechsler, et al. 1955), with measures Verbal IQ, Performance IQ, and Full-Scale IQ, Verbal Comprehension, Working Memory, Perceptual Organization, and Processing Speed; 2) the Barthel assessment scale (Collin, et al. 1988), that consists in self-reported scores related to the level of independence of the patient in Activities of Daily Living (ADLs), and 3) the Star Cancellation Test (Halligan, et al. 1989), a screening tool that was developed to detect the presence of unilateral spatial neglect.

All evaluations were performed at baseline and at the end of the treatment. In addition, we extracted the maximum level reached by each patient in each difficulty parameter, we averaged them across the sessions, and later we summed the averaged values to obtain a compound score that approximates the patient’s global capability to perform the task.

During the days following the last session, an acceptability questionnaire was filled by the patient. This questionnaire included 11 items grouped in 5 evaluation categories:

1) Memorability
   • Do you remember the last time you did this activity?
   • Could you perform this activity without receiving further information?
   • Do you think somebody who never did this activity could perform it well after a brief explanation?

2) Difficulty
   • Evaluate the difficulty of this activity (Too easy/Adequate/Too difficult) (1-5 score)

3) Perceived Length
   • The sessions are (Too short/Adequate/Too long) (1-3 score)

4) Enjoyment
   • Did you find this task fun? (Boring/Normal/Very Fun) (1-10 score)

5) Adherence
   • Is it difficult to be focused during the whole activity?
   • Do you think this activity is more motivating for you than the other activities you did today?
   • Would you recommend this activity to a friend?
   • If you could choose, how long would you do this activity? (2 times a week/ 4 times a week/ every day/ 2 times a day)
We conducted a between-groups comparison of demographic and clinical evaluations at baseline using the Wilcoxon rank-sum test (Table 1). Both groups were balanced for all demographical items, primary and secondary outcomes. Given the small sample size, we used non-parametric tests to compare the change in the selected clinical scales. Specifically, we applied Wilcoxon rank-sum and signed-rank tests for between-and within-groups comparisons respectively. To explore the co-variation of outcome measures we calculated the Spearman’s rank correlation coefficient. We set the significance level α at 0.05.

III. RESULTS

A. Validation of adaptive mechanisms

We evaluated the Spheroids difficulty parameters in all the participants in the RGS group, and we observed that the Difficulty Controller maintained average success rates between 0.6 and 0.8 thresholds in 92.5% of the sessions for the dominant arm and 83.5% of the sessions for the non-dominant arm. We extracted the maximum level reached by each patient in each difficulty parameter, we averaged them across the sessions, and later we summed the averaged values to obtain a compound score that approximates the patient’s global capability to perform the task. We found that these compound scores were strongly correlated with the participants’ clinical scores in the Box and Blocks Test (Spearman’s \( r=0.80, \ p=.02 \)) and the Barthel assessment scale (Spearman’s \( r=0.85, \ p=.01 \)) (Figure 3). However, we did not find any significant correlation between the levels reached in the RGS difficulty parameters and the participant’s performance in the Star Cancellation test, which may suggest that mild visuospatial attentional impairments do not necessarily interfere with the performance in the Spheroids scenario.

B. Acceptability and Usability Evaluation

We analyzed the score compounds for each of the 5 categories included in the acceptability questionnaire: Memorability, Difficulty, Perceived length, Enjoyment, and Adherence (Figure 4). We found no differences between groups in any of these acceptability items (p>.37, Wilcoxon Rank-sum). We explored the correlation between the reported scores and the participants’ impairment severity. The reported difficulty correlated positively with the level of independence (Spearman’s \( r=0.53, \ p=.05 \)). We also found a marginally significant inverse correlation between the memorability of the activities performed in conventional therapy and the patient’s independence (Spearman’s \( r=-0.84, \ p=.057 \)).

C. Clinical effectiveness

Clinical scores related recovery in motor and attentional function (i.e. hemineglect) did not show improvements for any of the groups (Table 2). Differences between groups in motor recovery scores from the Box & Blocks evaluation were not statistically significant, neither for the non-dominant (p =.12, Wilcoxon Rank-sum) nor for the dominant hand (p =.24, Wilcoxon Rank-sum). Similarly, improvements in visual attention were not different between the control and the RGS group (p=.98, Wilcoxon Rank-sum).

IV. DISCUSSION AND CONCLUSIONS

The majority of adolescents and adults living with CP experience a drastic decline in gross motor function before reaching 35 years old (Strauss, et al. 2004; Day, et al. 2007). While the effectiveness of conventional therapy and VR-based training in children affected by CP has been largely studied, the effects and benefits of these rehabilitation approaches in adults remain unclear. To the best of our knowledge, clinical trials exploring the benefits of VR-based systems for preventing deterioration in adults with CP are non-existent. In this pilot study, we conduct a randomized controlled trial to explore the effects of a one-month VR-based therapy for adolescents and adults with CP. We first validate the adequacy of the adaptive difficulty algorithm integrated into the Rehabilitation Gaming System. Secondly, assess its effectiveness in promoting motor function and preventing deterioration in comparison to conventional therapy. And lastly, we evaluate the system’s acceptability in terms of Memorability, Difficulty, Perceived length, Enjoyment, and Adherence.

<table>
<thead>
<tr>
<th>TABLE 2 MEASURES OF IMPROVEMENT IN CLINICAL SCALES</th>
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<tr>
<td><strong>Within-group analysis</strong></td>
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<tr>
<td>End of treatment (T1)</td>
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<td>Mean(SD)-Median(IQR)</td>
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<td><strong>Box &amp; Blocks (dominant arm)</strong></td>
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<td>EG</td>
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<td>CG</td>
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<tr>
<td>Both</td>
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<td><strong>Box &amp; Blocks (non-dominant arm)</strong></td>
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The findings of this pilot study support that the RGS is a suitable tool for motor training in patients affected by CP, presenting hemiparesis (MACS < IV) and mild to moderate spasticity (Ashworth scale proximal < 3). The RGS automatically adjusts the virtual task parameters according to specific performance descriptors, allowing the patient to experience optimal levels of success while training. These task parameters correlate significantly with the patient’s scores in standardized clinical scales related to motor function (i.e. Box & Blocks), independence (i.e. Barthel), and visual attention (i.e. Star Cancellation). The analysis of the participant’s answers to an acceptability questionnaire did not show significant differences between therapies in regards to effort and enjoyment. Overall the acceptability measures were not different between groups. However, we observed that those patients with lower levels of independence reported finding the RGS sessions more demanding and conventional therapy activities more difficult to remember. In the field of usability, the term memorability refers to the degree to which a user can return to a system’s interface and remember how to use it. A user that attributes a low memorability to an activity may be less prone to perform it frequently, consequently hampering its adherence. In regards to the effectiveness of the system to induce recovery and prevent deterioration, we did not find any significant effects neither within nor between groups. This lack of effects could be due to lack of power (n=14) as well as to the low intensity and short length of the treatment (3 sessions a week for four weeks). Large clinical randomized controlled trials should further analyze the potential of VR-based therapy as an adjunct to usual treatment for the prevention of deterioration in adolescents and adults with CP.

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REFERENCES


