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The Feasibility of Functional Electrical Stimulation to Improve Upper Extremity Function in a Two-year-old Child with Perinatal Stroke: A Case Report

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ABSTRACT

Aims: To evaluate the effectiveness and feasibility (i.e. tolerability, adherence) of functional electrical stimulation (FES) for the upper extremity (UE) in a two-year-old child with perinatal stroke. **Methods:** Forty hours of FES over eight weeks was prescribed. FES to the hemiplegic triceps, extensor carpi radialis longus and brevis, extensor carpi ulnaris and extensor digitorum was timed with reaching during play. Assessments were performed before, during, and two months post-intervention. UE function (Melbourne Assessment 2 (MA2), Assisting Hand Assessment (AHA)) and spasticity (Modified Tardieu with electrogoniometry and electromyography) were measured. The mother completed a semi-structured interview post-intervention. Descriptive statistics were used for adherence and UE measures. A repeated-measures ANOVA compared Modified Tardieu parameters (e.g. catch angle) over time. Conventional content analysis was used for the interview data. **Results:** The child completed 39.2/40 hours. Immediately post-intervention, improvements were observed on MA2's Range of Motion subscale and catch angle (Modified Tardieu, $p < 0.001$). Two months post-intervention, improvements were observed on MA2's Accuracy and Fluency subscales. No change in AHA score occurred. Three themes emerged from the interview: (1) Ingredients for program success; (2) Information about the FES device; and (3) The child's response. **Conclusions:** UE FES was feasible in a two-year-old child with hemiplegia.

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One out of every 2,300–5,000 infants suffers an ischemic perinatal stroke, which occurs between 20 weeks gestation and 28 days after birth (Raju et al., 2007). Over half of these children are eventually diagnosed with hemiplegic cerebral palsy (Basu, 2014; Golomb et al., 2008). In the hemiplegic upper limb, muscle weakness, spasticity, limited joint range of motion (Klingels et al., 2012), and impaired bimanual coordination (Greaves et al., 2010; Hung et al., 2004) result in inefficient and abnormal movement strategies (Chen et al., 2013). Secondary complications, such as contractures and osteopenia (Presedo et al., 2007), may also develop,

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limiting movement further. Since upper extremity (UE) function is known to be especially important for activities of daily living (James et al., 2015; Kuiper et al., 2010), UE hemiplegia can lead to lifelong dependence in daily activities, reduced function at school, unemployment, reduced quality of life and decreased self-esteem (Damiano et al., 2006; Riad et al., 2013; Russo et al., 2008; van der Slot et al., 2007). Thus, facilitating the recovery of UE function is of utmost importance.

Treatment of UE impairments in children with perinatal stroke is predominantly symptom-focused, as disease-modifying options are limited (Basu, 2014). There is evidence that constraint induced movement therapy (CIMT) (Chen et al., 2014; Sakzewski et al., 2014), intensive bimanual training (Gordon et al., 2011), splinting (Jackman et al., 2014), virtual reality gaming (Chen et al., 2014; James et al., 2015), and standard occupational therapy (James et al., 2015; Sakzewski et al., 2015) are beneficial for the UE, with the majority of these treatments targeting school-age children or adolescents.

There is, however, a need for an effective, early intervention for the UE of young children with perinatal stroke (Basu, 2014; Kirton, 2013). The importance of early intervention is linked to the occurrence of “maladaptive plasticity,” or atypical development of the corticospinal tracts (Kirton, 2013; McIntyre et al. 2011; Yang et al., 2013), which is associated with greater impairment and poorer function later in childhood (Eyre et al., 2007; Kirton et al., 2008; Staudt et al., 2004). Work in young animals has shown that corticospinal tract development can be modified through motor or neural activity (Friel et al., 2012; Martin et al., 2011; Salimi et al., 2008). This has been referred to as a critical period, or window of opportunity, that is likely present in the first few years of life (Basu, 2014; Kirton, 2013; Yang et al., 2013). By intervening early after perinatal stroke, physical interventions may facilitate more normal development of motor pathways (Basu, 2014; Kirton, 2013) and consequently, better functional outcomes (Eyre et al., 2007; Kirton et al., 2008).

One intervention used in adults, though rarely applied to children, is functional electrical stimulation (FES). FES involves applying a low-level, electrical current to the skin overlying a muscle. This induces contractions of weakened or paralyzed muscles and facilitates repetitive practice of functional movements, such as reaching. In adults with stroke, FES has been shown to improve UE function (Page et al., 2010; Persch et al., 2012; Thrasher et al., 2008), increase muscle strength (Glanz et al., 1996), and reduce spasticity (Sahin et al., 2012), with associated neuroplastic changes (Page et al., 2010). Despite its documented efficacy in adults, there has been little research examining surface FES in children with stroke (Auchstaetter et al., 2015; Kapadia et al., 2014) or in children under the age of six years (Karabay et al., 2012; Xu et al., 2012). Hence, our objectives were to evaluate, in a two year-old-child with perinatal stroke, the: (1) feasibility of FES treatment of the upper extremity (e.g. tolerability and adherence to intervention prescription); and (2) effect of FES on this child’s UE function, as measured by the Melbourne Assessment 2 and the Assisting Hand Assessment.

Methods

Participant

This case report involved a trial of FES intervention in a single child. Ethical approval was obtained from the Research Ethics Board of the University of Alberta. The participant was a 2.6-year-old female with a history of a unilateral, arterial-presumed, perinatal ischemic stroke (focal brain injury on MRI: left posterior frontal, anterior parietal, superior posterior temporal lobes, insular cortex, external capsule and basal ganglia), and resulting right

hemiplegia. To confirm the child's appropriateness for FES, the contraindications for surface electrical stimulation (Houghton et al., 2010) and the training requirements were reviewed with the mother. Exclusion criteria included presence of an implanted electronic device or peripheral nerve damage in the affected upper extremity, a history of epilepsy, injection of botulinum toxin to the affected upper extremity within the past six months, and the parent being unable to follow instructions in English. The mother's goal was for her child to be able to use the hemiplegic arm in everyday activities. The child lived with her two parents and twin brother in a large city (population >800,000 people). The child's mother had a Bachelor of Science degree in Occupational Therapy and worked full-time as an occupational therapist (OT) with adult populations.

A researcher (PM), who is also a physical therapist (PT), then completed a physical exam and a single-session of FES to assess the child's appropriateness for and tolerance of, respectively, the proposed intervention. The researcher confirmed that the child did not have UE contractures and was capable of producing some active movement in the affected wrist. It was noted that the child had reduced active finger extension range of motion (AROM), especially in digits 3 and 4, and reduced wrist and elbow extensor AROM in the right arm. Increased tone in the right elbow, wrist and finger flexors was also noted, with greater severity distally. The researcher applied motor-level electrical stimulation to the child's triceps, extensor carpi radialis longus and brevis, extensor carpi ulnaris and extensor digitorum of the affected arm to ensure a motor contraction could be produced and that the child could tolerate the sensation.

FES Intervention

FES was delivered as a home program under the supervision of a PT with FES experience (KM). The first three sessions occurred at the University of Alberta with the researchers (KM and PM). At these initial sessions the mother was instructed on how and where to apply the self-adhesive electrodes. She was provided with photographs of the proper electrode placement on her child's arm. The safety precautions for FES were reviewed with the mother, such as frequent skin inspection and what warrants discontinuation. Subsequent sessions occurred in the child's home, led by the mother. A researcher (KM) observed and assisted with a minimum of one FES session/week via a video chat application. FES was administered as a home program in order to facilitate compliance with the targeted training dosage (outlined below). A home program was deemed feasible in this case, given the mother's OT background and her demonstrated ability to apply the intervention safely.

FES was delivered to the hemiplegic elbow, wrist and finger extensors using a 2-channel FES device (Empi 300PV, Empi, St. Paul, Minnesota). Stimulation was elicited with a hand switch (controlled by the mother) and timed to occur during the performance of functional tasks in sitting, standing and walking. The functional task targeted was reaching for toys, stickers, etc. with the affected arm (see Table 1). During the first week of FES intervention, weight-bearing through the hemiplegic UE, while manipulating crayons and food with the unaffected arm, was also practiced to encourage the child to use the hemiplegic upper extremity as a support when performing tasks typically performed by the unaffected or dominant upper extremity. The child preferred to practice open chain activities with the hemiplegic arm. Thus, the amount of time spent with the hemiplegic arm in a weight-bearing position was gradually reduced over the first week of the intervention. The targeted dosage of FES was 40 hours, or 2 sessions of 30 minutes per day, 5 days/week for 8 weeks. The prescribed dosage was based on that shown to be effective in adolescents (Kapadia et al., 2014). It also exceeds the minimum dosage recommended for UE training in children (Sakzewski et al., 2015). We aimed to elicit

Table 1. Example activities performed during the FES intervention.

Unilateral Activities	Bimanual Activities
<ul style="list-style-type: none"> • Reaching and grasping toys or food • Weight-bearing through affected arm while coloring/eating with unaffected UE • Reaching and touching screen of tablet • Turning door knobs during scavenger hunt • Manipulating small toys, stickers and food with pinch grasp 	<ul style="list-style-type: none"> • Pushing a toy grocery cart or doll stroller • Stabilizing a piece of paper with the affected UE while the unaffected UE colors • Grasping a cup or large toy with both hands • Rolling play dough with a rolling pin

30 repetitions of wrist/finger extension and 30 repetitions of elbow extension per session with FES. These two movements were often performed simultaneously during reaching, resulting in about one repetition per minute (30 reps over 30 minutes).

The stimulation parameters were set by a researcher (KM), who reviewed them weekly and assisted the mother in making any modifications, as needed. One channel of stimulation targeted the wrist and finger extensors. The cathode was placed over the motor point of the wrist extensors, and the anode was placed more distally to also target the finger extensors. A second channel targeted the triceps muscle (cathode on triceps motor point, anode placed just proximal to elbow joint). An asymmetric, biphasic waveform was used for both channels of stimulation. Frequency was set at 30 pulses/second, and the pulse duration ranged 200–300 μ s. Ramp up and down times ranged 1–3 seconds. Longer (i.e. 3 seconds) ramp up times were used initially to decrease the likelihood of increasing spasticity in the antagonists (i.e. wrist and elbow flexors). The mother was taught how to gradually increase the amplitude and set it to an appropriate level (i.e. comfortable and elicited the desired range of motion). During the two months following the intervention period, the participant did not receive any UE intervention.

Measures

To evaluate adherence to the FES intervention, the mother was asked to maintain a daily training log. In the log, the number of sessions/day and the number of wrist and elbow extensions performed/session were recorded. Adherence to the FES prescription was expressed as a percentage (e.g. (total number of repetitions performed/total number of repetitions prescribed) \times 100%). Performance on semi-quantitative clinical scales was assessed at multiple time points throughout the study: (1) two baseline assessments (before training), spaced two weeks apart, (2) after 2 and 4 weeks of FES (during training), (3) after eight weeks of FES (end of training), and (4) at two months post-training (follow-up). The assessments were administered by an OT who was blind to the intervention being applied. The OT had been formally trained in the administration and scoring of the clinical scales (i.e. Melbourne Assessment 2 and Assisting Hand Assessment). The following clinical scales were used:

Melbourne Assessment 2 (MA2) assesses the quality of affected arm movements in children as young as two years of age (Randall et al., 2001, 2008). It consists of four subscales (range of motion, dexterity, accuracy, and fluency) and involves performing 14 tasks, such as reaching, grasping and manipulating objects. The MA2 is valid (Randall et al., 2008, 2012), and has high interrater reliability and good test-retest reliability (Randall et al., 2001) in children with neurological conditions. Published standard error of measurement (SEM) values for the raw Melbourne scores (Randall et al., 2014) were used to calculate the minimally important

difference ($MID = 2 \times SEM$) for each subscale. Calculated MID values were as follows: range of motion = 4, accuracy = 4, dexterity = 2, and fluency = 4.

Assisting Hand Assessment (AHA) was designed for use in children with a hemiplegic arm, aged 18 months – 5 years (Krumlinde-Sundholm & Eliasson, 2003). It measures how well children use their affected arm in bimanual activities, such as spontaneous handling of toys during play. The validity, reliability and responsiveness of the AHA have been demonstrated in children with hemiplegic cerebral palsy (Krumlinde-Sundholm et al., 2007). Raw AHA scores were converted to a logit-based 0–100 scale in AHA units (Krumlinde-Sundholm, 2012). Five AHA units represent the smallest detectable change (Krumlinde-Sundholm, 2012).

Modified Tardieu Scale (Instrumented): Biceps Spasticity

An instrumented assessment of *biceps spasticity* was administered at the same time points as the clinical scales with the exception that only one baseline assessment was performed due to the length of this measurement. This spasticity assessment was based on the Modified Tardieu Scale (Ansari et al., 2008). This scale has been used in children with cerebral palsy as young as 3 years old (Yam & Leung, 2006), with adequate interrater reliability (Fosang et al., 2003). A researcher moved the child's affected elbow through the available range of motion at slow and fast speeds. Multiple trials were performed at each speed. A twin-axis electrogoniometer (Penny and Giles, Biometrics, Blackwood Gwent, UK) recorded the joint angle of the affected elbow and electromyography (EMG) (AMT-8, Bortec Biomedical, Calgary, Alberta) was used to record biceps muscle activity, all A/D converted on-line (Axoscope, Molecular Devices, Sunnyvale, CA). These assessments were videotaped.

The video was reviewed offline to identify trials in which the child was actively moving. These trials were excluded as the Tardieu test requires passive movement of the arm. Data from included trials were analyzed with custom-written programs (Matlab, The Mathworks, Inc.). The angle of “catch” (i.e. R1 in Tardieu Scale) was manually digitized from the goniometer data; it was identified as the first peak in the joint velocity trace. The magnitude of biceps EMG was calculated by subtracting the rectified background EMG (i.e. mean EMG magnitude over the 500 ms preceding the onset of stretch) from the EMG magnitude averaged over the 500 ms following stretch onset). The trials were matched across study time points for speed. Trials in which the stretch velocity was between 150–200 degrees/second were chosen for analysis. This speed corresponds with the V2 speed (i.e. the speed of the arm falling under the influence of gravity) on the Modified Tardieu Scale (Mackey et al., 2004). A repeated-measures (RM) ANOVA was used to compare the stretch velocity, catch angle and the magnitude of biceps EMG across time points. Alpha was set to 0.05 and the Bonferroni correction was used for post-hoc comparisons.

Parent-Completed Questionnaire and Interview

Following the 8-week FES intervention, the mother completed a questionnaire rating her experience using FES with her child (Table 2). It was based on a pre-existing questionnaire assessing parental satisfaction with CIMT for children with hemiplegic cerebral palsy (Wallen et al., 2008). At this time, the mother also participated in a semi-structured phone interview with a researcher (Table 3). The interview was digitally recorded and transcribed verbatim off-line. A conventional content analysis (Hsieh & Shannon, 2005) was used to interpret the interview data. Two researchers independently coded the transcript and through discussion reached a consensus about the themes.

Table 2. Parent questionnaire completed post-intervention.

Question	Response
1) How worthwhile was FES treatment?	<input checked="" type="checkbox"/> Very worthwhile <input type="checkbox"/> Worthwhile <input type="checkbox"/> Unsure <input type="checkbox"/> Not worthwhile
2) Would you participate in home-based FES treatment for your child again?	<input checked="" type="checkbox"/> Yes <input type="checkbox"/> Unsure <input type="checkbox"/> No
3) How did your child tolerate the electrical stimulation?	<input type="checkbox"/> Tolerated very well <input checked="" type="checkbox"/> Tolerated <input type="checkbox"/> Disliked
4) How easy was it to apply the electrodes to your child's skin?	<input type="checkbox"/> Very easy <input checked="" type="checkbox"/> Easy <input type="checkbox"/> Neither easy or hard <input type="checkbox"/> Difficult <input type="checkbox"/> Very difficult
5) How easy was it to carry out the FES treatment?	<input type="checkbox"/> Very easy <input type="checkbox"/> Easy <input checked="" type="checkbox"/> Neither easy or hard <input type="checkbox"/> Difficult <input type="checkbox"/> Very difficult
6) How easy was it to complete 2 sessions/day, 5 days/week?	<input type="checkbox"/> Very easy <input type="checkbox"/> Easy <input type="checkbox"/> Neither easy or hard <input checked="" type="checkbox"/> Difficult <input type="checkbox"/> Very difficult
7) How is your child's arm now compared with before FES treatment?	<input checked="" type="checkbox"/> Much better <input type="checkbox"/> Better <input type="checkbox"/> Neither better or worse <input type="checkbox"/> Worse <input type="checkbox"/> Much worse

Results

Adherence to FES Prescription

Adherence to the training prescription was high. The child completed 39.2 hours of the prescribed 40 hours. The mean number of weekly sessions completed was 9.8 ± 1.8 (range of 8–12) out of a targeted 10 sessions/week. The mean number of wrist extension repetitions performed per session was 33.3 ± 6.2 (range of 15–60) out of a targeted 30 repetitions/session. Similarly, the mean number of elbow extension repetitions performed per session was 31.9 ± 8.4 (range of 15–60) out of a targeted 30 repetitions/session.

Melbourne Assessment 2

Baseline scores on the four subscales of the MA2 were mostly consistent over the baseline period (Figure 1). After the first four weeks of FES intervention, there was no change in score on the Range of Motion subscale; however, at the end of the eight-week intervention, this score increased by four points, which is the MID. Performance on this subscale subsequently dropped by one point at the follow-up assessment. Performance on the Dexterity subscale decreased by two points after two weeks of FES intervention, but returned to baseline performance at the end of the eight-week intervention. Improvement in dexterity was seen only at the follow-up assessment. Like the Dexterity subscale, improvements in the Accuracy and

Table 3. Semi-structured interview guide.

Interview with Mother
We would like to hear about your/your child's experience with FES.
1. What went well? (INTERVIEWER PROBES as necessary for details—for information related to schedule, results, level of assistance from therapist as appropriate.)
2. What was challenging?
3. Did you notice changes in your/your child's abilities or play habits over the FES trial? If yes, please describe. (INTERVIEWER PROBE as necessary for details—what changes occurred, about when they occurred etc.)
4. What was your/your child's reaction to using FES? Did your/his/her reaction change over the length of the trial?
5. How do you feel your child tolerated the FES intervention?
6. What were your goals for participation? Do you think you reached those goals?
7. Do you have suggestions for things we could do differently, if we were to do this program again with another child? (If suggestions have been provided already, INTERVIEWER acknowledges those suggestions and asks if there is anything further.)

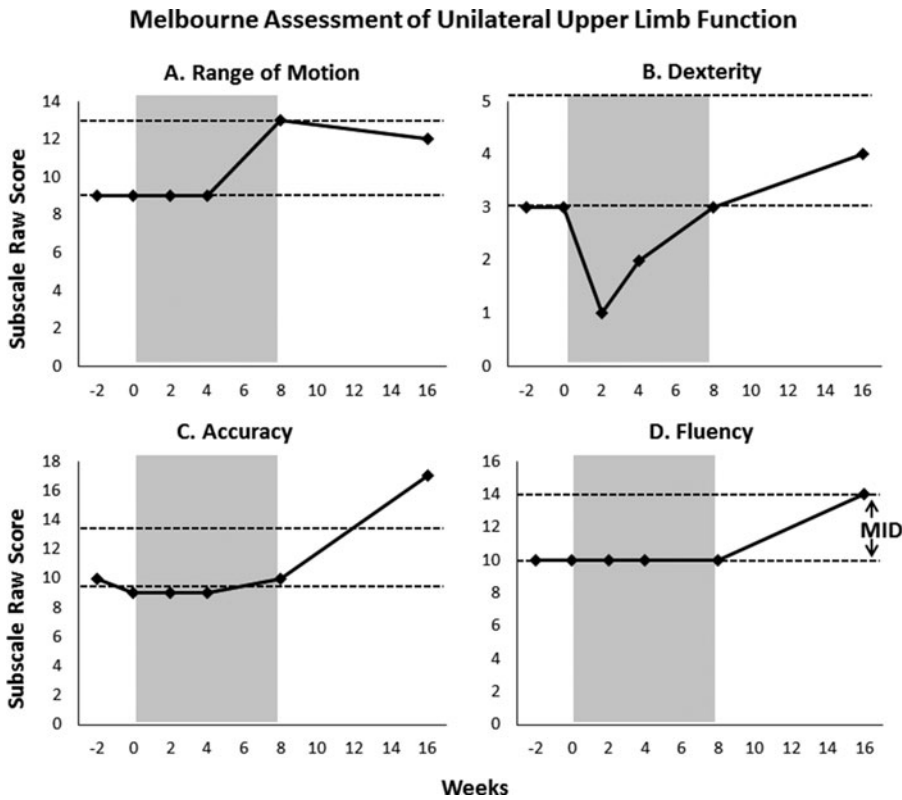


Figure 1. Melbourne assessment of unilateral upper limb function. Each plot shows the raw score over time on 1 of the 4 Melbourne subscales: A. range of motion (total possible score 27), B. Dexterity (total possible score 15), C. Accuracy (total possible score 25), D. Fluency (total possible score 21). The 2 baseline tests were performed at weeks -2 and 0. The FES intervention was initiated at week 0, and lasted for 8 weeks (grey shaded region). The vertical space between the 2 dashed lines on each plot represents the minimally important difference (MID) for that subscale.

Fluency subscales were not seen until two months post-training. At this time point the gains achieved on both subscales either met or exceeded the MID.

Assisting Hand Assessment

Overall there was no improvement in the child's AHA score throughout the study (Figure 2). Baseline scores on the AHA were not stable. The AHA score dropped during the first two weeks of training, and then returned to baseline values by the end of the eight-week training period. No change was seen during the follow-up period.

Modified Tardieu Scale (Instrumented): Biceps Spasticity

There were a sufficient number of stretch trials at 150–200 deg/s for comparison at the baseline, two week and eight week (i.e. end of intervention) time points. No significant difference was found between the mean stretch velocities of the trials across time points ($F = 0.223$, $p = 0.81$). A significant main effect was found for catch angle (RM ANOVA: $F = 9.08$, $p = 0.33$), but not for biceps response ($F = 0.75$, $p = 0.53$) (Figure 3). Post-hoc tests revealed

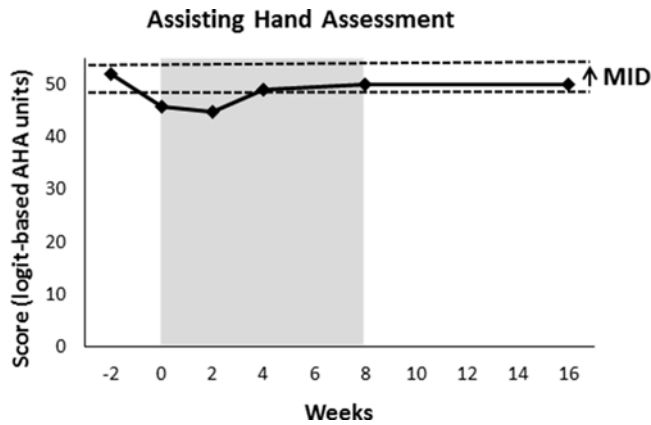


Figure 2. Assisting hand assessment (AHA). The AHA score, plotted in logit-based AHA units, recorded during baseline, intervention and follow-up phases. The 2 baseline tests were performed at weeks -2 and 0 . The FES intervention was initiated at week 0 , and lasted for 8 weeks (grey shaded region). The vertical space between the 2 dashed lines on each plot represents the minimally important difference (MID) for the AHA.

a significant reduction in catch angle between baseline and the end of FES (i.e. 8 weeks) ($t = 6.13$, $p < 0.001$), indicating that the catch occurred at a more extended elbow position.

The Parent's Experience with FES

The questionnaire responses from the mother are shown in [Table 2](#). The mother felt that her child tolerated the FES treatment and she rated the intervention as ‘very worthwhile’; however, she did report some difficulty with adhering to the schedule of two sessions/day for 5 days/week. The mother rated her daughter’s arm as “much better” following the FES intervention. Analysis of the interview data identified three themes: (1) Ingredients for program success; (2) Information about the FES device; and (3) The child’s response.

Ingredients for Program Success

The mother provided insight into factors that influenced the success of the intervention. These factors were grouped into three sub-categories. First, scheduling influences success. The mother felt that acknowledgement and acceptance of the program’s intensity was required by all involved. She stated that adhering to the intervention schedule was challenging at times

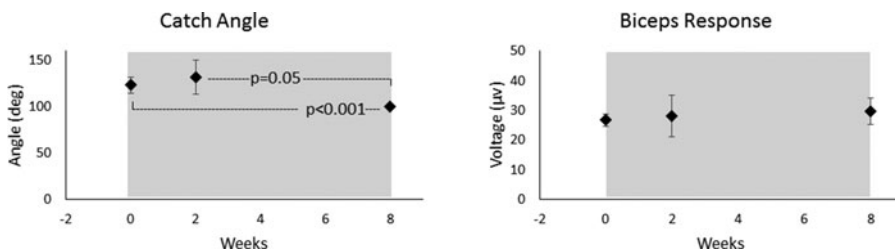


Figure 3. Mean (± 1 SD) catch angle plotted over time (left plot). There was a significant decrease in the catch angle from baseline to the end of training (8 weeks). Note: the SD of the 8-week data point is 3.4 deg. There was no significant change in the response of the biceps muscle over the training period (right plot). Grey shaded regions indicate the period of FES intervention.

due to her child's young age and "moods": "So keeping up with ...five-times-a-week kind of scenario is challenging sometimes. It was pretty intense." Likewise, flexibility in the schedule is also required. For example, the mother's adherence to the dosage became easier once she changed the schedule to include two 30-minute evening sessions (with at least one hour between sessions) rather than one 30-minute session in the morning and one 30-minute session in the evening. This eliminated the need to set up the electrodes and training activities twice/day. The mother felt that two sessions per day were feasible if "...flexible to work within the family schedule."

Second, certain parent characteristics can facilitate FES success. The mother felt it important to be adaptable and a good problem-solver. She discovered ways to increase her child's participation and interest during a training session: "I did find that having a list of activities that I could go to, and not having a set activity I wanted to do, but having a group of things that I could pick from and being able to change ...if something wasn't working and wasn't motivating her to participate, I'd have to kind of change things quickly and move to something else." The mother also felt that "with some patience and some discipline [FES is] something that would be easy to incorporate into a child's routine and that it's easy to incorporate into whatever kind of play or home activities you do with them anyways."

Third, the clinician plays a role in the program success. The mother spoke of the importance of providing the parents with sufficient information: "...providing written information beforehand about ...what FES is, things that they could expect, ...pros and cons of doing it, and then what are some things that might be easy, and what things might be hard, just to kind of prepare people in advance," as well as "...being able to teach parents or give them resources of like ideas of things that they can do and how to adapt an activity. There might be some teaching involved ...". The mother also stated that it would be very helpful if the clinician helped parents with land-marking, to ensure appropriate placement of electrodes on the child.

Information About the FES Device

The mother provided feedback on using a FES device with a young child. The set-up and operation of the device was not difficult for her. "Once I got ...the placement of electrodes down it was easy to put on, easy to use." In order to keep her daughter engaged during the sessions, the mother changed activities frequently and allowed her daughter to move around the home. In this respect, there were some challenges with the FES device; for example, "...the leads themselves were a bit too short." She also reported difficulty with the timing of stimulation: "I tried to be as accurate as possible ...But it's so quick like you know she's reaching for something and grabbing and it's just so quick ... I felt like sometimes I was working against her."

The Child's Response

The discussion of the child's response can be grouped into two sub-categories: reaction to the FES and the perceived effects of FES. First, the mother described how the child's reaction to FES changed over the course of the trial: "...she was more into it in the first three weeks I would say. And then at the midway point I started to notice that it was maybe getting to be more of like a tedious task for her." In addition, the mother reported that one brand of electrodes was sticky and caused some discomfort for the child upon removal. The mother stated that FES was more acceptable to her daughter than CIMT, which had caused her daughter "emotional distress."

Second, the mother reported numerous positive effects of FES on her child's function. For example, spontaneous use of her hemiplegic (i.e. right) arm in situations where protective reactions were needed: "...sometimes she'd be running at something and she'd lose her balance and her right arm would come out ...she would brace her arm against the wall or against the couch or the floor or whatever and like use [the right arm] to push herself up. And she wasn't doing that before at all. She would just totally switch over to the left or just crash into a wall with her body." The mother also reported increased use of the child's right hand during bimanual activities, like clapping her hands together for the first time and feeding: "...eating is one of the things where I noticed a big change in her in terms of how much she's using her right hand ...she'll hold onto a bowl for example with her right hand and use her utensils with the left and she wasn't doing that before." The mother also stated that the child showed more awareness of movement following FES. The mother used holding a cup as an example, and reported that after FES "...she'll figure out that she's not holding it well enough and she'll actually let go, open her hand, and place it on the cup. That was one of the huge things that I saw." The mother also noticed an increase in ROM of elbow extension and supination, and reduced spasticity in the hemiplegic arm following FES.

Discussion

The case report described here demonstrates the feasibility of UE FES in a child with perinatal stroke aged two years. Adherence to the prescribed number of hours and repetitions of FES was high, and the child tolerated the intervention. After eight weeks of FES, ROM increased and spasticity decreased in the child's hemiplegic arm. Increases in the accuracy and fluency of movement in the affected arm, as measured with the MA2, did not emerge until the follow-up assessment. In contrast, function of the affected UE during bimanual tasks, as measured with the AHA, did not improve during the intervention or follow-up period. Through a semi-structured interview, the mother reported functional gains achieved by her child that were not reflected in standardized measures of UE function, such as clapping the hands and using the hemiplegic UE for eating and during protective reactions when balance was lost. The FES intervention was completed at home by the child's mother, who as an OT had above average therapeutic knowledge. Hence, the generalizability of the findings is limited. Yet the positive results suggest that FES in young children warrant further investigation.

UE Interventions for Young Children with Hemiplegia

Research concerning UE interventions for children with perinatal stroke is described as "extremely limited" (Kirton, 2013). Two methods previously investigated in children as young as two years old are bimanual intensive training (Gordon et al., 2011) and CIMT (Taub et al., 2007). Both methods have been shown to improve UE function (Chen et al., 2014; Gordon et al., 2011; Sakzewski et al., 2014), and induce neuroplasticity in the affected motor cortex (Bleyenheuft et al., 2015; Inguaggiato et al., 2013; Walther et al., 2009). CIMT, however, has been criticized for not promoting the acquisition of bimanual tasks (Charles & Gordon, 2006; Kirton, 2013), and for not facilitating normal movement patterns (Taub et al., 2007). Furthermore, children may feel frustration with CIMT, and as a result, compliance with the protocol can be a challenge (Brady & Garcia, 2009; Martin et al., 2008).

Compared with CIMT, FES may cause less frustration for the child, and result in greater compliance with the protocol and prescription, as observed in this case example. FES also enables the practice of bimanual tasks; another potential advantage over CIMT. We intended

to target both unimanual and bimanual activities during the FES intervention; however, we did not record the amount of training time engaged in these two types of activities. It is possible that there was less focus on bimanual function. This may explain why score on the AHA did not improve over the course of training or follow-up. In addition, the baseline scores of the AHA were variable (Figure 2). The difference between the two baseline scores (six units) exceeded the magnitude considered attributable to measurement error by one unit (Krumlinde-Sundholm, 2012). This baseline variability may have masked any true change in AHA score (Lang et al., 2008). It would have been preferable to collect data from more than two baseline sessions. Despite the lack of improvement in AHA score, the mother reported gains in bimanual function during the interview (e.g. eating, clapping hands).

To our knowledge, this is the first report of the application of surface FES to the hemiplegic UE of a young child. Kapadia and colleagues (2014) studied the effect of FES for the affected arm in four children with chronic stroke, all aged 13 years. The participants completed 48 one-hour sessions of FES therapy over 16 weeks, and showed improvement on several measures of UE function (Kapadia et al., 2014). Similarly, our study participant showed improved function of the affected UE, as measured with the MA2. Other FES application have been used in pediatric populations; for example, FES for gait (Damiano et al., 2013; Prosser et al., 2012) and cycling (Johnston et al., 2011; McRae et al., 2009). Neuromuscular electrical stimulation (NMES), which is the application of stimulation in an exercise or non-functional context, has also been used in children with cerebral palsy (reviewed in Merrill, 2009). Despite this growing body of literature, it was recently reported that some physical therapists believe FES is not appropriate or feasible in children (Auchstaetter et al., 2015). The research presented here may help to dispel this belief.

Delayed Benefit of FES Intervention

Scores on the Accuracy and Fluency subscales of the MA2 did not increase until after the completion of the FES intervention (i.e. at the follow-up assessment). The child did not participate in any UE rehabilitation during the two-month follow-up period. This finding may suggest a delayed benefit of FES. Perhaps the improved tone and ROM in the affected UE enabled improved function in the hemiplegic UE; however, practice was needed to achieve functional improvement. A similar phenomenon was observed in adults with stroke who received a trial of UE NMES (Hsu et al., 2012). A delayed benefit of therapy has also been reported in another pediatric case report examining CIMT in a three year old child (Martin et al., 2008). In this case, improvements in grip strength did not appear until the follow-up assessment.

Limitations and Future Directions

This study has a number of limitations given that it is a case report involving a single participant. It is not possible to generalize the study findings to other children with hemiplegia. In particular, this study found that FES could be successfully administered at home by a parent; however, the mother's experience as an OT likely contributed greatly to this success. Furthermore, having two parents in the home was likely an advantage as the father was able to care for the sibling during the FES intervention and/or assist with the intervention as needed. Considerably more training and education would be required for other parents. Only two baseline assessments were completed despite variable performance on one of the outcome measures. Baseline assessments could have been repeated until performance stabilized; however, this practice is often impractical, especially when working with young children.

Given the positive outcomes of this study, a larger feasibility study investigating UE FES in young children is warranted. Future work could include more than one follow-up assessment, and a measure of hemiplegic UE use in daily life (e.g. the Pediatric Motor Activity Log (Uswatte et al., 2012) or accelerometry (Sokal et al., 2015)). In conclusion, UE FES was effective and feasible in a young child with hemiplegia.

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Declaration of Interests

The authors report no declarations of interest.

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