

Assistive and Rehabilitative Effects of the Playskin Lift™ Exoskeletal Garment on Reaching and Object Exploration in Children With Arthrogryposis

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Importance: Children with arthrogryposis multiplex congenita are often delayed in their development of reaching and object exploration, which can place them at risk for associated delays in motor and cognitive development.

Objective: To evaluate the longitudinal assistive and rehabilitative effects of the Playskin Lift™ (hereinafter Playskin), a novel exoskeletal garment, on reaching and object exploration abilities in children with arthrogryposis.

Design: Single-case ABA design with a 1-mo baseline, 4-mo intervention, and 1-mo postintervention.

Setting: Home environment.

Participants: Seventeen children with arthrogryposis (ages 6–35 mo at first visit; 5 boys).

Intervention: Participants used the Playskin daily for 30 to 45 min while participating in structured intervention activities to encourage reaching for objects across play spaces larger than they were typically able to.

Outcomes and Measures: Participants were tested biweekly throughout the study with and without the Playskin using a systematic reaching assessment. Coding of reaching and object exploration behavior was performed using OpenSHAPA software; statistical analyses were conducted using Hierarchical Linear and Nonlinear Modeling software. Feasibility of the Playskin for daily home intervention was evaluated with a parent perception questionnaire.

Results: Positive assistive effects (improved performance when wearing the Playskin within sessions) and rehabilitative effects (improved independent performance after the Playskin intervention) were observed with increased active range of motion, expanded reaching space, improved grasping with the ventral side of the open hand, and greater complexity and multimodality and intensity of object exploration.

Conclusions and Relevance: The Playskin may be a feasible, effective assistive and rehabilitative device to advance object interaction and learning in young children with arthrogryposis.

What This Article Adds: The novel exoskeletal Playskin garment improves reaching and object exploration in young children with arthrogryposis.

A rthrogryposis multiplex congenita is a nonprogressive neuromuscular condition. It manifests in significant muscle contractures, weakness, and movement impairments across multiple joints (Bamshad et al., 2009; Staheli et al., 1998). Children with arthrogryposis have difficulty raising their arms against gravity and are at risk for delays in reaching and object exploration (Babik, Cunha, & Lobo, 2019; Babik et al., 2016; Corbetta & Snapp-Childs, 2009; Gibson, 1988; Needham et al., 2002; Zuccarini et al., 2017). Both delayed and impoverished object exploration may negatively affect children's cognitive outcomes, self-care efficacy, and quality of life (Bornstein et al., 2013; Jouen & Molina, 2005).

Therapy can improve motor function for children with arthrogryposis (Sells et al., 1996). In addition, interventions using exoskeletons (Haumont et al., 2011; Rahman et al., 2007) or robots (Heo et al., 2012; O'Neill et al., 2017) have

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improved upper extremity range of motion, motor control, and independent function in adolescents and adults with neuromuscular disorders. However, similar devices have not previously been available to younger populations.

Recently, a pediatric version of the Wilmington robotic exoskeleton (P-WREX; Nemours Alfred I. DuPont Hospital for Children, Wilmington, DE) was designed and tested longitudinally in an 8-mo-old with arthrogryposis (Babik et al., 2016). The P-WREX improved the child's visual attention, reaching space, and bimanual object exploration within sessions (assistive effect) as well as unassisted reaching and object exploration across time (rehabilitative effect). Although the device improved functionality, parental reports suggested it was limited in size, weight, bulk, comfort, ease of use, aesthetics, and other factors.

These limitations inspired the design of the first soft exoskeleton, the Playskin Lift™ (hereinafter referred to as *Playskin*), to support arm mobility in young children (see University of Delaware Move 2 Learn Innovation Lab, 2014, for instructions on how to fashion a Playskin garment). Pilot testing of the Playskin's assistive effects with a 23-mo-old child with arthrogryposis suggested that the Playskin improved the child's visual attention and reaching within a session and was reportedly comfortable, easy to use, discreet, and attractive (Lobo et al., 2016).

Another study evaluated the Playskin in 10 young infants born preterm, some with brain injury (Babik, Cunha, Moeyaert, et al., 2019), using a multiple-baseline design. The Playskin improved infants' reaching ability, promoted open-handed grasping with the ventral side of the hand, and facilitated multimodal exploration of objects within sessions. These positive effects were especially pronounced during the intervention phase (Babik, Cunha, Moeyaert, et al., 2019). Moreover, the Playskin improved infants' independent (i.e., unassisted) reaching and object exploration performance across sessions during the intervention phase, with positive effects preserved during postintervention (Babik, Cunha, Moeyaert, et al., 2019). Although participants in this study had impaired upper extremity function, they had full active range of motion (AROM) against gravity in their arms. Therefore, this study is not informative about whether the Playskin might be a feasible and effective tool for children with significant upper extremity weakness.

Our study is the first to evaluate the feasibility and effectiveness of the novel Playskin exoskeletal garment for young children with arthrogryposis and limited shoulder AROM against gravity. The three aims of this study were to evaluate (1) feasibility of intervention with the Playskin in children's natural environment, (2) assistive effects of the Playskin on reaching and object exploration when donned within sessions, and (3) rehabilitative effects of the Playskin intervention on independent reaching and object exploration across time.

Method

Participants

Power analysis for this study was conducted using General Linear Mixed Model Power and Sample Size (GLIMMPSE [Version 3.0.0]; software that calculates sample size for longitudinal designs; Kreidler et al., 2013). Using the target power of 0.8 and Type 1 error rate of .05 as well as implementing the Hotelling–Lawley trace statistical test, we found that the recommended sample size was 16.

Participants were 17 children with arthrogryposis and no other neuromotor or musculoskeletal conditions (5 boys; 5.8–35.0 mo old at the first visit, $M = 13.9$, $SD = 8.7$). The sample was 11.7% African-American, 82.4% Caucasian, and 5.9% Asian. Household annual income spanned from \$0 to \$14,999 (11.8%) to more than \$80,000 (35.3%). All children had independent head control; 12 sat independently.

Participants were recruited through word of mouth or social media from March 2014 to August 2016. Data from local families (6) were collected in person; data for geographically distant families (11) were collected by caregivers using standardized written instructions. Five distant participants dropped out of the study reportedly because of the effort required for data collection (self-recording videos and sharing them via Dropbox [Dropbox, Inc., San Francisco, CA]) after 4 to 13 biweekly data collections ($M = 7.4$, $SD = 3.71$). The remainder of the participants had no more than 5% data missing ($M = 1.82$, $SD = 1.76$).

All participants completed the informed consent process, and activities were performed in line with the regulations enforced by the University of Delaware's internal review board. Study participants received monetary compensation.

Material: Playskin

The Playskin is a soft exoskeletal garment that assists antigravity shoulder movement for children from birth to age 4 yr (Lobo et al., 2016). It uses spring inserts to assist users' arms up to 90° of shoulder flexion, thus increasing the reaching space for object exploration and play (Figure 1).

Study Design

An ABA single-case design was used (for more information on single-case designs, see Lobo et al., 2017). During the baseline phase (A₁; 1 mo duration), performance of baseline reaching and object exploration was assessed; the Playskin was used only during assessments performed every 2 wk. During the intervention phase (B; 4 mo duration), the effects of daily Playskin intervention on reaching and object exploration were assessed. Caregivers were asked to use the Playskin daily for 30 to 45 min in structured intervention activities. Activities encouraged children to reach for objects in larger play spaces; across hip, chest, and eye level; and to the right and left while wearing the Playskin garment (Lobo & Galloway, 2008). Within each intervention session, caregivers were asked to vary the amount of assistance provided by the Playskin by switching among spring inserts that positioned the arms at approximately 30°, 60°, or 90° of shoulder flexion.

During the postintervention phase (A₂; 1 mo duration), carryover effects of the intervention were evaluated. Because of ethical considerations, families were allowed to keep their Playskin, but the prescribed intervention activities were discontinued.

Procedures and Measures

Children were tested in their home environment. Feasibility of intervention was evaluated by caregiver-reported daily intervention times (i.e., number of days per week and minutes per day) and by caregiver responses on a parent perception questionnaire devised for this study (Table 1), collected 2 wk, 2 mo, and 4 mo into the intervention.

This questionnaire used a 5-point rating scale (1 = *very difficult*, 2 = *difficult*, 3 = *neutral*, 4 = *easy*, 5 = *very easy*).

To assess changes in muscle strength across time, shoulder flexion AROM against gravity was measured (in degrees) separately for the right and the left arm at the first visit, at the end of the baseline phase, 2 and 4 mo into the intervention phase, and at the end of the postintervention phase. Maximum AROM was measured from videos taken in the coronal plane (side view) for each participant, while seated on the floor, with the validated two-dimensional motion analysis system Kinovea®

Figure 1. Child wearing the Playskin Lift™ exoskeletal garment during a reaching assessment (A); garment consists of vinyl tunnels under the sleeves, spring support inserts (bottom) that are placed in the tunnels, and circular arm straps to maintain inserts directly under the arms for optimal support (B).

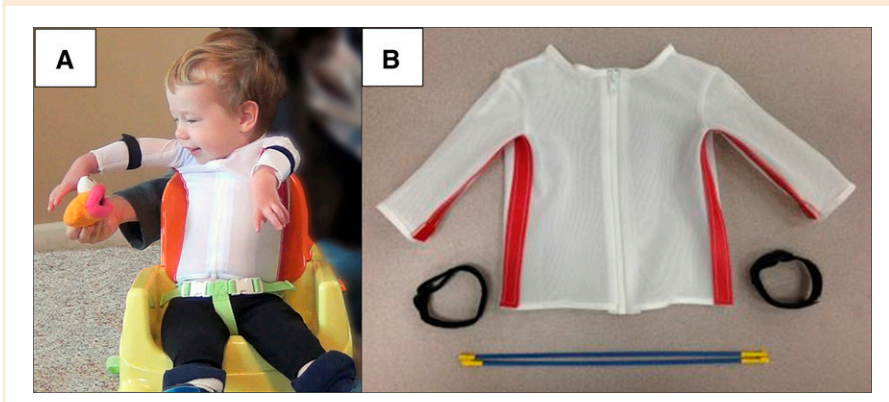


Table 1. Parent Perception Questionnaire Questions and Outcomes

Question	Rating, <i>M</i> (<i>SD</i>)
How easy is the exoskeleton to put on?	4.4 (0.4)
How easy is the exoskeleton to take off?	4.6 (0.4)
How would you rate the user's comfort level in the exoskeleton after 5, 15, and 60 min?	
5 min	4.5 (0.5)
15 min	4.7 (0.4)
60 min	4.5 (0.7)
What is the longest time [in min] the user has tolerated wearing the exoskeleton in one instance?	112.7 (76.4)
How easy is it to take the user places outside of your home while wearing the exoskeleton?	4.2 (0.7)
Within your home, how limited is the user in performing typical daily activities while wearing the exoskeleton?	4.0 (0.4)
How limited is the user's ability to move between positions, like rolling, sitting, crawling, and standing, while wearing the exoskeleton?	4.2 (0.7)
As a parent, how would you rate the overall appearance of the exoskeleton?	3.9 (0.7)

Behaviors (i.e., outcome variables) were coded using OpenSHAPA software (GitHub, San Francisco, CA). Custom software (Filemaker, Version 16, Santa Clara, CA) was used to determine instances of co-occurring behaviors. We coded the behaviors as follows:

- *Unimanual contact*: One hand of child contacts the object.
- *Bimanual contact*: Both hands of child contact the object.
- *Ventral contact*: Ventral side of child's hand contacts the object.
- *Open-handed contact*: Child's fingers extend more than 50% upon object contact.
- *Looking at the toy*: Child's eyes focus on the object.
- *Looking during unimanual contact*: Child's eyes focus on the object while one hand contacts it (multimodal exploration).
- *Looking during bimanual contact*: Child's eyes focus on the object while both hands contact it (multimodal exploration).
- *Bouts of exploration*: Number of transitions from one behavior to another per minute (behavioral intensity).
- *Combined behaviors*: Child uses more than one behavior simultaneously to act on the object (behavioral complexity).

Intra- and interrater reliabilities were established by recoding 20% of the data. After coding five visits, coders recoded one of their own visits for intrarater reliability and coded a visit previously coded by the primary coder for interrater reliability. Agreement within and among coders was calculated using the following equation: $[\text{Agreed}/(\text{Agreed} + \text{Disagreed})] \times 100$. Intra- and interrater agreements were $\geq 85\%$.

Statistical Analyses

The data used in the current analyses are available upon request; privacy and ethical considerations prevent public availability of the data. Results were considered significant at $\alpha \leq .05$ and considered trending toward significance at $\alpha \leq .10$.

Feasibility of the Playskin for Daily Home Intervention

IBM SPSS Statistics (Version 18.0.3; IBM Corp., Armonk, NY) was used for feasibility analyses. Caregiver-reported time spent performing the intervention was averaged across all participants ($M \pm SD$), and caregivers' responses to items on the parent perception questionnaire were evaluated ($M \pm SD$).

(Version 0.8.15; Joan Charmant, <http://www.kinovea.org>).

To assess reaching and object exploration, we tested children every 2 wk with (*on*) and without (*off*) the Playskin in a structured reaching assessment (Babik, Cunha, Moeyaert, et al., 2019). The assessment involved presenting a small (2×4 in.) toy to the child within reach at eye, chest, or hip level (for 60 s/level). For the *on* condition, spring inserts supporting children's arms throughout 70° of shoulder flexion were used. The order of *on* and *off* conditions was alternated at each visit to control for the effects of task familiarity or fatigue. Children sat in a highchair or booster seat, depending on sitting ability, and were assessed only when they were in a positive or neutral mood. A frontal view camera recorded all the assessments.

Assistive Effects of the Playskin on Reaching and Exploration

Hierarchical Linear and Nonlinear Modeling software (HLM; Raudenbush et al., 2004) was used to account for nonindependence of multiple observations per participant. To evaluate differences between children's performance in off versus on conditions, each outcome variable was regressed on the condition variable (0 = off; 1 = on) across three object presentation levels (hips, chest, eyes) for the baseline, intervention, and postintervention study phases.

Rehabilitative Effects of the Playskin on Active Range of Motion, Reaching, and Exploration

AROM data were averaged within each of the following periods: baseline, first 2 mo of intervention, second 2 mo of intervention, and postintervention. Repeated-measures analysis of variance (ANOVA) using IBM SPSS Statistics (Version 25) was conducted to determine whether AROM (separately for the right and left arm) changed across these periods.

Reaching and exploration data analyses were conducted with HLM using off data to evaluate the change in unassisted performance across time. To evaluate change in children's motor function during the study, each outcome variable was regressed on a set of dummy-coded phase variables (B/I, which compares outcomes between the baseline and the intervention phase, and B/PI, which compares the baseline with the postintervention phase, with the baseline serving as the reference).

Results

Feasibility of the Playskin for Daily Home Intervention

Intervention logs showed that the Playskin was used 96.8 ± 81.4 min/day (range = 31.7–285.0) on 5.8 ± 1.2 days/wk (range = 3.8–7.0) throughout the 4-mo intervention phase. Results from the parent perception questionnaire showed caregivers' positive assessment of the Playskin's function, comfort, ease of use, and aesthetics (see Table 1). No adverse effects occurred as a result of using the Playskin.

Assistive Effects of the Playskin on Reaching and Exploration

Summarized observed data comparing infants' performance with and without the Playskin during baseline, intervention, and postintervention are reported in Supplemental Figure 1. Statistical parameters for assistive effects are summarized in Figure 2 and reported in detail in Supplemental Table 1. (To access supplemental content, go to <https://ajot.aota.org>, navigate to the top of this article, and click on "supplemental"). During the baseline phase, significant improvements associated with wearing the Playskin were found in unimanual contact (chest and eye level), ventral contact (chest level), open-handed contact (chest and eye level), looking during unimanual contact (chest and eye level), bouts of exploration (chest and eye level), and combined behaviors (chest and eye level). Marginally significant improvements were identified in bouts of exploration (hip level).

During the intervention phase, significant improvements were observed in unimanual contact (chest level), ventral contact (hip and chest level), open-handed contact (hip and chest level), looking at the toy (eye level), looking during unimanual contact (chest level), bouts of exploration (chest and eye level), and combined behaviors (all levels). Marginally significant improvements were found in unimanual contact (hip level) and bouts of exploration (hip level).

During the postintervention phase, significant improvements continued in unimanual contact (eye level), bimanual contact (chest level), ventral contact (chest level), open-handed contact (chest and eye level), looking at the toy (eye level), looking during unimanual contact (eye level), looking during bimanual contact (chest level), bouts of exploration (eye level), and combined behaviors (chest and eye level). Marginally significant improvements were observed in ventral contact (hip and eye level) and bouts of exploration (hip level).

Rehabilitative Effects of the Playskin on Active Range of Motion, Reaching, and Exploration

Across all participants, maximum independent AROM against gravity in sitting at the first visit was $45.6 \pm 9.7^\circ$ for the right arm and $32.4 \pm 9.1^\circ$ for the left arm. The average change ($M \pm$ standard error [SE]) in AROM throughout the study

Figure 2. Assistive and rehabilitative effects of the Playskin Lift™ on the outcome variables (i.e., behaviors) at hip, chest, and eye level.

Behaviors	Assistive Effects								
	Baseline			Intervention			Postintervention		
	Hips	Chest	Eyes	Hips	Chest	Eyes	Hips	Chest	Eyes
Unimanual contact									
Bimanual contact									
Ventral contact									
Open-handed contact									
Looking at the toy									
Looking during unimanual contact									
Looking during bimanual contact									
Bouts of exploration									
Combined behaviors									

Behaviors	Rehabilitative Effects					
	Baseline to Intervention			Baseline to Postintervention		
	Hips	Chest	Eyes	Hips	Chest	Eyes
Unimanual contact						
Bimanual contact						
Ventral contact						
Open-handed contact						
Looking at the toy						
Looking during unimanual contact						
Looking during bimanual contact						
Bouts of exploration						
Combined behaviors						

Assistive effects compare *on* and *off* performance during the baseline, intervention, and postintervention phases. Rehabilitative effects compare change in mean performance from baseline to intervention and from baseline to postintervention. Black cells = significant effects ($p \leq .05$), gray cells = marginally significant effects ($p \leq .10$), and white cells = no significant effect.

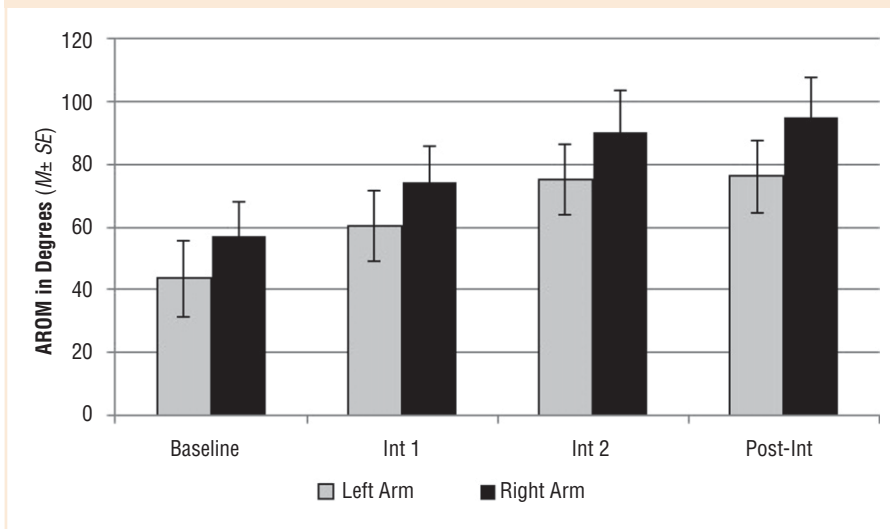
is reported in Figure 3. For the right and left arms, repeated-measures ANOVA with a Greenhouse–Geisser correction for nonsphericity showed a statistically significant change in AROM throughout the study (right: $F[1.3, 14.4] = 35.70$, $p < .0001$, $\eta_p^2 = 0.76$; left: $F[1.1, 12.3] = 24.92$, $p < .0001$, $\eta_p^2 = 0.69$). Bonferroni-corrected post hoc comparisons revealed an increase throughout the first 2 mo of intervention (right: $p = .007$; left: $p = .001$), the second 2 mo of intervention (right: $p < .001$; left: $p = .002$), and the postintervention (right: $p < .0001$; left: $p = .002$), compared with baseline. There was an increase in AROM from the first to the second half of intervention (right: $p < .0001$; left: $p = .002$), but no change during the postintervention (right: $p = .169$; left: $p = .999$).

Statistical parameters for rehabilitative effects are summarized in Figure 2 and reported in detail in Supplemental Table 2. During the intervention phase, compared with baseline, children’s independent performance improved significantly for unimanual contact (all levels), ventral contact (chest and eye level), open-handed contact (all levels), looking during unimanual contact (hip and eye level), bouts of exploration (hip level), and combined behaviors (hip and chest level). Improvements in children’s independent performance were also observed during the postintervention phase, compared with baseline, for unimanual contact (all levels), ventral contact (all levels), open-handed contact (all levels), looking at the toy (chest level), looking during unimanual contact (all levels), bouts of exploration (all levels), and combined behaviors (all levels). No improvements were found for bimanual contact or looking during bimanual contact.

Discussion

The goal of this study was to evaluate the assistive and rehabilitative effects of the Playskin exoskeletal garment on reaching and object exploration in young children with arthrogryposis and to assess the feasibility of parent-guided home intervention with the Playskin.

Figure 3. Degree of independent active range of shoulder flexion from baseline to postintervention.



Note. AROM = active range of motion; Int1 = first 2 mo of intervention; Int2 = second 2 mo of intervention; Post-Int = postintervention. Error bars indicate standard error (SE).

Feasibility of the Playskin for Daily Home Intervention

According to caregivers, the Playskin was used at a level that often considerably exceeded the 30 to 45 min/day that parents were requested to perform. The level of intervention performance reported in the current study was much greater than that previously reported in research using the Playskin for intervention with infants born preterm, some with a brain injury (Babik, Cunha, Moeyaert, et al., 2019). This difference might reflect a higher level of motivation for Playskin use by parents of children with arthrogryposis because these children have more significant, observable limitations in their arm movement and function.

According to parental ratings, the Playskin was very easy to don and doff with a high level of comfort even through 1 hr of wear. Although some children used the Playskin for a combined total of 5 hr per day, the longest single continuous bout of wear reported was about 3 hr. For some children, especially at the beginning of the study, the Playskin afforded them their only opportunity to play with toys, so parents allowed their children to wear the Playskin for prolonged periods of time. Moreover, parents reported that the Playskin did not restrict their children's daily activities or transitions among postures. Parents found the Playskin to be attractive and reported use outside of their homes. Overall, the Playskin was easy to use, comfortable, aesthetically pleasing, and feasible for families to incorporate into their daily lives.

Assistive Effects of the Playskin on Reaching and Exploration

At the beginning of the study, many participants were able to reach for objects presented at hip level, but not at chest or eye level. Therefore, during the baseline phase while wearing the Playskin, no significant improvements were found in performance at hip level. In contrast, the Playskin significantly improved children's unimanual reaching toward objects presented at chest and eye level, expanding their reaching space into areas that enable better visual-manual coordination, which might further improve children's manual abilities as they observe and learn from their actions with objects (Corbetta & Snapp-Childs, 2009; McCarty & Ashmead, 1999; Petkovic et al., 2016). Indeed, wearing the Playskin facilitated children's visual-manual coupling, allowing enriched multimodal object exploration (Bahrick et al., 2004; Corbetta & Snapp-Childs, 2009; Gibson, 1988; Wilcox et al., 2007).

Interestingly, providing support proximally to children's shoulders resulted in several changes distally in hand function, including improved ability to make contact with objects with the ventral side of the hand and with the hand open. These grasping changes enable improved manual exploration, manipulation, and information gathering with objects (Lobo & Galloway, 2013; Needham et al., 2002). Furthermore, wearing the Playskin allowed children to perform more complex, multimodal behavior and to increase the intensity of their object exploration, changes that would be expected to further advance cognitive development (Lobo et al., 2015).

The same beneficial effects of the Playskin on children's reaching space, hand position while grasping objects, visual-manual coupling, multimodality, and intensity of object exploration were also observed during the intervention

and postintervention phases. Note that during the postintervention phase, while wearing the Playskin, children also showed significantly more bimanual reaching at chest level, which creates opportunities for more sophisticated object manipulation (Babik & Michel, 2016). Bimanual behavior with objects typically involves hand differentiation; for example, one hand may support an object while the other feels and manipulates its parts. Therefore, this activity informs children on how to plan complex, coordinated actions between their hands. It also provides them with opportunities to interact with more than one object to learn about important concepts, such as in and out and cause and effect, that are imperative for children's language and cognitive development (Bonawitz et al., 2010; Iverson, 2010; Kimmerle et al., 2010).

Rehabilitative Effects of the Playskin on Reaching and Exploration

During the intervention phase, children improved unassisted, independent shoulder AROM against gravity; unimanual reaching; visual-manual coupling; and the ability to make contact with objects with the ventral side of the open hand. They also increased the complexity and intensity of behavior at the hip and chest levels. These findings suggest that a 4-mo-long intervention with the Playskin improved independent reaching and object exploration for children with arthrogryposis. More important, these improvements persisted into the postintervention phase. Throughout the study, all children showed improvements in active range of shoulder flexion, which allowed them to increase their reaching and play space. These results suggest that parent-guided, play-based interventions using tools such as the Playskin in children's natural environments may positively affect the development of reaching and object exploration in children with significant muscular weakness.

Study Limitations

One confounding factor in studies with pediatric populations is maturation. It might be hypothesized that the improvements in children's reaching and object exploration behaviors in this study could be attributed to maturation rather than intervention with the Playskin. The study design aimed to strengthen our ability to infer causal effects of the intervention on the outcome measures. First, performance during the intervention and postintervention phases was compared with the baseline level of performance to account for each child's typical developmental level. Second, the intervention effects were tested across multiple participants ($N = 17$). It is important to note that children were enrolled in this study at the ages of 6 to 35 mo, many with significantly limited manual abilities, whereas the typical age of reaching onset is 3 to 5 mo (Michel & Harkins, 1986) and that of complex object manipulation is 6–12 mo (Kimmerle et al., 2010). Significant positive changes from the baseline to intervention and postintervention phases across multiple participants suggest that the observed improvements can be attributed to the intervention with the Playskin. The replication of these positive effects across multiple participants who entered the study at varying ages, well after the behaviors assessed would be expected to emerge, further supports this argument.

Implications for Occupational Therapy Practice

This study has the following implications for occupational therapy practice:

- The Playskin can be an effective assistive and rehabilitative device to improve reaching and object exploration behaviors in children with muscular weakness.
- By advancing children's reaching and object exploration behaviors, the Playskin might potentially improve their motor and cognitive development, self-care efficacy, and quality of life (Bornstein et al., 2013; Jouen & Molina, 2005; Nelson, 1999).
- Future research should test the Playskin in other populations of children with muscular weakness.

Conclusion

This longitudinal study is the first to test the feasibility and effectiveness of intervention with the Playskin garment to improve reaching and object exploration in young children with arthrogryposis. The results provide important, novel information suggesting that the Playskin is an easy-to-use, comfortable, and aesthetically appealing garment that is feasible for parent-guided home-based intervention in children younger than age 4 yr. ■

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