

The effects of massage therapy on symptoms related cerebral palsy in children with cerebral palsy: A systematic review

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Abstract

The purpose of this review was to systematically present the effectiveness of massage therapy on cerebral palsy related symptoms in children with cerebral palsy based on existing evidence. A comprehensive systematic literature search was conducted from inception to February 2020 using the following electronic databases: PubMed, Google Scholar, Science Direct, Cochrane Library Database of Reviews of Effectiveness, Physiotherapy Evidence Database. To evaluate the articles concerning eligibility, a screening process was carried out by two reviewers. Summary of the studies including outcomes, measures, and results, and methodological quality of the studies were described according to the American Academy for Cerebral Palsy and Developmental Medicine guidelines. Five studies with different conducting design met inclusion criteria were included to document the current body of evidence of massage therapy in children with cerebral palsy. Almost all of the included articles reported that massage therapy (with or without conventional physiotherapy) is effective to improve spastic/hypertonic muscles and gross motor functions. Besides, it was also found that massage therapy could be effective on comorbidities including pain, sleep disorder, and lower perception of life satisfaction. Some of the included studies established the effects of massage therapy on comorbidities related to the cerebral palsy, whereas some of the studies directly demonstrated the effects of massage therapy on secondary problems related to The International Classification of Functioning, Disability and Health body structures/functions, and activity. Consequently, performing massage therapy (with or without physical therapy and/or routine therapy), would be effective to improve most of the symptoms related to cerebral palsy.

Key words: cerebral palsy, spasticity, massage therapy, pain, children

ЕМДІК МАССАЖДЫҢ БАЛАЛАРДАҒЫ ЦЕРЕБРАЛЬДЫ САЛ АУРУЫМЕН БАЙЛАНЫСТЫ БЕЛГІЛЕРГЕ ӘСЕРІ: ЖҮЙЕЛІ ШОЛУ

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ТҰЖЫРЫМДАМА

Бұл шолудың мақсаты қолда бар мәліметтер негізінде церебральды сал ауруы бар балалардағы церебральды сал ауруының белгілері үшін емдік массаж тиімділігін жүйелі түрде ұсыну болды. 2020 жылдың басынан ақпан айына дейін келесі электронды деректер базасын қолдана отырып, әдебиеттерді кешенді жүйелі түрде іздеу жүргізілді: PubMed, Google Scholar, Science Direct, Кохран кітапханасының тиімділігін шолу дерекқоры. Физиотерапиялық дәлелдер дерекқоры. Қатысу құқығына байланысты мақалаларды бағалау үшін рецензиялау процесін екі рецензент жүргізді.

Зерттеу нәтижелері, қорытындысын қоса отырып, көрсеткіштері мен нәтижелері және зерттеу әдіснамалық сапасы Американдық Церебральды сал ауруы академиясының және Даму медицинасының нұсқауларына сәйкес сипатталған. Церебральды сал ауруы бар балалардағы емдік массаж туралы қолда бар деректерді құжаттау үшін әр түрлі әдістері бар және енгізу критерийлеріне сәйкес келетін бес зерттеу енгізілді. Барлық мақалалардағы барлық дерлік массаж терапиясы (қалыпты физикалық терапиямен немесе онсыз) спастикалық/гипертониялық бұлшықеттер мен мотордың жалпы функцияларын жақсартуда тиімді болып табылады.

Сонымен қатар, массаж терапиясы ауырсыну, ұйқының бұзылуы және өмірге қанағаттанушылықты қабылдауды төмендету сияқты ауруға қарсы жағдайлар үшін тиімді деп табылды. Бірқатар зерттеулер массаж терапиясының мидың сал ауруымен байланысты жанама ауруларға әсерін анықтады, ал кейбір зерттеулерде Медициналық қызметке, мүгедектікке және денсаулықты қорғаудың халықаралық жіктемесінің денесінің құрылымымен / функциясымен байланысты екінші проблемаларға массаж терапиясының әсері анықталды.

Сондықтан массаж терапиясын (физиотерапиямен және/немесе кәдімгі терапиямен немесе онсыз) жүргізу церебральды сал ауруымен байланысты көптеген белгілерді жақсартуда тиімді болады.

Негізгі сөздер: церебральды сал ауруы, серпімділік, терапиялық массаж, ауырсыну, балалар

ЭФФЕКТ ЛЕЧЕБНОГО МАССАЖА НА СИМПТОМЫ, СВЯЗАННЫЕ С ЦЕРЕБРАЛЬНЫМ ПАРАЛИЧОМ У ДЕТЕЙ: СИСТЕМАТИЧЕСКИЙ ОБЗОР

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РЕЗЮМЕ

Целью данного обзора было систематически представить эффективность лечебного массажа при симптомах, связанных с церебральным параличом, у детей с церебральным параличом на основании имеющихся данных. Комплексный систематический поиск литературы проводился с начала до февраля 2020 года с использованием следующих электронных баз данных: PubMed, Google Scholar, Science Direct, База данных обзоров эффективности Кокрановской библиотеки, База данных физиотерапевтических доказательств. Для оценки статей, касающихся права на участие, процесс рецензирования был проведен двумя рецензентами. Резюме исследований, включая исход, показатели и результаты, а также методологическое качество исследований были описаны в соответствии с руководящими принципами Американской академии церебрального паралича и медицины развития. Пять исследований с различной методологией и соответствующие критериям включения, были включены для документирования имеющихся данных о лечебном массаже у детей с церебральным параличом. Почти во всех включенных статьях сообщается, что лечебный массаж (со стандартной физиотерапией или без нее) эффективен для улучшения спастических/гипертонических мышц и больших моторных функций. Кроме того, было также обнаружено, что лечебный массаж может быть эффективен при сопутствующих заболеваниях, включая боль, нарушение сна и снижение восприятия удовлетворенности жизнью. Некоторые из включенных исследований установили влияние лечебного массажа на сопутствующие заболевания, связанные с церебральным параличом, в то время как некоторые из исследований прямо продемонстрировали влияние лечебного массажа на вторичные проблемы, связанные со структурами/функциями тела Международной классификации функционирования, ограничений жизнедеятельности и здоровья. Следовательно, выполнение лечебного массажа (с физиотерапией или без нее и/или обычной терапией) будет эффективным для улучшения большинства симптомов, связанных с церебральным параличом.

Ключевые слова: церебральный паралич, спастичность, лечебный массаж, боль, дети

Introduction

Cerebral palsy (CP) is the most common form of motor disorder of childhood [1] and is characterized by a group of disturbance of the development of movement and posture, inducing activity limitation through non-progressive impairments that emerge in the developing fetal or infant brain [2]. Although cerebral palsy stems from a primary disruption in the central nervous system (CNS), clinic symptomatology is largely observed in the peripheral neuromuscular system [3], especially in the skeletal muscles [4]. From the International Classification of Functioning, Disability and Health (ICF)'s perspective, CP compromise a person's 'functioning', (containing body structures [e.g. upper limbs], body functions [e.g. psychological functions], activities [e.g. manual ability], and participation [e.g. park activity]), which may lead to 'disabilities' such as limb involvements, activity and participation restrictions, respectively [5,6]. Children with CP largely experience a wide range of problems (impairments) with motor and sensory functions such as muscle stiffness, spasticity and/or dystonia, reduced muscle length, musculoskeletal deformity [7]; loss of proprioceptive input, light touch perception, two-point discrimination, and weight perception [8]. Spasticity is the most common motor disorder [9], following damage in descending motor pathways [8].

Spasticity can induce muscle stiffness/hypertonia, limitation at the joint movements, muscle contracture, impairments in motor function, and lower functional profile [10]. Moreover, spasticity

frequently leads to pain in children with spastic CP, especially in those classified within the Gross Motor Function Classification System (GMFCS) V [11]. The presence of pain is related to a worse quality of life in children with CP [12]. Of children with moderate to severe CP, 40-73% have various sensory deficits leading to decreased use of the extremity, impaired hand function, and motor ability, diminished spatial perception, or position of the joints [13,14]. Besides, cerebral palsy is generally accompanied by comorbidities, which might be a problem as important as the physical impairments, such as behavior disorder, bladder incontinence, sleep disorder, blindness, non-oral feeding deafness and epilepsy [15]. Therefore, there is a wide range of potential problems a person with cerebral palsy may experience such as motor or sensory deficits, sleep disorder, non-oral feeding, and bladder incontinence.

Although there are various comorbidity-specific treatment approaches in cerebral palsied individuals, management of the most common symptoms mentioned above might be expected to be related to spasticity [10,16], because abnormal physiological features of the skeletal muscle itself might single-handedly lead to most common problems seen in CP.

Management of spasticity includes a major challenge to health professionals. Various types of treatment approaches are available to decrease the severity of spasticity/hypertonia, such as botulinum toxin, several medications, intrathecal baclofen, massage therapy, manual stretching, casting, selective dorsal rhizotomy, and complementary and alternative therapy. Given that these available intervention approaches are effective in

treating spasticity [17], each of these can be applied to spastic muscle depending on the choice of health professionals

However, there is yet no adequate evidence demonstrating comprehensively the advantages of the current intervention approaches regarding reducing of spasticity/hypertonia [18]. Massage therapy, as one of the intervention approaches, was reported to be effective in reducing the severity of spasticity/hypertonia in children with cerebral palsy [19]. Massage therapy might be effective in improving motor functions, sensory deficits, pain, and other symptoms "of children with CP, by regulating muscle tone or stiffness [20]. It reduces the severity of spasticity/hypertonia by stretching the muscle-tendon unit and inducing the Golgi Tendon Organ that could suppress alpha motor neurons [17]. Finally, in their study which reported the current evidence about intervention approaches for children with CP, Novak et al [21] showed massage therapy at the "Yellow Light" level, which stands for either lower-level proof establishing its effectiveness or inconclusive evidence. Based on the premise mentioned above, the purpose of this review was to report whether that massage therapy, as a complementary treatment, is effective on cerebral palsy symptoms in children with CP. To answer these questions, we planned to provide a summary of the body of the available evidence regarding massage therapy, and we hypothesized that the massage therapy might be effective on CP-related symptoms in children with CP.

Material and methods

Study design

This review was carried out by the guidelines of the American Academy of Cerebral Palsy and Developmental Medicine (AAPDM) methodology for reporting systematic reviews of intervention approaches [22], and Preferred Reporting Items for Systematic Review and Meta-Analysis Protocols (PRISMA-P) [23]. Ethical approval was not provided because the manuscript did not request human participation.

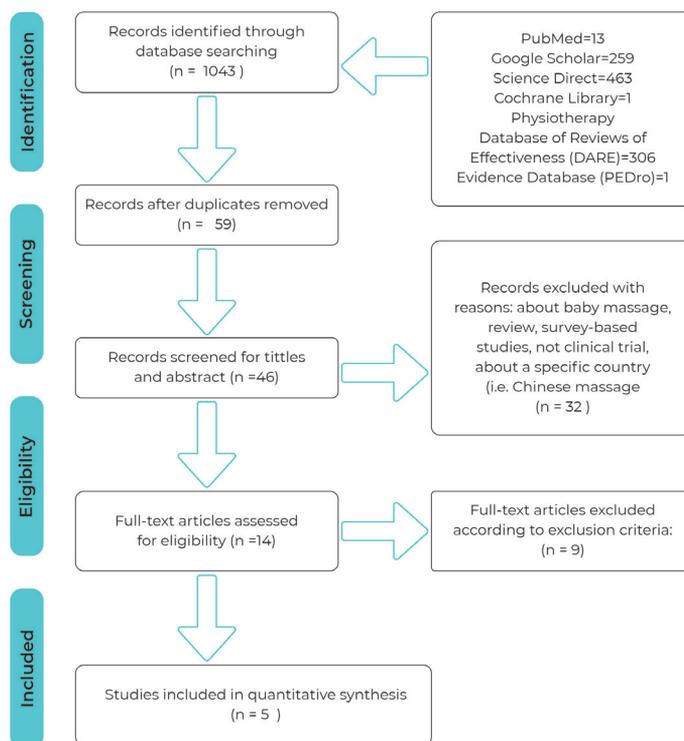
Search Strategy

In June 2019, a comprehensive systematic literature search on all articles regarding massage therapy published from database inception to June 2019 was conducted by searching the following databases: PubMed, Google Scholar, Science Direct, Cochrane Library Database of Reviews of Effectiveness (DARE), and Physiotherapy Evidence Database (PEDro). Besides, literature searches were updated in February 2020. Keywords by The Medical Subject Headings (MeSH) terminology were specified using: (1) most recognized websites such as Developmental Medicine and Child Neurology (www.aacpdm.org), CanChild (www.canchild.ca), (2) notable articles of the field, and (3) articles selected for this review. The keywords 'cerebral palsy', 'spasticity', 'massage therapy', and 'children' were used as collective search terms. The keyword "cerebral palsy" was searched as a Population Descriptor or Sample Descriptor and then, this keyword was combined with other keywords to reach all relevant studies. 59 articles retrieved from all databases were examined by two authors in terms of their titles and content. Full-text articles qualified with the inclusion criteria or the works that were carried out as clinical studies were selected. The screening process was conducted by two authors, and when the authors could not reach a consensus, they consulted the third author. Each electronic database was searched on an individual basis using the P.I.C.O. Model for Clinical Questions [patient/problem, intervention, comparison, and outcome] search terms [24].

Inclusion and exclusion criteria

All retrieved articles were reviewed for content, methodology, feasibility, target population, generalizability (generic versus specific to region), clinic-based outcome (rather than case-report), and ICF domain (body function, body structure, or activity and participation) [25]. Taken altogether, the trials were included if (1) they had text published in English, (2) subjects of the study were children or adolescents with CP (aged between 31 months and 17 years), (3) they were conducted in a clinical setting (not survey-based), and (4) the effect of massage therapy on improving muscle tone was investigated using at least one outcome measure. Articles featured (1) systemic or non-systematic reviews, (2) only about the baby or adult massage, (3) about a specific country (e.g. Chinese massage), (4) unpublished studies or grey literature publications were excluded. After detailed searches in given databases, a total of 59 articles were initially screened for relevance following inclusion/exclusion criteria. At the first stage, 13 articles were deported due to various reasons. Subsequently, the remaining 46 works were reviewed by authors considering the inclusion criteria. After a detailed examination of each article, the authors agreed on five articles for systematic review. A flow diagram of the retrieved articles, illustrating the inclusion and exclusion process, is shown in Figure 1.

Figure 1 - Prisma Flow Diagram



Data collection procedure and quality assessment

Two authors, independently from each other, retrieved data from each included paper by utilizing the convenient AAPDM 'study data extraction summary form', and any disagreement was discussed to reach consensus by the third author. These forms were eligible for both study designs including group or single-subject designs. A total of five articles consisting of two single-subject design studies and three group design studies were

included in this study. The tables of summary for each article documented data regarding participants, intervention, outcome measure(s), level of evidence and conduct rating, ICF domain, and the results. For each article studied as subject design or group design, the questions were answered by adhering to the

AACPDM guideline [26].

For the studies conducted as a single subject design, changes between the pre- and post-interventions of each participant involved in the study were recorded separately (Tables 2, 3), while changes in participants in group design

Table 2 Summary of studies: outcomes, measures, and results

Single Subject Study	Level of Evidence and Conduct Rating	Participants	n	Ages	Intervention Group	Control group	Outcome of interest	Measure	ICF Component	Results
2006 Macgregor BSc et al.23	-IV, Non-randomized controlled	-Adolescents with CP (spastic diplegia) -GMFCS I-IV	Total=5	12-15 y (mean age=14)	-Stretching calf muscle transversely rather than longitudinally with a standardized sequence of Swedish massage strokes + -Strong Stretch exercise -The data were collected at 1st week before intervention, and 5th week, and 17th week after intervention -Duration of intervention: 14 minutes, twice a week, for 5 weeks.	no	-Range of passive and voluntary movement -Stretch reflex -Motor skills	Goniometer -EMG -GMFM-66	•B(b710) mobility of joint function •B(b750) Motor reflex function •B(b789) Neuromusculoskeletal and movement-related functions, other specified	-consistently increased dorsiflexion of only two ankles among ten ankles - Elongation or shortening in the calf muscles of the individuals was observed. -increase in GMFM score of four participants -In the first two sessions, a stretch reflex in the soleus muscle of participant 2 began at a more dorsiflexed angle. -decreased phasic stretch reflexes - inhibition of the soleus muscle during stretch in participant 1 in sessions 4 and 5 as a diminution of background EMG activity. -Participant 1 made striking gains in the freedom of movement of both ankles after the third massage session.

Abbreviations: GMFCS, Gross Motor Function Classification System; EMG; electromyography; GMFM; Gross Motor Function Measure

Table 3 Summary of studies: outcomes, measures, and results

Single Subject Study	Level of Evidence and Conduct Rating	Participants	n	Ages	Intervention Group	Control group	Outcome of interest	Measure	ICF Component	Results
2010 Nilsson ⁴²	-I, M (9/14), Randomized Controlled Trial	-Unilateral spastic CP (1) -Bilateral spastic CP (3) -Dyskinetic CP (2) -GMFCS II-V	Total=6	3-17y	-Massage therapy starting with the right leg, and continued with the foot, right arm, hand, left arm, hand, and finally the left leg and foot. -Duration: 30 min	Resting for 30 minutes	-Sleep quality -Well-being -Hearth rate -Pain	-Parent's report -VAS (Visual Analog Scale) -CHQ-PF50 (Child Health Questionnaire Parent Form 50) -Pulseoxymeter -CAS (Coloured Analogue Scale) -Qualitative report	B (b280-b289) Pain B (b134) Sleep functions B (b410) Heart functions	-no difference in heart rate between before and after massage therapy -decreased heart rate in the control group -low pain in all participants -no statistically significant difference between the study groups concerning pain intensity, well-being, or sleep quality.

Abbreviations: M, Moderate; GMFCS, Gross Motor Function Classification System; CHQ, Child Health Questionnaire Parent Form 50; CAS, Colored Analogue Scale

Table 5 Summary of studies: outcomes, measures, and results

Single Subject Study	Level of Evidence and Conduct Rating	Participants	n	Ages	Intervention Group	Control group	Outcome of interest	Measure	Components of Health	Results
2017 Rasool ²³	II, M (4/7) Small sample size	-Spastic and diplegic cerebral palsy -the presence of mild mental retardation	Total=60	3-9 y (mean= 6.03±1.73)	Routine therapy + -Cross-friction massage on both the legs at soleus and gastrocnemius along Achilles tendon. -Conventional physiotherapy (hot pack and Bobath Therapy) Duration:30 minutes, 5 times a week lasting 6 weeks	Routine therapy	-Spasticity -Mobility (ambulation level)	-Modified Ashworth Scale (MAS) -A 9- pointed Scale for defining functional level	B (b735) Muscle tone functions B (b789) Movement functions, other specified and unspecified	reduction in the severity of spasticity -no significant improvement in gross and fine motor function

Abbreviations: M, Moderate; MAS; Modified Ashworth Scale

Table 6 Summary of studies: outcomes, measures, and results

Single Subject Study	Level of Evidence and Conduct Rating	Participants	n	Ages	Intervention Group	Control group	Outcome of interest	Measure	Components of Health	Results
2018 Bingöl ⁴⁰	II, W (3/7) Small sample size	Quadriplegic=13 Diplegic=4 Hemiplegic=3	Total=20	5-12 y (mean=8,15±1.7)	- 10-minute FM to adductors, hamstring muscles, calf and hip flexors at each lower extremity	Conventional physiotherapy	-Spasticity -Gross motor function -Mobility level	- MAS -GMFM GMFCS	B (b735) Muscle tone functions D (d410-d429) Changing and maintaining body position D (d450-d469) Walking and moving (d450-d469)	- significant improvement in muscle spasticity at hamstring group muscles and triceps surae in both group Significant reduction in hip flexor muscles spasticity only for experimental group - significant increase in GMFM Score and GMFCS level for both group

Abbreviations: W, Weak; FM, Functional Massage; MAS, Modified Ashworth Scale; GMFCS, Gross Motor Function Classification System; GMFM, Gross Motor Function Measure

Table 4 Summary of studies: outcomes, measures, and results

Single Subject Study	Level of Evidence and Conduct Rating	Participants	n	Ages	Intervention Group	Control group	Outcome of interest	Measure	Components of Health	Results
2007 Hernandez-Reif ¹⁷	-II, M (5/7) -Small RCT	-Cerebral Palsy (spastic, athetoid, ataxic) -GMFCS I-IV	Total=20	Mean age=31 months	-Massage Therapy to head, face, scalp, shoulders, arms, hands, hips, legs, feet -in the form of 30 minute- sessions twice per week for 12 weeks	Reading attention	-Spasticity values of both arm, elbow, and wrist -Spasticity values of right and left leg, hip, and ankle -Range of motion of hip extension and abduction -Perceptual/ fine motor; gross motor; self-care (feeding, toileting, dressing/hygiene skills), social/emotional, language and cognition	Modified Ashworth Scale -Arms, legs, and trunk muscle tone scale (ALT muscle tone scale). -Goniometer -Developmental programming for infants and young children (DPIYC)	B (b735) Muscle tone functions B (b710) Mobility of joint functions B (b789) Movement -D(d910) Community life -D (d530) Toileting -D(d540) Dressing -D(d550) Eating	reduced arm spasticity -improved muscle tone for the overall body -improved right hip and left hip extension -improved: (a) cognition (b) fine motor (c) gross (d) dressing and (e) social functioning -positive facial expressions

Abbreviations: RCT; Randomized Controlled Trial; GMFCS, Gross Motor Function Classification System; M, Moderate

Table 1 Conduct of Study Design

Single Subject Study	Level/Quality	1	2	3	4	5	6	7	8	9	10	11	12	13	14
2006 Macgregor BSc et al.23	IV, SSRDs	-	-	-	-	-	-	-	-	-	-	-	-	-	-
2010 Nilsson ⁴²	I, M (9/14)	yes	yes	yes	no	yes	no	no	yes	yes	yes	yes	no	no	yes
Group Study	Level/Quality	1	2	3	4	5	6	7	8	9	10	11	12	13	14
2007 Hernandez-Reif ¹⁷	II, M (5/7)	no	yes	yes	yes	yes	No	yes	-	-	-	-	-	-	-
2017 Rasool ²³	II, M (4/7)	yes	yes	no	yes	yes	no	no	-	-	-	-	-	-	-
2018 Bingöl ⁴⁰	III-W (3/7)	yes	yes	yes	no	no	no	no	-	-	-	-	-	-	-

Abbreviations: SSRD, Single Subject Study; M, Moderate; W, weak

studies were recorded as the mean value. The quality assessment of each article included was detected in two steps (Tables 5, 6). The first step was to determine the study quality according to the classification of the AACPDM methodology based on the description of Lynne Romeiser (for single-subject design study) and Dr. David Sackett (for group design study) [26]. The second step was to assess the conduct of the studies. For group studies, this step is a quality assessment consisting of seven conduct

questions. Each question was responded "yes" or "no". The quality of the design of an individual trial was evaluated as Strong ('yes' response on 6-7 of the inquiries), Moderate (point 4 or 5) or Weak (point <3) (Table 4). For single-subject design studies, each included study was rated using a 14-item questionnaire, and the quality of design of an individual work was evaluated as Strong ('yes' point on 11-14 inquiries), Moderate (point 7-10), or Weak (point <7) (Table 1).

Results

Types of participants

Studies were involved in this systematic review if they predominantly included children or adolescent participation diagnosed with any subtype of CP. Motor type of CP enrolled in the included studies consisted of various subtypes. In this context, the participants diagnosed with spastic unilateral or bilateral CP, and spastic athetoid or dyskinetic type, and aged between 31 months to 17 years, and with any mobility level (Gross Motor Function Classification System [GMFCS] score I to V) were incorporated in the current review

This systematic review is based on literature search in PubMed, Google Scholar, Science Direct, Cochrane Library Database of Reviews of Effectiveness (DARE), and Physiotherapy Evidence Database (PEDro) utilizing the key phrases “the efficacy of massage therapy on muscle tone/spasticity in children/individual with CP” with an emphasis on clinical studies. Using the search strategy, a total of 1043 articles were identified; of which 46 were screened for title and abstract. The full text of 14 studies were assessed for eligibility, and only five met inclusion criteria. Among these five articles, two of them involved single-subject design studies (evidence level I, IV), while the other three (evidence level II, III) included group study design studies. Considering the characteristics of the articles, tables were created separately according to the study design (single-subject design or group design) by the AACPDm methodology. Hence, most of the included articles provided low-level evidence by to Romeiser's (for single-subject design study) and Sackett's (for group design study) criteria. Table 1 describes the methodological quality of the included articles. The contents of each article associated with a citation, level of evidence and conduct rating, treatment type, subjects, findings, assessment tools, and coding concerning the ICF-Children & Youth Version [27] were summarized based on the AACPDm principles [26]. Types of massage therapy of which its effectiveness was investigated across reviewed studies were as follows: Swedish massage (in three studies) [28-30], deep cross friction massage (in one study) [31] and functional massage (in one study) [32]. In two studies included, the control groups received usual care, whereas there was no reported intervention approach for control groups of other studies. In two of 5 included articles, participants in the experimental group were delivered only massage therapy, while in two of two included articles, massage therapy was performed as adjunctive therapy. In contrast to these four, massage therapy was integrated with stretch exercise (simultaneously with stretching exercises) in another study.

Efficacy of Different Types of Massage Therapy Across Main Domains of the ICF

The effectiveness of five retrieved studies was elaborately evaluated and abstracted according to the relevant ICF subdomains under the AACPDm guidelines (Table 2-6). In their randomized controlled study, Hernandez Reif et al. carried out a trial with 10 participants in the experimental group. The participants of the study were delivered a 30-minute massage therapy integrated with the children routine therapy including occupational therapy, music therapy, and speech therapy two times per week for 3 months. They revealed a significant reduction in arm spasticity, improved muscle tone for the overall body, increased extension of right and left hip joint, and improved (a) cognition (b) fine motor, (c) gross motor (d) dressing and (e) social functioning in the massage therapy group [28]. According to the study of Rasool et al. conducted with a group design in which 60 participants were distributed

randomly into the experimental and control group, both groups were applied conventional physiotherapy including 10-minute hot pack application and Bobath Therapy followed by a stretch exercise to the Achilles tendon; however, the treatment group was additionally delivered with cross-friction massage on soleus and gastrocnemius of both the side along Achilles tendon. A significant improvement both in the severity of spasticity and in the functional level was revealed in the experimental group [33]. Another study conducted by Nilsson et al. through single-subject design which consisted of a total of six participants who had undergone orthopedic surgery at one or two limbs. Three participants in the experimental group received massage therapy for 30 minutes before the physiotherapy session. In contrary to this, the control group took a rest for 30 minutes before the conventional therapy. After seven intervention sessions, it was found a low score for pain intensity and discomfort in either experimental and control groups. However, participants in both groups demonstrated similar statistical differences in variables regarding pain intensity, well-being, or sleep quality. Besides, there was not found out the statistically meaningful difference in heart rate immediately post-intervention for the experimental group, while there was a significant change in the same variable for control group [30]. In the study conducted as single-subject design by Macgregor [29] which explored the effectiveness of massage therapy on the mechanical properties of muscles in children spastic diplegia, each participant's calf muscles were moved transversely rather than longitudinally with an adjusted sequence of classic massage lasting for 14 minutes, twice a week for 5 weeks. After the massage therapy, it was found that the range of movement increased, but not consistently. Also, the participants within the Gross Motor Function Classification System (GMFCS) Levels I and II exhibited continued improvements in Gross Motor Function Measure-66 scores.

Consequently, the authors of this study reached a consensus that calf muscles could either shorten or lengthen when passively moved. For stretch reflex, in the first two session periods, a stretch reflex in the calf muscle of one subject started at a more dorsiflexed angle after massage therapy, suggesting a decreased responsiveness. On the other hand, in the subsequent massage sessions, reduced phasic stretch reflexes were noted soon after the beginning of stretch. As for voluntary movement, freedom of movement of both ankles after the third massage session was developed significantly for another subject. One subject developed a more modest movement in one limb, also after the third massage session. Besides, one subject made no evident improvements; but, another one subject developed, especially in the last session when forced to contact the calf muscle, rather than inverting and evert the ankle. Subsequently, one subject could not act either ankle voluntarily. Concerning changes that occurred in gross motor function, by the 5th week of the intervention, the ambulant participants developed their GMFM-66 scores by an average of 6.4%, but the score of the non-ambulant subject was unaltered [29]. Finally, another study conducted by Bingöl et al [32] suggested that stretch exercise integrated with Swedish massage, described as functional massage, might be more effective on muscle spasticity and gross motor function than forcible exercise alone.

Discussion

Management of CP includes a major challenge for health professionals, and currently there is no standardized treatment option for children with CP [34]. From the ICF perspective, CP commonly compromises a person's body structure/functions and activity/participation [35]. Also, it is often accompanied

by pain, sensory deficit, impaired perception and cognition, communication, and behavioral problems [15].

However, increased muscle tone/spasticity which has serious consequences concerning body position (i.e. joint deformity, joint subluxation, and contracture), functional ability, quality of life, and financial cost is the underlying factor of most notable problems. Thus, it is well known that most of the intervention strategies towards body structures/functions are based upon the management of abnormal muscle tone or spasticity. Many treatment approaches are used to manage increased muscle tone/spasticity in children with CP such as oral medications, physical and occupational therapy, splinting and casting, botulinum toxin, selective dorsal rhizotomy, intrathecal baclofen, orthopedic surgery [36-39]. Massage therapy, as reported in a systematic review by Novak et al [35], was indicated as one of the intervention approach which is used to regulate abnormal muscle tone in the management of CP. When we examine the history of massage therapy, it is seen that published research studies on massage therapy date back to the 1930s, when human and animal studies were popular [28].

Massage therapy is a relatively simple [40], inexpensive and non-invasive treatment, and has been applied to treat stroke subjects for many years [17,41]. In this context, massage therapy is utilized to improve spastic muscle, to augment the elastic quality of muscles and connective tissue, to reduce muscle pain, improve range of motion and muscle contracture, and support relaxation [42,43]. Based on the issues mentioned above, we suggested that massage therapy may be effective on CP-related symptoms and motor function by improving the abnormal muscle tone/spasticity in individuals with CP safely and efficacious. Any intervention to improve spasticity/abnormal muscle tone, either complementary or evidence-based practices, should be assessed in terms of their efficacy on body structures/functions and, activities and participation, as well as comorbidity. Considering our purpose in this systematic review, we selected articles whose resultants were predominantly based on ICF's s body structure/function domains (muscle tone function, mobility of joint function, and pain), ICF's activity domain (gross motor function) and as well as well-being. Hence, the main purpose of this review was to summarize the clinical studies investigating the effectiveness of massage therapy on symptoms related to CP in children with cerebral palsy.

To our knowledge, this pooled study from both group and single-subject designed studies are the first to document the body of evidence of the effectiveness of the massage therapy on symptoms associated with CP within all main subdomains of the ICF. Most of the articles summarized in this review demonstrated level III (moderate) evidence. Almost all of the included articles reported that applying massage therapy to spastic/hypertonic muscles, with or without conventional physiotherapy, is effective for reducing increased muscle tone or

spasticity. Besides, four out of the included articles revealed that massage therapy is effective for improving gross motor function. Besides, one study demonstrated that massage therapy affected fine motor functions. Also, two studies found out significant gains in the range of motion at joints in the participants being subjected to massage therapy. Subsequently, one study reported meaningful findings regarding independence in daily activities. Finally, 1 out of the included articles reported notable changes in satisfaction related to well-being, sleep, and pain.

Accordingly, applying massage therapy for sufficient dosage, for instance, 5 to 12 weeks and with a duration of 14 to 30 minutes per session on average, can improve symptoms related to CP including increased muscle tone, gross motor function, and well-being. This could be explained by the presence of the effect of massage therapy on increased stretch reflex stiffness which leads to spasticity. Given that spasticity causes increased muscle tone that induces a diminished range of motion, muscle contracture, restricted movement patterns, and releasing of pain, it might be expected that relaxation of spasticity improves muscle hypertonia, motor functions, and physical or psychological well-being. Consequently, some included studies established the effects of massage therapy on comorbidities related CP, whereas some of the studies directly demonstrated the effects of massage therapy on secondary problems related to ICF's body structures/ functions, and activity.

Study limitation

This study has some limitations. Only studies with low to moderate quality of evidence were included in the review. Besides, we also eliminated nine studies because they featured specific to region or countries. This led to a limited number of studies being included for reporting the effectiveness of massage therapy on symptoms in children with cerebral palsy. Furthermore, the studies included in this review consisted of small sample size which negatively affect the reliability of the studies' findings. Thus, further studies are warranted, before concluding that massage therapy is beneficial.

Conclusion

Performing massage therapy (with or without physical therapy and/or routine therapy), would be effective to improve most of the symptoms related to CP. If so, rather than performing a pure challenging stretch exercise which may lead to disturbance, pain, tendon tear, and an increase in tendon reflex, we recommend that it may be better to perform stretch exercise integrated with massage therapy. Nevertheless, because of the heterogeneity in the study designs of the included studies, this implication should be interpreted also in the light of healthcare professionals' own clinical experiences.

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