Serial casting for equinus deformity in children with cerebral palsy: A systematic review

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Abstract
Aim: The primary cause of gait deformity in children with cerebral palsy (CP) is equinus foot. Children with equinus foot typically receive physiotherapy as part of their care, as serial casting. In this study, we aimed to systematically examine the impact of serial casting procedure in children with CP with equinus deformity.

Material and Methods: Articles were identified from 2008 to 2019 by literature searches using Medline (MEDLINE), physiotherapy evidence base (PEDro), and Cochrane database. Randomized trials focused on CP children with equinus deformity were included. Data were extracted from the included studies and methodological quality of these data was evaluated using the PEDro scale. The modified Sackett scale was used to evaluate the level of evidence of each intervention.

Results: Four trials with good quality methodology were identified with good quality methodology. It was examined whether studies regarding intervention techniques were heterogeneous or not. Findings were analyzed in qualitative terms. This review revealed moderate evidence in three articles about the effectiveness of serial casting application and the ineffectiveness in one article, in addition to traditional physiotherapy programs. Meta-analysis was applied for homogeneous studies and it showed that serial casting could be used as a method to improve deformity of the equinus.

Discussion: The current systematic review analyzed four randomized controlled trials, applying strict inclusion selection criteria. The present evidence supports the use of serial casting for improving equinus deformity and modulates spasticity in children with CP and it is considered a moderate evidence of the efficacy of serial casting application. Although the findings of this review support the effectiveness of serial casting application in CP children with equinus deformity, additional randomized control trials with a larger sample size are still required to confirm the present evidence.

Keywords
Serial casting; Deformity of equinus; Cerebral palsy; Toxin of botulinum; Abnormality of gait; Scissoring

DOI: 10.4328/ACAM.20264   Received: 2020-06-27   Accepted: 2020-09-04   Published Online: 2020-10-28   Printed: 2021-02-01   Ann Clin Anal Med 2021;12(2):222-227
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Introduction
Equinus foot is a disorder of the musculoskeletal system often seen in patients with cerebral palsy (CP) and can restrict activities such as walking function [1]. Equinus is the most frequent problem in ambulatory children with spastic CP, resulting in an unstable and inefficient gait pattern. Without careful early treatment, it may evolve into permanent foot deformities which may require multiple surgical procedures. A variety of conservative approaches are currently available for managing Equinus in children with CP, such as bracing, stretching exercises, electrical stimulation, and casting/serial casting [2].

Through physical therapy, such as stretching and muscle strengthening exercises, antispastic medications, BTX-A injection, ankle-foot orthoses, and serial casting, or surgery, such as tendon elongation of Achilles, Equinus foot can be handled. BTX-An injection and serial casting are among such treatments with limited side effects relative to drug therapy [3]. Serial casting is described as using a series of progressive casts to increase the length of the muscle using low load prolonged stress on contracted tissues [4]. Serial casting aims to improve a pattern of the equinus gait in children with spastic CP (SCP) [5].

Numerous clinical trials have shown that casting is a useful adjunct to Botulinum toxin A (BoNT-A) for enhancing results, using either serial or single casts. In reducing spastic hypertonia, the combination of BoNT-A and casting was superior to BoNT-A alone [6]. This systematic review aimed at verifying the effect of the application of serial casting on equinus deformity for children with CP.

Material and Methods
Search strategy
This study was based on the recommendations of the Statement on Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) [7]. Eligibility criteria were defined as follows: (a) Participants: children with CP accompanied by equinus deformity aged from 2 to 17 years of both genders, (b) Interventions: the study group received serial casting after BoNT-A and traditional physiotherapy program, (c) Outcomes: Passive range of motion (PROM), spasticity degree, ankle kinematics during gait, gross motor function level and (d) Study design: Randomized controlled trials (RCTs). An electronic search was done from 2008 to 2019, in the Cochrane Central Register of Controlled Trials, PEDro and PubMed databases using the following keywords: “Serial casting” and/or “Equinus deformity” or “Cerebral palsy” or “Botulinum toxin” or “Scissoring” or “Gait abnormality.” The search was limited to RCTs only which published from 2008 to 2019. Studies were excluded according to the following criteria: (a) Non RCTs, (b) Participants aged over 17 years, (c) If no English translation is available and/or (d) Unpublished studies. Two authors independently evaluated each title and abstract identified in the search against the eligibility criteria. The full text was obtained for complete analysis.

Data extraction
One reviewer extracted data from the included articles, and a second reviewer cross-checked it. The data extraction form included authors and year of publication, the characteristics of the participant, measures of intervention, and outcomes [8].

Quality assessment
Two authors applied the PEDro scale [9] separately to determine the quality of the trials and the third author resolved any disagreements.

Data analysis
The following classification was used for the quantitative quality rating: PEDro score < 4 indicated poor quality; 4-5 indicated fair quality; 6-8 for good quality and 9-10 indicated excellent quality. The modified Sackett scale [10] was used for assessing the level of evidence as follows:

- Level 1a (Strong) = Well-designed meta-analysis or 2 or more ‘high’ quality RCTs (PEDro Scale scores ≥ 6) that show similar findings.
- Level 1b (Moderate) = One RCT of ‘high’ quality (PEDro Scale score ≥ 6).
- Level 2a (Limited) = At least one ‘fair’ quality RCT (PEDro Scale score 4-5).
- Level 2b (Limited) = At least one well-designed non-experimental study: non-RCT, quasi-experimental studies; cohort studies with multiple baselines; single-subject series with multiple baselines.
- Level 3 (Consensus) = Agreement by an expert panel, a group of professionals in the field or a number of pre-post design studies with similar results.
- Level 4 (Conflicting) = Conflicting evidence of two or more equally designed studies.
- Level 5 (No evidence) = No well-designed studies: “Poor” quality RCTs with PEDro scores ≤ 3; only case studies/case descriptions or cohort studies/single subject series with no multiple baselines.

Results
Search results
The search identified 54 trials until July 2019. After screening titles and abstracts and removing duplicates, four studies [Dai and Demiryürek, 2017 [11]; Dursun et al., 2017 [12]; Kelly et al., 2019 [13] and Abd El-Monem et al., 2019 [14]] were included in this review. Search results are presented according to the flow chart (Figure 1).

Characteristics of the included studies
All included studies are RCTs. The summary of the included studies is presented in Table 1. The clinical homogeneity between some of the included trials allowed the quantitative analysis of their data.

Qualitative analysis
Participants
The sample size ranged from 10 to 80. There were a total of 176 children participating across the four RCTs, diagnosed with CP, including both genders and aged from 2 to 17 years.

Interventions
The study versus the control groups in four of the included RCTs received botulinum toxin type A in addition to traditional sessions of physiotherapy.

Outcome measures
In the reviewed studies, spasticity testing was performed using modified Ashworth scale (MAS) and modified Tardieu scale (MTS), passive range of motion (PROM) was assessed using...
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Table 1. Summary of the reviewed studies

<table>
<thead>
<tr>
<th>Study design</th>
<th>Participants</th>
<th>No. of participants</th>
<th>Intervention (study)</th>
</tr>
</thead>
<tbody>
<tr>
<td>RCT Level II</td>
<td>Children with CP</td>
<td>Control = 40 Study = 40</td>
<td>- BoNT-A injection and serial casting in adductor position of the lower extremities. - Both groups received physiotherapy</td>
</tr>
<tr>
<td>RCT Level III</td>
<td>Spastic equinus foot due to CP</td>
<td>Control = 17 Study = 34</td>
<td>- BoNT-Ato planter flexors and serial casting - Both groups received physiotherapy for 3 weeks</td>
</tr>
<tr>
<td>RCT Level II</td>
<td>Children with spastic CP and equinus gait</td>
<td>Control = 10 Study = 10</td>
<td>- Injections into spastic Triceps surae muscles with Serial casting - Both groups received Physiotherapy</td>
</tr>
<tr>
<td>RCT Level II</td>
<td>Children with hemiplegic CP and equinus deformity</td>
<td>Control = 12 Study = 13</td>
<td>- 3 consecutive casts applied for 5 days each and removed in the last 2 days in each week to conduct the same physical therapy program</td>
</tr>
</tbody>
</table>

Table 2. Methodology assessment of studies according to the Physiotherapy Evidence Database (PEDRO) scale

<table>
<thead>
<tr>
<th></th>
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<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>1-Specified eligibility criteria*</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
</tr>
<tr>
<td>2-Random allocation of participant</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
</tr>
<tr>
<td>3-Concealed allocation</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>4-Similar prognosis at baseline</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
</tr>
<tr>
<td>5-Blinded participant</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
</tr>
<tr>
<td>6-Blinded therapists</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>7-Blinded assessors</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>8-More than 85% follow-up for at least one key outcome</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
</tr>
<tr>
<td>9-‘Intention to treat’ analysis</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
</tr>
<tr>
<td>10-Between group statistical analysis for at least one key outcome</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
</tr>
<tr>
<td>11-Point estimates of variability for at least one key outcome</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
</tr>
<tr>
<td>12-PEDro score</td>
<td>7</td>
<td>7</td>
<td>7</td>
<td>6</td>
</tr>
<tr>
<td>Quality</td>
<td>Good</td>
<td>Good</td>
<td>Good</td>
<td>Good</td>
</tr>
</tbody>
</table>

*Item 1 does not contribute to the total score

(MTS), gross motor function level was assessed using GMFM-66 and gait abnormalities were assessed using observational gait scale (OGS).

Quality of the included studies and the level of evidence

The methodological quality of included four studies is presented in Table 2. The quality of the studies is good with a mean PEDro score (range 6 to 7). The four included studies had similar groups at baseline, analyzed the between-group difference. The four studies carried out an intention-to-treat analysis, none of the studies blinded participants.

Evidence of serial casting application interventions

The results of the 4 reviewed trials (Dai and Demiryürek, 2017, Dursun et al., 2017, Kelly et al., 2019, and Abd El-Monem et al., 2019) [11-14] which investigated the effects of serial casting application for CP children with equinus deformity are summarized in Table 3.

Statistical analysis

Only three articles (Dai and Demiryürek, 2017, Dursun et al., 2017, Kelly et al., 2019) [11-13] have homogeneity in all four components (participants, intervention, outcome (modulation of spasticity) and the outcome measures) so only one meta-analysis was performed and this meta-analysis favors the use of serial casting application for modulation of spasticity in children with spastic CP.

Description and Interpretation of Forest plot of RevMan

The Forest plot is composed of (from left to right):
1. The names of the included studies.
2. The data of the study and control groups including the mean difference between pre- and post-intervention, SD of the paired
Table 3. Summaries means of study groups and control groups and the difference between this means

<table>
<thead>
<tr>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>- Decrease spasticity</td>
<td>- Decrease spasticity</td>
<td>- Decrease of spasticity and hypo-extendibility of gastrocnemius-talus complex</td>
<td>- Decrease of spasticity and hypo-extendibility of gastrocnemius-talus complex</td>
<td></td>
</tr>
<tr>
<td>- Reduction contracture due to spasticity</td>
<td>- Increased PROM</td>
<td>- Improvement of PROM</td>
<td>- Improvement of PROM</td>
<td></td>
</tr>
<tr>
<td>- Gain additional ROM</td>
<td>- Improved gait</td>
<td>- Gross motor function improvement</td>
<td>- Improvement of PROM</td>
<td></td>
</tr>
</tbody>
</table>

Means of the control group

<table>
<thead>
<tr>
<th>Outcomes</th>
<th>Pre= 42.26</th>
<th>Post= 64.57</th>
<th>Pre= 3.81</th>
<th>Post= 2.78</th>
<th>Pre= 44.29</th>
<th>Post= 64.65</th>
</tr>
</thead>
<tbody>
<tr>
<td>- GFMM 66 mean</td>
<td>- MAS mean</td>
<td>- MTM mean</td>
<td>- OGS mean</td>
<td>- MTS mean</td>
<td>- OGS mean</td>
<td>- MTS mean</td>
</tr>
<tr>
<td>Pre= 41.18</td>
<td>Post= 77.37</td>
<td>Pre= 3.74</td>
<td>Post= 1.88</td>
<td>Pre= 42.66</td>
<td>Post= 75.95</td>
<td></td>
</tr>
</tbody>
</table>

Means of the study group

<table>
<thead>
<tr>
<th>Outcomes</th>
<th>Pre= 42.26</th>
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<tr>
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<td>Pre= 3.74</td>
<td>Post= 1.88</td>
<td>Pre= 42.66</td>
<td>Post= 75.95</td>
<td></td>
</tr>
</tbody>
</table>

Differences between means

<table>
<thead>
<tr>
<th>Outcomes</th>
<th>Control group</th>
<th>Study group</th>
</tr>
</thead>
<tbody>
<tr>
<td>- GFMM 66</td>
<td>12.51</td>
<td>12.51</td>
</tr>
<tr>
<td>- MAS</td>
<td>-1.6</td>
<td>-1.6</td>
</tr>
<tr>
<td>- MTM</td>
<td>-0.9</td>
<td>-0.9</td>
</tr>
<tr>
<td>- OGS</td>
<td>4.1</td>
<td>4.1</td>
</tr>
<tr>
<td>- MTS</td>
<td>1.52</td>
<td>1.52</td>
</tr>
</tbody>
</table>

PROM: Passive range of motion. GFMM: Gross Motor Function Measurement Scale. MAS: Modified Ashworth Scale. CHQ: Caregiver Health Questionnaire. MTS: Modified Tardieu Scale. OGS: Observational Gait Scale. PEDI: Pediatric Evaluation of Disability Inventory. MTS mean: Pre= -3.6 | Post= 12 | MAS mean: Pre= 2.6 | Post= 1.3 | GMFM 66 mean: Pre= 72.7 | Post= 76.5 | PEDI mean: Pre= 74.6 | Post= 81.2

Figure 2. Forest plot of comparison: 1 Comparison between study and control groups, outcome: 1.1 Difference in modified Ashworth scale between pre and post interventions.

Statistical methods:
We analyzed data from the included studies using Review Manager (RevMan – version 5.2, The Nordic Cochrane Center, The Cochrane Collaboration, Copenhagen, Denmark), and Microsoft Excel 2010 (Microsoft Corp., Redmond, WA, USA). A formal meta-analysis was conducted for our outcome (difference in MAS between pre- and post-intervention). The pooled MAS values were expressed as the mean difference (MD) between study and control groups, with 95% CI. We explored and quantified between-study statistical heterogeneity using the I2 test. Since I2 was 0%, we used the fixed-effect model. We considered 2-sided statistical analysis testing setting the α-error level at 0.05 (Figure 2).

Figure 2. Forest plot of comparison: 1 Comparison between study and control groups, outcome: 1.1 Difference in modified Ashworth scale between pre and post interventions.
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Discussion

The aim of the current review is to determine the efficacy of serial casting on the deformity of the equinus in children with CP. The analysis covers studies conducted between 2008 and 2019 searched via PubMed, PEDro, Cochrane library, and Google scholar databases in the Medline database that most likely contain a large number of papers published annually. The current systematic review analyzed four randomized controlled trials, applying strict inclusion selection criteria. All trials satisfied at least six PEDro-scale criteria. Three studies, including Dai and Demiryürek (2017) [11], Dursun et al. (2017) [12] and Kelly et al. (2019) [13], underwent meta-analysis. The scoring of each study of the three studies with the PEDro scale is seven after collecting data based on AACPDM sheet items. The higher the number of scores of factors measuring the study's efficiency, the greater the study's efficiency.

The research design of the three studies is randomized controlled trials with evidence level two; children involved in the three studies were spastic CP with ages ranging from 2 to 17 years. The age range in Dai et al. ‘s study (2017) [11] was 2-4 years, in Dursun et al. ‘s study, (2017) [12] was 3-17 years and in Kelly et al. ‘s study (2019) [13], was from 2-7 years.

In the study by Dai and Demiryürek (2017) [11], the intervention was a 3-week serial casting application once a week and consisted of a dual cast which consisted of a standard long leg plaster cast and a circular cast from below the knee to above the knee, while in the study by Dursun et al. (2017) [12], the intervention was three consecutive serial casting applications for three weeks once a week and consisted of a dual cast which consisted of a standard short leg plaster cast and a circular cast from below the knee to above the knee, with a follow-up period of 12 weeks. Moreover, the intervention in the study by Kelly et al. (2019) [13] was six weeks once a week and consisted of a dual cast which consisted of a standard short leg plaster cast.


In the study by Dai and Demiryürek (2017) [11], GMFM and child health questionnaire scores (CHQS) were markedly elevated in both groups following Botulinum toxin type A treatment, MAS scores decreased for 12 weeks in the Botulinum toxin type A injection with serial casting group (p<0.05), however no marked reduction in MAS scores in the Botulinum toxin type A injection only group. In the study by Dursun et al. (2017) [12], there was a significant improvement in MAS, PROM of both groups (p<0.001 for all parameters of each group). The mean MAS and PROM of the casting group were better than those of the control group at week 4 (p=0.006, p=0.013) and week 12 (p=0.015, p=0.013) with significant improvement in OGS in both groups (p=0.001 in both groups).

Regarding Kelly et al. ‘s study (2019) [13], there is a significant improvement of MTS of dorsiflexion with knee extension (p<0.001) and dorsiflexion with knee flexion (p<0.001), a significant improvement for GMFM-66 (p=0.002) and all PEDI domains except social function caregiver assistance (p=0.009-<0.001).

There was no significant difference in ankle ROM, the distribution of spasticity grades of calf muscle tone, and the total score of OGS (p=0.64, p=0.44, p=0.64) in Abd El-Monem et al. (2019) [14].

Conclusion

Although results from this analysis support the efficacy of using serial casing application for children with CP associated with equinus deformity, there is still a need for additional RCTs with larger sample sizes to validate the evidence.

Scientific Responsibility Statement

The authors declare that they are responsible for the article’s scientific content including study design, data collection, analysis and interpretation, writing, some of the main line, or all of the preparation and scientific review of the contents and approval of the final version of the article.

Animal and human rights statement

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. No animal or human studies were carried out by the authors for this article.

Funding: None

Conflict of interest

None of the authors received any type of financial support that could be considered potential conflict of interest regarding the manuscript or its submission.

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11. Dai AI, Demiryürek AT. Serial casting as an adjunct to Botulinum Toxin Type A

How to cite this article: