Physical therapy intensive mobility training recommendations for children and young adults that have undergone childhood cerebral hemispherectomy

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1. Background Information

Cerebral hemispherectomy is a complete surgical resection and/or disconnection of one hemisphere performed to stop life-threatening drug-resistant seizures (Cook et al., 2004). After surgery 50% to 80% of the individuals post cerebral hemispherectomy are seizure free (Devlin et al., 2003; Hemb et al., 2010). This procedure represents about 16-20% of all pediatric epilepsy surgeries (Harvey, Cross, Shinnar, & Mathern, 2007). With more national centers performing this dramatic procedure, and performing it at younger age (< 1 year), the likelihood of treating a child after hemispherectomy for health professionals is increasing. Children after cerebral hemispherectomy present with chronic hemiparesis and functional deficits resulting from the complete loss of innervation by a corticospinal tract that originated in the resected hemisphere. Clinical presentation resembles individuals post stroke, as well as children with spastic hemiplegic cerebral palsy. Preliminary evidence suggests that rehabilitative techniques existing for individuals with stroke and cerebral palsy may be efficient in children and young adults after cerebral hemispherectomy (Fritz, Merlo, et al., 2011; Fritz, Rivers, Merlo, Mathern, & de Bode, 2011)

The objective of the following review is to guide physical therapists in the application of an evidence based program in Intensive Mobility Training (IMT), which has been utilized for a series of patients with neurological deficits. The World Health’s Organization’s International Classification of Functioning, Disability, and Health (ICF) is an internationally recognized interdisciplinary framework, which will be utilized below to assist in describing health and health-related conditions as well as suggested outcome measures and treatment (Organization, 2007). We will also briefly describe further research needed to better understand implications of chronic disability occurring early in life.

2. Body Functions, Activities and Participation

The ICF provides practitioners a guide to identify primary factors (“body functions and structures”) impacting an individual’s performance within activities and their participation in various contexts. Impairments resulting from neurological pathologies such as stroke, cerebral palsy and cerebral hemispherectomy include muscle weakness, incoordination, poor endurance, pain, spasticity and poor balance leading to persistent difficulties with walking (Eng & Tang, 2007). Figure 1 below is an example of the ICF application in a case study of a 21-year old girl after left cerebral hemispherectomy. Improved walking ability is one of the important priorities for this ambulatory young woman similar to many patients after cerebral hemispherectomy. It is even more critical for a minority of this population without independent ambulation. Research suggests that children who are able to walk are more successful in social roles as well as activities of daily living (ADL) than children who use a wheelchair (Le Page et al 1998). Thus, gait training interventions have potential to improve walking ability across all the 3 levels of functioning (Body Functions and Structures, Activities and Participation).
Figure 1 Utilization of the ICF for a 21 year old woman after cerebral hemispherectomy at 5 years of age.
3. Gait Training

Many of the deficits in individuals with cerebral hemispherectomy may be irreversible because their primary sensorimotor cortex has been unilaterally resected during hemispherectomy. Although poorly understood, it is clear, however, that partial recovery of motor functioning after hemispherectomy is possible as a result of major brain reorganization utilizing brain plasticity mechanisms (Bates & Zadai, 2003). The mechanism and specific deficits associated with the complete removal of one hemisphere are now being investigated (J. T. Choi, Vining, Reisman, & Bastian, 2009; J.T. Choi, Vining, Mori, & Bastian, 2010). With few animal models of hemispherectomy existing (Burke, Zangenehpour, & Ptito, 2010) we rely upon the models of stroke and assume that recovery of some functions would be achieved utilizing the ability of one part of the brain to support the function of the damaged part (Slavin, Laurence, & Stein, 1988). In cases of cerebral hemispherectomy, neural reorganization must rely upon the sensorimotor cortex of the remaining hemisphere and its ipsilateral connections, as well as subcortical mechanisms, to support motor functioning of a hemiparetic side (Benecke, Meyer, & Freund, 1991; Bernasconi, Bernasconi, & Lassonde, 2000; Bittar, Ptito, & Reutens, 2000; Dijkerman, Vargha-Khadem, Polkey, & Weiskrantz, 2008; Pilato et al., 2009; Samargia & Jacobson, 2009). Following the analogy with stroke survivors we suggest that rehabilitation facilitates neural plasticity to maximally involve the remaining hemisphere in partial compensation of functional loss. To be effective, the main principles of neurorehabilitation should be met ensuring that a protocol used is based on active motor learning which causes the cascade of neuroanatomical changes including formation of new synapses, increase in astrocytic volume and dendrite sprouting and angiogenesis (Kleim & Jones, 2008; Kleim et al., 2007; Mulder & Hochstenbach, 2001). Mere physical activity whether passive or lacking voluntary drive is unlikely to result in efficient rehabilitation (Lotze, Braun, Birbaumer, Anders, & Cohen, 2003). Further, to achieve neuroanatomical change, therapy should incorporate functionality, repetitive and intensive practice (Arya, Pandian, Verma, & Garg, 2011, Adomaitis & Varrada, 2005 #415). Initially shown to be effective for retraining gait in persons following spinal cord injury, body weight support treadmill training (BWSTT) has proved to be beneficial for a variety of subjects with other neurological conditions, such as cerebrovascular accident. (Berhman 2000, McNevin, Coraci, & Schafer, 2000; Schindl, Forstner, Kern, & Hesse, 2000; Suteerawattananon, MacNeil, & Protas, 2002). Although stronger research is needed to confirm findings, systematic reviews suggest that BWSTT may be effective in improving gait and functional abilities in children with chronic, long term neurological deficits such as cerebral palsy (Damiano & DeJong, 2009; Mattern-Baxter, Bellamy, & Mansoor, 2009; Mutlu, Krosschell, & Spira, 2009; Zwicker & Mayson, 2010). We suggest that the Intensive Mobility Training (IMT) which focuses on mass practice of walking and active, voluntary gait-related activities may be a promising mode of intervention in population post-hemispherectomy (Dahl et al., 2008; Field-Fote & Roach, 2011; Hesse, Werner, Frankenberg von, & Bardeleben, 2003).

3.1 Intensive Mobility Training, IMT

Intensive Mobility Training (IMT) is an example of a comprehensive treatment option designed to address gait, balance and mobility deficits in chronic neurological deficits (Fritz 2007). It borrows concepts from other successful therapies such as constraint induced movement therapy (CIMT), (DeBow, Davies, Clarke, & Colbourne, 2003; Gordon, Charles, & Wolf, 2005). With the addition of Body Weight Support Treadmill Training (BWST), it was found to be a feasible option for children status post hemispherectomy (Fritz, Merlo, et al., 2011; Fritz, Rivers, et al., 2011).

IMT is a 10-day, 30 hours intervention based on the principles of massed practice and repetitive task specific training. This is comparable with other effective rehabilitation studies have used longer training periods over several weeks but with actual intervention time between 12 and 30 hours. Therefore,
having individuals perform a shorter, more intense 10-day intervention does not significantly alter the amount of time (in hours) that the individual engages in intensive massed practice that has been shown to underlie successful therapeutic approaches (Bastille & Gill-Body, 2004; Mount, Bolton, Cesari, Guzzardo, & Tarsi, 2005; Plummer, Behrman, & Duncan, 2007; Pohl M, 2002; Schmidt & Lee, 2005 Werner, 2002.)

4. Target Population

4.1. Etiology

On the basis of a primary lesion associated with intractable seizures patients may be classified into three etiological subgroups (Devlin et al., 2003; van der Kolk et al., 2013):

(1) ‘developmental’ (hemimegalencephaly(HME), cortical dysplasia or other);

(2) ‘acquired’ (stable and non-progressive brain lesions, occurring perinatally or early in postnatal life, mostly consisting of ischemic or hemorrhagic pathology);

(3) ‘progressive’ (Rasmussen’s encephalitis or Sturge—Weber Syndrome).

Previous literature suggests functional impact may be related to etiology and/or age of surgery (Van Empelen, Jenneks-Schinkel, Buskens, Holders, & Van Nieuwenhuizen, 2004). For example children with developmental etiology (cortical dysplasia and HME) more often have significantly diminished distal arm strength and hand function and show less improvement in gross motor function compared to those with acquired pathology (infarcts). Two years after surgery, gross motor development had improved in patients with acquired and progressive brain lesions, but not in children with the disorders of cortical migration (van der Kolk et al., 2013).

4.2 Considerations and precautions:

After a thorough examination (the patient/client history, the systems review, and tests and measures, see Interactive Guide to Physical Therapist Practice) the physical therapist may identify a need for referrals to other specialists such as: a Neurologist; Orthopedist, Endocrinologist, Psychologist, Neuro-Ophthalmologist, Occupational Therapist, Speech and Language Therapist, and/or Orthotist due to presenting motor, sensory, cognitive and behavioral deficits.

Delayed hydrocephalus can develop at any time, even several years (>10) post-hemispherectomy. Most often the symptom is recurrence of seizures, vomiting and headaches (Lew, Matthews, Hartman, & Haranhalli, 2013). If a shunt has been placed, one must be cautious of shunt malfunctions (cases of infection, overdrainage, etc have been reported). Signs may be of any gradual changes such as a deterioration of school performance.

Inclusions and Exclusions:

Inclusions:

- Current published literature suggest feasibility of IMT 1-22 years after a cerebral hemispherectomy from ages 5 to 25 years old.
- Clinically, IMT has been utilized for children 6 months after a cerebral hemispherectomy, ages 18 months to 21 years old at Whole Hearted Pediatric Physical Therapy, Long Beach CA, under the constant supervision of a licensed physical therapist.
- Prior to participating in an IMT program the patient must be medically stable and cleared by their surgical team, neurologist and pediatrician.
• A communication system should be established in order for a child to express their needs and follow directions or simple commands.
• A patient requiring an assistive device, such as a walker, may participate but should be able to weight bear on bilateral lower extremities without pain and be able to demonstrate or tolerate facilitation of reciprocal gait pattern.
• A patient and family must agree to level of participation.

Exclusions:
• medically instability;
• inability to participate in purposeful play or functional activity;
• behavioral problems that would jeopardize patient safety;
• contractures that limit functional lower extremity use or inability to bear weight;
• uncontrolled seizure activity

5. Pre-therapy evaluation and intervention protocol
5.1 Evaluation

Within the evaluation process a central concern is how children with disabilities participate in daily routines that promote their health and well-being (Goldstein, Cohn, & Coster, 2004). According to the ICF model this participation is the result of the interaction of body structure, function, activity demands, contexts, and the person’s goals and desires (see Figure 1). Following a physical therapist’s synthesis of this information collected from the examination process, Fritz et al (2011) suggest establishing a list of activities specific to each client prior to following an IMT program. This list is referred to throughout intervention to address gait, balance and functional goals specific to the patient’s activity and participation level. For example, an 11 year old boy participating in IMT who is independently walking throughout all environments will have a different list of activities than an 11 year old girl who primarily uses her wheelchair for community mobility. The list is to have to ensure structured therapy sessions that has little interruptions in therapy to develop therapeutic activities. These activities should be able to be advance or regressed as needed according to patient level of function.

5.2 Intervention: Intensive Mobility Training Description and Frequency

In Fritz et al. 2011, IMT was performed 3 hours per day for 2 weeks (10 consecutive weekdays), for a total of 30 hours. The sessions focused on encouraging participants to use their more-affected lower-extremity in a massed practice setting. The goal for each 3-hour session was to have 1/3 of the session dedicated primarily to locomotor training (LT), 1/3 to therapeutic interventions aimed at improving balance, and the final 1/3 to muscle coordination, strengthening, ROM and other activities. See Figure 2. While there were not one-hour blocks of each activity, the total amount of time for each sub-group totaled one-hour, allowing standardization of the intervention across participants. Rest time was limited to 30 minutes and divided evenly between of the 3 categories. Therefore an overall goal of 150 minutes of daily treatment is suggested. The activities were documented in a daily activity log (See Figure 3) and individualized to fit each participant’s level of function. Throughout the therapy sessions, the participants received continual verbal feedback. A specific training protocol for LT was adapted from a previous investigation, and the following main objectives were addressed during training: 1) approach normal temporal parameters of gait; 2) maintain upright trunk; 3) approximate normal joint kinematics for lower extremity joints; and 4) avoid excessive weight bearing on the upper extremities (Berhman 2000).
If an individual was unable to accomplish these main goals independently on the treadmill, body weight support was used. The amount was determined in an effort to maximize bilateral limb loading without subsequent knee buckling. If the participant was unable to generate the stepping motions independently, then manual cues were used to assist. Once optimal gait kinematics was achieved first, BWS was decreased as well as manual assistance and then speed of walking was increased (Berhman 2005). In order to maintain appropriate intensity all activities were monitored to always challenge the patient. Progression of therapeutic activities was made by increasing time, distance, or height as applicable, as well as changing a support surface or reducing amount of support.

5.3 Outcome Measures
Initially, improvements associated with IMT were identified for toe in/out, step length of unaffected leg, Dynamic Gait Index, Berg Balance Scale, Fugl Meyer (lower extremity and balance score) and 6-minute walk. Normative values of 6 measures that were most influenced by the intervention were combined to create a Combined Functional Index (CFI) to assess global impact of therapy in gait and balance. Patients improved from an average of 77.3% to 82.7% of normal following IMT. Improvements in CFI were greater in patients five years or younger at time of surgery compared with older patients, (Fritz, Rivers, et al., 2011). We have further elaborated outcome measures that are both sensitive to change and have normative data in healthy populations in our recent work on the effects of repeated therapy pulse. We suggest that adequate gait, balance and mobility primary measures with normative data available are Dynamic Gait Index and 6-minute walk together with measures of velocity and the Activities-specific Balance Confidence (ABC) Scale. The secondary measures may include the Fugl Meyer scale, Berg Balance Scale, Bruininks-Oseretsky Test of Motor Proficiency, Gross Motor Function Measure, Peabody Developmental Motor Scales and Bayley Scales of Infant and toddler development.
Figure 2. This is an illustration of how Intensive mobility Training was used by Fritz et al 2011 for 19 children with status post hemispherectomy. The goal for each 3-hour session was to have 1/3 of the session dedicated primarily to locomotor training (LT), 1/3 to therapeutic interventions aimed at improving balance, and the final 1/3 to muscle coordination, strengthening, ROM and other activities. While there were not one-hour blocks of each activity, the total amount of time for each sub-group totaled one-hour, allowing standardization of the intervention across participants.
<table>
<thead>
<tr>
<th>TIME</th>
<th>ACTIVITIES DONE DURING THIS PERIOD</th>
</tr>
</thead>
<tbody>
<tr>
<td>7:55</td>
<td>standing calf stretch, hamstring, tennis ball massage, plank, row</td>
</tr>
<tr>
<td>8:06</td>
<td>R. arm ST massage</td>
</tr>
<tr>
<td>8:16</td>
<td>treadmill prep</td>
</tr>
<tr>
<td>8:23</td>
<td>treadmill calf stretch on break</td>
</tr>
<tr>
<td>8:17</td>
<td>treadmill sideways (R&amp;L) and backwards</td>
</tr>
<tr>
<td>9:27</td>
<td>red ball bouncing, throwing and passing (60 degree turns both sides)</td>
</tr>
<tr>
<td>9:37</td>
<td>obstacle course: balance beams (2), step ups (17), wedge, trampoline,</td>
</tr>
<tr>
<td>9:48</td>
<td>break from obstacle course, STM on R. hand</td>
</tr>
<tr>
<td>9:53</td>
<td>bathroom break</td>
</tr>
<tr>
<td>9:59</td>
<td>balance beam and 10&quot; side step ups and over to other side</td>
</tr>
<tr>
<td>10:00</td>
<td>balance beam and 8&quot; step up and over backwards 5X</td>
</tr>
<tr>
<td>10:09</td>
<td>wobble board: with PT guard, single limb stand with toe touch forward, side, and back (both legs) see note</td>
</tr>
<tr>
<td>10:15</td>
<td>proprioceptive hand test</td>
</tr>
<tr>
<td>10:17</td>
<td>washed blue marker off hand</td>
</tr>
<tr>
<td>10:21</td>
<td>knee flex/ext ankle DF/PF, anterior/posterior medial/lateral shifts (SITTING) R. leg ball or blue squashy under foot</td>
</tr>
<tr>
<td>10:24</td>
<td>same as previous, but standing led both legs</td>
</tr>
<tr>
<td>10:27</td>
<td>quad (knee ex): R. leg, 20lb (1 sets of 5), 25lb (2 sets of 5), knee mobilization by PT</td>
</tr>
<tr>
<td>10:34</td>
<td>bathroom break, with high marches to and fro, Wrist extension in water</td>
</tr>
<tr>
<td>10:41</td>
<td>Abduction (55 lb. Graps), with SIJ massage on R. hand during</td>
</tr>
<tr>
<td>10:47</td>
<td>standing quad weight bearing</td>
</tr>
<tr>
<td>10:49</td>
<td>marching on Z blue foam, single limb stance (EOIEC)</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Total Balance Time</th>
<th>Total Core Time</th>
<th>Total Gait Time</th>
<th>Total Break Time</th>
</tr>
</thead>
</table>

**Figure 3** Daily Logs may be recorded to track minutes devoted to each area of treatment. Fritz et al utilized this to track daily activities, timing and breaks. Here is an example from Pagan and DeBode (in submission) for an 11 year old boy status post hemispherectomy due to Rasmussen’s encephalitis at age 4 years old.
Parent Education/Home exercises
Prescribed home exercises during participation of the IMT program has not been fully investigated. Like CIMT programs, patients may benefit from prescribed exercises which increase awareness of the hemiparetic side. Parent participation is highly recommended especially for younger clients, such as toddlers, to integrate suggestions and discourage non use throughout the child’s day.

Due to persistent hemiplegia and spasticity of muscles, infants and children are at risk and have been observed to acquire secondary conditions such as torticollis, leg length discrepancy and joint pain. Education for families of children with cerebral hemispherectomy should include social, cognitive and psychological impacts of growing and aging with a chronic neurological condition such as cerebral hemispherectomy. Appropriate recommendations on orthotics as well as assistive devices such as standers, walkers and wheelchairs may be required to enhance quality of life as well as promote independence.

Future Research Agenda
The highest level of evidence-based research include randomized control trials (RCT). (Sackett 2000) At this time there are no RCT to warrant efficacy of IMT on gait, mobility and balance. However, due to the fact it is a relatively small population it is possible that an RCT would never be feasible. Nevertheless, future research and use of control groups is needed to establish:

- The effectiveness of IMT versus traditional therapy dosages (1 hour of therapy 1-2x/week or monthly consultations) to determine optimal schedule
- More investigation is required on the progress and ongoing functional level as well as fitness level of chronic hemispherectomy clients as a child ages. Our preliminary IMT results collected in 4 individuals after 6 years of no therapy suggest decrease in functioning that has been reversed with a repeat pulse of IMT (Pagan & de Bode, 2014 in submission)
- The clinical presentation of children with cerebral hemispherectomy and its impact on treatment protocol such as sensory dysfunction
- Pain and the effects of chronic disability
- Appropriate dosage of IMT as well as how to retain results
References


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