**TITLE: What is the evidence for upper limb splinting in children with cerebral palsy?**

**Prepared by:** Angeline Loh

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## CLINICAL SCENARIO

Cerebral palsy [CP] is a group of conditions whereby an injury to the developing brain during the foetal, perinatal or early infancy stages causes non-progressive but lifelong problems of movement, muscle tone and coordination (Yamamoto, 2007). Hand splints help to maintain a functional position, prevent deformity and increase use of the affected hand (Dodd, Imms & Taylor, 2010). In New Zealand, paediatric occupational therapists often prescribe hand splints to hemiplegic children both in preparation for school and during their school years to increase functional hand use in common bilateral occupations such as drawing, writing and using scissors. In this CAT, the evidence for the effectiveness of hand splints in these children will be located and appraised.

## FOCUSSED CLINICAL QUESTION

In children aged 0-18 with CP, do upper limb [UL] splints improve hand function?

## SUMMARY OF SEARCH, 'BEST EVIDENCE' APPRAISED, AND KEY FINDINGS

A search was conducted on four databases chosen for being relevant to occupational therapy and having studies of high quality. Studies were initially shortlisted based on title and/or abstract. Articles were located online or in the library for comparison against inclusion and exclusion criteria. Five articles with acceptable quality were found. The systematic review/meta-analysis found little to some evidence supporting casting combined with neurodevelopmental therapy [NDT]. Another systematic review (Boyd et al, 2001) found some evidence for occupational therapy and serial casting. A randomised controlled trial [RCT] found strong evidence for static night casting following botulinum A toxin [botox] injections (Sakzewski et al, 2009). A trial using randomised crossover design with washout (Law et al, 1997) found no evidence for intensive NDT plus casting over regular occupational therapy [OT]. Kinghorn and Roberts (1996) found inconclusive evidence for a weight-bearing splint on a single child's tone and function.

**CLINICAL BOTTOM LINE**

Based on the systematic review by Sakzewski et al (2009) and the quantitative study by Kanellopoulos et al (2009), there is some evidence that upper limb splints improve hand function when combined with NDT or botox.

**LIMITATION OF THIS CAT**

This critical appraisal has been peer reviewed by one lecturer as part of an assignment.

**SEARCH STRATEGY**

The search strategy was developed to locate any study that focussed on children from 0-18 years old with CP and use of splints.

Terms used to guide search strategy were:
Patient: child* AND cerebral palsy
Intervention: (splint* OR cast* OR orthos*) AND ("upper limb" OR "upper extremity" OR hand)
Outcome: hand function OR hand use

| Databases searched | Search Terms | Limits used |
|---|---|---|
| Cochrane Library | "cerebral palsy" AND (splint* OR cast* OR orthos*) | None |
| OT Seeker | splint* OR cast* OR orthos* | Diagnosis/ Subdiscipline: Cerebral palsy |
| CINAHL | "cerebral palsy" AND splint* OR cast* OR orthos* | None |
| PubMed | "cerebral palsy" AND (splint* OR cast OR casting OR orthos*) | None |

**INCLUSION CRITERIA**

- Diagnosis of CP
- Children aged 0-18
- Splinting, casting or orthoses were used as an intervention for UL/hand

- Research studies or systematic reviews

**EXCLUSION CRITERIA**

- Studies not published in English
- Not accessible electronically or in hard copy through University of Otago Library networks

**RESULTS OF THE SEARCH**

Five studies were located and ranked (Polit & Beck, 2008, p.31) as shown in Table 1.

**Table 1:** Summary of Study Designs of Articles retrieved

| *Study Design/ Methodology of Articles Retrieved* | *Level* | *First Author (Year)* |
|---|---|---|
| Systematic review and meta-analysis | Ia | Sakzewski (2009) |
| Systematic review | Ib | Boyd (2001) |
| Randomised controlled trial | IIa | Kanellopoulos (2009) |
| Randomised crossover design with washout period | IIa | Law (1997) |
| Single-case study | IIb | Kinghorn (1996) |

**BEST EVIDENCE**

The first article is a systematic review/meta-analysis by Sakzewski et al (2009). This was the most rigourous systematic review because it contained a meta-analysis and only included studies of high quality (systematic reviews and RCTs with good methodological quality). This study was directly relevant to the population of interest and focussed on the UL.

The second article was a RCT by Kanellopoulos et al (2009) that was directly relevant to the population and intervention of interest and was not included in the chosen systematic review.

**SUMMARY OF BEST EVIDENCE**

Sakzewski, L., Ziviani, J. & Boyd, R. (2009). Systematic review and meta-analysis of therapeutic management of upper-limb dysfunction in children with congenital hemiplegia. *Paediatrics, 123*(6), e111-e1122. DOI: 10.1542/peds.2008-3335.

**Aim/Objective of the Systematic Review**

To systematically review the efficacy of nonsurgical UL therapeutic interventions for children with congenital hemiplegia.

**Study Design**

Systematic review and meta-analysis

**Search Strategy**

Comprehensive searches using computerised bibliographic databases: Medline (1950-July 2008), CINAHL (1982-July 2008), Embase (1988-July 2008), Cochrane Central Register of Controlled Trials (Issue 3, 2008), AMED (1985-July 2008), PsychINFO (1967-July 2008), and Web of Science (1945-July 2008).

The search used exploded MeSH terms and text words:
1) “cerebral palsy” OR “hemiplegia”
2) AND “child” OR “adolescent” OR “infant”
3) AND “physical therapy” OR “occupational therapy” OR “movement therapy” OR “neurodevelopmental therapy” OR “splinting” OR “casting” OR “botulinum toxin” OR “functional electrical stimulation” OR “strength training” OR “conductive education” OR “virtual reality” OR “constraint induced movement therapy”
4) AND “upper limb” OR “arm” OR “hand”
5) AND “randomized, controlled trial” OR “ramdom sampling” OR “double blind method” OR “single blind method” OR “placebo” OR “systematic review”

**Study Selection Criteria**

Inclusion:
*Trial Design*: Systematic reviews with/without meta-analysis and randomized or quasi-randomized, controlled trials
*Participants*: Children aged 0-18 years with congenital hemiplegia with spasticity affecting UL function
*Interventions*: Nonsurgical, to improve UL function and participation, including OT, physiotherapy, NDT, movement training, motor learning, conductive education,

constraint induced movement therapy [CIMT], strength training, virtual-reality training AND/OR pharmocologic agents including botulinum toxin. 50% of intervention content had to focus on UL.
*Outcomes*: UL activity and/or participation outcomes

Exclusion:
*Trial design*: Non-randomized, no control group
*Participants*: No subset of children with hemiplegia
*Outcomes reported*: Impairment only
*Language*: Studies not published in English

Eligibility for inclusion/exclusion was independently assessed by Sakzewski and Boyd. Sakzewski reviewed initial search results based on title/abstract and both assessors reviewed full text articles meeting inclusion criteria.

**Method**
*Trials*: Methodological quality was independently evaluated with PEDro[1] scale by Sakzewski and Boyd. Those with ≥5 (out of 11) had adequate internal validity and were included in meta-analysis.
*Systematic reviews*: Methodological quality was assessed with AMSTAR[2].

**Results**
Search strategy yielded 177 references. 20 RCTs and 8 systematic reviews met inclusion criteria and were assessed independently by Sakzewski and Boyd.
References of excluded studies and reasons for exclusion were provided:
7 trials were excluded for: not hemiplegia (n=2), not RCT (n=1), no control (n=3), impairment outcome (n=1) and two articles reported the same trial.
1 systematic review was excluded because the focus on UL was <50%.

Participants
7 trials targeted 222 children with congenital hemiplegia.
5 trials used broader sample of 176 children with CP with subset with hemiplegia.
1 study had predominantly children with hemiplegia.
Age range across trials: 18 months to 16 years

[1] PEDro– Physiotherapy Evidence Database
[2] AMSTAR – Assessment of Multiple Systematic Reviews

Types of interventions

Within the included studies which studied a range of nonsurgical UL interventions, two RCTs specifically pertained to UL casting.

Outcome measures

Standardised and non-standardised assessments were used, assessing UL function, self-care and individualised outcomes. No outcomes measuring participation were reported.

Primary outcomes

*UL Function:* Children who received NDT and casting (n=88) compared to control group who received regular OT or regular NDT (n=84) scored SMD (standardised mean difference) of 0.4 (95% CI: 0.10 – 0.71, p=.009) on QUEST[3]. Same groups compared on PFMS[4] achieved SMD of 0.15 (95% CI: -0.15 – 0.45, p=.34).

**Original Authors' Conclusions**

Meta-analysis of casting plus intensive NDT showed medium treatment effects for UL movement quality, measured by QUEST and small treatment effects for acquisition of motor skills (measured by PFMS). QUEST is primarily a measure of impairment with a few items that measure activity performance. Improvement in these results may indicate gains in range of motion and biomechanical alignment due to casting. Results suggest that older children may have a greater response to casting and intensive NDT.

**CRITICAL APPRAISAL**

**Validity**

The review had a clear focus. The search strategy was comprehensive and synonyms were used to ensure studies were not missed. There was no mention of searching reference lists of relevant studies. The authors only wanted to include high level evidence such as systematic reviews and RCTs. They used the PEDro scale to assess methodological quality of trials and only included those with a score of ≥5 for meta-analysis. The systematic reviews were assessed with AMSTAR and all rated ≥6 (out of 11). The face and content validity of AMSTAR was stated as good but the validity of PEDro was not mentioned, although the interrater reliability was stated as adequate. This was important as two authors independently rated the trials using

[3] QUEST - Quality of Upper Extremity Skills Test
[4] PFMS - Peabody Fine Motor Scales

PEDro. There was clear inclusion and exclusion criteria and when articles were excluded specific reasons were given. Only Sakzewski was involved in the initial selection of studies that fit the inclusion criteria, so bias may have been introduced then. Data analysis methods were identified and a fixed-effect model was used to pool treatment effects when trials had similar outcome measures and populations. This method helped strengthen the results of the two trials which specifically looked at NDT and casting.

**Interpretation of Results**

Two studies were relevant to UL casting in children with CP. One was an RCT that compared intensive/regular NDT with casting against controls of intensive/regular NDT with no casting. The second was a crossover design study comparing regular NDT with casting against controls of regular OT. Meta-analysis of these showed small to moderate treatment effects. However the outcomes measures described did not include functional hand use in school-related occupations. There two studies differed in age groups and the older age-group trial was reported to have experienced a greater response. One systematic review was reported to have clinical inference of small effect for OT and casting but this was not synthesised with any other trial focussing on casting. There was no mention of statistical significance of any of the trials.

Overall, there was not a lot of information about casting specifically.

**Conclusion: Relevance to Clinical Question of this CAT**

After meta-analysis, the trials that were relevant to the clinical question showed small (statistically insignificant, p=.34) to medium (statistically significant, p=.009) treatment effects. However because this systematic review focussed on all nonsurgical UL interventions in children with CP, there was relatively little information. A systematic review of studies that only focussed on UL splinting, casting or orthoses may provide information that is more relevant to this clinical question.

Kanellopoulos, A. D., Mavrogenis, A. F., Mitsiokapa, E. A., Panagopoulos, D., Skouteli, H., Vrettos, S. G., Tzanos, G. & Papagelopoulos, P. J. (2009). Long lasting benefits following the combination of static night upper extremity splinting with botulinum toxin A injections in cerebral palsy children. *European Journal of Physical Rehabilitation Medicine, 45,* 501-6.

**Aim/Objective of Study**

To evaluate the necessity and effectiveness of a static night splint following outpatient botulinum toxin A [botox] treatment in children with UL spastic cerebral palsy [CP].

**Study Design**

Randomised controlled trial

**Setting**

Department of Pediatric Orthopedics of Athens University School of Medicine

**Participants**

Sample comprised 20 spastic CP hemiplegic children with upper extremity involvement. The mean age was 7 years (range 2.5-12 years). There were 13 boys and 7 girls. All children were under the care of the senior authors.
Inclusion criteria included having a UL primary contracture that limited reaching, grasping, releasing and manipulation and affected the appearance of the limb. Exclusion criteria included neuromuscular disorders, previous neurosurgical procedures or intrathecal baclofen.

**Method**

The required dose of botox was calculated (formula given) and injected into all children. Injection sites and method of location were given. Injections were administered at ≥2 UL muscles. Following injection, children were randomly allocated into two groups – group A (n=10) received a thermoplastic night splint for 6 months; group B (n=10) received no further treatment. It was not mentioned who administered the injections. The splints were described as “easy to modify” but exactly who modified them to fit the children was unclear. Pictures and a general description of the splint were provided. Following treatment, children were discharged from hospital and families were given instructions for ≥3 sessions of occupational therapy per week and participation in a mobility programme. Measurements were taken at baseline, at 2 and 6 months after injection using QUEST. A reference was given for QUEST and

it was described as standardised, valid and reliable for upper extremity function in four domains (listed) and that it "may assess changes following treatment" (p.502). The statistical analysis method was identified (one-way ANOVA test).

**Results**

The similarity of the groups at baseline was not mentioned. After injection, all children showed improvement in muscle tone, range of motion and motor function as measured by QUEST. At 2 months, group A showed 15.4% improvement over baseline scores while group B showed 12.2% improvement – the difference between groups (measured by F ratio[5]) was not statistically significant. At 6 months the treatment group had 15.9% improvement of UL function over baseline compared to 4.2% improvement in the control. The difference between groups, measured by F ratio, was statistically significant (p=0.000).

**Original Authors' Conclusions**

This study showed a "definite treatment effect" (p.505) for the combination of botox and static night splinting to decrease spasticity and increase function for the target population. The night splints were well-tolerated and combined with occupational therapy can help to improve function and maintain improvements from botox treatment for ≥6 months.

**CRITICAL APPRAISAL**

**Validity**

This study had a clear focus which was addressed by the method. While the sampling process was more purposive than random, the allocation process to splint or no splint was randomised. The population of interest was a very specific group and hence while the sampling was not random, it could be seen as being representative of the population, ie children with CP, hemiplegia and UL involvement, though the limitations must be considered, for example this may only be generalised to other children fitting this criteria within this particular hospital or geographic location. There were no dropouts reported. The literature review was relevant and appropriate, undertaken both in the introduction and within the discussion. The literature reviewed at the start described spasticity as the most common condition in CP and described deformities arising from spasticity as well as previous studies that used botox and casting for spasticity. The authors justified the need for this study, stating it would

[5] Ratio of *between group mean square* with *within group mean square.*

contribute knowledge regarding the use of casting and botox for children specifically with UL involvement. It was unclear who administered the injections and who fit the splints on group A. As the children were known to the senior authors, bias could have been introduced if they were the administrators of treatment; administrator-blinding was not mentioned. It would be difficult to blind the participants as they either received a splint or not and no other placebo was offered. No mention about how similar the groups were at the start of the trial was made either. This is an important consideration when weighing up the treatment effect as differences between the groups could account for part or all of the difference measured after treatment. Ethical approval was not mentioned but the parents gave "written informed consent" (p.502). No details were given about what the parents were informed about.

**Interpretation of Results**

The results of the study showed a 6-month difference in improvement over baseline between the groups to a statistically significant level (p=0.000). However, there was insufficient information about the homogeneity of groups at the start. Other than diagnosis of CP hemiplegia and age, no other demographic information about participants is included. The mean age of both groups was identical, but whether the spread and variance of ages was statistically significant was not addressed (group A ranged from 2.5-12 years while group B ranged from 5-11 years). Although CP is non-progressive, the age of the child will affect how much prior treatment and therapy they may already have received and their developmental stage. Minimisation of bias is not mentioned and the sampling method may have introduced bias, while the small sample size may limit generalisation.

**Conclusion: Relevance to Clinical Question of this CAT**

The clinical question of this CAT is related to UL splinting in children with CP and functional outcomes. This study showed that one type of splinting (static, night, thermoplastic) following botox injections can produce statistically significant results at 6-month follow-up. While this information does not contribute directly to the clinical question, it informs readers that splinting is an effective adjunct to botox in this population. Function was described as "improved" but no details were provided. The fact that children who wore splints improved more than those without cannot directly isolate the effectiveness of splints in the absence of botox. Hence this study is only relevant to children who receive botox treatment for UL spasticity. As botox is not part of OT treatment this supports multidisciplinary treatment of CP.

## IMPLICATIONS FOR PRACTICE, EDUCATION AND FUTURE RESEARCH

The search for evidence for the clinical question of this CAT led to a mix of articles of various types, designs and quality. The five articles identified for this CAT were a good representation of the variety – from a systematic review/meta-analysis of systematic reviews and RCTs to single case studies. Generally there was limited research on splinting/casting as the main intervention for the upper limb.

Based on the two articles appraised, there is some evidence for casting combined with NDT and strong evidence for static night splinting following botox. The clinical implication is that a multidisciplinary combination of treatment and therapy modalities may be the best approach to children with cerebral palsy. However, there is insufficient focus on school-related occupational outcomes in the current literature and more research is needed that specifically looks at these as compared to simply measuring performance components, which cannot be assumed to translate into functional gains.

For research to be useful in supporting or changing clinical practice, not only must the study methodology be rigourous, but the research question should also be directly relevant to the area of interest. The cost of implementing any change and the risk of unpredictable effects on clients need to be outweighed by sound, relevant research. These two studies were of good quality, but research more specifically related to the question of UL splinting would be preferable to justify any major changes in this area of practice.

These two articles could be of use to the occupational therapist whose client is from the target population and is likely to receive NDT or botox. In these cases the therapist might consider what benefit the child might receive from casting/splinting. There is no guarantee that any particular child with CP will respond to any treatment as others in previous studies have done, so occupational therapists are best to adhere to client-centred principles and collaborate with their clients to discover what is best for them individually, using their professional expertise to weigh up the research evidence. If over time a therapist finds that a great proportion of their clients in a certain population respond well to a particular treatment they could establish the empirical evidence to support the anecdotal if they saw a need for justifying their practice or recommending changes to practice guidelines.

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