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Diversity of practices in telerehabilitation for children with disabilities and effective intervention characteristics: results from a systematic review

Chantal Camden^{a,b} , Gabrielle Pratte^a, Florence Fallon^a, Mélanie Couture^{a,b} , Jade Berbari^b and Michel Tousignant^a

^aSchool of Rehabilitation, Sherbrooke University, Sherbrooke, QC, Canada; ^bResearch Center of the Sherbrooke University Hospital, Sherbrooke, QC, Canada

ABSTRACT

Purpose: To describe the characteristics and effectiveness of pediatric telerehabilitation interventions offered to children 0–12 years old or to their families.

Methods: A systematic review was conducted on randomized control trials published between 2007 and 2018 involving at least one rehabilitation professional who provided services remotely. Information was extracted about key study, participants and intervention characteristics. The percentage of outcomes that improved were computed per study, and per intervention characteristic.

Results: Out of 4472 screened articles, 23 were included. Most studies were published after 2016 and evaluated outcomes related to the child's behavior (n ¼ 12, 52.2%) or to the parent (n ¼ 10, 43.5%), such as parental skills or stress. Overall, 56.1% (SD: 38.5%) of evaluated outcomes improved following telerehabilitation. A great diversity of population and teleintervention characteristics was observed. Effective interventions tended to target parents, centered around an exercise program, used a coaching approach, focused on improving children's behavioral functioning, lasted >8 weeks and were offered at least once a week.

Conclusions: Intervention characteristics that appear to yield better outcomes should inform the development of future telerehabilitation studies, especially in populations for whom telerehabilitation is currently understudied (e.g., children's with physical functioning difficulties). Future trials should compare telerehabilitation interventions to well-described evidence-based face-to-face interventions, and document their cost-effectiveness.

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KEYWORDS

Telehealth; children; rehabilitation; online intervention

> IMPLICATIONS FOR REHABILITATION

- Despite a great variety in practices, telerehabilitation might be as effective as face-to-face interventions, across disciplines, for a variety of clinical outcomes.
- Telerehabilitation might be more effective when coaching approaches are used, especially to achieve outcomes related to children's behavior or parental skills.
- Further research is required to better understand the characteristics of effective telerehabilitation interventions, and to determine how these characteristics may differ for specific populations and outcomes.

Introduction

Rehabilitation professionals working with children with disabilities are increasingly encouraged to consider the child's development and focus on activity and function [1–3]. Current thinking suggests that rehabilitation professionals should partner with families and stakeholders to share information, build capacity, foster self-management and create opportunities for children to practice and develop skills [1,3–6]. In parallel, technologies offer unprecedented opportunities to support the implementation of these best practices, by providing platforms where families can access online information at their own pace, whenever and wherever they want [7–9]. Moreover, technology can also be used to “bring services closer to patients” [10,11] and to increase the accessibility and the cost-effectiveness of services [12–14]. Rehabilitation

professionals have however been relatively slow in embracing the opportunities provided by such technologies despite the avenues they offer as a means to delivering cost-effective services [15].

Technologies can be used in a variety of clinical ways to work with children and their families. Online modules presenting evidence-based information [16–18] or applications aimed at fostering physical activity, healthy lifestyle or chronic condition management [19,20] are examples of how the Internet has been used to disseminate general reliable information. For specific treatment goals, virtual reality systems [21] and web-based games [19] might offer interesting and effective alternatives to traditional one-on-one interventions. Despite promising results, these applications offer limited opportunity for therapist–client interactions. These interactions are required to implement best practices

promoting activity and participation, and helping families manage the child's health condition. When technology is used to support therapist–client interactions or interactions among professionals, the term telehealth is generally used [22].

Telehealth can be applied in very different ways, ranging from supporting interprofessional meetings to treating patients [23,24]. Telemedicine, another common term, generally refers to doctors treating patients remotely. Telerehabilitation is the most common term used when rehabilitation professionals interact with patients at a distance, through information and communication technologies, to provide rehabilitation services [25,26]. The technology used by rehabilitation professionals can be diverse, ranging from simple day-to-day applications (e.g., contact via phone calls or by email) to complex technologies (e.g., specialized equipment installed in a clinical setting and at home). Telerehabilitation might be particularly well suited to implementing best practices for children with disabilities when the focus of the therapies is on supporting the children and their families, problem-solving with them to foster the child's development and functioning.

Systematic reviews have documented the use and the effectiveness of telerehabilitation with a wide range of clients [27,28]. Most systematic reviews have been conducted for specific populations, such as stroke [29,30] or multiple sclerosis [31] patients. Telerehabilitation studies tend to focus on adult populations and relatively little is known about how this approach is used and how effective it could be in pediatric rehabilitation. Promising results have been reported in literature reviews conducted on specific pediatric populations (e.g., autism) [32]. However, no systematic review has described the variety of practices among high-quality telerehabilitation studies and the key intervention characteristics that positively impact effectiveness. Given the interdisciplinary nature of disability management and the similarities in best practices across disciplines and populations, we conducted a systematic review of all relevant telerehabilitation interventions in pediatrics to inform the development of future telerehabilitation studies for children with disabilities. In this study, we used the World Health Organization (WHO) definition of rehabilitation and disability, and thus include all interventions optimizing functioning and aiming at reducing impairments, activity limitations, and participation restrictions for children with chronic conditions [33,34]. We chose to focus on children 0–12 years of age and to exclude interventions specifically targeting teenagers, as these interventions often target a very specific set of objectives, related to life transition. The aim of this study was thus to describe the characteristics and effectiveness of pediatric telerehabilitation interventions offered to children 0–12 years old or to their families.

Methods

This systematic review is reported based on the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement [35].

Identification and selection of studies

An electronic search was completed in CINAHL, PsychInfo, Francis, and PubMed (MeSH database) in March 2018. The search strategies included synonyms for our three main components: rehabilitation (e.g., “physiotherapists”), children (e.g., “pediatric,” “parents”) and telehealth (e.g., “web-based,” “online services”). A detailed list of keywords is presented in the [Supplementary Materials \(Supplementary Table S1\)](#). Zotero was used for

screening, removing duplicates and classifying articles along the eligibility process.

Studies had to meet the following criteria: (1) including 0 to 12 year old children or their family members, (2) using a telerehabilitation intervention, defined as an intervention aimed at improving children's functioning involving at least one rehabilitation professional providing all or part of their services remotely, (3) using an experimental randomized trial design reporting results for at least one outcome measure, (4) having been published between January 2007 and March 2018, in English or French, to focus on recent technologies, and (5) scoring 5/10 or more on the Physiotherapy Evidence Database (PEDro) checklist as this cut-off was previously used to determine studies that were of moderate to high quality [36]. Qualitative studies, abstracts or literature reviews were excluded. The references listed in existing systematic reviews were hand searched for additional relevant articles. No limit was placed on the type of technology, the duration of the telerehabilitation intervention or the outcomes evaluated. All childhood disabilities were included as per the WHO definition, including not only neurodevelopmental disabilities and acquired injuries, but also emotional disturbance or medical conditions leading to functional limitations. Interventions fostering the general development of typically-developing children were excluded. Two reviewers independently reviewed the titles and abstracts of the first 10% of identified studies. As article selection was consistent between both reviewers, the rest of the process was completed by a single reviewer. The same approach was adopted for the quality assessment using the PEDro checklist, and for the final selection of relevant articles. A third reviewer was available at all times to adjudicate in case of disagreement.

Data extraction and analysis

Study characteristics (i.e., quality, design, publication date, nature of the control group, outcomes assessed), participant characteristics (i.e., number of participating families, age and condition of the child) and key intervention characteristics (i.e., type of intervention, participants targeted, interaction style with parents, technology used, frequency and duration of the intervention), including information about the professionals who provided the intervention, were extracted for every relevant study. To acknowledge the diversity of populations included in our study and explore any potential differences among categories of populations and interventions, we classified studies into two categories: those targeting children with neurodevelopmental or acquired disabilities, and those including children with emotional dysfunctioning or chronic medical conditions. We also classified interventions as either focusing on behavioral functioning (e.g., when authors reported that the aim of the intervention was to improve children's behavior, or parent–child relationship), or physical functioning (e.g., to improve motor skills or bimanual function).

A thematic analysis [37] was performed on the extracted data, where codes were assigned to the extracted information in order to create general categories and characterize the studies, participants, and interventions. Frequencies and percentages were later computed. This process was undertaken and validated by two reviewers, assisted by an adjudicator in case of disagreement. For each study, among outcomes evaluated pre- and post-intervention, we computed a percentage of improved outcomes, defined as outcomes reported to have significantly improved over the control group or over time. To explore patterns leading to greater effectiveness across studies, a percentage of improved outcomes was associated with each key intervention characteristic, reflecting

the mean percentage of improved outcomes for all studies that incorporated this intervention characteristic.

Results

Study selection

Figure 1 presents the flow chart of the identification, screening and selection process. The titles and abstracts of 4472 articles were screened; of these, 4340 articles were excluded because they did not meet the inclusion criteria for age or design, or did not pertain to a genuine telerehabilitation intervention according to the definition used for the present review. After reading the full text of the 132 remaining articles, an additional 109 studies were excluded based on our inclusion criteria or because they were neither accessible through the University library, nor publicly available (e.g., on the Web or on Pubmed), nor available after directly contacting the authors. No relevant article scored below the quality assessment threshold on the PEDro scale. Twenty-three articles [19,20,38–58] were included in the systematic review. The study, participant and intervention characteristics are summarized in Table 1.

Quality of methodological reporting

The mean score on the PEDro scale was 6.8 (SD ¼ 0.9). All studies met the first PEDro criteria (specifying the eligibility criteria) and

had similar groups prior to intervention (Criteria #4). Only one study respected PEDro Criteria #5 (blinding of subjects) and no study respected PEDro Criteria #6 (blinding of therapists). Nine studies (39.1%) included a blinded assessor (Criteria #7). The last four PEDro criteria were respected by most studies: Criteria #8 (one key outcome for at least 85% of the subjects), #9 (intention-to-treat analysis), #10 (statistical comparisons) and #11 (point measures and measures of variability) were fulfilled in 18 (78.3%), 19 (82.6%), 23 (100.0%) and 22 (95.7%) of studies, respectively.

Study, participant and intervention characteristics

Study characteristics

All studies were RCTs, but a few were described as pilot RCTs (n¼ 14, 17.4%). More than half were published since 2016 (n¼ 17, 73.9%). Control groups most frequently used a waitlist approach (n¼ 10, 39.1%). Other control groups included face-to-face interventions without telerehabilitation (n¼ 4, 17.4%), educational groups (n¼ 3, 13.0%), usual treatment (e.g., consultation only) (n¼ 3, 13.0%) or online resources (n¼ 4, 17.4%). Studies evaluated a mean of 5.5 outcomes pre- and post-intervention, ranging from 1 to 8 different outcomes. Primary outcomes were not always clearly stated and most interventions had multiple expected outcomes, as reflected by the broad diversity of objectives assessed, classified into seven categories: 1) child’s behavior (n¼ 12 studies included the child’s behavior as an outcome, 52.2%), 2) parental

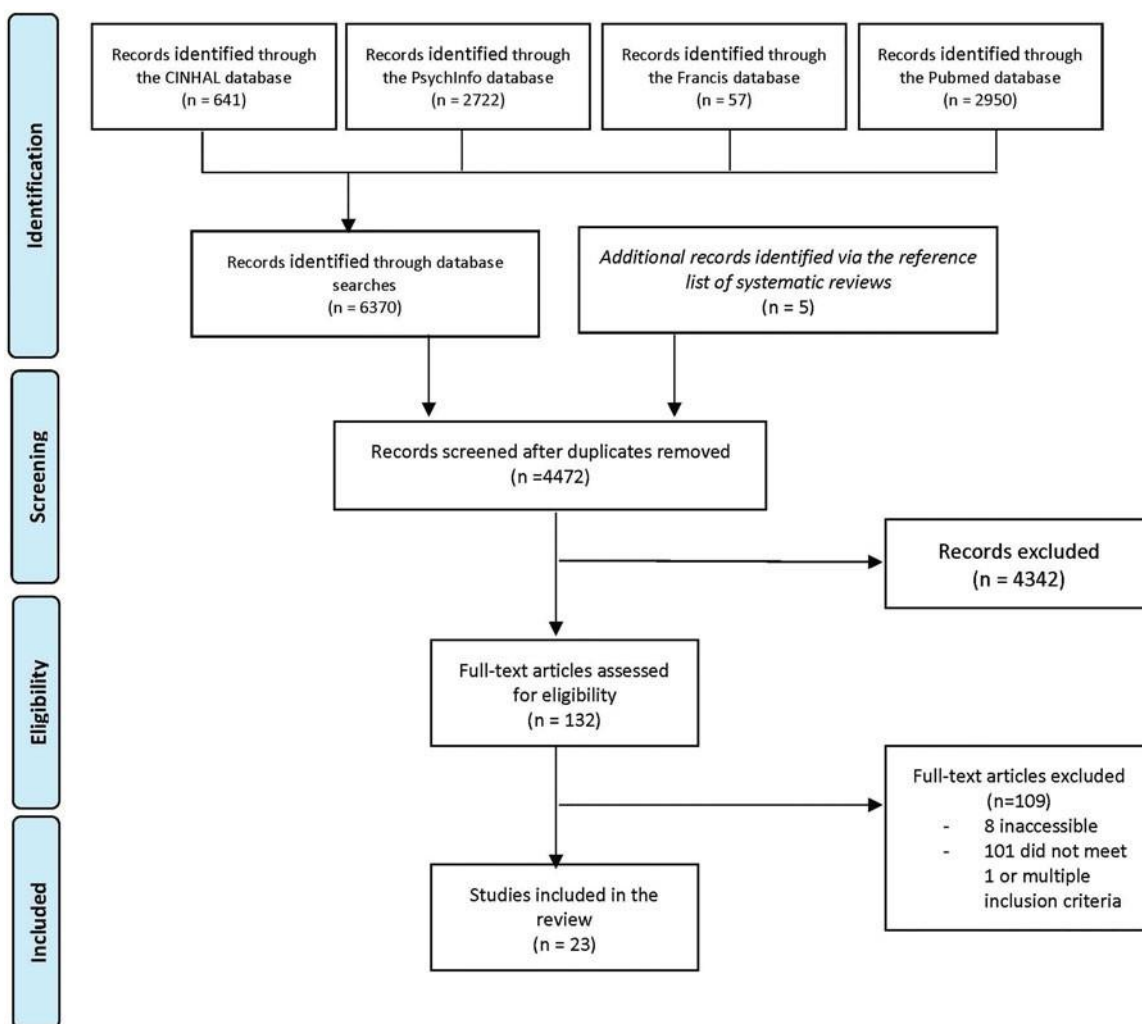


Figure 1. PRISMA flow chart.

Table 1. Summary of studies characteristics and outcomes.

Author (year)	Study design (PEDro score)	Total n	Professionals	Diagnosis	Child age	Intervention group	Control group	Main outcomes	Improved outcome (%)
Baque and al. (2017) [36]	RCT (7)	60	Physiotherapist	Acquired brain injury	8–16 years old	“Move it to improve it” a web-based therapy to improve motor skills – 30min, 6 times a week for 20 weeks and a weekly contact with a professional via phone, email or videoconferencing	Waitlist control	<ul style="list-style-type: none"> Function: Functional strength ^m, 6MWT σ =, High-level Mobility Assessment Tool σ =, TUG σ =, Go Test σ =, Habitual physical activities σ =, 28-item Mobility Questionnaire parent report questionnaire σ ¼ 	14.3%
Comer and al. (2017a) [38]	Pilot RCT (8)	22	Psychology masters-level trainees	Obsessive compulsive disorder	4–8 years old	Family based-CBT via video-conference to teach parents coaching skills; computer game to enhance children’s understanding of treatment concepts-12 sessions within 14 weeks	In person treatment	<ul style="list-style-type: none"> Parental outcomes: FAS-PR “=, CGI-S “¼ Severity: CY-BOCS “=, OCD CSR “=, CGI-S “¼ Function: CGAS “¼ Satisfaction: High Retention: High 	100.0%
Comer and al. (2017b) [37]	RCT (7)	40	Psychologist	Disruptive behavior disorders	3–5 years old	I-PCIT a videoconferencing behavioral parent-training program to coach parent into improving their parent–child relationships – 60min, once a week for 10 to 14 weeks	In-person treatment	<ul style="list-style-type: none"> Behavior: ECBI intensity score “¼, CBCL externalizing “¼ Severity and function: CGI σ =, CGAS σ ¼ Parental outcomes: ECBI problem score “=, BTPS “^m 	66.7%
Conaughton and al. (2017) [39]	RCT (7)	42	Psychologist	Autism spectrum disorder and anxiety disorder	8–12 years old	Internet-based cognitive behavioral therapy (CBT) intervention (BRAVE-ONLINE) with a weekly 60-min online contact for support, 10 to 14 weeks long	Waitlist control	<ul style="list-style-type: none"> Satisfaction: High Severity: CSR “=, ADIS-C/P “¼ Function: CGAS “¼ Behavior: CBCL “¼ Other (anxiety symptoms): SCAS-P “¼ 	100.0%
Ferre and al. (2016) [40]	RCT (8)	24	Occupational therapist	Unilateral cerebral palsy	2–10 years old	H-HABIT (parental supervision) via webcam-based software at home to improve bimanual function – 60 min once a week for 9 weeks	At home supervision	<ul style="list-style-type: none"> Satisfaction: High Function: COPM-P “^m/-S “¼ Motor: AHA σ =, BBT “^m 	75.0%
Fossum and al. (2018) [41]	RCT (7)	464	Health care professionals or semi-professionals	Disruptive behavior disorders	4 years old	Internet-based intervention with a 45-min weekly telephone call to improve child externalizing behavior and parenting skills	Educational control group	<ul style="list-style-type: none"> Behavior: CBCL “? Parental outcomes: Barkley’s Adult AD/HD Quick Screen “?, DASS-21 “? 	100.0%

(continued)

Table 1. Continued.

Author (year)	Study design (PEDro score)	Total n	Professionals	Diagnosis	Child age	Intervention group	Control group	Main outcomes	Improved outcome (%)
Grogan-Johnson and al. (2010) [42]	Cross-over RCT (6)	24	Speech language therapist	Language disorder	4–12 years old	Individual therapy via video-conference with the child to attain child's objectives – 4 months followed by usual therapy for 4 months	Waitlist control	<ul style="list-style-type: none"> Behavior: CBCL "? GFTA-2 σ¼ Satisfaction: High Retention: missed session < 1% 	0.0%
Hinton and al. (2017) [43]	RCT (6)	98	Trained practitioner: Social workers, psychologists, doctors, counselors	Developmental, intellectual and physical disabilities	2–12 years old	Triple P Online – Disability (TPOD-D): a platform web using modules education with weekly contact (phone or email) with a professional for 6 weeks to improve child's behavioral and emotional problems, parenting practices and family adjustment	Usual treatment	<ul style="list-style-type: none"> Behavior: DBC-P "m, CAPES-DD "m Parental outcomes: PAFAS "m Satisfaction (CSQ): High 	100.0%
Ingersoll and al. (2016) [44]	Pilot RCT (6)	28	Masters level trainee	Autism spectrum disorder	19–73 months	ImPACT therapist-assisted group: 12 self-directed lessons of approximately 75min to 30min-contact with a professional via videoconferencing for coaching session twice a week for 6 months	Self-Directed access to ImPACT Online sessions	<ul style="list-style-type: none"> Parental Outcomes: Fidelity "m, PSOC " =, FIQ "¼ Other (Positive perceptions): "m Communication: Language Targets "m, MCDI " =, VABS-II σ¼ Function and Motor: σ ¼ 	75.0%
James and al. (2015) [19]	RCT (7)	102	Occupational therapist, physiotherapist, psychologist	Unilateral cerebral palsy	8–18 years old	"Move it to improve it" a web-based therapy to improve motor skills – 20 to 30 min, 6 or 7 times a week for 20 weeks and contact with professionals (email, phone or skype) once a week	Usual treatment: consultation only	<ul style="list-style-type: none"> Function: COPM "m Motor: AMPS "m, JTTHF-impaired limb σ =, MUUL σ =, AHA σ¼ Other (vision): TVPS-3 "m 	50.0%
Kierfeld and al. (2013) [45]	RCT (6)	48	Psychologist	Hyperactive or oppositional children	3–6 years old	Self-administered behavioral intervention (read one chapter of a book each week) followed by phone consultation – < 20 min once a week for 11–13 weeks	Waitlist control	<ul style="list-style-type: none"> Behavior: CBCL "m Parental outcomes: HSQ "m, PS "m, PSBC "m, PPS σ =, DASS " =, PCC σ =, QJPS "m 	75.0%
Kuravackel and al. (2018) [46]	RCT (6)	33	Psychologist and Doctoral level psychology student	Autism spectrum disorder	3–12 years old	Collaborative Model for Promoting Competence and Success for Hope (C-HOPE): an 8-week parent intervention program delivered via telehealth to improve child educational outcomes using four 120-min group sessions and four 60-min individual telehealth sessions once a week	Waitlist control and face-to-face intervention	<ul style="list-style-type: none"> Behavior: ECBI "m Parental outcome: PSI-4-SF σ =, BPS σ¼ Satisfaction: Very high Other (therapeutic alliance): High 	33.3%

(continued)

Table 1. Continued.

Author (year)	Study design (PEDro score)	Total n	Professionals	Diagnosis	Child age	Intervention group	Control group	Main outcomes	Improved outcome (%)
March and al. (2009) [47]	RCT (8)	73	Psychologist	Anxiety disorders	7–12 years old	“BRAVE for children-Online” an internet based CBT (reading, exercise, game, quiz) to teach anxiety management strategies – once a week automatic mail and two phone contacts	Waitlist control	<ul style="list-style-type: none"> Behavior: CBCL^m Parental outcomes: CES-D σ^4 Severity: SCAS-C “=, -p^m Satisfaction moderate 	75.0%
Mast and al. (2014) [48]	Pilot RCT (6)	7	Psychology doctoral students	Abusive head trauma	3–9 years old	I-InTERACT program: online sessions live coaching to parents (via Skype or Movi Client and wireless ear-piece) – 10–14 sessions over 5 months one initial in-home visit.	Online resources	<ul style="list-style-type: none"> Behaviour: ECBI σ^m/σ^4 Communication: DPICS σ^m/σ^4 	0.0%
Mitchell and al. (2016) [20]	RCT (7)	101	Physiotherapist, occupational therapist, neuropsychologist	Unilateral cerebral palsy	8–17 years old	Training program monitored by professional via email, telephone or videoconference to improve gross motor activities – Training: 30min, 6 times a week for 20 weeks, contact with professional as needed	Usual treatment	<ul style="list-style-type: none"> Function: LIFE-H σ^4 Motor: MobQues28 $\sigma^=$, Strengh σ^m, 6MWT σ^m, ActiGraph σ^4 Retention: High 	0.0%
Piovesana and al. (2017) [49]	RCT (7)	60	Physiotherapist, occupational therapist, neuropsychologist	Acquired brain injury	8–16 years old	“Move it to improve it”: a 6-times a week web-based therapy to improve motor skills with a weekly contact (phone or email) with professional for monitoring during 20 weeks	Waitlist control	<ul style="list-style-type: none"> Function: MACS$\sigma^=$, GMFCS$\sigma^=$, WISC-IV$\sigma^=$, D-FEFS$\sigma^=$, CTMT$\sigma^=$, TOL$\sigma^=$, TEA-Ch$\sigma^=$, BRIEF σ^4 Satisfaction: Moderate to high Other: Daily intake “^m, Height “^m, Weight σ^4 Satisfaction high 	0.0%
Powers and al. (2015) [50]	RCT (9)	78	Dietician and psychologist	Cystic fibrosis	2–6 years old	Individual nutritional advice and parental training for behavioral child-management skills with telephone follow-up – Once a week for 8 weeks monthly for 4 months	Education and behavioral placebo	<ul style="list-style-type: none"> Parent outcomes: GSI “^m, CES-D $\sigma^=$, PSI $\sigma^=$, CSES σ^4 	66.7%
Raj and al. (2015) [51]	RCT (6)	37	Psychologist	Traumatic brain injury	3–9 years old	I-InTERACT: self-guided online sessions live parental coaching while playing with their child (via Skype or Cisco Movi and wireless earpiece) – 10–14 sessions over 4–6 months (including one in-person session at home).	Online resources	<ul style="list-style-type: none"> Parent outcomes: GSI “^m, CES-D $\sigma^=$, PSI $\sigma^=$, CSES σ^4 	25.0%

(continued)

Table 1. Continued.

Author (year)	Study design (PEDro score)	Total n	Professionals	Diagnosis	Child age	Intervention group	Control group	Main outcomes	Improved outcome (%)
Ricketts and al. (2016) [52]	RCT (8)	20	Psychologist and psychiatrist	Chronic Tic Disorders	8–16 years old	Comprehensive Behavioural Intervention for Tics via Skype – 60–90 min, 8 sessions over 10 weeks	Waitlist control	<ul style="list-style-type: none"> Severity: YGTSS ^m, PTQ ^m Satisfaction: High 	100.0%
Sourander and al. (2016) [53]	RCT (6)	464	Licensed health care professionals	Disruptive behavior disorders	4 years old	Strongest Families Smart Website (SFSW): Online sessions (e.g.: exercises, video) and coaching-call to improve parent skills and parent–child relationships – One online session and one 45 min call per week for 11 weeks	Education control group	<ul style="list-style-type: none"> Behavior: CBCL ^m, ICUS ^m Parental outcomes: PS^m, DASS \emptyset/4 Satisfaction: very high Retention: 75–77% 	75.0%
Storch and al. (2011) [54]	RCT (8)	31	Psychology doctoral students	Obsessive compulsive disorder	7–16 years old	Family-based cognitive-behavioral therapy delivered via web-camera (W-CBT) and email to coach parents and to improve child's symptoms and developmental level – 60–90 min, 14 sessions over 12 weeks	Waitlist control	<ul style="list-style-type: none"> Severity: CY-BOCS ^m, CGI-S/I ^m, COCIS-C/P^m Other (child anxiety and depression) MASC \emptyset=, CDI \emptyset/4 Satisfaction: High 	60.0%
Vismara and al. (2018) [55]	RCT (5)	61	Trained therapist	Autism spectrum disorder	18–48 months	Telehealth parent training in the Early Start Denver Model (P-ESDM) via 12 weekly 1.5h-videoconferencing sessions website access to P-ESDM learning resources	Usual community treatment, follow-up, online resources	<ul style="list-style-type: none"> Behavior: ^m Other (fidelity): ^m Other (program website use): ^m Satisfaction: High^m 	100.0%
Williams and al. (2016) [56]	Pilot RCT (6)	18	Psychologist	Leukemia	2–8 years old	Triple P via group videoconferencing (Go to meeting on Ipad) and individual calls to improve parent skills – once a week for 8 weeks	Waitlist control	<ul style="list-style-type: none"> Behavior: SDQ \emptyset/4 Satisfaction: High Retention: moderate 	0.0%

Effectiveness symbols: " indicates improvement over time in participants having received the telerehabilitation intervention; \emptyset indicates no difference over time; ^m indicates greater improvements in the telerehabilitation group compared to the control group; [?] indicates no statistical difference between groups; ? indicates no information provided.

ActiGraph: ActiGraph GT3X^B tri-axial accelerometer; ADIS-IV-C/P: Anxiety Disorders Interview Schedule for Children and Parents for DSM-IV; AHA: Assisting Hand Assessment; AMPS: Assessment of Motor and Process Skills; BBT: Box and Blocks Test; BRIEF: Behaviour Rating Inventory of Executive Functioning; BTPS: barriers to treatment participation scale; CAPES-DD: Child Adjustment and Parent Efficacy scale; CBCL: Child Behaviour Checklist; CDI: Child depression inventory; CES-D: Centre for epidemiological studies for depression scale; CGAS: Children's Global Assessment Scale; CGI-S/I: Clinical Global Impression-Severity/Improvement scale; COCIS-C/P: Child obsessive compulsive impact scale-child and parent versions; COPM-P/S: Canadian Occupational Performance Measure Performance/Satisfaction; CSR: Clinical severity rating; CSES: Caregiver Self-Efficacy Scale; CTMT: Comprehensive Trail Making Test; CY-BOCS: Children's Yale-Brown Obsessive Scale; CIS-P: Columbia impairment scale, parent-report version; DASS: Depression Anxiety Stress Scale; D-FEFS: Delis-Kaplan Executive Functioning System; DPICS: Dyadic Parent-Child Interaction Coding System; DBC-P: Developmental Behaviour Checklist - Primary Carer version; ECBI: Eyberg Child Behaviour Inventory; FAS-PR: Family Accommodation Scale-Parent Report; FIQ: Family Impact Questionnaire; GFTA-2: Goldman-Fristoe Test of Articulation; GMFCS: Gross motor function classification scale; GSI: Global Severity Index; HSQ: Home situation questionnaire; ICUS: Inventory of Callous-Unemotional Scale; JTTHF: Jebsen-Taylor Test of Hand Function; MASC: Multidimensional anxiety scale for children; MCDI: MacArthur-Bates Communicative Development Inventory; MobQues28: 28-item Mobility Questionnaire; MUUL: Melbourne Assessment of Unilateral Upper Limb; LIFE-H: Assessment of Life Habits; OCD CSR: Obsessive-compulsive disorder Clinical Severity Rating; PAFAS: Parenting and Family Adjustment Acales; PPC: Parent Problem Checklist; PPS: Parent practice scale; PS: Parenting scale; PSBC: Problem setting and behaviour checklist; PSI: Parenting stress index; PSOC: Parental sense of Competence Scale; PTQ: Parent Tic Questionnaire; QJPS: Questionnaire on Judging Parental Strains; SCAS-S/P: Spence Children's Anxiety Scale, Child or Parent version; SDQ: Strengths and difficulties questionnaire; Tea-Ch: Test of Everyday Attention for Children; TOL: Tower of London; TUG: Time up and go; TVPS-3: Test of Visual Perceptual Skills; VABAS-II: Vineland Adaptive Behaviour Scales Second Edition; WISC-IV: Wechsler Intelligence Scale for Children; YGTSS: Yale Global Tic Severity Scale; 6MWT: 6min walk-test.

outcomes, including parents' skills and stress, and parent-child interactions (n = 10, 43.5%), 3) diminishing the severity of symptoms in relation to the diagnostic criteria (n = 5, 21.7%), 4) communication skills (n = 3, 13.0%), 5) functional abilities (n = 8, 34.8%), 6) motor skills (n = 4, 17.4%), and 7) other measures, such as vision or therapeutic alliance (n = 5, 21.7%). Some studies also reported on the implementation of the intervention and documented satisfaction with intervention (n = 12, 52.2%) and retention rates (n = 5, 21.7%). Most of these reported high or very high parental satisfaction (n = 10, 83.3%), however only two studies reported high retention (40.0%). No information whatsoever about intervention costs was reported in any of the included studies.

Participants characteristics

Total sample sizes, including both intervention and control group participants, varied between 7 and 464, with a mean of 84.1 (SD = 123.0) participants. All interventions were designed for children aged between 0 and 12 years old, although six studies (26.1%) also included older children. Fourteen studies (60.9%) targeted children with neurodevelopmental disabilities or acquired injuries; the most frequent of these conditions were autism spectrum disorders (n = 4, 17.4%), traumatic or acquired brain injury (n = 4, 17.4%) and unilateral cerebral palsy (n = 3, 13.0%). In the remaining nine studies including children with emotional or medical conditions, the most frequent condition was disruptive behavior disorder (n = 3, 13.0%).

Key intervention characteristics

The general goal of most interventions was to improve behavioral functioning (n = 16, 69.6%). The types of interventions varied greatly, but can be classified into three categories: interventions centered around an exercise program to be implemented by the parents (n = 14, 60.9%), interventions providing some form of real-time treatment for children (n = 5, 21.7%), and interventions limited to the sharing of information (n = 4, 17.4%). Some telerehabilitation interventions targeted only the parents (n = 12, 52.2%), while others also included the children (n = 11, 47.8%). In most studies except one, parents interacted directly with a therapist. Those synchronous contacts were almost all pre-scheduled rather than happening on an as-needed basis (n = 22, 95.7%). Interactions with the therapist could either entail a fully fledged coaching intervention (n = 11, 47.8%) or only giving information and technical feedback (n = 11, 47.8%). The technology most often used was videoconferencing (n = 16, 69.6%), and most interventions used at least two technologies including the telephone, emails, web platforms or online forums (n = 19, 78.3%). In 13 studies (56.5%), families had access to an asynchronous web-based intervention (e.g., access to online modules or sessions). For the frequency of the intervention, in most cases, telerehabilitation sessions were offered at least weekly (n = 18, 82.6%). In most studies, the duration of the intervention was for a period of 8 weeks or more (n = 20, 87.0%). Most of the interventions were provided wholly or in part by psychologists (n = 16, 69.6%), while some studies included occupational therapists (n = 4, 17.4%) or physiotherapists (n = 4, 17.4%). In four studies (17.4%), the intervention was provided by more than one type of professional. Offering training to the therapists prior to the intervention was observed in fewer than half the studies (n = 10, 43.5%).

Characteristics most frequently associated with significant improvements

Globally, as presented in Table 1, most studies (n = 14, 60.9%) reported a significant improvement over the control group or overtime for more than 50% of study outcomes. Only five articles (21.7%) reported no significant improvement on any outcome measure. The mean outcome improvement across studies was 56.1% (SD = 38.5%). Figure 2 presents the percentage of improved study outcomes for the key intervention characteristics in bold above. When the general aim of the study was to improve behavioral functioning, the mean improvement per study was 68%, compared to 23% when the focus was on physical functioning. The type of intervention that seemed to lead to greater effectiveness was an intervention/exercise program to be implemented by the parent (Mean improvement per study = 67%). Interventions targeting predominantly the parent as opposed to focusing on the child (Mean = 60%), and interventions fostering a coaching approach as opposed to providing only information or feedback (Mean = 71%) also appeared as characteristics leading to a greater percentage of improvements.

With regards to the type of technology, studies that did not include videoconferencing reported a greater percentage of outcome improvement than those that did (70% vs 50%). Exploring which specific technologies were related to higher apparent effectiveness was not conclusive. The fact that more than one technology was used (e.g., email and videoconferencing, etc) did not seem to have an impact on outcome improvement. The frequency and duration of the intervention did have an influence on outcomes improvement. Having a contact with parents at least once a week (Mean = 65%) and offering the intervention for more than 8 weeks (Mean = 58%) led to greater improvements. The percentage of outcomes that improved was greater when psychologists were involved compared to studies where a physiotherapist or occupational therapist was involved. Across all studies, we found a greater percentage of outcomes improvement when authors reported having provided training to their therapists.

Discussion

This study described the variety of practices within current high-quality telerehabilitation RCT studies, and explored whether some key intervention characteristics were more frequently associated with significant outcome improvements. A diversity of practices and outcomes was surveyed within the high-quality telerehabilitation RCT studies. It might not be surprising that psychologists are the most frequently involved professionals and that behavior and parental skills are the most commonly assessed outcomes, especially for children with autism or behavioral challenges, where the effectiveness of telerehabilitation studies have already been studied more extensively [32]. When psychologists were involved, interventions seemed to be more effective, but this should be interpreted with caution, as psychologists were most often involved in studies focusing on improving behavioral functioning as opposed to physical functioning, and the former had higher outcomes improvement than the latter. In our review, telerehabilitation interventions aiming to improve physical functioning, most often for children with motor difficulties, reported improvement for some outcomes, but current evidence for improving function and motor skills via telerehabilitation appears weaker than for other outcomes. For children with motor difficulties, future telerehabilitation interventions reflecting best practices – by coaching families and fostering knowledge transfer and capacity building

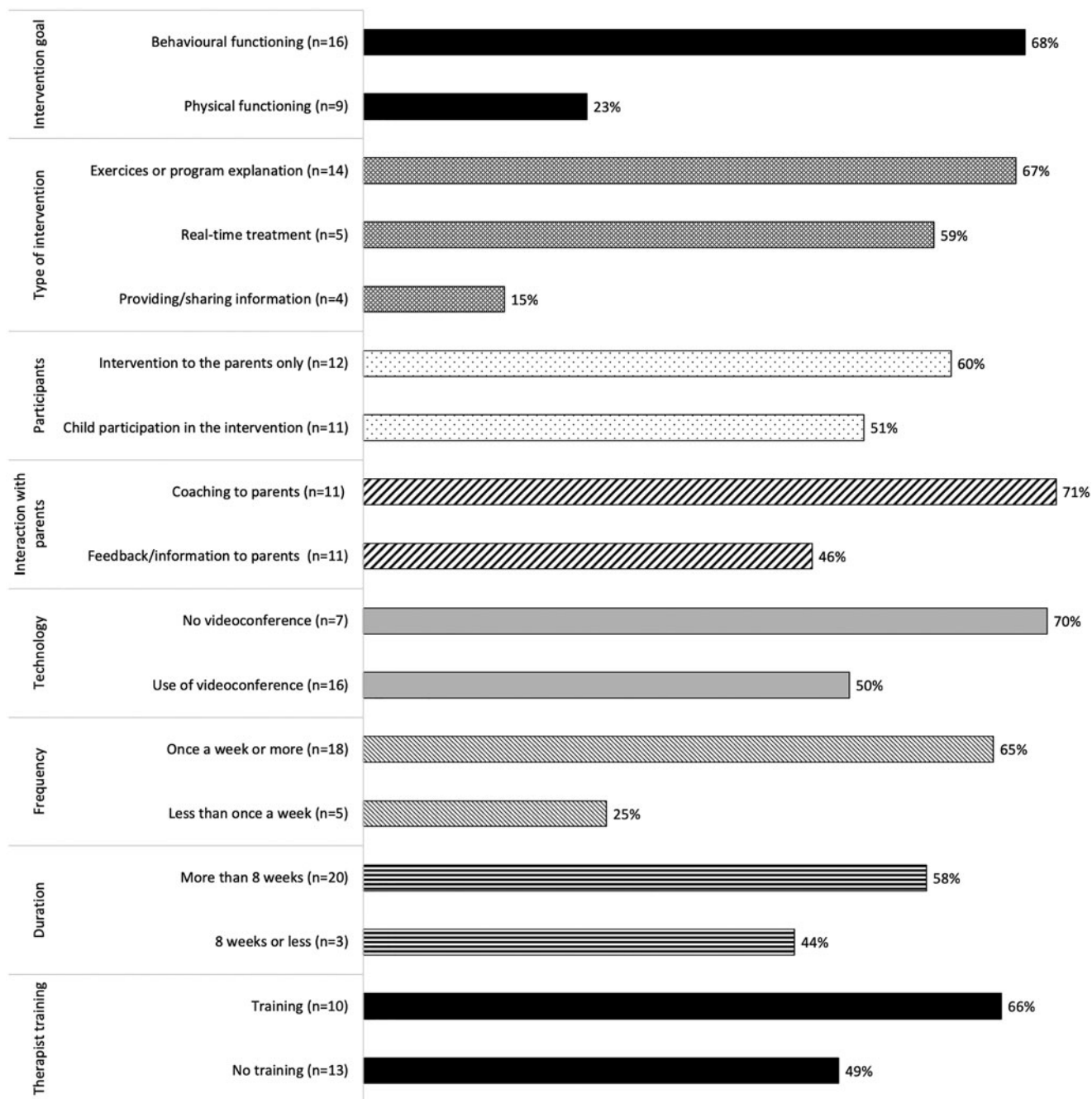


Figure 2. Percentage of outcomes that improved over time by categories of study characteristics.

[1–3] with the aim to increase parental skills to improve the child’s functioning – might prove more effective while delivered online, instead of interventions providing direct treatment to the child. This, however, remained to be explored.

Using a coaching approach was identified as being more frequently associated with outcome improvement, compared to providing information. Although providing information to families is part of best practices and family centred care [59], it might not be sufficient to engage families of children with disabilities and to build their capacity to manage disability – what diverse coaching approaches might be able to achieve, even if currently, inconsistencies in definitions and components challenge rigorous evaluations of coaching approaches [60]. Similarly, coaching approaches described by authors in the included articles varied greatly, ranging from self-reporting the use of a non-defined coaching

approach to using a previously developed coaching intervention that built on established coaching approaches. Most of the studies included in this review, and especially the ones focussing on improving physical functioning, appeared to use traditional coaching approaches, where the therapists explain to families what to do, since coaching was often used in combination with implementing an exercise programs (another intervention characteristic associated with greater outcome improvement). Other coaching approaches fostering greater families involvement, where families problem-solve to identify effective strategies to manage the child’s condition, were less frequently reported. When these approaches were used, they were part of interventions aiming at improving behavioral functioning. It would be interesting to explore in greater depth the key characteristics of the online coaching interventions that lead to greater effectiveness,

particularly as coaching seems to lead to better results than simple information sharing but the distinction between the two might be quite subtle.

Somewhat surprisingly, including children in interventions did not seem to lead to greater effectiveness for telerehabilitation. This result needs to be interpreted with caution, since telerehabilitation best practices should be aligned with the intervention goals. Most effective interventions seem to be associated with coaching approaches and parent-implemented programs. However, intervention developers should carefully consider the need to include children in the online intervention or not, especially given that real-time treatment provided to children was also associated with a significant percentage of outcomes improvement.

Another result that was somewhat surprising is the fact that the technology used did not appear to influence outcomes improvement. Videoconferencing is sometimes perceived as the gold standard, since professionals can see and interact in real time with families. Our results do not support this perception. Likewise, a multimodal approach (i.e., a combination of technologies such as videoconferencing, phone, email, online module) does not appear to influence effectiveness, even if providing multiple ways to access information is known to work best to induce behavioral change. Still, having multimodal telerehabilitation strategies including videoconferencing might accommodate different families' preferences and styles. Caution should be taken to avoid one-size-fits all interventions, and not all families might be willing or able to fully participate in telerehabilitation interventions, and Internet or system issues might limit the use of videoconferencing. Again, the choice of technology should probably be guided by the intervention's goals, but also by the families' preferences. Caution with telehealth interventions has even been suggested, since they could increase social inequities by failing to reach vulnerable families [61].

The results with regards to the frequency and duration associated with greater effectiveness (i.e., at least once a week, for more than 8 weeks) are, in counterpart, not surprising. Intensity is perceived to be associated with both engagement in the rehabilitation process and with outcomes [2]. Results suggested that, for online interventions, having preset scheduled sessions as opposed to adopting a needs-based approach (i.e., families can contact the therapist when they want) might be more effective and might help families to better identify and address their needs. That being said, many studies did include an intervention in which the therapist was able to adjust their schedule depending on the family's needs, some with a baseline frequency of sessions, others without.

It is somewhat surprising that interventions provided by multiple disciplines did not lead to greater outcome achievements. This might be explained by the fact most interventions targeted very specific goals, and that teamwork may not have been required to achieve these outcomes. Yet, for interventions addressing broader or multiple goals, a multidisciplinary team might be required. This warrants the future study. Likewise, an evidence base in physical telerehabilitation (occupational therapy and physiotherapy) is only just emerging and current studies report lower outcome improvement rates. This might be explained by the fact that changes in motor outcomes might be harder to achieve online – or by the lack of suitable outcome measures to detect changes in children with motor difficulties involved in telerehabilitation interventions [62].

Finally, many studies reported high adherence rates and satisfaction with telerehabilitation, which is coherent with other

qualitative studies that explored parental satisfaction [11]. It is important to note that no study reported a deterioration over time or better results for face-to-face interventions compared to telerehabilitation. This is a very interesting finding, given that telerehabilitation interventions are generally not perceived by therapists as being as effective as traditional interventions, and are often considered convenient stop-gaps in the absence of face-to-face interventions [10,11]. Our results demonstrate that telerehabilitation might be as effective as face-to-face interventions. We could not however confirm or discredit the perception that telerehabilitation is less costly than face-to-face interventions [10–14], since none of the included studies reported a comparative cost assessment. This might be due to our inclusion criteria and the fact that, when conducted as part of trials, economic analyses are often reported in separate articles. In fact, a recent study conducted alongside one of the RCTs included in the present systematic review [19] evaluated the cost of the online intervention and concluded that costs were minimal compared to its incremental effectiveness [63]. The costs were however not compared between the telerehabilitation group and the other group. Future telerehabilitation studies should explore cost-effectiveness since, overall, our results demonstrate that telerehabilitation interventions might be effective in a variety of contexts.

The principal limitations of this study were related to the challenges of synthesizing the diversity of populations, intervention characteristics and outcome measures in the included RCT studies, while only relying on the information provided by the authors. In many studies, information was lacking to clearly distinguish the differences between the intervention and control groups, and some approaches were described very vaguely (e.g., the use of coaching was often reported, but rarely described in details particularly in contrast with only providing information). This limits the possibility of results generalization, especially with regards to establishing the effectiveness of specific intervention characteristics. The approach we used to synthesize this diversity of practices is however also a strength of the study, since it allowed us to explore common intervention characteristics of effective telerehabilitation interventions, across rehabilitation and disability fields. As more telerehabilitation studies are published and the description of interventions increases, we might be able to explore the effectiveness for more specific telerehabilitation intervention characteristics and use stronger methodology (e.g., correlation between effectiveness and characteristics, or meta-analysis). An interpretation bias might also have occurred during data extraction. This bias was however addressed by the rigorous data extraction and analysis validation process described in the Methods section. This validation process is a clear strength of this study, along with the efforts made to present meaningful information for practice and for future research. The choice of including only RCTs might also be a limit, since other designs could provide some relevant information. However, this criteria and the application of the PEDro quality assessment scale provided clear cut-offs and promoted the inclusion of high-quality studies. PEDro scores should however be interpreted with caution, as the scale might prejudice some research questions and does not inform the user about the clinical significance and applicability of study results.

Conclusion

Telerehabilitation appears to be a promising approach for the provision of rehabilitation services to a pediatric population. Specifically, telerehabilitation might be particularly effective when

a coaching approach is used. Further research is required to explore the specific contexts, populations, and interventions that render telerehabilitation most effective and most cost-effective. To advance the field, we encourage authors of future trials to thoroughly detail their intervention, in particular with regards to the type of intervention, the interaction style with parents and the training provided to therapists. We do not envision telerehabilitation replacing in-person services, but given preliminary data on its effectiveness and its societal acceptability, we would recommend that rehabilitation professionals consider the use of innovative remote interventions as possible service delivery options.

Declaration of interests

The authors report no declaration of interest.

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ORCID

Chantal Camden  <http://orcid.org/0000-0002-5503-3403>

Mélanie Couture  <http://orcid.org/0000-0002-4919-573X>

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