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Telerehabilitation for Children with NeuroDevelopmental Disorders during Covid-19 Pandemic: A Scoping Review

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Abstract

Background: Children with neurodevelopmental disorders across the world receive pediatric rehabilitation services. The Coronavirus (COVID-19) pandemic challenged the rehabilitation community providing services for these children. The aim of the study was to outline the number, variety and characteristics of telerehabilitation services for children with neurodevelopmental disorders in the era of Covid-19.

Methods: To identify relevant literature, the scoping review method was used. Our results were presented according to PRISMA Extension for Scoping Reviews guidelines. The nine-step updated scoping review method by Peters et al. including some pre-search, search and post-search steps was conducted. A structured search of three databases of the Web of Science, PubMed, and Scopus was conducted for the studies published during 1 January 2020 to 12 December 2022. The extracted literature was reviewed in two steps by two independent reviewers.

Results: Twenty-seven articles met the inclusion criteria. Most of the studies were conducted in the United States. Services provided by telerehabilitation were mostly tele-intervention (n=19, 70.37%) followed by tele-evaluation (n=5, 18.52%). More than half of the studies used tele-conferencing for providing their services (n=15, 55.56%).

Conclusion: Although the variety of telerehabilitation service deliveries seems to have increased and even the number of participants in the studies has increased, still considering the rapid spread of the pandemic, the studies seem not to be adequately organized in terms of methods.

Key Words: Covid-19, Neurodevelopmental Disorders, Scoping Review, Telerehabilitation.

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1- INTRODUCTION

Children with Neuro-Developmental Disorders (NDDs) across the world receive pediatric rehabilitation services. NDDs are a set of disorders with the nature of growth and development that affect brain functions such as cognition, movement, language, learning behavior. Reasons cited for developmental disorders include genetic, perinatal, and These affect environmental. motor. cognitive, emotional functions and communication disorders, which require relevant interventions (1, 2). NDD is a broad term including a large number of disabilities that have some form of brain development disorder. This broad term defines a group of disorders that differ in phenotype and etiology. Disorders in this group include autism spectrum disorders (ASD), infrequent genetic syndromes, cerebral palsy (CP), congenital neurodegenerative disorders, and attention deficit hyperactivity disorder (ADHD) (3).

The group of practitioners providing direct and face-to-face habilitation and rehabilitation services to NDD children included occupational therapists. physiotherapists, and speech-language pathologists. The pandemic of coronavirus (COVID-19) affected the provision of interventions needed for this and children, group of with announcement of quarantine and the mandatory stay at home in order to create social distance and reduce the spread of this virus, the therapists who worked with these children, faced a new challenge that they never had before and in other words, they had no plan for it (4). However, with reduction in the number rehabilitation and habilitation intervention sessions and the change in the quality of services, it seems that serious harm was caused to these children. In the same way, none of the related authorities provided reliable news about the end of the pandemic and the subsequent laws related

to social distancing. Some therapists began to use their available technologies to compensate for these issues; so that, the services were provided in such a way that the therapist was located at a point far from the child, and according to the of the World definition Health Organization (WHO), the service recipient was at one point and the service provider was at another point (2). Providing services using this model is described as telerehabilitation defined as evaluation, monitoring. prevention. assessment. education, intervention. supervision, consultation and coaching through a variety of communication technologies (5).

There has been reported a large increase in the number of telerehabilitation research articles. Additionally, there is evolving support the feasibility, evidence to and effectiveness acceptability, telehealth interventions in pediatric populations (6-11). Also evidence showed high rates of telerehabilitation satisfaction (12). A systematic review analyzed various studies on children with disabilities reporting that over half of the outcome measures improved telerehabilitation, often through coaching interventions aimed at caregivers and utilizing exercise programs (13). It seems that the amount and nature of research in the use of this model in the field of rehabilitation of children with NDDs have not been investigated in the era of Covid-19. Therefore, the purpose of this review was to outline the dimensions and nature of telehealth services in the rehabilitation of children with NDDs in the era of Covid-19, and it seems that the scoping review method was the most suitable for the purpose of this study.

2- MATERIALS AND METHODS

In this study, to determine the number, variety and characteristics of studies related to telerehabilitation of children with NDD, the updated scoping review method by Peters et al. was used. Compared to the systematic review method, three major differences are mentioned, which are related to the registration of the search protocol in the relevant databases, critical review and synthesis of findings (14, 15). The updated method of Peter et al. is a 9-step method with some additional steps compared to the previously presented methods. These steps generally organize the pre-search, search and post-search steps. For example, in the pre-search section, the exact determination of goals, criteria and search approach is placed. And implementing the determined search approach, the next step includes selecting, extracting, analyzing, presenting and summarizing the evidence (14) (16). Then reporting and presentation were prepared by the use of Preferred Reporting Items for Systematic Reviews and Meta-(PRISMA) Analyses Extension Scoping Reviews guidelines (17).

2-1. Determining questions

- a) What is the number (size) of telerehabilitation research for children with NDDs during Covid-19 pandemic?
- b) What is the variety and characteristics of telerehabilitation research for children with NDDs during Covid-19 pandemic?
- c) Which part of methodological issues of telerehabilitation research for children with NDDs is more interesting for researchers during Covid-19 pandemic?
- d) Which kind of telerehabilitation technologies is more attractive to researchers during Covid-19 pandemic?
- e) Which area of telerehabilitation research for children with NDDs during Covid-19 pandemic is unexplored?

2-2. Determining criteria

Original peer reviewed articles related to the implementation of different telerehabilitation methods in providing services (assessment, intervention, and counseling) for children with NDDs that were published in English were included. Developmental and feasibility studies, qualitative studies, review articles. guidelines and recommendations, protocol studies, and case reports, studies on adults with disabilities or normal developing children: and those published in a **English** language other than were excluded.

2-3. Determining search approach

Searching the three databases including the Web of Science, PubMed, and Scopus for the studies published between 1 January 2020 and 12 December 2022. related articles were recognized. The search terms were ("COVID-19" OR "SARS-CoV-2") **AND** ("telerehabilitation" "telehealth") OR ("Intellectual Disability" AND OR "Developmental Disabilities" OR Disabilities" "Learning OR "Neurodevelopmental Disorders" OR "Autistic Disorder" OR "Autism Spectrum Disorder" OR "Attention Deficit Disorder with Hyperactivity" OR "Cerebral Palsy"). The search was performed with MeSH Terms in PubMed, with topic search in Web of Science and with title/abstract search in Scopus. The PubMed syntax is presented in **Appendix 1**.

2-4. Selecting related articles

A total of 535 articles were found by searching the databases. After that, duplicate articles (N=137) were removed. Then, two independent reviewers (Z.GH, F.H) screened the remaining articles (n=398) in two steps. At the first step, they screened the titles and abstracts of the articles. At the second steps, they screened the full texts of the studies to confirm compliance with entry criteria. In case of disagreement between the two in the final decision, a third reviewer (Z.N) was consulted.

2-5. Extracting, analyzing, presenting and summarizing

In this stage, where the emphasis is on extracting, analyzing and presenting information, with several meetings between two researchers (F.S, Z.N), the required data of each article including the first author, country, study design, sample size, age, measurement tools and main results were extracted, analyzed, and presented in **Table 1**.

3- RESULTS

Fig. 1 illustrates a PRISMA flow diagram for the process of searching and selecting eligible studies. With the

application of appropriate search filters, we found 535 studies, in PubMed (158 articles), Scopus (268 articles), and Web of Science (109 articles), by the use of adapted syntaxes to search each database; and 137 articles were removed as duplicates. Titles and abstracts of studies (n = 398) were screened. Irrelevant articles (n=304) were excluded through the stage of screening titles and/or abstracts. Full text screening of articles (n=94) showed that 67 studies did not meet the eligibility criteria, and so were excluded. After the full-text review, 27 articles were, finally, included in this scoping review.

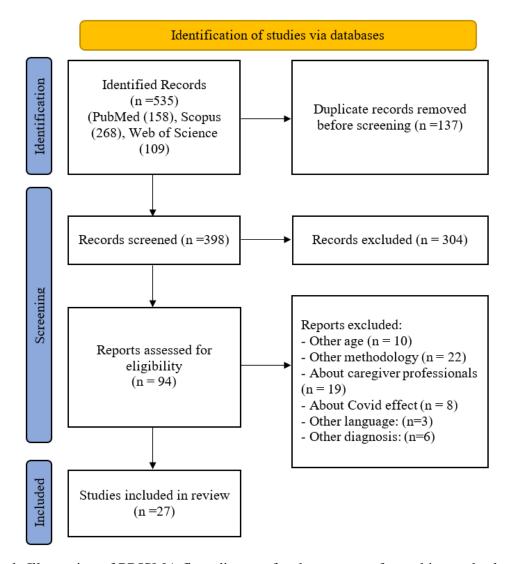


Fig. 1: Illustration of PRISMA flow diagram for the process of searching and selecting eligible studies

a) What is the extent and nature of telerehabilitation research for children with NDDs during Covid-19 pandemic?

Among the total of 27 studies published in 2021 and 2022, fifteen studies were published in 2021 and 12 in 2022. Most of

the studies were conducted in the United States (n=10, 37%) followed by Italy (n = 6, 23%). The distribution of published articles in each continent is shown in **Fig.** 2

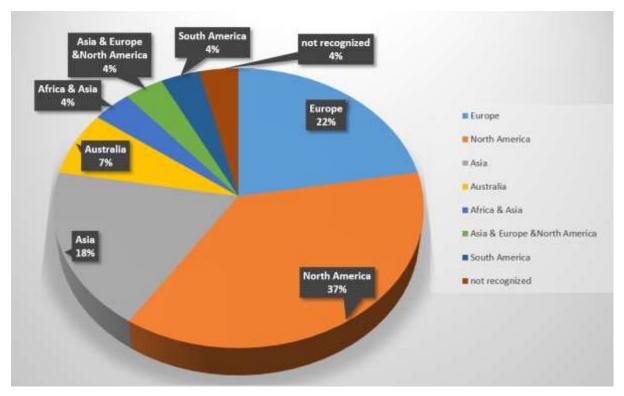


Fig. 2: Number of Articles Published in Each Continent

Populations of the selected studies were children with clinical diagnoses of ASD (n=9, 34%) developmental delay (n=3, 12%), cerebral palsy (n=3, 12%), student with disabilities (n=2, 7.4%), tic disorder (n=2, 7.4%), Rett syndrome (RTT) (n=1, 3.7%), down syndrome (n=1, 3.7%), fragile x (n=1, 3.7%), ADHD (n=1, 3.7%), social anxiety disorder (n=1, 3.7%), obsessive-compulsive disorder (OCD), disruptive behavior (n=1, 3.7%), and visually impairment (n=1, 3.7%).

In some studies tele-intervention was provided to both children and their caregivers (18-27). Some others provided the intervention to the child (28-31) or to the parents or family (32-35). It was

reported that children with ASD can participate in tele-intervention sessions and the intervention was effective in increasing engagement in online learning (25, 28).

The Program for the Enrichment and Education of Relational Skills (PEERS) intervention confirmed decreases in core autistic symptoms with a large effect size (18). The Internet-delivered format of The Coaching Approach behavior and Leading by Modeling (CALM) led to significantly greater reductions than waitlist in child discomfort with a large effect size (21). Also clinically meaningful reductions in tic severity and improved youth global impairment and functioning was reported

with large effect size. This reduction was maintained over a 6-month follow-up period with a very large effect size (27). Also significant improvements were reported in sleep quality of children with developmental delay and reduction of disruptive behavior in children with externalizing and internalizing problems.

b) More interesting methodological issues of telerehabilitation for researchers during Covid-19 pandemic

Services provided by telerehabilitation were mostly tele-intervention (n=19, 70.37%) followed by tele-evaluation (n=5, 18.52%) and tele-consultation (n=1, 3.7%) and two were case series.

Tele-intervention was used to provide services for relational skills and communication (18, 22, 32), reading, writing, mathematical skills and engagement in online learning (20, 25, 28, 29, 31), behavioral problems (23, 26, 30), Neromotor and neuropsychology (34-37), tic and anxiety (21, 27), visual impairment (33) and music therapy (19).

Tele-evaluation was used to evaluate parent-child interaction (38), mental health and loneliness (39), ASD scores symptoms (40), cognitive function (41), and sleep behavior (42).

c) More attractive telerehabilitation technologies to researchers during Covid-19 pandemic

More than half of the studies used teleconferencing for providing their services (n=15, 55.56%). Also web-based and chrome-based tools, web/iPad app, internet-based programs, MP4 videos, smart watch were used to provide services.

d) Unexplored telerehabilitation methods for children with NDDs during Covid-19 pandemic

As it was mentioned in the above part, tele-conferencing systems were most popular between researchers to administer their tele-rehabilitation research. It seems that other methods of providing tele-rehabilitation services such as web-based intervention, app-based intervention, and game based interventions as well as using devices or tools that have synchronous or asynchronous data sharing information with central data-bases are little explored. Also the evaluation part using visual error tools and apps applied for home visit and home setting evaluation are rarely examined by researchers.

4- DISCUSSION

The spread of coronavirus pandemic in recent decades has been unprecedented and it has had a significant impact on human life. The closure of schools, clinics and social programs imposed pressures on families and their children. However, this pressure was more challenging families and children with NDDs (43). Rehabilitation centers and schools made changes in their service delivery model and used tele-rehabilitation models. It has been reported in systematic reviews that the results obtained from tele-evaluation and tele-intervention delivery can be compared with face-to-face evaluation and intervention delivery (44, 45). Also, satisfaction with this way of providing services has been reported both in the group of users and in the group of providers (46).According to knowledge this study is the first scoping review of telerehabilitation for children with NDDs during Covid-19 pandemic.

Our findings revealed that Most of the selected publications are associated with children with ASD. The prevalence rate of NDD subgroups was reported as follows: intellectual disability (ID) (0.63 percent), ADHD (5–11 percent), ASD (0.70– 3 percent), specific learning disorder (SLD) (3–10 percent), communication disorders (CDs) (1–3.42 percent), and motor disorders (MDs) (0.76–17 percent) (1). Children with ASD are a broad and specific subgroup of children with NDDs.

Table-1: Extracted data from the selected articles

First Author (Year)	Country	Study Design	Sample Size, Male (N or %) and Diagnosis (N or %)	Mean Age (Year) or Age Range	Telerehabilitation Services	Measurement Tool	Results
Adler (18) (2022)	USA	pre- and post- intervention	22 Autism.	14 (1.50)	The PEERS® intervention teleconferencing and using materials Weekly 90-min session for 16 weeks.	Social Responsiveness Scale (SRS) The Quality of Socialization Questionnaire (QSQ) Test of Adolescent Social Skills Knowledge (TASSK)	Decreases in core autistic $t(21) = 5.511 p < 0.001$ Cohen's D = 1.18 increase in total invited and hosted get-togethers $t(20) = -5.975 p < 0.001$ Cohen's D = 1.30
Bailey (28) (2022)	Australi a	exploratory mixed methods design	21 autism	5–12 years	ABRACADABRA book reading activities 16 h over 8 weeks.	Neale Analysis of Reading Ability (NARA-3) Castles and Coltheart Test (CC-2) Wide Range Achievement Test (WRAT-4) Test of Everyday Reading Comprehension (TERC)	no significant improvements
Battistin (33) (2021)	Italy	before and after	106 visually impaired		Video Conferencing 30- 60 min	VAS	high levels of satisfaction p-value <0.001

First Author (Year)	Country	Study Design	Sample Size, Male (N or %) and Diagnosis (N or %)	Mean Age (Year) or Age Range	Telerehabilitation Services	Measurement Tool	Results
Bompard (19) (2021)	Italy	before and after	14 developmental delay,	< 12 years	visual 3 times a day for 12 consecutive days.	Sleep Disturbance Scale for Children (SDSC)	SDSC total score (P=0.010) SBD (P=0.012) SWTD sub-scores (P=0.032)
Bullard (38) (2021)	USA	online surveys	20 FXS	6-11 years	Semi-structured interactions videoconferencing. online questionnaires	interviews and observations	comfortable using technology (77% strongly agreed) videoconferencing software easy to use (69% strongly agreed)
(20) (2021)	USA	pilot intervention	10 ADHD	10 - 15 years	Digital Health Intervention (DHI) 6 week	interviews	successful use of the smart watch
Comer (21) (2021)	southeas tern United States	waitlist- controlled trial	40 SocAD	3.0 - 8.9 years (M = 6.2, SD = 1.8)	ICALM 12-session 16 weeks	The Child Behavior Checklist (CBCL) Children's Behavior Questionnaire (CBQ) The Child Anxiety Impairment Scale (CAIS)	decrease in: child anxiety symptoms fear discomfort anxiety-related social impairment Between-groups effects: anxiety symptoms: d = 0.31 child fear: d = 0.55 child soothability: d = 0.66 child discomfort: d = 0.89
Corona	USA	pre- and post-	49 (in person)	16 - 33	Six intervention sessions	Clinical global	across groups Differences:

First Author (Year)	Country	Study Design	Sample Size, Male (N or %) and Diagnosis (N or %)	Mean Age (Year) or Age Range	Telerehabilitation Services	Measurement Tool	Results
(32) (2021)		intervention	46 (telemedicine) 20 (hybrid) ASD evaluation	months	60-90 min	impressions of improvement (CGI-I) MacArthur-Bates Communicative Development Inventory (MCDI) Communication and Symbolic Behavior Scale Developmental Profile (CSBS DP)	play (Kruskal-Wallis H (2) = 6.77; p < .05) nonverbal communication (Kruskal-Wallis H (2) = 7.11; p < .05) social interactions (Kruskal-Wallis H (2) =6.61.; p < .05)
Cristinzia no (37) (2021)	Italy	an observational study	53 cerebral palsy	6 months - 12 years	Home-based TR Each session (50 minutes)	GMFM scores	significantly increased in median t0-t1 GMFM-66 score (P=0.000001) The t1-t2 score increased from (P=0.000005;
Day (31) (2022)	United States	one group pretest- posttest study	13 students with disabilities,	5th grade students	11 instructional lessons 45 min each session three weeks	number of total words written number of transitional words writing quality	statistically significant: total number of written words (Z = -2.045, p = .041) the number of transition words ($t(12)$ = -6.015, p ½ .001) the holistic writing quality (Z = - 2.659, p = .008).

First Author (Year)	Country	Study Design	Sample Size, Male (N or %) and Diagnosis (N or %)	Mean Age (Year) or Age Range	Telerehabilitation Services	Measurement Tool	Results
Gannon (49) (2021)	USA	A case series	Jane obsessive-compulsive disorder	15 year old	2-3 hours of group treatment 3 times a week		Decreased anxiety better tolerate social phobia
Garcia (22) (2021)	USA	pretest- posttest study	86 disruptive behavior	2 - 8 years	the course of 18 weeks Therapists coach caregivers on their parenting skills	ECBI Intensity Scale DPICS-IV Child Compliance Rate BASC-3 Internalizing Subscale	improved ECBI scores (t (83) = 10.19, p < .01) increased child compliance rate (t (32) = 6.13, p < .01) improved child BASC-3 Internalizing Problems Composite t scores (t (53) = 4.42, p < .01) medium to large effects
Graucher (23) (2022)	Israeli	open trial	55 autism and DB	3-9 years	12-weeks RUBI protocol 11 core sessions	Aberrant Behavior Checklist Home Situations Questionnaire	ABC Hyperactivity (t [31] = -2.84, p < .01) Irritability (t [31] = -2.44, p < .05) Overall ABC scale (t [30] = -2.22, p < .05). significant decrease in the HSQ overall score (t[25] = -3.55, p < .01)
Hegde (24) (2022)	India	Case Series	7 tic disorder	4-10 years	CBT	Yale Global Tic Severity Scale (YGTSS)	Improvement in severity improvement in the comorbid conditions
Houghto n (39) (2022)	Australi a	Online survey	476 with-or-without NDDs	adolescent s	Online assessments	The Perth A- loneness Scale Children's	elevated levels of adverse mental health

First Author (Year)	Country	Study Design	Sample Size, Male (N or %) and Diagnosis (N or %)	Mean Age (Year) or Age Range	Telerehabilitation Services	Measurement Tool	Results
						Depression Inventory-2 The Warwick- Edinburgh Mental Wellbeing Scale The Strengths and Difficulties Questionnaire	
Kim (25) (2022)	USA	multiple baseline design	3 students with disabilities	Second grade	teacher implemented a task analysis that listed five daily assignments		increase engagement in online learning
Logrieco (40) (2022)	Italy	A cohort	243 ASD	2–15 years	online survey	Pediatric Quality of Life Inventory TM PedsQL	Worsening of specific ASD core symptoms
Lotan (34) (2021)	Israel	pilot study	5 RTT	5–18 years	meetings via Skype the physical therapy program once a month over a 6- month	GAS	Progress in 16 of 20 goals
Matano (26) (2021)	Japan	pilot study	4 ASD	3 - 5 years	I-PCIT	Eyberg Child Behavior Inventory (ECBI)	effective in improving behavioral problems
Meguid (41) (2022)	Egypt	a cohort prospective study	135 Down syndrome	8 months— 15 years	online therapy sessions home-based training by virtual online sessions 45 min. 2 sessions/week.	Portage assessment of the child's skills	in home training: less delay in cognitive abilities (-5.9 ± 1.7 vs. $-$ 8.3 ± 1.5) less delay in language

First Author (Year)	Country	Study Design	Sample Size, Male (N or %) and Diagnosis (N or %)	Mean Age (Year) or Age Range	Telerehabilitation Services	Measurement Tool	Results
							performance $(-3.5 \pm 1.8; -8.5 \pm 1.7)$ less delay in motor abilities $(-5.2 \pm 2.2 \text{ vs.} -11.4 \pm 1.3)$
Rachami m (27) (2022)	Israel	RCT	41 Tic disorder	7 - 18 years	Internet-based, self-help CBIT program (ICBIT)	Yale Global Tic Severity Scale	reductions in tic severity improved youth global impairment and functioning primary outcome measure (Cohen's d = 0.91) at post- intervention (Cohen's d = 2.25) 6 months after
Sarti (29) (2021)	Italy	Online treatment	56 Specific Learning Disorders (SLD) Cerebral Palsy (CP)	130 months	SLD :2 online treatment sessions a week (45 min) CP :1 online treatment sessions (45 min) 13 weeks	Comprehensive Inventory of Thriving for Children Scale of Positive and Negative Experience	highest scores on the Learning scale
Scarselli (42) (2022)	Italy	survey	73 ASD	2.1-6 years	Questionnaire via email	Children's Sleep Habits Questionnaire (CSHQ)	89.3% had a score above 41
Schlichti ng (35) (2022)	Brazil	pre-post feasibility experimental study	15 delayed neuromotor development	3- 18 months	home-based program five times a week over 12 weeks	GMFM-88 Alberta Infant Motor Scale (AIMS)	increased GMFM-88 total score (Z=-2.701; p = 0.007) increased AIMS percentile (Z=-1.973; p = 0.049)
Singh		single-case	3	adolescent	The telehealth consultant	visual analysis	reductions in the frequency

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First Author (Year)	Country	Study Design	Sample Size, Male (N or %) and Diagnosis (N or %)	Mean Age (Year) or Age Range	Telerehabilitation Services	Measurement Tool	Results
(30) (2021)		experimental study	ASD	S	via Zoom		of Self-injurious behavior
Sivarama n (50) (2021)	(Belgiu m, India, Mexico, Costa Rica)	nonconcurren t multiple- baseline design	6 ASD	2 - 12 years	coaching caregivers via video-calling		all participants wore a face mask for a period of 10 min
Shierk(36) (2021)	USA	one-group pretest- posttest design	5 UCP	33 - 43 months	eight, 1 hour group sessions	Assisting Hand Assessment (AHA), Canadian Occupational Performance Measure (COPM)	significant changes in bimanual performance (AHA) and satisfaction (COPM)

In recent studies, the impact of quarantine on children with ASD and their parents has been investigated and high psychological distress has been reported (43, 47).

The educational and rehabilitation needs of these children were suddenly cut off after the quarantine was imposed. The social relationships of these children were also severely affected. In this situation, the family had not received the necessary education and training on how to communicate with these children and how to provide education to these children. Moreover, many of these children did not have access to services even for their initial diagnosis with the start of the pandemic. Studies have been conducted to carry out tele-diagnosis for these children. In this situation, many caregivers whose children were diagnosed with autism and lost access to common services are in great need of alternative services. Also, the therapists were faced with a situation where they did not know how to provide remote rehabilitation services to a child with special needs such as autism. Then, it seems that this challenge along with the needs of families, children and therapists have created the desire to conduct studies in this field. The age ranges of the participants in selected studies are very diverse. From three months to 18 months in one study to adolescence in the other; however, it seems that most study participants have been children above five years old. This might be attributed to the issue reported in a study on caregivers of ASD children indicating that children who were under five years of age had the most problems in receiving tele-services, and enjoyed the least amount of benefit. In another study, it was reported that Children under the age of 5, primarily those with better gross motor function levels showed the greatest GMFM-66 value variations. No statistically

significant increase was reported in the level V group (37).

COVID-19 pandemic 229 affected countries and territories. Among these countries the United States took the first place. And the majority of selected publications, in the present study, were conducted in the United States followed by Italy. The outbreak of the COVID-19 pandemic, which was very widespread in Italy, forced the Italian Government to immediately adopt very restrictive and severe containment measures; and Italy was the first European country to impose such measures (29, 33, 40). Obviously, events like this pandemic have affected the use of telerehabilitation model of service delivery and subsequently its publication in this country, though it was reported that Italy had no publication in the field of telehealth and occupational therapy (6).

Remote rehabilitation and habilitation services have been introduced as teleevaluation, tele-assessment, telemonitoring, tele-intervention, telesupervision and tele-consultation. In the present study, 5 studies were conducted tele-evaluation in the form of observation with video conferences and questionnaires (38), online assessments via self-report measures (39), online surveys (40, 41) and via email (42). The accuracy and reliability of tele-evaluation was also examined in some studies (48). In the present study, it seems that compared to remote intervention, researchers have shown less interest in remote evaluation.

Assessment, intervention, counseling, supervision, and monitoring can be provided remotely in various ways, e.g., through video conferencing, web-based, virtual games with remote monitoring and other methods. In line with the findings reported in our previous study, video conferencing was the most popular method in providing services (6).

Although the variety of telerehabilitation service deliveries seem to have increased, and even the number of participants in the studies has increased, considering the rapid spread of the pandemic, still the studies seem not to be adequately organized in terms of methods.

5- COMPETE OF INTERESTS

None.

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