

Economic evaluations of physiotherapy interventions for neurological disorders: A systematic review

Abstract

Purpose: To identify the existing evidence evaluating the cost-effectiveness of physiotherapy treatments for people with neurological disorders.

Methods: Multiple databases were searched from database inception until July 2018. Studies estimating the cost-effectiveness as incremental cost-effectiveness ratios, cost per quality-adjusted life year, cost per disability-adjusted life year and cost per other measurable results were included. PEDro scale and Consensus on Health Economic Criteria (CHEC) list were used for rating the quality of the evidence.

Results: Ten studies involving 1462 participants were included. Aerobic training, progressive strength training, and a pragmatic physiotherapy programme (combination of stretching, strength and balance training) were reported as potentially cost-effective for older adults with vascular cognitive impairment, falls prevention in Parkinson's disease and multiple sclerosis respectively. Physiotherapy as an adjuvant for pain control was also reported as cost-effective for reflex sympathetic dystrophy. One study testing extra physiotherapy-by-physiotherapy assistant in cerebral palsy and two studies testing extra therapy using a robotic arm and Wii therapy for hand rehabilitation in stroke were reported as not cost-effective.

Conclusions: There are limited studies that have evaluated the cost-effectiveness of physiotherapy treatments in neurological disorders. Three studies that combined extra physiotherapy-by-physiotherapy assistant and novel interventions with conventional physiotherapy were found not cost-effective.

Keywords: Cost-effectiveness; economic analyses; physical therapy treatments; neurorehabilitation, QALY, DALY.

Introduction

Neurological disorders result in a wide spectrum of physical and mental disabilities requiring long-term care. Neurological disorders contribute towards 6.3% of the burden of disease globally[1]. They have a significant economic burden on both the patient and the healthcare system. It is estimated that neurological disorders resulted in 250.692 million disability-adjusted-life-year (DALY) which comprises 10.2% of global DALY [2]. DALY is defined as the sum of years of life lost (YLL) due to disability and years lived with disability (YLLD)[3]. Physiotherapy treatment is an integral part of rehabilitation for clients with neurological disorders. Rehabilitation of clients with neurological disorders lasts for several months, and conditions like cerebral palsy may require rehabilitation for the most part of a person's life. Owing to the expanding population and increase in life expectancy in people with neurological disorders, the need for specialist long-term care such as physiotherapy continues to grow. Therefore, studies on cost-effectiveness of different physiotherapy treatment techniques for neurological disorders are important in minimizing the expenditure on healthcare services.

Economic evaluation is a “comparative analysis of alternative courses of action in terms of both costs and consequences” [4]. Studies on economic evaluations are centered not only on cost-effectiveness but also on the quality of intervention outcome since managing cost should not warrant lowering the standard of care. They are conducted in either healthcare or societal perspective [5]. The results of cost-effectiveness studies provide evidence for the government, non-governmental organizations (NGOs), healthcare departments and practitioners to make decisions on healthcare policies or interventions. However, there is a limited number of studies on cost-effectiveness. Rather, the majority of clinical trials focus more on evaluating the therapeutic effect of the intervention.

In physiotherapy, studies on cost-effectiveness of physiotherapy treatment techniques in neurological rehabilitation are limited. To our knowledge there are no previous systematic reviews reporting on the cost-effectiveness of physiotherapy treatment techniques in neurological rehabilitation. Therefore this systematic review aims to 1) to identify the existing evidence evaluating the cost-effectiveness of physiotherapy treatment techniques for people with neurological disorders and 2) provide future implications for research on cost-effectiveness evaluation for physiotherapy treatment techniques in neurological rehabilitation.

Materials and methods

Study design

This systematic review was developed and reported in accordance with the PRISMA guidelines [6].

Search strategy

A comprehensive search strategy was used to identify potentially relevant studies. Databases, Allied and Complementary Medicine (AMED), Ovid Medline, Embase, Health Technology Assessment (HTA), NHS Economic Evaluation Database (NHS EED) and Cumulative Index to Nursing and Allied Health Literature CINAHL were searched from the date of database inception to July 2018. Search terms were constructed with three themes: neurological disorders, cost-effectiveness, and physiotherapy interventions. Related terms for each theme were identified and used for the search. The search strategy was modified for each search engine. Search strategy for Medline is outlined in [supplementary material 1](#). In order to enhance the rigor of literature search we hand-picked and included 16 common neurological disorders resulting in

physical disabilities that are treated by neurological physiotherapist. Neurological disorders considered for this systematic review are listed in table 1. The inclusion and exclusion criteria (PICOS model) are summarized in [supplementary material 2](#). Three reviewers (SHL, HSL, and HYL) independently screened the articles for the title, abstract and full text. Disagreements were resolved by collectively revisiting the screening criteria. A fourth reviewer was consulted (SW) for unresolved discrepancies.

Insert table about here

Eligibility criteria

Randomized controlled trials (RCTs) evaluating the cost-effectiveness of physiotherapy treatment techniques for neurological disorders were included for this review. Neurological disorders were restricted to those listed in Table 1. To be included, the treatment should be delivered either by a physiotherapist or under the supervision of a physiotherapist. Studies that compared physiotherapy treatment versus no treatment without any restraints, no treatment with restraints, treatments other than PT, placebo and sham physiotherapy were included. This systematic review included both cost-effectiveness and cost-utility studies. Studies were excluded if they were 1) protocols, systematic reviews and conference abstracts; 2) participants undergoing surgery related to their neurological disorder during the study; 3) studies evaluating a multidisciplinary team rehabilitation where physiotherapy was not the focus of intervention, 4) patients in the intervention group do not receive physiotherapy, and 5) non-English studies. The outcome measures for this systematic

review include incremental cost-effectiveness ratios (ICER), cost per quality-adjusted life year (QALY), cost per disability-adjusted life year (DALY) and cost per other measurable results.

Quality assessment and data extraction

The quality of economic evaluation of included studies was assessed using the Consensus on Health Economic Criteria list (CHEC-list), and the methodological quality using the Physiotherapy Evidence Database (PEDro) scale. The CHEC-list allows measurement and comparison of the quality of economic evaluations among studies in systematic reviews [7]. The CHEC-list has 19 items relating to cost-effectiveness and is scored as ‘yes’ or ‘no’. CHEC-list is scored out of 19. Studies scoring more than 14 (>75%) were graded as high quality, studies scoring between 10 to 14 (50-75%) were graded as moderate and those with scores below 10 were scored poor quality [8]. Items were scored ‘no’ if the information was not available or reported. PEDro scale is a reliable and valid tool to quantify the methodological quality of RCTs [9 10]. PEDro scale has 11 items. We rated methodological quality of the included studies as ‘high’ if they scored ≥ 6 or ‘low’ if studies scored ≤ 5 . [11] Quality assessments were carried out independently by three reviewers (SHL, HSL, and HYL).

The following data were extracted: Author’s name, year and country of publication, participants’ demographic characteristics (neurological disorder, mean age, and sex), sample size, type of intervention including duration of treatment, frequency and duration of follow-up, type of economic analysis (cost-effectiveness, cost-utility), perspective (societal, healthcare), cost data (direct medical cost, indirect medical cost, non-medical cost), economic outcome measure, study currency, discounting and author’s conclusion. For consistency, all the study currencies were

converted into US dollars using a conventional currency converter (<http://www.xe.com/en/currencyconverter/>).

Results

The search yielded 225 articles, of which 51 were duplicated studies. Following the full-text screening, ten studies were included for this systematic review [12-21]. Figure 1 illustrates the flow of studies through the review process. Summary of the included studies is reported in table 2. All included studies were published after the year 2000. Heterogeneity among the included studies in terms of outcome measures, study population and intervention tested, made meta-analysis impossible. A combination of generic and disease-specific outcome measures was used among the included studies making it difficult to pool data. Six studies used European Quality of Life-5 Dimensions (EQ-5D) to quantify the quality of life for cost-effectiveness analysis [12 13 16 19-21]. Incremental cost-effectiveness ratio (ICER) per health-related change, which is the ratio of the cost difference between two interventions to the corresponding difference in health outcome, was used for cost-effectiveness analysis in seven studies, while three did not report ICER [12 15 19]. Either ICER per QALY or ICER per Gross Motor Function Measure (GMFM) was used for cost-effectiveness analysis. Six of the included studies conducted economic evaluations among neurological disorders requiring long-term care such as stroke [12 20], subcortical ischemic vascular cognitive impairment (SIVCI) [13], multiple sclerosis [16], cerebral palsy [18] and Alzheimers disease [21]. Follow-up assessments of these studies ranged between 4 and 9 months.

Insert figure 1 about here

Insert table 2 about here

Quality appraisal

Methodological quality and the quality of economic evaluation according to PEDro and CHEC-list are reported in tables 3 and 4 respectively. The methodological quality of seven studies [12-14 18-21] was high, while three [15-17] were of low quality. The quality of economic evaluation was high for two studies [18 20], seven were of moderate quality [12-14 16 17 19 21] and one was of low quality [15]. Since the study protocol of six included studies [13-17 19] was separately published, they were scored ‘no’ for item 2 on describing the competing alternatives. Incremental cost-effectiveness ratio was not available for three studies [12 15 19]. Five of the included studies did not perform a sensitivity analysis for the economic evaluation [12 15 17 19 21]. Since none of the included studies conducted follow-up over 12 months, item 14 on discounting was marked ‘not applicable’ (NA) for all studies.

Insert table 3 about here

Insert table 4 about here

Characteristics of included studies

The included studies were conducted in the United States of America (USA) [17], Canada [13], The Netherlands [15 19], New Zealand/Australia [14], Denmark [21] and the United Kingdom [12 16 18 20]. The ten studies included 1462 participants with sample size ranging between 56 and 235. The mean age of participants ranged between 19.8 months and 74 years. Seven [12 14-17 20 21] of the ten studies included more than 100 participants. Included studies assessed cost-effectiveness of physiotherapy treatment in participants with spastic cerebral palsy [18], reflex sympathetic dystrophy [15 19], stroke [12 17 20], multiple sclerosis [16], subcortical ischemic

vascular cognitive impairment [13] Alzheimer's disease [21] and Parkinson's disease [14].

Among the included studies, three [12 17 18] tested the cost-effectiveness of alternative or extra physiotherapy against physiotherapy in participants with cerebral palsy and stroke. One study [15] compared physiotherapy to occupational therapy and social work, and the remaining seven studies compared specific physiotherapy treatment protocols against usual care. In terms of intervention follow-up, the longest was for 12 months [15], one study [21] followed-up at 4 months and the remaining were more than 6 but less than 12 months.

Physiotherapy intervention parameters of the included studies

Duration and frequency

Duration of intervention ranged from 6 weeks [12] to 12 months [18], with frequency ranging from 1 session per day [12] to 1 session per month [14]. Cost-effectiveness of physiotherapy treatment in neurorehabilitation was not related to duration and frequency of intervention, in that interventions were found to be cost-effective that were performed frequently [13], least frequently [14], short duration [16] as well as longer duration [15].

Perspective

Two [15 16] of the four [15-18] studies that assessed economic evaluations from a societal perspective reported cost-effective and one study [17] reported a tentative lack of cost-effectiveness. In terms of the healthcare perspective, three [13 14 19] of the six studies [12-14 19-21] reported cost-effective physiotherapy treatment techniques.

Conventional and novel neurorehabilitation

Robotics and Wii for participants with stroke were compared to conventional exercises in the studies by Wagner et al. [17] and Adie et al. [12] respectively. Wii™ plus conventional tailored arm exercises was found to be more expensive than conventional tailored treatment for improving arm function, whereas the cost-effectiveness of robotics was unclear. Novel physiotherapy intervention or novel interventions as an adjunct to conventional exercises were found not to be cost-effective among two of the included studies in this systematic review.

Cost-effectiveness of physiotherapy treatment in neurological rehabilitation

The range of ICER (per QALY, GMFM or fall prevented) was between US\$ 43.8425 and US\$ 450255. Table 5 reports the summary of cost and cost-effectiveness of the included studies. One study [12] reported negative ICER. Six studies [13-16 19] concluded the physiotherapy treatment tested was cost-effective or potentially cost-effective, one reported unclear cost-effectiveness [17], while four [12 18 20 21] reported as not cost-effective. Two studies [13 16] of high PEDro and moderate CHEC-list quality found aerobic training and pragmatic exercise intervention as cost-effective for vascular cognitive impairment and multiple sclerosis respectively. The former study conducted an economic evaluation of a 6-month (thrice weekly) progressive aerobic exercise training for 70 adults with mild vascular cognitive impairment and found aerobic training to be potentially cost-effective [13]. The later study estimated the cost-effectiveness of a pragmatic exercise intervention programme including static stretching, aerobic exercise, strength and balance training for 120 participants with mild to moderate multiple sclerosis and found the pragmatic exercise programme as cost-effective. One study [14] of moderate CHEC-list quality and high methodological quality reported that physiotherapy treatment involving balance training,

lower limb strengthening exercise and gait training as potentially cost-effective for preventing falls in people with Parkinson's disease. Among studies testing participants with reflex sympathetic dystrophy, one study of low PEDro and CHEC-list quality found that physiotherapy for pain control and self-management as cost-effective [17] and one study of high methodological quality and moderate CHEC-list quality reported Pain Exposure Physical Therapy as a less costly intervention than conventional physiotherapy for pain relief. Three [12 17 20] studies on stroke evaluated the cost-effectiveness of Wii Sports™ [12], conventional physical therapy plus functional strength training or movement performance therapy [20] and robotic-assisted training to for improving upper limb function [17]. Studies testing conventional physiotherapy plus Wii Sports™ and conventional physiotherapy plus robotic-assisted training, both [12 17] of moderate CHEC quality, however, one [12] was of high PEDro quality; reported additional physiotherapy as not cost-effective. One study [18] of high PEDro and CHEC-list that compared the cost-effectiveness of extra physiotherapy (by physiotherapy assistant), group physiotherapy, and physiotherapy by family support worker for spasticity in children with cerebral palsy found physiotherapy-by-physiotherapy assistant as not cost-effective. One study [21] of high PEDro and moderate CHEC-list score reported that directly supervised group therapy for improving physical activity in participants with mild Alzheimer's disease as not cost-effective.

Insert table 5 about here

Discussion

This systematic review aimed at providing a comprehensive evidence on cost-effectiveness

evaluation of physiotherapy treatment techniques in neurological rehabilitation. This systematic review included ten studies estimating the cost-effectiveness of physiotherapy treatments among seven neurological disorders. Five of the included studies found physiotherapy treatment as cost-effective [13-16 19]. Aerobic training, progressive strength training, and a pragmatic physiotherapy programme (combination of stretching, strength and balance training) were reported as potentially cost-effective for older adults with vascular cognitive impairment, falls prevention in Parkinson's disease and multiple sclerosis respectively. Physiotherapy as an adjuvant for pain control was also reported as cost-effective for reflex sympathetic dystrophy. One study testing extra physiotherapy-by-physiotherapy assistant in cerebral palsy and two studies testing extra therapy using a robotic arm and Wii therapy for hand rehabilitation in stroke were reported as not cost-effective.

Heterogeneity among the included studies made it difficult to pool results for meta-analysis. The country of origin of the included studies was diverse. Variations in the country of origin of study, duration of intervention and follow-up of studies assessing the cost-effectiveness of interventions makes it difficult to draw conclusions on cost-effective physiotherapy treatment techniques in neurological rehabilitation. In spite of the variations in the included studies, this review provides evidence for the available literature on the cost-effectiveness of physiotherapy treatment techniques and scope for future research.

Four studies [13 14 16 21] reported cost-effectiveness of physiotherapy treatment in neurorehabilitation using an acceptability curve. It is relatively a new approach in a cost-effectiveness analysis that illustrates the willingness to pay extra to obtain an additional QALY health benefit for health condition [21]. Reiterating recommendation from Korthals-De Bos et al. [22], this systematic review highlights the need for reporting acceptability curve in conjunction

with ICER per health-related change in future studies on cost-effectiveness analysis, in order to provide a comprehensive guide to the policy makers [22]. Oerlemans et al. [15] reported the economic evaluation data graphically without descriptions of data and discussion making it difficult to interpret the results. The quality of economic evaluation for their study was rated ‘poor’ due to insufficient data. This systematic review strongly recommends future cost-effectiveness studies to report a detailed description of the data and interpretation for transparent interpretation of findings.

Weindling et al. [18] and Adie et al. [12] drew a negative conclusion for cost-effectiveness of interventions tested. However, it is worth noting that *Weindling* et al. [18] concluded additional intervention by family or physiotherapy assistant in children with spastic cerebral palsy to be cost ineffective. Likewise, Adie et al. [12] compared novel physiotherapy to conventional physiotherapy (Wii against traditional arm exercise) for rehabilitation of stroke patients. Considering the cost-ineffectiveness of providing additional therapy to conventional intervention, future studies testing the benefits of additional therapies such as the Wii or extra therapy need to consider conducting a cost-effectiveness estimation extension to RCT to provide evidence for the cost-effectiveness of the proposed modification to conventional physiotherapy treatment.

Despite cost-effectiveness analysis gaining importance, there are a limited number of studies evaluating cost-effectiveness in literature. This systematic review found only ten studies that investigated the cost-effectiveness of limited physiotherapy treatment techniques in neurological rehabilitation. We acknowledge that our study adopted stringent criteria for screening included studies. Such measures involved excluding studies that administered physiotherapy treatment as part of a larger multidisciplinary care. Nonetheless, a limited number of studies (n=10) included in this study may serve as an indicator that studies on cost-effectiveness of physiotherapy

treatment in neurological rehabilitation are limited. Among the countries of origin of the included studies, none were from Asia and Africa region. Economic evaluations are largely conducted in developed countries [23] in particular the Scandinavian countries, Great Britain and Netherlands [24]. Among the low and middle-income countries, lack of resource, lack of good quality economic studies [25], poor acceptance of economic studies among decision-makers [25] and lack of researchers with expertise in cost-effectiveness evaluation have been identified as barriers for conducting and implementing economic evaluations. Diversity in culture, economic dynamics and healthcare policies adopted are country specific. Hence generalizing the available findings is limited to Asian or African countries. Therefore, more studies on economic evaluation of physiotherapy treatment techniques are needed in developing countries where healthcare systems are struggling to provide quality care for the populace.

Our systematic review has the following strengths: 1) this study is the first of its kind as no systematic reviews in the past have investigated the cost-effectiveness of physiotherapy treatment techniques in neurological rehabilitation, 2) the literature search of this systematic review was specific to physiotherapy, 3) we adopted high quality methodological procedures for screening articles and lastly 4) the use of CHEC-list and PEDro scale as appraisal tools are specific and standardized for estimating the quality of method [9] and economic assessment [7].

Some of the limitations of this systematic review include: 1) studies were restricted to the English language, we may have missed studies published in languages other than English, 2) we did not include grey literature including conference abstracts and thesis which might have excluded some high-quality economic evaluation studies, and 3) since the articles included for this review were heterogeneous with regard to population, disease, and intervention tested pooling results was

not possible making it difficult to draw conclusions on cost-effectiveness of physiotherapy treatment techniques for neurological disorders.

Conclusion

There is a limited number of studies testing the cost-effectiveness of physiotherapy treatment techniques in neurological rehabilitation. All the existing literature on cost-effectiveness studies is restricted to North America, Europe, Australia and New Zealand. There is a need to conduct cost-effectiveness studies in Asia and Africa region as the generalizability of findings of previously published studies is limited to the healthcare system of the country of origin. Findings based on three studies combining extra physiotherapy-by-physiotherapy assistant and novel interventions with conventional physiotherapy were found not cost-effective.

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