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TELEREHABILITATION DURING COVID-19 LOCKDOWN AND GROSS MOTOR FUNCTION IN CEREBRAL PALSY: AN OBSERVATIONAL STUDY

Running title: Efficacy of telepractice in cerebral palsy

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Abstract

Background: COVID-19 (Coronavirus Disease-2019) refers to a mainly respiratory disease, caused by a new SARS-CoV-2 virus predominantly transmitted through direct or indirect contact with mucous membranes of eyes, mouth, or nose. The main control measures are physical distancing, use of specific protective devices, hand hygiene and disinfection of environments and tools. During this health emergency, telemedicine and telerehabilitation guaranteed patients to receive continuity of care through a virtual support while maintaining physical distance.

Aim: The aim of this study is to evaluate the effects of telerehabilitation on gross motor skills in children with Cerebral Palsy (CP) during Covid-19 lockdown.

Design: observational study

Setting: Pediatric Outpatient Neurorehabilitation Service

Population: 53 children with Cerebral Palsy aged between 6 months and 12 years classified according to the Gross Motor Function Classification System (GMFCS).

Methods: variation on the Gross Motor Function Measure-66 (GMFM-66) score calculated before and after the telerehabilitation period was analyzed.

Results: after telerehabilitation there was a statistically significant increase in the median value of GMFM scores both on the total sample (from 54.82% to 63.18%, p-value 0.000005) and in the subgroups. Specifically, in children classified as level I and II at the GMFCS, this value increased more after the telerehabilitation period. Only the GMFCS level V group did not show statistically significant changes and only in two cases a decrease in the GMFM score after the telerehabilitation phase occurred.

Conclusion: telerehabilitation can be considered an efficient tool that can temporarily replace the in person therapy. It can allow the patient or caregiver to acquire skills in performing home exercises and to integrate and implement activity carried out at the rehabilitation center.

Clinical Rehabilitation Impact: this study shows a positive effect of telerehabilitation on gross motor function in children with cerebral palsy

Key words: cerebral palsy; telerehabilitation; gross motor function; GMFM-66; Covid-19

TEXT

Introduction

COVID-19 refers to a mainly respiratory disease, caused by a new SARS-CoV-2 virus. At the end of 2019, several cases of pneumonia with unknown etiology in Wuhan (China) emerged. The infection quickly spread throughout China and overseas [1]. Due to the uncontrolled spread, on January the 30th a public health emergency of international concern (PHEIC) was declared by the World Health Organization (WHO) [2] and on the 11th of March SARS-CoV-2 infection was defined as a Pandemic. SARS-CoV-2 is predominantly transmitted through direct or indirect contact with mucous membranes of eyes, mouth, or nose [3 4]. The main control measures are physical distancing, use of specific protective devices, hand hygiene and disinfection of environments and tools. During this health emergency, telemedicine guaranteed patients to receive continuity of care through a virtual support while maintaining physical distance. Telemedicine, in fact, entails the remote exchange of data between patients and health care professionals as part of diagnosis and management [5]. Telerehabilitation (TR) is a subfield of telemedicine described as the delivery of rehabilitation services via information and communication technologies. It includes rehabilitation services such as assessment, monitoring, prevention, intervention, supervision, education, consultation, and counseling [6].

Thanks to their beneficial effects, in recent years, home-based programs have received increasing attention in the CP child rehabilitation field. Home-based programs offer the opportunity to intensify aspects of physical therapy and to repeat specific exercises several times, thus increasing the intensity and efficiency of the therapeutic plan [7 8]. Moreover, TR improves caregiver's involvement resulting in a greater collaboration with therapists thus allowing both parties to learn, to share useful information and mutual perspectives on the child's rehabilitation [7]. Furthermore, home programs are useful in specific circumstances, as in the case of excessive distance from the rehabilitation center [7 9].

Many previous studies proved the positive effects of telerehabilitation on motor functions in children with different disabilities. A non-randomized study conducted on children with CP highlighted the contribution of TR in improving the resistance of the lower limbs to specific tests [10]. The efficacy of TR has also been reported in a large randomized controlled study conducted by James et al. in 2015, showing an improvement in the ADL performance, in processing capacity, in visual perception and in the dexterity of upper limbs in children with CP [11]. Similar results were observed by Bilde et al [12]. The effect of TR in increasing the sense of agency and the consequent positive impact on CP children functional performance was also studied [13].

The aim of this study is to evaluate the effects of telerehabilitation on gross motor skills in children with Cerebral Palsy aged between 6 months and 12 years, comparing the variations of GMFM-66 scores [14 15], before and after the execution of the telerehabilitation period during Covid-19 lockdown.

Materials and methods

Design

An observational study was conducted in order to evaluate the effects of TR on gross motor skills in children with Cerebral Palsy aged between 6 months and 12 years. Gross motor skills of all children were assessed through the GMFM-66 administered both before and after the TR period delivered during Covid-19 lockdown.

Inclusion and exclusion criteria

Inclusion criteria were: diagnosis of Cerebral Palsy, ages between 6 months and 12 years, participation in the TR treatment proposed during Covid-19 lockdown. Patients with other diagnoses and younger than 6 months or older than 12 years were excluded.

Ethics

This study followed the STROBE Guidelines (strengthening the reporting of observational studies in Epidemiology Statement), it was approved by the Independent Ethics Committee of the Research Institute of the Santa Lucia Foundation in Rome (Italy) and was conducted in accordance with the

Helsinki Declaration principles. Research participation consent was signed by parents or guardians of all children.

Participants

Fiftythree children were recruited (30 males and 23 females). The GMFM-66 rating scale was administered to each child three times, every six months: in autumn 2019 (t0), in February 2020 (t1) and in late summer 2020 (t2). During the first period (between t0 and t1) the usual face-to-face treatment at the rehabilitation center was carried out, while in the second period (between t1 and t2), due to the lockdown, a home-based TR plan was proposed and performed with the aid of caregivers. Each treatment session (50 minutes) was carried out under the remote simultaneous supervision of the therapist, who remained the same as before. The number of sessions per week varied for each child depending on the individual treatment plan. Each child maintained the previous number of weekly sessions.

Each child was classified according to the Gross Motor Function Classification System (GMFCS). Sample grouping based on the five GMFCS levels is shown in table I.

	I	II	III	IV	V	TOT
M	6	11	2	2	9	30
F	7	6	1	2	7	23
TOT	**13**	**17**	**3**	**4**	**16**	**53**

Table I: Sample Grouping by GMFCS level

Statistical analysis

Non-parametric statistics, the Wilcoxon test, was applied to research data since the GMFM scores are ordinal values. The significance threshold was set at 0.05 (p-value). Variations among t0-t1, t1-t2 and t0-t2 GMFM-66 scores were compared, both relative to the total sample and to the GMFCS levels subgroups. Statistical analysis was not applied to the III e IV GMFCS levels subgroups, due to the small sample.

Results

To begin with, the variation of the total sample median GMFM-66 scores relative to the three periods (t0-t1, t1-t2 and t0-t2) was analyzed.

Data showed a statistically significant increase of median scores in all periods examined. In particular, the median t0-t1 GMFM-66 score significantly increased from 48.44% to 54.82% (p-value 0.000001) The t1-t2 score also increased from 54.82% to 63.18% (p-value 0.000005); this improvement was higher than the one recorded in the first period. Overall, the t0-t2 GMFM-66 median value statistically increased (p-value 0.0000006) (Figure 1).

As for the t1-t2 period, only two cases showed a GMFM-66 score decrease, while 31 children (58%) showed an improvement and 20 (38%) stabilized (Table II).

Figure 1: Box plot of GMFM-66 values of the total sample at t0.t1 and t2

GMFM Value	T1-T2
Increased	58%
Reduced	4%
Unchanged	38%

Table II: percentage of increase, reduction and stability of GMFM-66 value between t1-t2

Children under the age of 5, primarily those classified as I or II GMFCS levels showed the greatest GMFM-66 value variations.

More specifically, in the I GMFCS level children group a statistically significant increase of GMFM-66 median value occurred (from 89.02% to 93.87%), both between t1 and t2 (p-value 0.008) and between t0 and t2 (p-value 0.012). Although an increase in the median value between t0 and t1 was not observed, dispersion of the scores around the central value changed resulting in a statistically significant variation also in this period (p-value 0.018). (Figure 2)

Furthermore, these children showed an over 90% GMFM-66 score at the age of 4 and a plateau at 9 years.

Figure 2: Box plot of GMFM values in GMFCS level I children at t0, t1 and t2

Second level GMFCS children showed a statistically significant variation of the GMFM-66 median value both between t0 and t1 and between t1 and t2. In particular, it increased from 79.36% to 81.14% between t0 and t1 (p-value 0.002) and mainly between t1 and t2, with a median GMFM-66 score of 84.98% (p-value0.001) (Figure 3).

Figure 3: Box plot of GMFM values in GMFCS level II children at t0, t1 and t2

No statistically significant increase was reported in the V level group during the t1-t2 period. In fact, while the GMFM-66 median value increased from 11.72% to 12.50% between t0 and t1 (p-value = 0.008), the increase between t1 and t2 was lower than the previous and it was not statistically significant (GMFM 13%, p-value = 0.5).

Despite that, a statistically significant variation was overall recorded between t0 and t2 (p-value 0.045) (Figure 4)

Figure 4: Box plot of GMFM-66 values in GMFCS level V children at t0, t1 and t2

Discussion

This study aimed to evaluate the effect of TR on gross motor functions in children with cerebral palsy (CP) assessed by the 66-item Gross Motor Function Measure (GMFM-66) rating scale. As shown by several studies, the development of gross motor functions in children with CP can be described and predicted by the GMFM, which highlights variations even in the order of a few months [15 16 17]. Furthermore, GMFM is frequently used to compare the effects of different therapeutic interventions [15], even when control groups are not foreseen: in these cases, a comparison of the GMFM scores before and after the treatment is carried out [18]. Based on the study of Alotaibi et al [18], an extreme variability of the values and evolution of GMFM score emerges both among children belonging to the same level of GMFCS [19], and among children belonging to different levels [15]. In particular, I e II GMFCS levels patients obtained higher GMFM scores and the median value reached by V level children did not exceed 20%. These data are in agreement with the 2007 Beckung et al.study that described the natural evolution of the scores on this scale [15]. According to literature, our results showed a plateau of GMFM values after 9 years of age [19].

Interesting results emerged when comparing the GMFM score trend during face-to-face therapy with that of TR. To begin with, in accordance with Russel et al. in 2000 [16] and with Hanna et al. in 2008 [19], the main GMFM value changes were recorded among children under the age of 5 and with better functional abilities (levels I and II). Furthermore, in both periods considered, there was a statistically significant increase in the median value of GMFM both in the total sample and in the subgroups. Specifically, in both the level I and level II groups this value increased more during the TR period. The group with level V is the only one among those analyzed that did not show statistically significant changes and in which there were two cases of decrease in the GMFM score after the TR phase. This is probably linked to the greater severity of the clinical-functional picture of the patients and to a reduced adherence to the new therapy modality. However, even during the face-to-face therapy, the variation in GMFM values was reduced compared to that recorded in the other levels. This trend is in line with the initial Rosenbaum study [20] and the successive study by Hanna et al. [19] who analyzed the progression of GMFM value in relation to the GMFCS level and created useful statistical curves.

According to these curves there is an exponential growth in levels I and II during the first years of life, reaching a plateau; proceeding from the III level onwards, however, growth rate reduces over time and after the age of 8 it depicts a slight deflection, indicating a decrease in the GMFM score.

In spite of the small sample size in this study, tele-rehabilitation proved to be a useful tool for maintaining and improving gross motor functions in children with CP, in accordance with previous numerous studies and reviews [21 22]. These benefits could be due to various factors. First of all, the therapists' ability to explain and teach the appropriate exercises even from a distance. Furthermore, greater family involvement was highlighted both by continuity of treatment and participation in therapy. As shown in the literature [5 23], one of the main advantages of TR is the possibility of carrying out therapy at home, especially important for patients who have to travel from great distances to reach the rehabilitation facilities. It is likely that, in these people, the convenience of carrying out the therapy at home increases adherence to treatment and continuity over time. Furthermore, during TR of pediatric patients, since caregivers assume the role of the therapist and learn the exercises, these can be repeated even outside the therapy session, thus increasing caregiver's sense of participation and involvement in the treatment plan [7 24 25].

Limits

Limitations of this study include reduced sample size and the dissimilar number and age of members at the different level subgroups. Moreover, the number of weekly sessions and type of treatment also differed, due to individualized treatment plans. The study did not foresee a control group, since all eligible children in charge carried out the TR treatment. It would have been interesting to compare research data with the trend of GMFM values of children who had not undergone therapy during lockdown.

Conclusions

The onset and uncontrolled spread of SARS-CoV-2 infection caused a health emergency and imposed restriction measures including physical distancing. Consequently, health related activities such as hospitalizations, outpatient visits, instrumental diagnostics, were delayed, reduced or even interrupted for months. In this critical period, TR allowed people with disabilities to continue rehabilitation, despite objective limitations due to the possible incorrect execution of the exercises even with therapist's remote supervision. Furthermore, TR entails availability and confidence with remote media. Despite these limitations, as reported in literature [10 11 12 13], TR has proved to be effective and useful in improving functional outcomes. According to this, the present study highlighted the benefits of a temporary TR treatment on gross motor skills in children with cerebral palsy. Most of them showed a significant increase of GMFM score except V level children, probably due to the greater severity of their clinical-functional conditions. In line with the literature [7 8 9], these positive results could be explained by the high adherence to the therapies, the caregivers' ability to learn the proposed home-exercises and to repeat them outside the therapy sessions and a greater caregiver involvement, in line with the family centered model. Results of the present study suggest that TR can be used as a temporary replacement to the in person therapy during a health crisis or when, for different reasons, the patient can't reach the rehabilitation facilities. Finally it sustains patients'/caregivers' involvement in the treatment, encouraging them to take an active role and promotes knowledge and skill in performing the required exercises enabling implementation and integration of the activity carried out at the rehabilitation center.

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NOTES

Conflicts of interest. The authors report no conflicts of interest.

Authors' contributions. **Martina Cristinziano** has taken care of study design, data interpretation and manuscript writing. **Carla Assenza** and **Clementina Antenore** have contributed to critical writing revision and interpretation data. **Leonardo Pellicciari** has given contributions to statistical analysis and interpretation data. **Calogero Foti** has supervised the study. **Daniela Morelli** has coordinated the study design and the interpretation of data. All authors read and approved the final version of the manuscript.

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Figure 1: Box plot of GMFM-66 values of the total sample at t0.t1 and t2

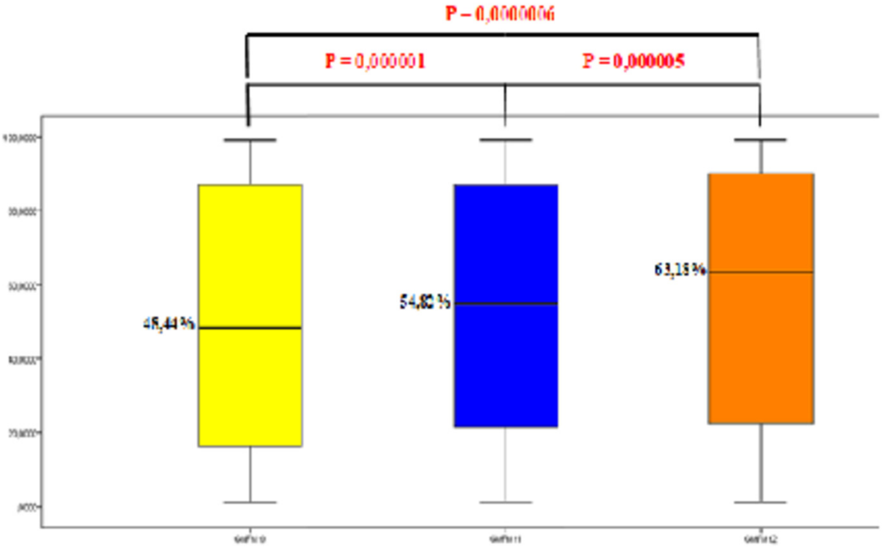

Figure 2: Box plot of GMFM values in GMFCS level I children at t0, t1 and t2

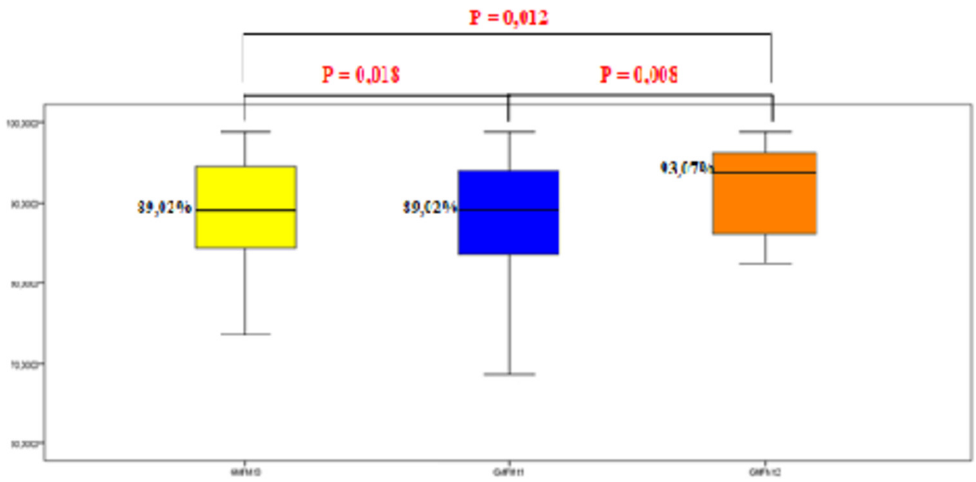

Figure 3: Box plot of GMFM values in GMFCS level II children at t0, t1 and t2

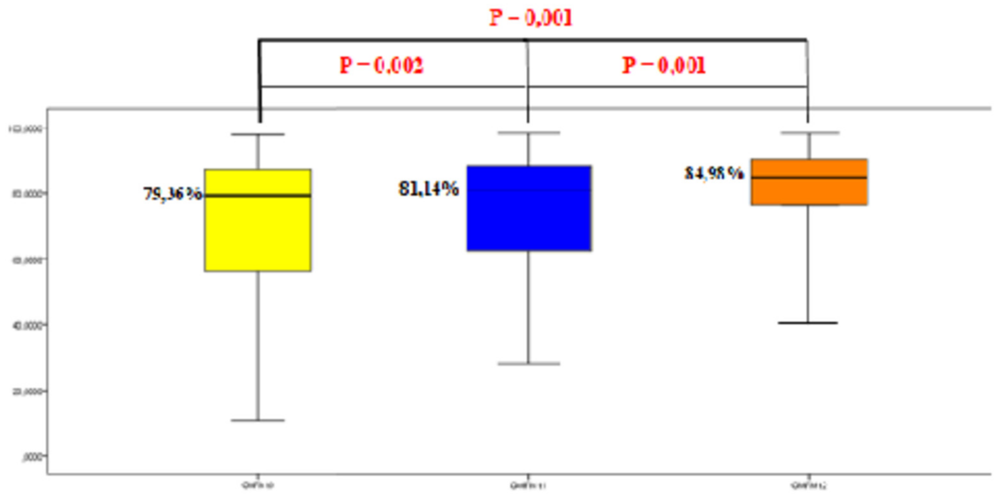

Figure 4: Box plot of GMFM-66 values in GMFCS level V children at t0, t1 and t2