A multi-site study of functional outcomes following a themed approach to hand–arm bimanual intensive therapy for children with hemiplegia

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AIM This study investigated the effects of a theme-based (‘magic’) variation of the hand–arm bimanual intensive therapy programme, in two different countries, in improving activity performance for children with hemiplegia, including those with severe movement restrictions.

METHOD Twenty-three children with spastic hemiplegia (13 males, 10 females; mean age 10y 7mo, range 7–15y; Manual Ability Classification System level I, two; level II, 13; level III, eight), participated in one of three, 2-week, summer camps. A within-participant experimental design was used with the Assisting Hand Assessment and Children’s Hand Experience Questionnaire as primary outcome measures. Evaluations occurred immediately before the first day, on the last day, and 3 months after intervention. Two groups underwent additional assessments 2 weeks before the camp.

RESULTS Significant intervention effects were seen on the Assisting Hand Assessment (p<0.002) and on the Children’s Hand Experience Questionnaire (p<0.001), the latter maintained at follow-up. The affected hand was reported to be used in 25% of bimanual activities before the camp, progressing to 93% after camp, and decreasing to 86% at follow-up. Severity of impairment did not influence progress.

INTERPRETATION This themed approach to intensive intervention showed positive results in bimanual use, with improvements in independence sustained at follow-up. Although children across camps and motor severity made progress, some questions remain about intensity and duration of intervention to optimize longer-term outcomes.

Children with hemiplegic cerebral palsy (CP) have early brain injuries resulting in predominantly unilateral motor impairments. These impairments frequently cause considerable functional disability in these children because of difficulties in performing two-handed tasks.1 Evidence of the effectiveness of traditional therapies for children with hemiplegia has been found to be lacking, or they require repetitive and laborious actions with reduced compliance limiting their potential effectiveness.2–4

Over the past decade, interventions such as constraint-induced movement therapy and intensive bimanual training (e.g. hand–arm bimanual intensive therapy [HABIT]), have been the focus of intense efficacy research and show promise in improving upper limb function.5–11 Despite the intensity, however, positive responses have not been achieved for all children.12–15 Limitations of these interventions relate to reduced intensity and duration of treatment, which may have been influenced by children’s frustrations and lack of motivation for sustained involvement.2,12,13 Furthermore, studies of these approaches have been limited to participants with mild to moderate hand impairments, and largely from Europe, North America, and Australia. It is unclear whether children with more severe limitations to hand function (severely restricted wrist and finger extension), older children, or those from different cultures would benefit from such intensive therapies.

Motivation has been related to improved behavioural performance in demanding tasks and rehabilitation outcomes.16 In particular, positive benefits have been shown in studies that incorporated magic tricks or magi-
cians within therapeutic programmes. As children had shown an interest in learning to be young magicians, a programme of specifically graded bimanual magic tricks was developed for children with hemiplegia. In developing this themed model we embedded the therapeutic tasks within magic tricks to engage children creatively in the practice of repetitive actions to achieve a ‘greater’ (magical) goal.

The current study investigated the effects of incorporating ‘magic’ hand tricks into an intensive intervention programme designed as a summer camp in two different settings: Israel and the UK. The International Classification of Functioning, Disability and Health provided a framework to evaluate the impact of movement disorders on activity performance. A within- and between-participant, single blind experimental design, in which evaluators were blind to the treatment phase, contrasted the influence on bimanual skills after participation in a 2-week summer camp. We hypothesized that mass practice in bimanual skills, within meaningful and fun activities using a magic theme for enhanced motivation, would be beneficial in showing changes in upper limb function, but that age and severity of movement problems might limit outcomes. We also explored any differences in outcome across two cultures, particularly as UK children do not traditionally attend summer camps.

METHOD
Participants
A multi-site study in the UK and Israel recruited children aged seven to 16 years using a convenience sample of young people attending movement therapy clinics and/or known to child development centres in the South East Thames, London, or Tel Aviv regions. Informed consent was obtained from children and their parents. The study was approved by the relevant research ethics committees (London: 08/H0804/159) and Ministry of Health (Israel: 0204-10-TLV), conforming to the Declaration of Helsinki.

Children were included if they had clinical signs of spastic hemiplegia, attended mainstream school, were interested in magic tricks (ascertained by direct questioning and/or response to demonstration), could remember simple sequences of actions, participate in group tasks, and were able, at onset of the study, to attend all sessions of the summer camp. Children were excluded if they had any overt seizure activity, new motor therapy treatments (e.g. new programme of physiotherapy), and/or musculoskeletal and tone management treatments such as botulinum toxin injections or surgery in the previous 6 months or anticipated in subsequent 6 months. Notably, children were not excluded, as in other HABIT studies, if severity of motor disorder showed severe limitations to range of movement of wrist (<20°) or fingers (<10° flexion) or active grasp capacity. Power analysis using the Levin procedure to account for multiple comparisons indicated a total sample size of 20 was required to determine a main effect of intervention (using within-participant analysis with each child acting as his or her own control for primary outcomes). An achieved sample size of 22 participants across both sites gave 80% power at the 0.05% significance level to detect an effect size of 0.80 (Cohen’s $d$; the effect size was determined from pooling effects sizes from a meta-analysis of bimanual training in adults with more recent studies in children using similar outcome measures).

Intervention procedures
Children attended one of three 2-week camps, one in Israel or either of two UK camps, over 10 out of 12 consecutive days during the summer holidays in a hospital facility. Children attended 6 hours daily, with homework targets involving practice of bimanual magic tricks set each day child each day. Homework diaries were reviewed each morning with practice and demonstration of previously learnt activities, and a star awarded in the diary if targets had been achieved.

The camp programme and activities followed procedures summarized by Charles and Gordon, using the HABIT principles to promote intensive practice and repetition in part- and whole-task movements. Modifications to the HABIT protocol were made through inclusion of specifically selected/modified magic hand tricks in collaboration with professional magicians. Magic tricks were scaled to require increasingly complex bimanual skills, in timing, accuracy, or fine manipulation. Individual and group work was equitable across sites (three staff, four participants).

Both sites used the same repertoire of magic tricks, introduced at similar stages. The format of the camps included pre-breakfast sessions with individualized support for learning/practice of tasks/tricks; breakfast preparation and eating as bimanual tasks; late morning sessions incorporating further magic practice and bimanual gross motor play; bimanual tasks during lunch; and afternoon activities related to theatrical studies, including making costumes and props for the magic show.

Occupational therapists, physiotherapists, and students assisted camp management, supervised by therapists with experience of intensive motor interventions and magic (DG and/or AM). The Israeli camp was assisted by a young magician throughout. Two professional magicians assisted on four half-days at each UK camp. All camps were ‘opened’ by a magic show from professional magicians. Children prepared and participated in their own magic show at the end of the camp, assisted by professional magicians. The Israeli camp performed on stage during an International Magician’s convention on the penultimate day of the camp. The UK children performed...
on the afternoon of the last day of each camp in a central hospital atrium equipped with a stage.

Measurement
Participants were evaluated twice before intervention (initial [2 wks before camp; Israeli and the second UK (UK2) camps only] and pre-camp [2 or 3 d before the camp start]) and twice after the intervention (immediately post-camp and at 3 months’ follow-up [3mo-fu]). Activity performance and participation in bimanual tasks were considered primary outcomes. Any coexisting conditions were verified by medical records. Severity of movement difficulties at baseline was considered to be reflected by higher classification on the Manual Ability Classification System (MACS), which ranks a young person’s ability to handle objects in important daily activities.22 The Modified Ashworth Scale (MAS) was used to document spasticity across elbow, wrist, fingers, and thumb.23 Practice in bimanual tasks during the camps was recorded by the allocated interventionist for each activity and confirmed in staff discussions at the end of the day (0 [absent/no bimanual participation] to 4 [maximum achievable]). Daily and overall averages were calculated. Practice of magic tricks (bimanual tasks) during the period between the end of the camp and the 3mo-fu was recorded based on reports of children/parents as ‘little’ or ‘none’, approximately once a week, or two or more times per week.

Activity performance
The Assisting Hand Assessment (AHA; version 4.3) is a standardized test of spontaneous use and performance of an affected hand during bimanual interactions in functional/play-based tasks with excellent validity and reliability.24 The AHA was used as a baseline and primary outcome measure of the effective use of the involved hand during bimanual activities. Comparison of AHA scores between initial and pre-camp assessments was also used to consider natural progress. Transformation of raw scores gives a logit unit scale score with a difference of 5 corresponding to the least detectable difference (1.96 × 1/2 × standard error of the mean).23 Assessments were undertaken by trained therapists and video scored by experienced evaluators, not involved in delivery of the therapy and blind to group assignment and other AHA scores. Interrater reliability was undertaken on 20% of videos (intraclass correlation coefficient 0.96, p=0.001).

Daily task activity performance
The Children’s Hand Experience Questionnaire (CHEQ) is a 29-item questionnaire of independence in typical daily bimanual activities using the affected/hemiplegic hand with good reliability and item validity reported.25 Questions also cover the skill, time, and extent to which a child may be bothered by the difficulties they experience performing a particular task. All children or parents completed the English version of the questionnaire and were fluent in English as a first or second language. The extent to which the child’s affected hand was used in and influenced independence in daily bimanual activities was calculated as a percentage of independent activities in which the affected hand was used to stabilize or grip items.

A further secondary outcome, the Jette-n-Taylor Test of Hand Function (JTTHF),26 a standardized test used to document the efficiency of grasp and release of items, was administered by therapists (some of whom had assisted with the delivery of the therapy but were blind to previous JTTHF scores) to document unimanual changes in activity performance. The JTTHF was modified by eliminating the writing task. A 3-minute time limit was used for each task; therefore, maximum time to complete all items was 1080 seconds.

Data analysis
Statistical analyses, using the Statistical Package for Social Scientists version 19 (SPSS Inc, Chicago, IL, USA), were undertaken relevant to the original aims using intention-to-treat principles. Descriptive statistics explored characteristics and performance of the children before and after participation. Repeated-measures analysis of variance on test sessions was performed on logit-transformed AHA scores and raw scores of the JTTHF and CHEQ, and Bonferroni pairwise post hoc mean comparison and confidence intervals (CI) were calculated. Between-group comparisons were explored as pilot data because of the small sample. Natural change was calculated by a 2 (group: Israel and UK2) × 2 (test session) analysis of variance of variance of initial and pre-camp scores. Homogeneity was tested by Levene’s test of equality of variance and Mauchly’s test of sphericity. Changes of at least 5 logit unit scale scores on the AHA were used to ascertain the numbers/percentages of children making substantial progress; binary logistic regression considered effects of age, severity, and reported practice after the camp on outcome. Using intent-to-treat principles, missing data at 3mo-fu was considered missing at random and, therefore, prorated as an average of each child’s score from pre- to post-camp testing to avoid assumptions of a future directional trend. Binary logistic regression considered influence of age, severity of hand skills (MACS, MAS, and initial JTTHF), additional behaviour, or learning problems on progress, indicated by an improvement of at least one least detectable difference on the AHA after treatment. The Hosmer–Lemeshow statistic was calculated to consider goodness-of-fit of our model to the data. Non-parametric analyses, Kruskal–Wallis, and post-hoc Wilcoxon signed-rank analysis with Bonferroni correction were conducted to account for small sample sizes between groups, with the same results for treatment effect and, therefore, parametric tests are reported here.

RESULTS
Participants
Tables I and II show group and individual characteristics respectively. Nine children participated in the Israeli camp
(six males, three females), seven children (three males, four females) in the first UK (UK1) camp, and seven (four males, three females) in the UK2 camp. UK2 was undertaken 1 week after UK1 to allow for post-camp assessments for UK1 and pre-camp assessments for UK2. Pre-treatment assessment 2 weeks before the camp was only achieved for children in Israel and UK2* (excluding CHEQ because of technical difficulties). Seven children in Israel and 13 children in the UK (seven in UK1 and six in UK2) were available for 3mo-fu. Mean age across camps was 10 years 7 months (SD 2y 7mo). Six children had additional behaviour or learning difficulties (as documented in medical records and reported by parents). Three children had missing data at 3mo-fu, one from each camp, and their data were therefore pro-rated. Analyses were run with and without these children with similar results and these children’s pro-rated scores were therefore included in all analyses. There were non-significant differences in age and movement skill between groups, with the Israeli group showing lower scores on the AHA ($F_{(2,21)}=2.73, p=0.07$). There were no violations of homogeneity between groups or testing sessions, therefore no corrections were required.

### Changes in activity performance

**Trend/natural progress**

Data were available from the initial assessment 2 weeks before the camp as well as pre-camp for eight children in Israel and seven children in UK2. There were no measurable changes in AHA scores ($F_{(1,13)}=0.30, p=0.60$, mean difference –0.55, CI –2.7 to 1.6) or JTTHF total time ($F_{(1,13)}=1.54, p=0.24$, mean difference –0.25, CI –2.2 to 22.2) during the period without treatment or group interactions (see Fig. 1a,b).

### Effect of intervention

Table II reports changes in individual performance across measures. A significant effect of intervention was seen on the AHA ($F_{(2,19)}=8.87, p=0.002, \eta^2=0.48$), with post-hoc analysis showing main differences occurring between pre-camp and post-camp but progress was not maintained at 3mo-fu (mean differences: pre- to post-camp 5.93, CI –9.52 to 2.34, $p=0.005$; pre-camp to 3mo-fu 1.24, CI –5.58 to 0.77, $p=0.50$; post-camp to 3mo-fu –3.52, CI –0.25 to 7.29, $p=0.15$; see Table II and Fig. 1c). There was a non-significant decline at 3mo-fu, but these scores were no longer significantly different from those at pre-camp. There were no differences in the treatment effect sizes between UK (combined group) or Israeli camps ($\eta^2=0.44$ and 0.48 respectively). The small sample size precludes detailed between-camp differences.

Binary logistic regression did not indicate influence of age or severity of hand skills (MACS, MAS, and initial

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### Table I: Characteristics of participants across camps

<table>
<thead>
<tr>
<th>Camp</th>
<th>Sex Male/Female</th>
<th>Mean (SD) age, y:mo</th>
<th>Mean MACS level (SD; range)</th>
<th>Mean MAS (SD; range)</th>
<th>Behaviour problems*</th>
<th>Attendance/Practice during/Practice after</th>
</tr>
</thead>
<tbody>
<tr>
<td>Israel (n=9)</td>
<td>6/3</td>
<td>11:4 (2:7)</td>
<td>2.2 (1.42; 1–3)</td>
<td>2.44 (0.67; 1–4)</td>
<td>AD(H)D n=2; ODD n=1</td>
<td>One child missed 1d/60%/little or none</td>
</tr>
<tr>
<td>UK1 (n=7)</td>
<td>3/4</td>
<td>10:0 (2:4)</td>
<td>2.4 (0.82; 2–3)</td>
<td>1.0 (0.53; 0–2)</td>
<td>Anx/OCD n=1; ASD n=1</td>
<td>100% attendance/75%/once a week</td>
</tr>
<tr>
<td>UK2 (n=7)</td>
<td>4/3</td>
<td>10:2 (2:7)</td>
<td>2 (1.46; 1–3)</td>
<td>2.3 (0.69; 1–5)</td>
<td>Learning difficulties n=1</td>
<td>One child missed 1d/80%/at least once a week</td>
</tr>
</tbody>
</table>

*Owing to delays in processing ethical and research and development approval paperwork, it was not possible to assess children attending UK1 2 weeks before the camp and complete the entire programme within the 6-week summer holidays in the UK.

### Table II: Performance on each outcome measure at pre-camp, post-camp, and 3 month’s follow-up, and number of children progressing one or more least detectable difference or performing 20% better than at baseline

<table>
<thead>
<tr>
<th>Camp</th>
<th>AHA logic scale score (mean [SD])</th>
<th>CHEQ independent activities using two hands (mean [SD])</th>
<th>JTTHF raw score, s (mean [SD])</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Pre-camp</td>
<td>Post-camp</td>
<td>3mo-fu</td>
</tr>
<tr>
<td>Israel</td>
<td>46.8 (13.4)</td>
<td>53.6 (9.1)</td>
<td>50.0 (13.6)</td>
</tr>
<tr>
<td>UK1</td>
<td>62.7 (10.0)</td>
<td>70.1 (10.4)</td>
<td>62.0 (10.7)</td>
</tr>
<tr>
<td>UK2</td>
<td>63.1 (20.4)</td>
<td>66.7 (19.8)</td>
<td>67.9 (16.7)</td>
</tr>
<tr>
<td>All</td>
<td>56.6 (16.5)</td>
<td>62.6 (14.9)</td>
<td>59.1 (15.4)</td>
</tr>
</tbody>
</table>

| Number | 1 – | 17 | 7 | 1 – | 16 | 14 | 1 – | 15 | 12 |

| ≥1 LDD/ ≥20% |

AHA, Assisting Hand Assessment; CHEQ, Children’s Hand Experience Questionnaire; JTTHF, Jebsen-Taylor Test of Hand Function; 3mo-fu, 3 months’ follow-up; LDD, least detectable difference.
JTTHF), or additional behaviour or learning problems, on progress \((p>0.05 \text{ in all cases})\). The Hosmer–Lemeshow statistic \((\chi^2(8)=7.4, p=0.49)\) indicated good fit of our model to the data. Six of the eight children in MACS level III made progress of at least one least detectable difference post-camp, one sustaining this at 3mo-fu, and three showing continued benefit at 3mo-fu. This older child with an MAS score of 4 also reported a greater amount of home-based practice after the camp, in contrast to two children with MAS scores of 4 who did not report any home-based practice and who did not maintain their initial progress on the AHA (one child with a MAS score of 4 who reported some practice showed progress at 3-month-fu and data were missing for the fifth child). Whereas 12 of the 13 children in MACS level II made progress post-camp, none maintained this at 3mo-fu. One of the two children in MACS level I made progress of one least detectable difference post-camp with continued progress at 3mo-fu, with a ceiling effect in the others’ scores.

**Changes in daily task activity performance**

The percentage of bimanual activities performed independently using the affected hand (progressing from one-handed to two-handed use) as reported on the CHEQ progressed significantly pre- to post-camp \((F(2,19)=12.93, p<0.001, \eta^2=0.58)\). These changes were maintained at 3mo-fu (mean differences: pre- to post-camp 22.18, CI -38.79 to -5.57, \(p=0.001\); pre-camp to 3mo-fu 15.66, CI -31.04 to -0.29, \(p=0.001\); post-camp to 3mo-fu 6.5, CI -0.60 to 13.63, \(p=0.68\)). There were no group × camp interactions or influences of age or severity. Before the camp, 25% of the 18 activities performed independently were with reported use of the affected hand, whereas at post-camp use of the affected hand had increased to 93%, decreasing to 86% at 3mo-fu. Figure 2 shows comparisons of independence and bimanual use in daily activities.
**Unimanual skills**

Across treatment camps, children improved their speed in unimanual performance in the JTTHF with improvements of over 2 minutes (128.9s) across the six tasks after treatment (F(2,19)=18.09, p<0.001, η²=0.48). There were no group × camp interactions or influences of age or severity. Improvements were maintained at 3mo-fu (mean differences: pre- to post-camp −128.5, CI −209.9 to −47.0, p<0.001; pre-camp to 3mo-fu −110.5, CI −177.2 to −43.8, p<0.001; post-camp to 3mo-fu 17.9, CI −18.8 to 54.7, p=0.27; see Fig. 1d).

**DISCUSSION**

This study explored the efficacy of incorporating ‘magic’ hand tricks within the HABIT protocol, including children with more severe disorders (i.e. MACS level III) than in previous studies, across the differing cultural contexts of two countries. We had hypothesized that mass practice in bimanual skills within motivating exercises would promote upper limb motor skill but differences in age and severity of movement problems would influence response to treatment.

Our results suggest that such a programme is beneficial for children with hemiplegia, with progress not being influenced by age, severity of movement restriction, or country. Although there was more limited maintenance in the quality of use of the affected hand in bimanual activities on the AHA at 3mo-fu, significant improvements in independent functioning and use of the affected hand in daily bimanual activities shown by the CHEQ results were retained at 3mo-fu. By the end of the magic intervention programme, 93% of daily tasks were performed using a bimanual approach and maintained at 86% 3 months later, compared with only 25% before intervention. Furthermore, children reduced the overall time taken to perform the six tasks of the JTTHF with the affected hand and this progress was also sustained.

Although these results provide encouraging evidence of the benefits of such a programme for improving independence in daily activities, the constraints of our follow-up programme and lack of verification of practice time (both home based during the camp and between the end of the camp and the 3mo-fu) limit our ability to comment on whether trends shown on the AHA would be reflected in other measures. It is possible, however, that the quality of use of the affected hand as measured clinically is ultimately less important than the fact that a child is performing more bimanual tasks independently and using the affected hand.

As expected, the extent of improvement in hand function was variable between the children. It is encouraging that children with severe motor difficulties (MACS level III with no or limited active wrist extension or active grasp) made substantial progress in the short term on the AHA and JTTHF, some sustaining progress at 3mo-fu. It is not possible from our data to determine whether a difference in regimen or duration of intervention would have supported longer-term gains for these children. On examination of individual responses, it appeared that children who continued to practise magic tricks after the camp sustained progress; however, the small numbers preclude further analysis. Furthermore, AHA gains after 2 weeks of HABIT have been shown to decline by the 1-month post-test, whereas the gains after a 3-week HABIT programme did not.

The cultural context of undertaking intensive camp-based therapy is an important consideration for designing therapy programmes that are sensitive to the needs and habits of families. It is encouraging from our pilot data that there were no differences in the response to the intervention programme between the Israeli camp and UK camps, despite the absence of a ‘camp’ culture in the UK. We were further encouraged by the positive feedback provided by children and families to the project, with all but one child asking to be involved in a camp the following summer. Although the choice of ‘magic’ as a theme to engage children in practice of bimanual exercises during the camp may have supported initial large gains in hand function, the difficulty of integrating magic trick practice into a regular routine subsequently may have compromised ongoing progress. This is in contrast to recent results reported by Cohen-Holtzer et al., who followed a programme of HABIT and constraint-induced movement therapy, and Gordon et al. using activities of daily living with better sustained progress overall on the AHA. Although the children receiving bimanual training in the study by Sakzewski et al., which used a novel circus theme, and those who received a combined constraint-induced movement therapy/bimanual training approach using a pirate theme maintained progress on the AHA at longer follow-up, our study differs from these studies with the inclusion of more children in MACS level III and/or with more severe movement restrictions. It is possible, therefore, that severity influenced the ability to maintain initial progress after a 2-week intervention, and a longer period for consolidation may have been warranted. However, results are dependent on which outcome measure is deemed to be of higher importance. According to the CHEQ, which reflects estimated activity performance in daily activities involving bimanual skills, the progress of children in the magic HABIT programme supported our hypothesis at the 3mo-fu.

There are several limitations to this study, including the small sample size and resultant reduced power to detect smaller, but potentially meaningful, clinical changes, and lack of randomization. However, blinded scoring of the assessments adds robustness to the results. Although only four children sustained progress on the AHA, questioning the cost efficiency of this intervention, the improved participation and independence of the children, as reported on the CHEQ, is considered an equally viable outcome. It is unclear if sustained progress on the AHA would have been supported by more family involvement, supervision of home-based practice, and/or whether children with severe
motor disorders needed more time to consolidate skills (i.e., >60h). Although frustrations with such intense therapies that do not incorporate a themed approach have been documented,11 the current study was not able to determine the extent to which the ‘magic’ incentive enhanced outcomes. Direct comparisons within similar cultures of themed and non-themed approaches are warranted. Furthermore, exploration of the potential psychosocial benefits of such group programmes is also needed.

CONCLUSION

A themed approach to intensive intervention showed positive results across all measures of hand function reflecting the International Classification of Functioning, Disability and Health domains of activity performance and participation. Although several questions remain about the intensity and duration of intervention required to optimize longer-term outcomes, progress was evident, with increased independence and use of the affected hand in bimanual daily activities, sustained at 3mo-fu.

REFERENCES


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