Upper limb rehabilitation in children with unilateral cerebral palsy

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Abstract

Children with unilateral cerebral palsy experience difficulties with unimanual and bimanual functions, impeding self-care independence in daily life. Hence, research aimed at improving upper limb function has increased tremendously throughout the last decade. In this manuscript, we will provide an overview of all evidence-based, non-surgical therapy models intended to improve upper limb function in children with unilateral cerebral palsy, described according to the International Classification of Functioning, Disability and Health framework. The strongest level of evidence refers to activity-based interventions like constraint-induced movement therapy, bimanual training and goal-directed training. Interventions targeting body structures or functions, such as muscle strengthening, taping, splinting or casting, are less well investigated and should at this point be considered as assistive interventions with therapy goals on the level of body structure and function. Limited evidence exists on the efficacy of participation-based interventions. Finally, environmental factors can further shape the therapy model by providing the therapy in the home setting or a camp environment, while personal factors may influence the response to treatment.

Introduction

The upper limb plays a crucial role in acquiring self-care independence in daily life enabling participation in home, school and leisure activities. The performance of such activities requires the skilled use of both hands together. Throughout daily life, the variety of upper limb functions is tremendous, with reaching and grasping being the first developmental milestone. Already during the first year of life, reaching and grasping develops into a skilful movement. As a matured motor activity, reaching and grasping looks fairly easy, yet it involves a complex neural action to control and coordinate the numerous degrees of freedom. Hence, a lesion during early brain development, such as in cerebral palsy (CP), may disrupt the fine-tuned coordination of upper limb movements, compromising the performance of daily life activities.

CP is a major cause of paediatric disability and is defined as "*a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing foetal or infant brain. The motor disorders of CP are often accompanied by disturbances of sensation, perception, cognition, communication and behaviour, by epilepsy, and by secondary musculoskeletal problems*" (1). The upper limb is most often investigated in children with spastic unilateral CP (uCP), in whom sensorimotor impairments are predominantly present at one side of the body. Studies have shown that particularly distal muscle weakness and somatosensory dysfunction negatively affects upper limb activity and self-care independence (2,3). This may cause the child to end up in a vicious circle. Increased muscle weakness and somatosensory dysfunction can contribute to a reduced hand use in daily life, further preventing the spontaneous daily stimulation of muscle strength and somatosensory input (2,3). Moreover, a recent five-year longitudinal study reported that from the age of nine years onwards, the spontaneous use of the impaired side in bimanual tasks decreases (4). Together these findings underline the importance of motor interventions to improve and retain function, even before the age of one year.

In the end, the ultimate goal of each clinician is to enhance the child's functional potential for which adequate treatment planning is imperative. Hence, in this manuscript, we will provide an overview of all evidence-based therapy models aimed at improving upper limb function in children with CP, described according to the ICF-framework (International Classification of Functioning, Disability and Health) (5). We will focus on the non-surgical interventions, which are recommended by Novak et al. in their traffic lights paper with a green light referring to strong positive evidence and an orange light to weak positive evidence (see Figure 1) (5). Where applicable, personal recommendations based on clinical experience and as performed at the University Hospitals Leuven, were added.

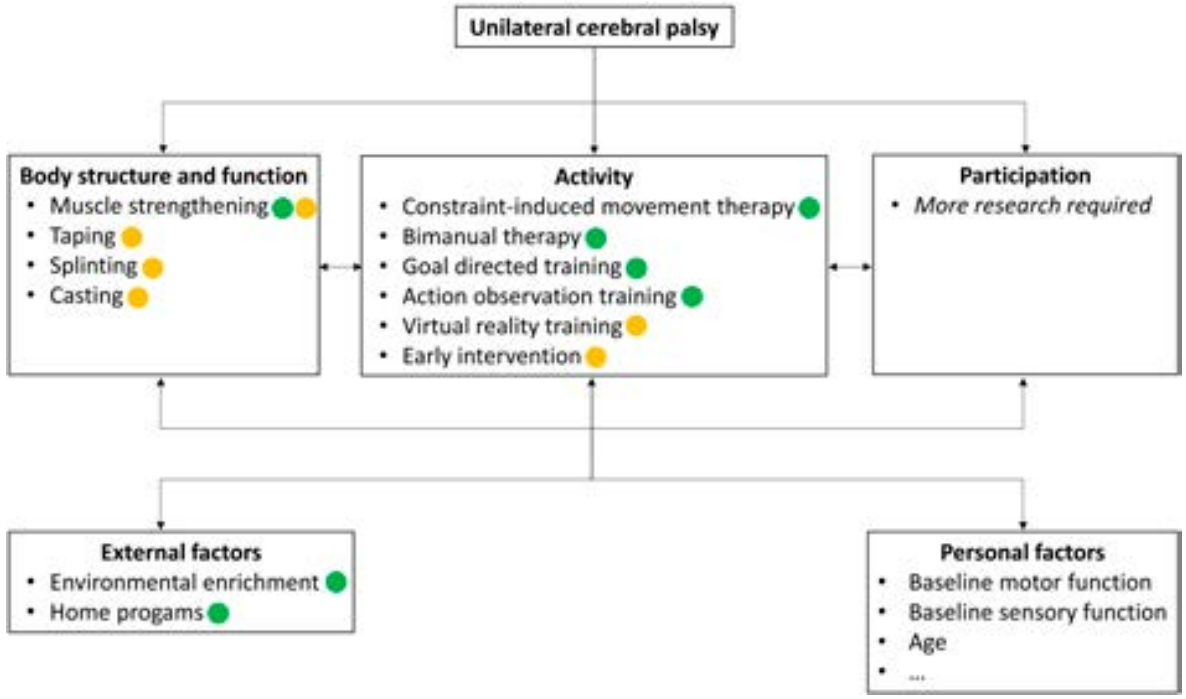
Interventions at the body structure and function level

It is recommended to perform muscle strengthening, taping, splinting and casting as an assistive intervention combined with task-specific motor training (5).

For muscle strengthening, there is strong evidence that it is effective in improving muscle strength (green light) (5). Moreover, practicing strength during functional tasks may result in a better transfer towards manual activities compared to practicing strength in non-functional positions (orange light) (6). The National Strength and Conditioning Association guidelines recommends for a muscle strengthening program in children, a duration of 12 weeks with a frequency of three times a week at 8–12 repetition maximum (6).

Recently, taping gains in popularity as an assistive intervention due to its low costs and easy application (7). There are two main kinds of tape. Kinesio tape, a flexible/elastic type of tape and the athletic tape which is a more rigid/inelastic tape that is more effective in limiting joint movement (7). In children with CP, taping is often used in the management of spasticity/hypotonia, facilitation of muscle function and joint stabilization (7). In combination with a task-specific motor training, taping may augment the treatment effects (orange light) (5). Though specifically for the upper limb, immediate effects are usually not visible, indicating that the taping needs to be applied long

Figure 1. Overview of interventions recommended by Novak et al.5 according to the ICF-model.



enough (7). Moreover, taping has additional benefits with respect to comfort and cosmesis compared to traditional orthotics (5).

Hand splinting is considered as standard practice in children with uCP. Though, less than 10 randomized controlled trials have been published on the efficacy of hand splinting. According to a meta-analysis, the use of non-functional hand splinting (i.e. splints worn at night) has a small beneficial effect on upper limb function in combination with therapy compared to therapy alone (orange light) (8). However, these benefits were diminished already two to three months after splint wearing was stopped. Moreover, the combined therapy that was used across these studies strongly differed (i.e. botulinum injections, neurodevelopmental therapy, goal-directed training), which makes it hard to draw clear conclusions. Only two randomized controlled trials investigated the efficacy of functional hand splinting (i.e. worn during the performance of functional tasks), but with conflicting results (9,10). According to our point of view, the main benefit of functional hand splinting is the stabilization of the wrist and carpometacarpal joint of the thumb in a functional position which facilitates a digital grip. Though for children with intact somatosensory function, the main disadvantage of hand splinting is that it restricts tactile input.

Upper limb casting may aid in the prevention of contractures (orange light) (5). In combination with botulinum toxin injections, the effects of casting can be enhanced (green light), which is also better tolerated by the children than applying casting without botulinum toxin injections (5). However, careful consideration is needed because casting can cause altered proprioception and secondary muscle weakness in particular with botulinum toxin injections (5). Hence, when the cast is removed, active and/or goal-directed motor interventions are crucial to regain these functions (5). Severe contractures cannot be treated with casting alone, and usually require orthopaedic surgery (5).

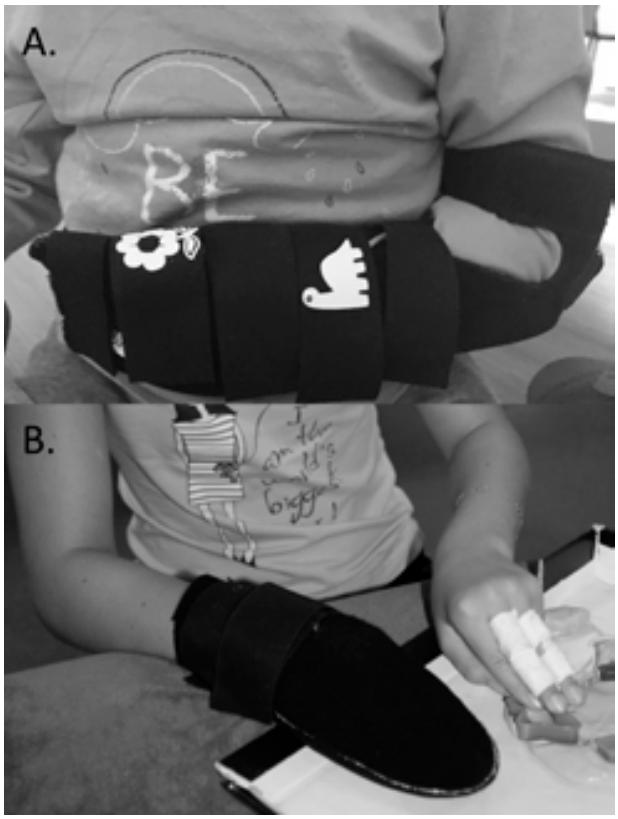
Interventions at the activity level

The strongest level of evidence for efficacious therapy models for the upper limb is situated on the level of activity (5). Novak et al. recommend to perform constraint-induced movement therapy, bimanual therapy, goal directed training, action observation training and virtual reality training (5). All these interventions are based on the theory of motor learning. Important motor learning principles are problem-solving, repetitive practice and structured feedback. Real-life tasks are practiced through self-generated movements at a high intensity and within a motivational setting, often aimed at achieving goals set by the child (or parent if necessary) (5). The most popular evidence-based therapy models are constraint-induced movement therapy (CIMT) and bimanual therapy.

The application of CIMT in children with CP was derived from promising results in stroke rehabilitation. CIMT involves restraining the less-impaired upper limb allowing motor stimulation of the impaired upper limb. However, the original model requested wearing the constraint during 90% of waking hours for at least two weeks, while providing upper limb therapy for at least three hours per day. In children, usually a more child-friendly approach is used (i.e. modified CIMT), which includes variation in type of restraint, the hours worn per day and duration in weeks (11). In literature, frequency varies from one hour per day to 24 hours per day, with a duration of two to ten weeks (11). Forced-use therapy is the restraint of the less-impaired upper limb without additional treatment for the impaired upper limb. The restraint can be a splint, a cast, a sling or a glove. In the University Hospitals Leuven, a sling or glove are used (see Figure 2), which have the advantage to be removed, limiting frustration outside the structured therapy moments. In general, we advise a sling in children younger than four years of age and in children with less functional abilities (i.e. children without an active grasp ability). A sling keeps the arm against the trunk preventing the child to use its less-impaired upper limb in any way for task execution. Working with a sling is preferably done in a sitting position since it can have a negative impact on balance and prevents a protective reaction in case of falling. A glove is used in children older than four-years of age and/or in children with better functional abilities. Nevertheless, this is not a strict guideline. It remains important to look at the individual child together with the parents and the therapist to consider whether a sling or a glove would be the best option. For example, in a six-year old child, also having ADHD, a sling might offer better results.

So far, CIMT is the most studied therapy model for the upper limb in children with uCP, in which strong evidence point towards clinical meaningful and sustained benefits (green light) (5,11). (Modified) CIMT can counteract the process of developmental disregard of the impaired upper limb (11). Children with uCP may disregard their preserved capacity as they may learn from an early age that using only the less-impaired upper limb or applying other strategies might be easier to complete daily life tasks. This may cause the child to end up in a vicious circle, which then can be interrupted by applying

Figure 2. Example of a sling (A) and a glove (B).



CIMT principles. The main disadvantage is that only unimanual functions can be practiced with CIMT, while in daily life most activities are bimanual.

Subsequently, intensive bimanual therapy models arose, often referred to as Hand-Arm Bimanual Intensive Therapy (HABIT) or Bimanual Intensive Therapy (BIT). These therapy models retained the intensive structured practice but instead of using a restraint they encourage the use of the impaired upper limb while performing bimanual activities. Although the amount of evidence is less abundant compared to CIMT, strong evidence points towards the efficacy of bimanual therapy to improve upper limb function (green light) (5). As such it is highly recommended to imply bimanual therapy models within upper limb therapy. Another therapy model added a lower limb component during HABIT (i.e. HABIT-ILE). This did not seem to attenuate the improvements of the upper limb (orange light) (5,12).

According to one systematic review, it could not be concluded whether CIMT or HABIT is superior (14). Though another systematic review acknowledged the task specificity of training, suggesting that CIMT results in better unimanual improvements, while bimanual training leads to better bimanual function (15). Hence, we often recommend a hybrid therapy model combining CIMT with bimanual training.

Both modified CIMT and bimanual therapy can also be applied in infancy. Such early intervention models are hypothesized to result in better motor outcomes compared to the same treatment at a later age due to the increased neuroplasticity of the young infant brain (13). Currently, there is mounting evidence pointing towards the efficacy of early interventions for improving upper limb function (orange light), including the absence of adverse events such as a negative impact of the restraint on the function of the dominant hand or on the child's gross motor development (5).

Goal-directed training addresses goals that children and their parents identified as important in their daily life. The training process consists of goal selection, task analysis, intervention and evaluation (16). This individualized approach has been shown to be efficacious for improving hand function in children with uCP with a range of physical and cognitive abilities (green light) (5,16).

In virtual reality training, video games are used to improve upper limb function. However, virtual reality should be considered as an assistive intervention, that when combined with task-specific motor training may enhance the positive effects (orange light) (5). Video games have the opportunity to create repetition, gradation and feedback in the game. The main advantage of virtual reality is the positive impact on the child's motivation. Moreover, video games could easily be implemented in the home setting to maintain rehabilitation benefits (17).

Finally, action observation training (AOT) is a novel approach aimed at stimulating the mirror neuron system. It is based on the principle that during the observation of a motor task, the same neurons are active as during the actual performance of that task (18). Hence, within AOT the child first observes a meaningful action repeatedly followed by the actual execution of the same task. AOT can include both unimanual as bimanual tasks, as long as the task itself is goal-directed. Although this is a fairly new method, there is strong evidence that AOT is effective for improving upper limb function (green light) (5).

Interventions at the participation level

Most interventions hope to further result in an improvement on participation level, yet there is only limited evidence available on the effect on participation outcomes. A participation-based intervention must address all personal, contextual and environmental factors. To the best of our knowledge, there is one intervention study that was specifically designed to target participation (19). This randomized controlled trial resulted in an increased perceived performance of leisure-time physical activity goals in children with CP. However, the actual level of physical activity only seemed to increase in children who already did not meet the physical activity guidelines (19). Although more research on this area is needed, adequate communication among the different stakeholders seems crucial in order to reduce barriers for participating in home, school and leisure activities.

Environmental factors

Environmental enrichment is a way to stimulate upper limb function already very early in infancy. The goal is to set up a motor enriched play environment at home to encourage self-initiated movements, exploration and task success. Hence, a variety of objects to stimulate grasp and reach behaviours is necessary. Toys must be selected carefully to match the desired motor task taking into account the motor and cognitive level of the child. Strong evidence reports that environmental enrichment improves upper limb motor function (green light) (5).

Moreover, environmental factors can further define the therapy model. The therapy can be provided in a private practice, at home, in a rehabilitation centre or in a camp environment.

From a family-centred perspective, the home environment is an interesting and natural place to stimulate the child in using his/her impaired upper limb with the main advantage of generalizing skills in daily life. However, also in this case, the therapist plays a crucial role in achieving success by guiding and supporting the parents to enhance their competency. Parents, grandparents and even caregivers in the nursery can be coached on how they can implement the therapy concept in the daily life of the child. Regular contact with the guiding therapist is crucial for support and motivation, while a manual with tips and tricks can provide additional guidance. Almost all therapy concepts can be implemented in a home programme. Home-programs are recommended for improving upper limb function (green light), and have also been shown to be feasible to perform (5,20).

Another environment for therapy delivery can be the rehabilitation unit of an hospital, where the child can receive ambulant therapy or can even be hospitalized for a short period of time (e.g. 2-3 weeks as performed in the University Hospitals Leuven) (11). A hospitalisation allows a boost of intensive rehabilitation, also providing multidisciplinary therapy and guidance.

Finally, camps have the advantage of being a group intervention usually including children of similar ages and motor difficulties. Hence, it has a more recreative feeling as well as an important social impact. These camps have shown its efficacy in research and, seeing its overall benefits, have been clinically implemented in Belgium (11).

Personal factors

Despite the proven effectiveness of activity-based therapy models, it has been reported that some children might benefit more than others (21). There is vast amount of variability in clinical characteristics amongst children with uCP that might contribute to the individualised treatment response. Factors like age, baseline motor function, brain lesions characteristics, cognitive ability, motivation… differ from child to child. In literature, there is most evidence that children with lower baseline function profit more from intensive therapy models like modified CIMT and bimanual training, and that modified CIMT seems more beneficial for children with a poor somatosensory function (22,23). With respect to age, literature tends more towards a non-relevant impact, suggesting that children from all ages benefit from intensive activity-based therapy models (22,23). However, from eight years onwards, the focus should be much more on goal directed therapy (22). In addition, treatment response seems independent of underlying brain lesion characteristics (23,24).

Conclusion

The strongest level of evidence to improve motor function of the upper limb is at the level of activity involving CIMT, bimanual therapy, goal-directed training and AOT. It has been suggested that it does not matter 'what' is done, but that the 'intensity' of the therapy model is more important as well as that repeating intensive therapy periods might be needed to maintain gains in function (21). Existing evidence has proposed following key ingredients of activity-based interventions: collaborative goal setting, whole-task practice (or part-task practice for building skills for whole-task practice), context (practice within real-life environments), increasingly challenging tasks, feedback, motivation and sufficient dosage (25). A minimum of 40 hours of practice is recommended to improve motor ability, while for improving individual goals, 14 to 25 hours of therapy might already be sufficient (25,26). Moreover, the type of the therapy model must be chosen and individualized based on the child itself (i.e. age, cognitive abilities, level of motor function, behavioural problems,…), whereby it is important to include potential goals the child and/or its parents might have.

The authors have no conflict of interest to declare.

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