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RESEARCH ARTICLE

Cost-effectiveness analysis of telerehabilitation for people who have sustained a stroke with return home without intensive rehabilitation: A systematic review

[Analyse coût-efficacité de la téléréadaptation pour les personnes ayant subi un accident vasculaire cérébral avec un retour à la maison sans réadaptation intensive : une revue systématique]

Lucien P. Coulibaly[1,2]
Christian M. Rochefort[1,3,4]
Dahlia Kairy[5,6,7]
Michel Tousignant[1,2]

[1] Université de Sherbrooke
[2] Centre de recherche sur le vieillissement
[3] Centre de recherche du CHUS
[4] Centre de recherche Charles-Le Moyne – Saguenay-Lac-Saint-Jean sur les innovations en santé
[5] Université de Montréal
[6] Centre de recherche interdisciplinaire en réadaptation du Montréal métropolitain
[7] Institut universitaire sur la réadaptation en déficience physique de Montréal

Correspondence:
Lucien P. Coulibaly, Centre de recherche sur le vieillissement, Institut universitaire de gériatrie de Sherbrooke, 1036 rue Belvédère Sud, Sherbrooke (QC), J1H 4C4, Canada

Email:
lucien.coulibaly@usherbrooke.ca

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Abstract: The number of people with stroke who do not require intensive functional rehabilitation and are directly discharged home after a hospitalization is on the rise, and represents a major economic burden for the healthcare system, patients, and their families. This systematic review aims at providing knowledge by summarizing the evidence on: a) studies comparing the cost-effectiveness of telerehabilitation (TR) versus face-to-face rehabilitation, and b) main clinical effectiveness variables used to calculate cost-effectiveness. Eight databases were consulted from January 1st 2005 to November 30th 2017 by an independent researcher and the selected articles were independently validated by two other researchers. The methodological quality and the level of evidence of the 15 articles included (out of a total of 166) was evaluated using validated instruments. Nine studies focused on the clinical effectiveness of TR, four on its economic effectiveness, and two were meta-analyses of the evidence. We found no study measuring the cost effectiveness of TR in stroke patients, but we did find evidence that this approach is less expensive to implement than face-to-face rehabilitation. Regarding the clinical effectiveness of TR, we found that for stroke patients, it is at least comparable to face-to-face rehabilitation for a number of outcomes (e.g., balance, mobility, motor recovery, pain reduction, verbal communication skills), and found no evidence suggesting its inferiority. TR thus appears to be an economically viable option in supporting stroke victims, but further studies are needed to measure its cost-effectiveness.

Keywords: telerehabilitation, stroke, cost analysis, economic evaluation, cost-effectiveness analysis, cost-benefit analysis.

Résumé : Le nombre de personnes ayant subi un accident vasculaire cérébral qui ne nécessite pas de réadaptation fonctionnelle intensive, et qui sont renvoyées à domicile après une hospitalisation, augmente et représente un fardeau économique pour le système de santé, les patients et leurs familles. Cette revue systématique vise à fournir des connaissances résumant les données probantes sur les : a) études comparant le rapport coût-efficacité (RCE) de la téléréadaptation (TR) versus la réadaptation en face à face, et b) principales variables d'efficacité clinique utilisées pour calculer le rapport coût-efficacité. Huit bases de données ont été consultées du 1er janvier 2005 au 30 novembre 2017 par un chercheur indépendant, et les articles sélectionnés ont été validés indépendamment par deux autres chercheurs. La qualité méthodologique des 15 articles inclus et leur niveau de preuve ont été évalués à l'aide d'instruments validés. Neuf études portaient sur l'efficacité clinique de la TR, quatre sur son efficacité économique, et deux méta-analyses ont été incluses. Il n'existait aucune étude ayant calculé le RCE de la TR chez les victimes d'AVC. Toutefois, il a été prouvé que la TR est moins coûteuse, et que son efficacité clinique était au moins égale à celle de la réadaptation en face à face pour certains résultats (par ex. : équilibre, mobilité, motricité, douleur, communication verbale). La TR semble être une option économiquement viable pour soutenir les victimes d'AVC, mais d'autres études sont nécessaires afin de déterminer son RCE.

Mots clés : téléréadaptation, accident vasculaire cérébral, évaluation économique, analyse de coûts, analyse coûts-efficacité, analyse coûts-bénéfices.

Introduction

In Canada, a person suffers a stroke every nine minutes, making it one of the main causes of disability [1]. Strokes represent a major economic burden for patients, their families, and the healthcare system [2] and are therefore a major public health problem. For example, costs associated with stroke care are estimated at $3.6 billion per year in Canada in terms of medical services provided, hospital costs, wages lost, and reduced productivity [3,4].

Every year, nearly 62,000 (1.7‰) people in Canada suffer a stroke, but only about 6,500 stroke survivors have access to rehabilitation services after their hospitalization [5]. Although we don't know the exact number of persons who can benefit from post-discharge rehabilitation at home, it is however well-documented that a vast majority of candidates for home rehabilitation receive insufficient services due to cost constraints or various barriers that limit the access to rehabilitation services (e.g., reduced mobility, geographical distance) [2]. In this context, there is a strong need for an innovative approach for the rehabilitation of stroke patients that simultaneously promotes accessibility to rehabilitation services, while improving quality of life in stroke patients, and being affordable.

In recent years, telerehabilitation (TR), defined as the use of information and communications technologies (ICT) to provide rehabilitation and long-term support to people with disabilities, has been proposed as a potentially cost-effective strategy to address these requirements [6]. This approach provided through videoconferencing technology makes it possible to establish a real-time remote communication between a patient and a healthcare professional [7]. Of note, TR is emerging as a potential alternative to face-to-face rehabilitation to increase access to rehabilitation services. Several studies have also suggested that TR can lead, in addition to health gains, to a wide range of benefits, such as cost-reduction, and the possibility to overcome the shortage of professionals [3,8].

Since the end of the 1990s and early 2000s, several studies have been conducted to evaluate the clinical and economic effectiveness of TR in general, as well as in stroke patients, in particular. However, two systematic reviews and meta-analyses in this literature have provided contradictory conclusions concerning the clinical effectiveness of TR. These were mainly attributed to the great variability of TR approaches, small sample sizes, large variability of clinical results investigated, and heterogeneity in patients' follow-up times [9,10]. Although TR represents a potential solution to increase the accessibility of rehabilitation services, the evidence regarding the cost-effectiveness of this solution remains scattered [9, 11]. We aim to contribute to this field of research by summarizing existing evidence from studies comparing the cost-effectiveness of TR to face-to-face rehabilitation. By this work, our ultimate goal is to support decision-making in the field of TR in stroke.

Objectives

The main objective of this systematic review is to provide knowledge by summarizing the evidence from studies comparing the cost-effectiveness of TR versus face-to-face rehabilitation, for people who have sustained a stroke and are directly discharged home without an intensive rehabilitation program. A second objective is to identify the clinical effectiveness variables that could be used to calculate cost-effectiveness.

Method

Design

We conducted a systematic review of the studies examining the cost-effectiveness of TR compared to face-to-face rehabilitation, with a special focus on the main clinical effectiveness variables used. Our methodological approach was based on the Preferred Reporting Items for Systematic Reviews and Meta-Analyses

(PRISMA) guidelines [12]. We provide a qualitative synthesis of the available evidence. A meta-analysis was not attempted, given the methodological heterogeneity of the retrieved studies.

Search strategy

Studies were identified in eight distinct databases: 1) MEDLINE, 2) Abstract in Social Gerontology, 3) AgeLINE, 4) CINAHL, 5) PsycINFO, 6) PsycARTICLE, 7) Cochrane Library and, 8) Canadian Agency for Drugs and Technologies of the Health (CADTH). Keywords used alone or in combination with Boolean operators "OR" and "AND" are outlined in Table 1. The reference period was from January 1st, 2005 to November 30th, 2017, and our search of the literature was last updated on August 31th, 2018. This reference period was selected to get the most recent evidence and to avoid the repetition of older studies already included in recent reviews or meta-analyses. To make the search more complete, and identify the maximum number of studies related to our research question, the reference list of the retrieved studies was also searched. In addition, the aforementioned databases were also searched for any prior or subsequent publications by the authors of the retrieved studies.

Inclusion and exclusion criteria

We included studies that: 1) were published in English or French; 2) reported an economic assessment of telerehabilitation (i.e., teleconsultation, telemonitoring and teleprocessing); 3) used any of the following approaches: cost-minimization analysis (CMA), cost-effectiveness analysis (CEA), cost-utility analysis (CUA), cost-benefit analysis (CBA) or cost-effectiveness analysis (CCA); 4) pertained to people who have sustained a stroke and are directly discharged home without intensive rehabilitation; 5) pertained to patients having experienced a stroke or any pathology requiring rehabilitation through TR; 6) were based on any of the following research designs: systematic review, meta-analysis; randomized or nonrandomized controlled trials, cohort studies); 7) included at least one comparator. Studies not meeting the aforementioned inclusion criteria were therefore excluded.

Study selection

The initial selection of articles was done by a researcher (LPC) based on reading titles and abstracts of potentially eligible studies identified in the database searches. The articles were read, and selected by applying the selection criteria. The accuracy of the study selection process was assessed by a second researcher (CR). Disagreements were resolved through discussion among members of the research team. Once the list of articles to include in the review was agreed upon, data was extracted into an Excel file.

Table 1: Search strategy

Database	Keywords
PubMed	("telerehabilitation"[MeSH]) AND ("stroke"[Mesh])
	("telerehabilitation"[Mesh]) AND ("cost-benefit analysis"[Mesh])
MEDLINE via EBSCO	telerehabilitation AND stroke or cerebrovascular accident AND cost-effectiveness
	telerehabilitation AND stroke or cerebrovascular accident AND cost benefit analysis
	telerehabilitation AND stroke or cerebrovascular accident
CADTH	telerehabilitation AND stroke
	telerehabilitation AND (stroke OR cost-effectiveness)
Cochrane Library	telerehabilitation AND stroke
	telerehabilitation AND (stroke OR cost-effectiveness)

Data extraction

The first author (LPC) extracted the following information from each of the selected studies: 1) authors/year/country 2) study design, 3) study population and sample size, 4) main intervention, and its comparator ; 5) confounding variables; 6) dependent variables; 7) perspective used for economic assessment, 8) follow-up duration, 9) main results and conclusions. For systematic reviews, we also extracted the number of studies included. The second author (CR) checked the extracted data for accuracy, and disagreements were resolved through discussion.

Data analyses

First, we provide descriptive statistics on the characteristics of the reviewed studies. Then, we provide a qualitative synthesis of the available evidence. We used $p < 0.05$ as the threshold to determine the statistical significance of the findings.

Evaluation of included studies quality

The methodological quality and the level of evidence provided by each study was assessed using several tools. Economic studies were assessed using ten items from Drummond's checklist [11]. Meta-analyses were assessed using the AMSTAR checklist (A MeaSurement Tool to Assess systematic Reviews) [13]. Finally, primary studies were evaluated using the randomized controlled trials and observational studies checklist developed by Downs and Black [14].

Results

Overall, 166 articles were retrieved from our search strategy. Out of the 37 articles selected for a more detailed evaluation, only nine were included in this systematic review (Figure 1). Subsequently, six additional articles were identified from searching the bibliographies of the identified studies, bringing the final number of studies included in this systematic review to 15 (Figure 1).

Study characteristics

Of the 15 studies, two (13.3%) were conducted in Holland, three (20%) in Canada, three (20%) in Australia, two (13.3%) in the USA, and one in each of the following countries: South Korea, Germany, Spain, Italy and Belgium. Out of the 15 articles included, nine (60%) were primary studies reporting on the clinical effectiveness variables of TR (Table 2), four (26.7%) were primary studies of the economic effectiveness of TR (Table 3), and two (13.3%) were meta-analyses (Table 5).

As for the nine studies (Table 2) examining the clinical effectiveness of TR, the most frequent clinical effectiveness variables that were reported were overall limb function, mobility, balance, quality of life, pain, and verbal communication. These studies were based on samples ranging from seven to 205 patients (Table 3) with a mean of 60 (SD: 57.7). The most typical study designs were: randomized controlled trial (RCT) (n=6, 66.7%), observational study (n=2, 22.3%) and Pre/Post intervention study (n=1, 11%). Interventions were applied with the same intensity (i.e., duration of session, number of sessions per week) in the TR group as in the control group in five studies (55.6%). However, TR intensity was higher in one study (11.1%), and lower in another one (11.1%). Out of the two observational studies, one (11.1%) did not provide information on treatment intensity, and the other (11.1%) gave only the number of hours of treatment over the entire study. The follow-up duration ranged between one to six months, and only three studies (33.3%) were multicentre investigations.

Out of the four studies (Table 3) reporting on the economic effectiveness of TR, two studies (50%) used the perspective of the healthcare system, one (25%) used society's perspective and that of the patient (n = 1), while the last study (n=1, 25%) did not specify the analysis perspective that was used. The sample size of these studies ranged from 30 to 197 patients with a mean of 134 (SD: 63.5). These studies were based on RCT designs (n=4, 100%) and two (n=2, 50%) were multicentre investigations. Concerning the type of economic analysis design, for the most part, these studies were based on cost-minimization analysis (n=3, 75%) and one (n=1, 25%) was a cost-utility analysis. Interventions were applied with the same level of intensity (duration of session, number of sessions per week) across groups in all studies. The follow-up duration was between seven weeks to 12 months. Direct and indirect costs were the only kind of costs included in these studies (Table 4) and authors used different time horizons (follow-up duration) in their economic analysis. Only two studies (50%) were multicentre.

Finally, regarding the two meta-analyses (Table 5), the first summarized the findings of a total of 12 distinct studies (n=1,236 patients) with a special focus on motor function disorders, whereas the second summarized the findings of a total of 13 distinct studies focusing on 1,697 patients with musculoskeletal problems.

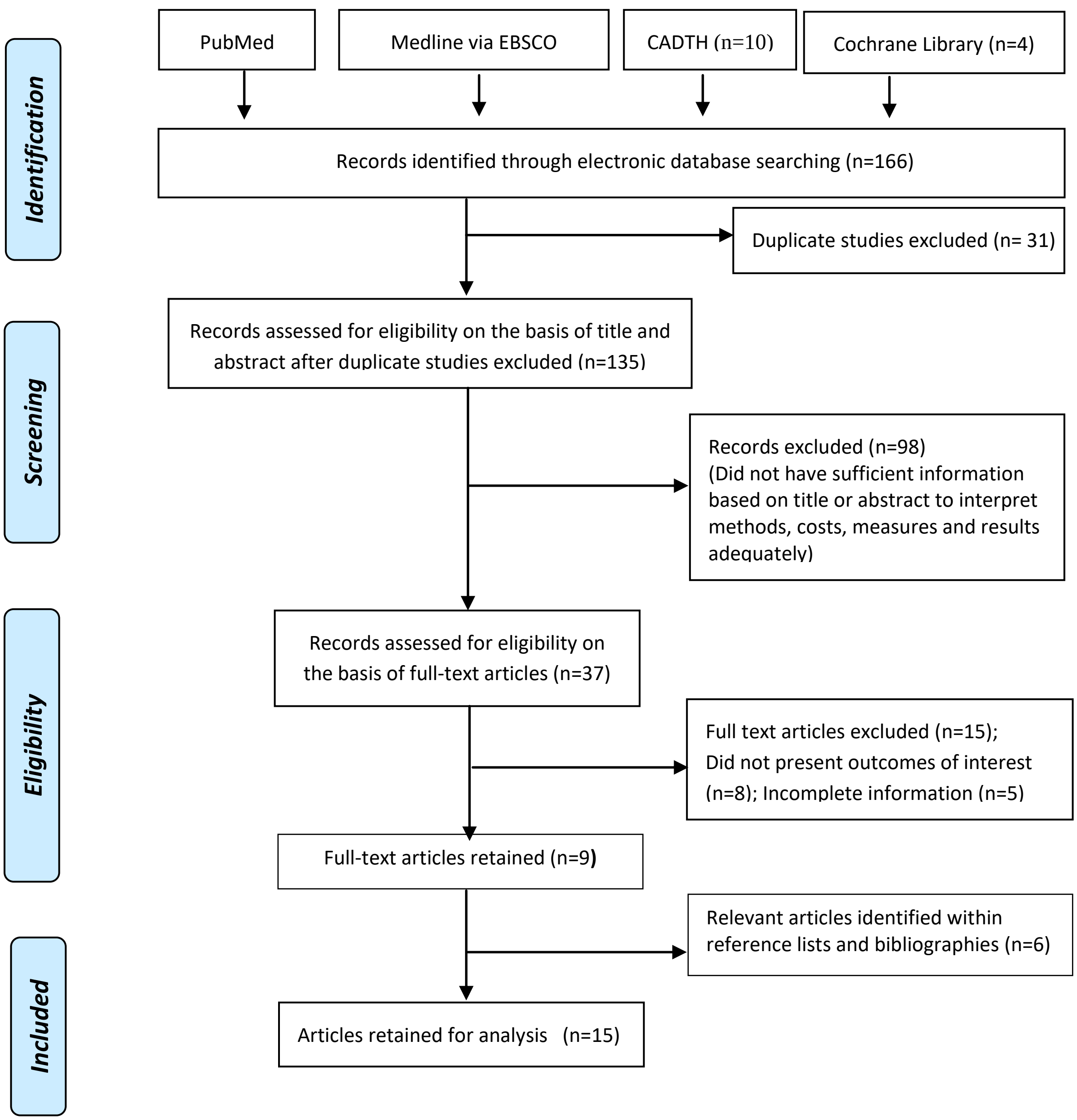

Figure 1. PRISMA flow diagram (November 30th 2017)

Table 2: Characteristics of primary studies included

Authors Year Country	Interventions *versus* Comparator	Study design Number of patients (interv/ctrl)	Intervention duration	Session duration and number of sessions (interv/ctrl)	Population	Practitioner	Variables (measured)	Results	Score Downs & Black
Huijgen et al. (2008) [22] Holland	TR *versus* face to face home' rehabilitation	Multicenter RCT n=81 (55/26)	1 month	30 min /45 min 5 sessions per week/ 3 sessions per week	Stroke patients	Physical therapist	* Arm functioning (ARAT) * Manual dexterity (NHPT)	Improvement is statistically insignificant in the use of hands and arms between the two groups	19/32
Kosterink et al. (2010) [15] Holland	TR *versus* face to face rehabilitation	Multicenter RCT (inter-countries) n=71 (36/35)	4 weeks	63 min /46 min	Patients with musculoskeletal disorders in the neck and shoulder	Physical therapist Osteopath Chiropractor Occupational therapist Stress manager	* Pain intensity (VAS) *Level of disability (PDI)	Teletreatment appears to be more effective; No statistically significant differences observed between the two groups	20/32
Tousignant et al. (2011) [18] Canada	TR *versus* face to face home' rehabilitation	RCT n=41 (21/20)	8 weeks	60 min in both groups 2 sessions per week in both groups	Patients who had total knee arthroplasty	Physical therapist	* Handicap (Berg Balance Scale Score; 30-s chair-stand test) * Function (WOMAC; TUG; Tinetti test); * Quality of life (SF-36)	Home TR is at least as effective as usual care	21/32
Russell et al. (2011) [17] Australia	TR *versus* face to face rehabilitation	RCT n=65 (31/34)	6 weeks	45 min in both groups 1 session per week in both groups	Patients following total knee arthroplasty	Physical therapist	* Lower limb function (WOMAC); * Specific function (PSFS); * Quality of life (SQLU) * Functional mobility (TUG) * Pain intensity (VAS)	Home TR is at least as effective as usual care	21/32
Chumbler et al. (2012) [23] USA	TR *versus* face to face home' rehabilitation	Multicenter RCT n=48 (25/23)	6 months	Three 1-hour televisits Participants' daily use of an in-home messaging device 5 telephone intervention calls between the teletherapist and the participant	Stroke patients	Teletherapist	* Functional Independence (FONEFIM) * Functional difficulty (LLFDI)	TR significantly improved physical function and functional capacity	22/32

Table 2 (continued): Characteristics of primary studies included

Authors Year Country	Interventions *versus* Comparator	Study design Number of patients (interv/ctrl)	Intervention duration	Session duration and number of sessions (interv/ctrl)	Population	Practitioner	Variables (measured)	Results	Score Downs & Black
Russell et al. (2013) [16] Australia	Data evaluation by TR *versus* face to face rehabilitation' data evaluation	observational study n=12	n.d	n.d	Patients with Parkinson's disease	Final-year physiotherapy and occupational therapy students	* Functional mobility (TUG) * Balance (Berg Balance Scale Score)	Using TR technologies can provide reliable data evaluation results	20/32
Langan et al. (2013) [24] USA	TR *versus* face to face home' rehabilitation	Pre/post intervention study n=7	6 weeks	60 min/session 5 sessions/week	Stroke patients	n.d	* Upper limb function (ULTrA)	TR bring a significant improvement in motor functions of upper limbs TR improves cognitive function	19/32
Moffet et al. (2015) [19] Canada	TR *versus* face to face home' rehabilitation	RCT n=205 (104/101)	2 months	45 to 60 min in both groups 16 sessions in both groups	Patients following total knee arthroplasty	Physical therapist	* Lower limb function (WOMAC) * Functions, quality of life(KOOS)	Non-inferiority of in-home TR	22/32
Choi et al. (2016) [21] South Korea	TR *versus* face to face rehabilitation	Observational study n=8	4 weeks	30.28 – 28.72 h	Patients with chronic post-stroke aphasia	Experienced physiatrist and speech–language pathologist	* Language Functions (K-WAB)	TR has improved verbal communication skills	19/32

ARAT: Action Research Arm Test; NHPT: Nine Hole Peg Test; WOMAC: Western Ontario and McMaster Universities Osteoarthritis Index; KOOS: Knee injury and Osteoarthritis Outcome Score; K-WAB: Korean version of the Western Aphasia Battery; ULTrA: Upper Limb Training and Assessment; TUG: Timed "Up and Go; FONEFIM: Functional Independence Measure; LLFDI: Late-Life Function and Disability Instrument; PSFS: Patient-Specific Functional Scale; SQLU: Spitzer Quality-of-Life Uniscale; VAS: Visual Analog Scale; PDI: Pain Disability Index; ctrl: control; interv: intervention; min: minute

Table 3: Characteristics of economic primary studies included

Authors Year Country	Interventions *versus* Comparator	Perspective	Study design Number of patients (interv/ctrl)	Practitioner	Follow-up duration	Session duration and number of session (interv/ctrl)	Economic analysis design	Results	Study quality according to Drummond checklist	Score Downs & Black
Körtke et al. (2006) [25] Germany	TR *versus* face to face clinical' rehabilitation	Healthcare system	Cardiac patients non-randomized control trial n=170 (100/70)	n.d	6 to 12 months	15–30 min daily in both groups	Cost-minimization analysis	Total cost of rehabilitation was 58% lower in intervention group compared to control group	4/10	19/32
Tousignant et al. (2015) [26] Canada	TR *versus* face to face home' rehabilitation	Healthcare system	Patients who had total knee arthroplasty Multicenter RCT n=197 (97/100)	Physical therapist	8 weeks	2 x 45-min sessions per week in both groups 16 sessions in both groups	Cost-minimization analysis	For each participant's total intervention, TR saves the health care system 18% of the costs incurred for conventional rehabilitation following total knee arthroplasty	7/10	23/32
Lloréns et al. (2015) [8] Spain	TR *versus* face to face clinical' rehabilitation	n.d	Stroke patients RCT n=30 (15/15)	Physical therapist	7 weeks	3 x 45-min training session per week in both groups 20 sessions in both groups	Cost-minimization analysis	TR allows to save time ; The patient transport services were private. The travel expenses represented 88% of the total cost of the in-clinic intervention	4/10	22/32
Frederix et al. (2016) [27] Belgium	TR + face to face rehabilitation *versus* face to face rehabilitation	Society and patient	Coronary disease Multicenter RCT n=140 (70/70)	n.d	24 weeks	2 x 45 to 60 min sessions per week in both groups 45 sessions in both groups	Cost-utility analysis	For an additional year of quality of life gained (QALY), a savings of 21,707 € is made per patient in the intervention group compared to a patient in the control group	7/10	20/32

Table 4: Kind of costs included in economic primary studies

Authors	Telerehabilitation group		Control group	
	Directs costs	Indirect costs	Directs costs	Indirect costs
Körtke et al. (2006) [25]	• Clinician salary	• Telemedical connectivity • Leasing and delivery of the bicycle ergometer • Transport of ergometer • Consultation, education	• Clinician salary	• Consultation, education
Tousignant et al. (2015) [26]	• Clinician salary • TR equipment procurement	• Internet • Installation/uninstallation of technology • Technical problems • Clinical and TR equipment amortization • Time of indirect treatment	• Clinician salary	• Travel to participant's home • Time of indirect treatment
Lloréns et al. (2015) [8]	• Salary for physical therapists • Instrumentation (Laptop; Kinect)	• Internet access	• Salary for physical therapists	• Transportation (round-trips)
Frederix et al. (2016) [27]	• Intervention (TR)	• Healthcare	• Intervention (TR+ face to face rehabilitation)	• Healthcare

Table 5: Characteristics of meta-analyses included

Authors Years Country	Reference period	Intervention *versus* comparator	Patients characteristics	Number of studies	Number of patients	Conclusion	Score AMSTAR
Agostini et al. (2015) [9] Italy	1946 to 2014	TR *Versus* face to face rehabilitation	Patients with motor function disorders	12	1,236	Not enough evidence on the efficacy of TR in motor function recovery.	8/11
Cottrel et al. (2017) [10] Australia	Until november 2015	TR *versus* face to face rehabilitation	Patients with musculoskeletal problems	13	1,697	There is evidence to conclude that post-operative TR is more effective or at least equivalent to face-to-face rehabilitation.	9/11

Methodological quality assessment

The quality of the two meta-analyses was deemed very good with AMSTAR scores of 8/11 and 9/11. As for the economic studies, their scores on Drummond's et al. (2015) evaluation grid ranged between 4/10 and 7/10. The quality of the primary studies was judged satisfactory with Downs and Black scores ranging between 19/32 and 23/32. Overall, all studies had low scores for blinding; the evaluators and the investigators being blinded to group assignment for the entire duration of the study in only three studies (33%). Last, concerning economic evaluation studies, patients were evaluated by a blind assessor before and after the intervention in three studies (75%). However, blinding of participants and clinicians was not performed in any study. Of all the studies included, there were only four studies for which the authors reported their conflicts of interest.

Findings related to the clinical effectiveness variables of TR

The characteristics of the studies examining the clinical effectiveness of TR are listed in Table 2. While these studies examined a variety of clinical outcomes, the most recurrent ones pertained to the rehabilitation of motor functions: functional mobility, limb function, and balance.

Generally

We found emerging evidence that the clinical effectiveness of TR is at least equivalent to that of face-to-face rehabilitation for a number of clinical outcomes. For example, according to the results of a RCT conducted by Kosterink et al. in the Netherlands, teletreatment appeared to be at least as effective as conventional care for treating pain in patients with neck and shoulder musculoskeletal disorders [15]. In fact, although the results of this multi-center study including 71 patients favored teletreatment, no statistically significant differences between the two groups were observed. In addition, the authors noted a high drop-out rate that probably affected the results.

Preliminary results from a pilot study by Russell et al [16] on remote physical assessments of 12 people with Parkinson's disease also indicated that the use of TR can provide reliable results (i.e. obtaining exactly the same results under the same conditions). However, the authors recommended that a larger scale study be conducted to confirm these findings, given the small sample of their pilot study (n=12 participants) [16]. Several studies have also examined the clinical effectiveness of TR in the case of knee arthroplasty. First, Russell et al. in Australia reported the results of a single-blind non-inferiority RCT, including 65 participants over six weeks [17]. A significant difference in some variables, such as lower limb function and functional mobility, was achieved in favor of the TR intervention. Despite these results, the authors concluded that TR at home is at least as effective as face-to-face rehabilitation in the same setting. Similarly, Tousignant et al. conducted a pilot study nested in one RCT including 48 patients followed at home for eight weeks after hospital discharge [18]. Disability and function were significantly improved for patients in both groups. However, for functional activities, face-to-face rehabilitation showed more improvement two months after the end of therapy. Hence, this study suggested that home TR was at least as effective as face-to-face rehabilitation to improve disability and physical function. These conclusions were confirmed by the same research team more recently in a larger study. They reported the results of a non-inferiority RCT on the clinical efficacy of TR versus face-to-face rehabilitation at home after knee replacement surgery in 205 patients [19]. The results demonstrated the non-inferiority of TR and the authors recommended its use as an effective alternative to face-to-face rehabilitation in the home.

In stroke patients

Theodoros conducted a study reviewing the current developments in TR applications. His study pointed out that TR improved verbal communication skills in adults following stroke or traumatic brain injury [20]. This finding was confirmed more recently by Choi et al. who conducted an observational study including eight patients with aphasia following a stroke [21].

A multicenter RCT involving 81 participants over a one-month period was conducted in Holland by Huijgen et al. [22]. This study compared the improvements generated by a TR program versus a face-to-face rehabilitation program at home on hand and arm functions in patients with stroke. There were no significant differences between the two groups on these functions. However, the study showed an improvement in the use of hands and arms in both groups. Also, the difference between the two approaches was not statistically significant.

For their part, Chumbler et al. conducted a six-month RCT in the United States. The purpose of their study was to determine the effects of a TR intervention, compared to face-to-face rehabilitation at home, on the physical function and functional capacity of 48 veterans with stroke [23]. The primary outcomes improved in the TR group and decreased for the control group, but the differences were not statistically significant. However the results indicated that TR significantly improved physical function and functional ability of participants compared to face-to-face rehabilitation at home. These improvements were incremental over time, and persisted until the third month; after which they remained constant until the sixth month. Therefore, the authors recommended TR as a complementary approach to face-to-face rehabilitation.

In addition, a pilot study conducted by Langan et al. in the United States on a sample of adults with chronic stroke showed that TR could lead to significant improvements in upper limb motor function [24]. Supporting these positive results observed in favor of TR, the authors noted that this intervention can also lead to an improvement in cognitive function.

In summary, there is emerging evidence that TR is at least equivalent to face-to-face rehabilitation to improve certain clinical outcomes, such as physical function and functional capacity. There is also evidence of the superiority of TR regarding balance and mobility. As for functional mobility, limb function, and balance, the results of existing studies are mixed. Further high-quality studies are required to better document the clinical effectiveness of TR on these outcomes.

Findings relating to the economic effectiveness of TR

The characteristics of the studies examining the economic effectiveness of TR are listed in Table 3.

Generally

Several studies have shown that the total costs associated with TR are lower than those of face-to-face rehabilitation [25-27]. For example, Körtke et al. conducted a pilot study with the objective of comparing the costs of TR to those of face-to-face rehabilitation in cardiac surgery clinic patients [25]. This study included 170 non-randomized patients and took place over a period of three months for the intervention group (n=100) and three weeks for the control group (n=70). The results showed that TR could result in a 58% reduction in total costs compared to the control group.

In addition, a cost-minimization multi-center study by Tousignant et al. aimed at comparing the costs between TR and face-to-face rehabilitation at home for patients having undergone knee replacement surgery [26]. This RCT included 197 patients and was conducted over an eight-week period. The authors found that, compared to face-to-face rehabilitation, TR allows the healthcare system to save 18% of the total costs incurred for patient rehabilitation [26]. However, they noted that under RCT conditions, a cost differential in favor of TR was observed only when the patient resided more than 30 km away from the rehabilitation center.

For their part, Frederix et al. conducted a multicenter RCT in Belgium over a 24-week period [27]. The main objective was to evaluate, using CEA, the cost-effectiveness of a combined TR-face-to-face rehabilitation intervention (n=70) versus face-to-face rehabilitation alone (n=70) in the cardiac rehabilitation of patients with coronary heart diseases. The results indicated that for an additional year of quality of life (QALY) gained, a savings of 21,707 € was achieved per patient in the intervention group, compared to a patient in the control group.

In stroke patients

We found only one study comparing cost differences between one group of stroke patients subjected to TR (n=15) versus patients receiving face-to-face rehabilitation in a clinic (n=15). This RCT was conducted in Spain by Lloréns et al. and included a total of 30 outpatients who had residual hemiparesis following a stroke [8]. The results indicated that TR saved time because the duration of the physiotherapist's intervention in the control group was significantly longer than in the experimental group (8.34 ± 0.36 hours vs. 1.63 ± 0.78 hours). In addition, in this study, the costs of transportation to the rehabilitation center were entirely supported by the patients, and these could represent up to 88% of the total costs of the face-to-face intervention.

Meta-analyses

Two meta-analyses, including 1,236 patients with motor function disorders and 1,697 patients with musculoskeletal problems, were performed (Table 5) [9,10]. These meta-analyses concluded that balance, mobility, and walking ability were the most common problems for patients who needed physical rehabilitation. However, their conclusions differed on the effectiveness of TR.

Indeed, Agostini et al. included 12 randomized trials that compared the clinical effectiveness of TR with that of face-to-face rehabilitation for the motor recovery of different types of patients (e.g., neurological diseases, knee replacement, heart problems) [9]. Their overall conclusion was that there currently is a lack of evidence to support the clinical effectiveness of TR over face-to-face rehabilitation. Interestingly, a sub-group meta-analysis performed by these authors demonstrated a large statistical heterogeneity (I2=84%), and they suggested that further high-quality studies are required.

In contrast, Cottrell el al. concluded that postoperative TR seems to be more effective or at least equivalent to face-to-face rehabilitation to improve on physical function [10]. This meta-analysis included 13 randomized studies of TR in patients with various musculoskeletal conditions. The duration of the intervention ranged from four to 52 weeks, depending on the musculoskeletal problem being treated. While aggregated results showed substantial statistical heterogeneity across studies, a stratified analysis demonstrated a moderate heterogeneity (I2=61%) for physical function and disabilities, and large statistical heterogeneity (I2=96%) for the pain subgroup.

Discussion

The objectives of this systematic review were to synthesize the evidence from studies comparing the cost-effectiveness of TR versus that of face-to-face rehabilitation for stroke patients. A second objective was to identify the main clinical effectiveness variables used to calculate cost-effectiveness. Overall, 15 studies met our inclusion criteria and were included. We will first discuss the results of studies examining the clinical effectiveness variables, then those of the studies examining the economic effectiveness of TR compared to face-to-face rehabilitation.

Clinical effectiveness variables of TR

Regarding the clinical effectiveness of TR for stroke patients, the most frequently measured variables included functional abilities, physical and motor recovery. However, study results are mixed based on the outcomes. These inconsistencies can be explained by several factors, including the nature of the TR intervention itself, which was often offered over varying time periods and using several different approaches [17]. This suggests that further standardization across the best evidence-based practices is still required. In addition, different levels of patient compliance with the follow-up instructions given by the physiotherapists at discharge could also explain the observed inconsistencies [18]. Further research on how to improve patients' compliance with physiotherapists' recommendations appears as a logical next step in the investigation.

We also found emerging evidence that the clinical effectiveness of TR is at least comparable to that of face-to-face rehabilitation for a number of outcomes (e.g., motor recovery, pain reduction, verbal communication skills.) While these findings are consistent with the conclusions of a recent meta-analysis performed by Cottrell et al. (2017), they nonetheless contrast with the results of another systematic review conducted by Agostini et al. (2015). However, these two systematic reviews included studies conducted over different time periods; with the most recent evidence being summarized in the Cottrell et al. (2017) study. As a result, it appears that the most recent available evidence supports the clinical effectiveness of TR compared to face-to-face rehabilitation in the recovery of motor function. However, there is still a need for studies based on robust research designs (e.g., RCTs) and larger sample sizes to strenghten the existing evidence.

Economic effectiveness of TR

Economic evaluations included in this review consistently measured both direct and indirect costs of TR (Table 4), and suggested that TR required lower costs per patient than face-to-face rehabilitation [8, 25-27]. This is mainly due to the large difference between the indirect costs generated by the two approaches. In fact, travel expenses, which are null in TR, represent up to 88% of the total cost of face-to-face rehabilitation [8]. Indeed, the main expense associated with TR is related to equipment purchase. Also other expenses are related to the installation/dismantling of the equipment (e.g., webcam, computer, large television screen) and technical support during teletreatments, which are minimal (estimated to range between CAD $0.00 - $1.25 for a typical treatment) [26]. Last, while there were important differences in follow-up times across studies, all were less or equal to a year. Consequently, there is currently no evidence on the long-term outcomes and benefits of TR (i.e., > 12 months) [11].

While we found emerging evidence for the cost effectiveness of TR, several important aspects still need to be documented [11]. Indeed, the clinical effectiveness of TR, relative to its costs, must be established to better support decision-making. Moreover, given the absence of cost-effectiveness studies in the field of TR for stroke patients, further studies in this specific area of investigation are required

It is very important to note that economic analyses of TR (as well as those of face to face rehabilitation) depend healy on the economic perspective adopted by the investigators [11]. For example, when an economic analysis is conducted from the perspective of the healthcare system, this means that only the costs relevant to the health centers are considered, therefore omitting any costs incurred by the patient [26]. Consequently, patient travel costs are not considered in this type of analysis, nor are those incurred by informal caregivers such as subsistence costs (e.g., meals). Therefore, a thorough economic assessment of TR should be conducted from a variety of perspectives (e.g., societal perspective, patient, system). [28].

On that note, the dual society/patient's perspective advocated by Frederix et al. appears to provide the most complete assessment [27]. This approach has several advantages, including taking into account the costs incurred or saved by patients or their relatives. It includes not only the direct costs of the intervention, but also some indirect costs related to illness (loss of productivity) and costs of additional years of life earned. Although this dual perspective requires the use of complex economic methods to comprehensively assess all costs associated with the intervention, we nonetheless recommend it for future research work.

Economic implications and potential influences for decision making

It is recognized that decision-makers regularly face budgetary constraints when promoting access to high-quality rehabilitation care for the greatest number of patients following a stroke. In this context, aside from the commonly used CMA, CEA could provide additional evidence on TR's economic effectiveness versus face-to-face rehabilitation and reimbursement policies could be required to account for it. Indeed, the lack of evidence is one of the most important barriers to generalizing TR. According to Theodoros and Russell (2008), this obstacle prevents often the reimbursement of TR services in healthcare systems around the world [29]. Moreover, differences in the intensity of TR programs which lead to differences in cost and probably differences in its effectiveness do not allow the establishment of reimbursement policies. Consequently, TR is not being consistently done. Thus, the additional evidence that CEA would provide would confirm the economic viability of this approach and allow the development of specific codes and procedures for the payment of TR [17,29]. It should also be noted that authors such as Frederix et al. (2016) confirm the innovative aspect and the impact that a cost-effectiveness study could have in the field of rehabilitation and decision-making at the level of resources assignment and priorities for research development in TR [27].

In addition, unlike CMA, CEA allows decision-makers to know the cost incurred for an additional unit of clinical efficiency gained. This information gives them a better understanding of the economic effectiveness of TR, with respect to the clinical gains achieved. This could influence their decision-making in favor of TR by informing them to leading priorities in terms of health policies in the field of physical rehabilitation following a stroke. In addition, CEA could demonstrate that with more patients in TR, economies of scale can be realized, resulting in monetary gains that could be reinvested in order to expand the pool of TR as part of their rehabilitation offer while increasing accessibility to rehabilitation services.

Strengths and limitations of this systematic review

Before concluding, some important strengths and limitations of this systematic review must be acknowledged. First, although this systematic review of evidence pertained specifically to the physical rehabilitation of stroke victims, we have nonetheless reviewed the evidence pertaining to the clinical benefits and cost-effectiveness of TR in other patient populations, therefore providing a more complete assessment of this technology. Second, in addition to the primary studies published between 2005 and 2018, we have also included two systematic reviews and meta-analyses of the evidence; again to broaden the scope of our conclusions. Last, while the overall methodological quality of the reviewed studies was judged as good, our systematic review is nonetheless limited by the limitations of the primary studies that were reviewed, which include selection bias, small sample sizes, the use of a single economic perspective (as opposed to Frederik's dual perspective), lack of standardization of TR interventions, short follow-up period, failure to consider costs associated with hospital readmissions, stroke-related emergency visits or indirect costs incurred by the patients or their relatives.

Conclusion

This review synthesized the evidence comparing the cost-effectiveness of TR versus face-to-face rehabilitation for stroke patients, highlighting the main clinical effectiveness variables used to calculate cost-effectiveness. In conclusion, this analysis has shown that, to the best of our knowledge, there are currently no studies evaluating the cost-effectiveness aspects of the use of TR for stroke patients. Also, results indicate that balance and mobility are the main clinical effectiveness variables that were considered in existing studies. Last, additional research in this area is urgently needed to better document the cost-effectiveness of TR as this approach could significantly increase the accessibility of rehabilitation services for people living in remote areas, and who are currently underserved.

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Conflicts of interest
The authors declare that they have no conflicts of interest.

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