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Article

FEASIBILITY OF A HOME MOTOR SKILL INTERVENTION FOR CHILDREN WITH CHARGE SYNDROME

Lauren J. Lieberman¹

Pamela Beach¹

Melanie Perreault¹

Ali Brian²

Luise Bebrisa-Fedotova²

¹State University of New York at Brockport

² University of South Carolina

Abstract: This study examined the effect of a home-based six-week motor skill intervention for children with CHARGE syndrome. Six families with children with CHARGE syndrome participated in this study. The children were assessed on their motor skills with the Test of Gross Motor Development-3 prior to the intervention and then again on the four most practiced motor skills after the intervention. Parents were also given a Child Movement Skills Research: Parent Questionnaire before and post intervention. The families were provided a Gross Motor Development Curriculum, videos, tip sheets, and equipment to support the intervention. Families maintained weekly logs of their specific training practices. Each family was interviewed, and the transcripts were analyzed for themes. The four themes that emerged were Personal goals, Improvement, Barriers, and Overcoming challenges. Results revealed that it is feasible to utilize this protocol to improve motor skills for children with CHARGE syndrome.

Keywords: *assessment, disability, visual impairment, deaf-blind, deafness, parent perceptions, motor development*

Feasibility of a Home Motor Skill Intervention for Children with CHARGE Syndrome

CHARGE syndrome refers to coloboma, heart defects, atresia choanae, growth restriction, genital abnormalities, and ear abnormalities (Blake et al., 2006). CHARGE syndrome encompasses multiple disabilities that typically affects more than one part of the body and most often multiple body systems (Blake et al., 2006). CHARGE syndrome generally affects one in every 10,000 births and occurs when a gene mutation presents itself during prenatal development (Thelin et al., 2006). Since CHARGE syndrome affects both hearing and vision, many children are identified as deaf-blind, which is one of the 13 disability categories under the Individuals with Disabilities Education improvement Act (IDEA, 2004). Due to the various medical complications inherent within CHARGE, approximately 40% of children will not develop functional communication skills and many more may exhibit difficulties with speech and oral communication (Thelin et al., 2006). The name CHARGE was from the characteristics of Coloboma of the eye, Heart defects, Atresia of the choanae, Retardation of growth and development, and Ear abnormalities and deafness. Currently, a diagnosis of CHARGE is made without the child necessarily demonstrating all of the traits.

In addition to the unique various medical characteristics, children with CHARGE syndrome often encounter motor skill delays impacting their flexibility, balance, and hand-eye coordination (Haibach-Beach et al., 2019; Haibach & Lieberman, 2013; Imel et al., 2020; Lalani et al., 2006; Möller et al., 2011; Perreault et al., 2020). For example, age of walking for children with CHARGE syndrome often occurs 14 months later than children with Usher syndrome, the second leading genetic cause of deaf-blindness, and approximately 25 months later than children without disabilities (Dammeyer, 2012). Age of walking is a critical underlying mechanism supporting locomotor skill development (Haibach-Beach et al., 2020). In addition, age of onset of walking is closely connected with other significant developmental outcomes such as sleep difficulties (Hartshorne, et al., 2009), adaptive behaviors (Salem-Hartshorne & Jacob, 2005), language delays (Dammeyer, 2012), and medications (Wachtel et al., 2007). Thus, it is critical to ensure stakeholders provide children with CHARGE syndrome a solid gross motor skill foundation during early development.

During early childhood, a solid gross motor foundation must include fundamental motor skills. According to Clark and Metcalfe (2002), fundamental motor skills are the foundational components of specific sport skills and are essential in promoting lifelong physical activity. Children with low competence in gross motor skills tend to choose a less physically active lifestyle partially due to avoidance of movement challenges (Wrotniak et al., 2006). There is also a relationship between limited motor development and language development which can influence social skills (Houwen et al., 2016). Unfortunately, limited involvement in physical activity during childhood will often lead to physical inactivity in adolescence and throughout adulthood (Telama, 2009). For these reasons, early opportunities and involvement in motor skill development, and therefore a physically active lifestyle, is critical. Moreover, fundamental motor skills are the foundation of recreational programming for all children including those who are deaf-blind (Lieberman et al., 2018). Without acquiring competence in fundamental motor skills, children with CHARGE syndrome may have difficulty participating in free time recreational

activities when they transition into adolescence and adulthood (Imel et al., 2020; Štěrbová et al., 2014). This also impacts social skills.

Difficulties with motor skill development are not surprising due to medical complications and extended hospitalizations experienced by children with CHARGE syndrome (Hartshorne et al., 2011; Salem-Hartshorne & Jacob, 2005). Medical complications necessitate multiple surgeries that include frequent hospitalizations interfering with natural development in motor milestones and educational programming. Even when children with CHARGE syndrome return from their medical leave, they are typically excused from physical education for long periods of time (Hilgenbrinck et al., 2020). Extended absences from physical education and skill development opportunities may greatly affect the gross motor development of children with CHARGE syndrome if they are not afforded individualized and adapted services and programs. Thus, interventions that focus on fundamental motor skills are warranted and necessary to steer children with CHARGE syndrome towards a positive developmental trajectory for health.

Currently, early intervention programs tend to include parents as a key part of the process; however, these programs typically focus on the improvement of communication skills with much less attention on motor skill development (Bremer et al., 2015). Interventions addressing the motor skill development of children with disabilities, particularly those including parents as facilitators, are limited at best (Colombo-Dougovito & Block, 2019; Hurley & Burt, 2015). In one study, Altunsöz et al. (2016) compared a Head Start intervention program with children with developmental delays and a parental supported home intervention program with a control group that received no intervention. Both intervention groups improved their locomotor skills in comparison to a control group. Interestingly, there were no differences with the parental support condition. In another study, Hamilton et al. (1999) demonstrated that children with developmental delays who received additional parental support (in combination with teacher instruction) did better than the students who only received instruction from their teachers. This is important because research has shown that although children with autism may participate in physical education at school, in some instances, the fundamental motor skills they acquire at school are not generalizing to the home environment and community recreation facilities (An & Goodwin, 2007; Chaapel et al., 2013). Lastly, in a large parent intervention by Scully et al. (2019), physical activity packs were delivered to 53 children, aged 5–15 years, with a visual impairment with 63% with additional disabilities. The activity packs included equipment and instructions and the intervention was three weeks long. Participants' experiences of physical activity prior to receiving this pack and after the intervention were documented. The study revealed inequitable experiences of physical activity related to limited opportunities, inaccessible programs, and discrimination due to the child's visual impairment. The First Steps pack helped to decrease these barriers which led to increased physical activity levels, perceived improvements in gross motor skills, and developments in children's social skills. In addition, the children's motor skills, social interactions, and confidence improved.

Parents should be part of the intervention process as their perceptions related to motor skills can play a critical role in the motor competence of their children with visual impairments or deaf-blindness (Authors, Under Review; Imel et al., 2020). As early as birth, parents are key players in a child's development (Harter, 1987; Reed, 1991). Parents create environments for

their children to engage with and practice motor tasks (Silva et al., 2017). Additionally, parents also provide toys and equipment in their immediate environment that improves competence in motor skills (Barnett et al., 2013; Cools et al., 2011). Parents may be a more critical component of their children's motor development if their child has a disability. For example, parents of children with visual impairments promote elevated levels of encouragement to their children by supporting them to be active and providing transportation and financial assistance (Linsenbiger et al., 2018). Alternatively, overprotecting their children from getting hurt may be a barrier to motor skill development (Lieberman et al., 2006). A study by McKittrick (2019) revealed valuable insights into the various and vital roles that parents of children who are deaf-blind play on their child's IEP teams. These include case management, advocacy, and knowledge sharing with team members.

Unfortunately, there is no intervention research (with or without the inclusion of parents) on motor development in children with CHARGE syndrome from which to create an evidence-based intervention (Hurley & Burt, 2015). As a result, we must first test the feasibility of a home-based intervention (Osmund & Cohn, 2015). A feasibility study is an assessment of the practicality of a proposed project with the intentional purpose to thoroughly understand all aspects of a project, so that we may become aware of any potential problems that could occur while implementing the project (Kenton, 2019). Moreover, feasibility trials must occur to determine if, after considering all significant factors, the project is viable and worth scaling up in future trials (Kenton, 2019). Thus, the purpose of this study was to examine the feasibility of a home-based gross motor program for children with CHARGE syndrome. The intervention feasibility indicators included 1) adherence to the intervention, 2) participant acceptability, and 3) intervention efficacy. We hypothesized that the majority of parents of children with CHARGE syndrome would adhere to the intervention protocols, families would be committed to the intervention, and that children would improve their motor skills as parents change their perceptions of their children's motor skill competence.

Methods

Participants and Setting

Participants included a convenience sample of children ($N = 6$; $M_{age} = 5.8$, $SD = 1.9$) and their parents who attended an International CHARGE Syndrome Foundation Conference. Pseudonyms were used throughout to retain confidentiality. All children could independently walk without assistance and possessed documentation of CHARGE syndrome. Participants revealed various degrees of severity regarding each of the six characteristics of CHARGE syndrome. See Table 1 for the overview of each child. Four of the participants communicated verbally and two of the participants communicated through American Sign Language.

Table 1*Demographic Information of Participants*

Sex	Age	Vision	Hearing	Semicircular canals	Atresia of Choanae	Heart defects	Growth Delay	# of surgeries	Age of independent walking
F	3	Low vision, B4	Right ear moderate, left ear severe	Left partially formed	Yes	No	Yes	7	24 months
F	6	B3 in both eyes	Both ears profound	Both partially formed	Yes	Yes	Yes	8	48 months
F	5	B3 in both eyes; bilateral optic nerve colobomas	Right ear normal; left ear profound	Both partially formed	No	Yes	Yes	10	24 months
F	6	Bilateral colobomas; Missing upper field; Right eye B3 and Left eye B4	Right ear profound; left ear moderate	Missing both	Yes	Yes	Yes	11	40 months
F	6	Nystagmus, colobomas and B3 in both eyes	Right ear severe; left ear is profound	Missing both	Yes	Yes	Yes	22	27 months
M	9	B3 in both eyes	Both ears moderately severe	Fully developed	Yes	No	Yes	5	24 months

Instrumentation***Test of Gross Motor Development, Third Edition (TGMD-3)***

The TGMD-3 is a process-based measure of 13 gross motor skills that are divided into two subtests: locomotor skills and ball skills. Locomotor skills require moving the body from one location to another (run, hop, skip, gallop, slide, jump) and ball skills include the handling of an object via force (overhand throw, kick, two-hand strike, underhand roll), interception (catch), or both (dribble, one-hand strike). Participants performed two trials of each skill and were scored on each trial based on the presence of specific criteria (i.e., 1 = criteria present; 0 = criteria absent). Each skill is assessed on three or four criteria, with a maximum score between six and eight for each motor skill. A trained researcher in the TGMD-3 assessed all of the performances from digital recordings. An expert in motor development assessment trained the researcher. The researcher and the trainer practiced analyzing the videos until reliability reached over 85%. The TGMD-3 shows robust psychometric properties in children ages 3 to 10 years, 11 months who are typically developing (Webster & Ulrich, 2017) and children up to age 18 years who are visually impaired (Brian et al., 2018).

Parent Perceptions Questionnaire

The Child Movement Skills Research: Parent Questionnaire was used to examine parent perceptions (Clancy et al., 2017; Jackson et al., 2012). This questionnaire includes 32 items and

is subdivided into three sections. The first section examines parental views on their child's motor abilities in 12 of the motor skills the children were assessed in the TGMD-3 in comparison to their perception of their child's skills related to same aged peers. These questions were rated using a 5-point Likert scale ranging from 1 = no confidence to 5 = complete confidence. The second section examined the importance of motor skills from the parents' view and how likely their child or other individuals are able to change their abilities with scores ranging on a 7-point Likert scale from 1 = strongly disagree to 7 = strongly agree. The third section examined parental support of their child in regard to developing and promoting their motor skills with scores ranging on a 5-point Likert scale from 1 = never or rarely to 5 = daily or always. A reliability analysis revealed excellent internal consistency for each set of items ($\alpha = .83 - .96$) except those measuring if parents feel that people in general can do much to improve their motor competence ($\alpha = .039$); thus, these items were removed from the analysis.

Intervention Programming

Families were provided a variety of tools to help with the home intervention that included curricula, videos, tip sheets, and equipment. Families were also provided with an e-book, *The Gross Motor Development Curriculum for Children with Visual Impairments* (Lieberman & Haibach, 2016). This manual provides descriptions of object control skills, such as batting, kicking, throwing, underhand roll and catching, and locomotor skills, such as running, skipping, sliding, leaping, jumping, and hopping. The curriculum manual describes the steps for performing these skills as well as a variety of strategies and specific modifications to teach children with visual impairments or deaf-blindness. Participants were also provided an accompanying video that parallels the content in the manual. The Gross Motor Development video is offered through the American Printing House for the Blind and on the Camp Abilities website (Camp Abilities N.D.). Additionally, participants were provided with tip sheets that provided a snapshot of the components of each of the locomotor and object control skills as well as the most commonly used modifications, cue words, and strategies. Each family was also provided three colorful plush balls and a bell kick ball. All materials and equipment were used as part of the intervention.

Procedures

Prior to the selection of the participants, the lead investigator's Institutional Review Board approved all procedures. Parents/guardians provided written consent prior to the start of the research. Out of the 14 families that were initially interested in participating in the study, six agreed to allow their child to be assessed and then to utilize the intervention materials and implement a six-week gross motor skill home intervention. First, parents completed a survey about their child's demographics and CHARGE syndrome characteristics and the Parent Perceptions Questionnaire. Then, members of the research staff assessed the motor skills of the children using the TGMD-3 and video-recorded all trials for scoring purposes. When needed, modifications made during the TGMD-3 included multiple demonstrations, sound source or clapping for locomotor skills, and a bell ball for catching. Children were communicated with using their preferred mode, which included American Sign Language for some children. The research staff consisted of four professors with expertise in working with children with

CHARGE syndrome, two were motor development specialists and two were adapted physical education specialists. The team also included four research assistants who had expertise in kinesiology and adapted physical education. Two of the researchers were fluent in sign language. After each child completed the TGMD-3, the parents were provided with materials and equipment to implement the home-based intervention.

Afterwards, parents conducted a six-week gross motor skill intervention program in their homes with their child with CHARGE syndrome utilizing the materials described above. Parents also completed a log which included the dates, durations, activities, equipment used per practice session, and a comment section for how it went each day. The researchers sent regular weekly email reminders to continue practicing and logging activities. Participants were also offered opportunities to ask questions to the researchers in regard to any of the activities or modifications. Most families worked on four main gross motor skills. These motor skills were ones that the parents felt the child would use most often, and also ones that they felt they knew how to teach comfortably. Following the intervention, parents were instructed to video record their child performing two trials of the four motor skills they worked on the most during the intervention and send it to the researchers to be scored based on the TGMD-3. Motor skills were chosen by the researchers based upon the four most frequently reported activities practiced according to their logs. Parents also completed the Parent Perceptions Questionnaire electronically post intervention. Please see Figure 1 for the overview of the feasibility process.

Feasibility Measures

Intervention Adherence

To assess intervention adherence, families were instructed to record every practice session (including activity type and duration) into activity logs. Families were instructed to practice two times per week for a minimum of 30 minutes each practice session for six weeks. All activities were considered safe activities, but families were recommended to report any accidents or injuries to the researchers.

Intervention Acceptability

Following the entire intervention, researchers set up phone interviews with a parent to examine the acceptability of the home intervention. During the interviews, parents were asked about facilitators of the home intervention, use of the materials and equipment provided, their intentions to continue working with their child on gross motor skills, and whether they would recommend the home intervention to other families of children with CHARGE syndrome. Parents were also asked about the activity log and their participation in the study. Two researchers were present during each interview to ask the questions and record information.

Researchers developed the interview guide with the advice of a panel of six experts. The experts included four professionals in the fields of adapted physical education and motor behavior with experience with deaf-blindness and CHARGE syndrome, one physical education teacher, and one parent of a child with CHARGE syndrome. The interview guide helped ensure that the line of questioning for the qualitative portion of this study was similar. Interviews lasted 15 to 45

minutes depending upon the duration of the responses. Interviewers took reflective notes during the interviews and then met to debrief after each interview. Research staff audio recorded all interviews and transcribed them verbatim. The interview data was coupled with the logs to ensure triangulation of the data. Data were analyzed for clarity of the training protocol.

Sample interview questions:

- Can you please explain to us how you used the motor development assessment for planning your intervention with your child?
- Did you look at or use any of the gross motor assessment curriculum that we sent you of the tip sheets or videos? Were they helpful?
- How often did you have the opportunity to work with your child on a typical week during this intervention?
- Who worked with your child on their motor skill intervention?
- What roles did each person play?
- What did you use the most? (Tip sheets, videos, book, balls, etc.)

Intervention Efficacy

Intervention efficacy was determined by assessing changes in the four most practiced motor skills of the child with CHARGE syndrome. In addition, changes in the parent's perceptions of their child's motor skill abilities from pre- to post-intervention were analyzed.

Analysis

Descriptive statistics were used to report the profiles of the participants. Measures of attendance and adherence were reported as frequency distributions. To analyze intervention adherence, members of the research staff analyzed and descriptively reported data from the parent's logs.

To assess intervention acceptability, the audio recordings of the parent interviews were transcribed verbatim by the researchers. Two of the researchers independently conducted a thematic line-by-line analysis of interview data with themes and subthemes of various aspects of the intervention. Once these themes and subthemes were created, the researchers compared their findings with each other to ensure each theme was included (Vaismoradi et al., 2016). The data was then reviewed and revised with the focus on the main themes with supporting quotes until consensus was reached between the researchers. A "critical friend" was included once the initial themes and supporting quotes were developed to ensure the data matched what was said in the interviews. The recommendations from the "critical friend" were reviewed and added upon consensus of all parties (Coughlan et al., 2014).

To assess changes in TGMD-3 skills before and after the six-week intervention, members of the research staff hand calculated Cohen's *d* effect sizes and reported all data descriptively. Cohen's *d* effect size interpretation includes .03 = small, .05 = moderate, and .08 = large (Cohen, 1986). Due to the lack of statistical power from the small sample, we were unable to conduct any inferential statistics.

Results

Six families of children with CHARGE syndrome recruited from the International CHARGE Syndrome conference participated in this study. From this sample, five parents participated in interviews, four submitted activity logs, and four completed post-testing assessments. Descriptive data on the children with CHARGE syndrome is included in Table 1. In this section we will review: (a) adherence to the intervention, (b) intervention acceptability, and (b) intervention efficacy.

Intervention Adherence

According to the logs, each parent adhered to the recommended dosage of six weeks, twice a week for 30 minutes each, and practiced locomotor and object control skills from the TGMD-3. See Table 2 for further description of total log activity. The most practiced activities were those that could easily be transformed into a game, sport, or activity. Table 3 provides a summary of activities, games played, equipment, resources, and who facilitated the intervention.

Table 2

Logs of the Activities and Time Spent in Each one

Skill	Frequency (n)	Time (mins)
Overhand throw	16	430
Kicking	15	445
Hopping	15	380
Underhand throw	12	335
Jumping	10	225
Sliding	9	270
Catching	7	225
Two hand strike	7	185
Galloping	4	120
Skipping	4	135
One hand strike	2	20
Run	2	32

Table 3*Log Reports Summary*

Parent	Equipment	Skills	Activities	Resources	Support
Mother of Kiera	squishy balls kickball bell ball goals tee ball Beanbag	Overhand throw Kicking Two hand strike Hopping Sliding Jumping	Threw small ball against hose, kick in a "Goal", two hand strike of the tee, hopping and sliding like dance moves with music, kicking ball with sister, jumping into beanbag	Tip sheets, Music	With older sister and herself
Mother of Latisha	Tee ball set Balls	underhand throw overhand throw Catching Kicking Two hand strike	Hot potato, catch Toss, cornhole Soccer into net, Tee ball	Tip sheets Music YouTube videos, Games, Stickers	Mostly herself, husband was there for 3-4 sessions
Mother of Sienna	Bean bags, Plastic clubs, Large rings, large balls, laundry basket, soft balls Elastic strips Tee ball set Bowling pins	Jumping Hopping Underhand throw Sliding one hand strike Overhand throw Gallop Kicking Skipping Catching Run Two hand strike	Obstacle course, Golfing Bean bag toss Kicking and throwing a ball changing distance, Bowling	Tip sheets Videos Colors sheet	Just herself
Mother of Clara	Exercise balls	overhand throw Hopping Skipping Kicking Jumping Running			Chad, private PT, school PT assistant, Two PE instructors, her intervener & herself

Intervention Acceptability

According to the interviews, parents reported intervention acceptance when discussing the activities, modifications, and interests of their child participating. The four themes that emerged from the parent interviews were: (a) I just want her to do anything she can do: Personal goals, (b)

Gains confidence every time: Improvement, (c) I don't know enough: Barriers, and (d) It is just play time: Overcoming challenges.

I just want her to do anything she can do: Personal goals

Many of the parents shared how they approached the intervention focusing on the personal, physical, and social goals for their children. Related to gait, the mother of Latisha, stated "I was really hoping to see her gait kind of adjust and change to be more typical, and it actually did. I'm totally contributing it to the study." This same mother said

She [Latisha] had her reevaluation with her therapist last week, and now we moved down to every other week instead of weekly because her gait has really come in. It's more stable. She is now stepping up by herself onto like a curb-sized step, jumping with both feet independently (Latisha's mother).

Kiera's mother envisioned her daughter doing typical activities that her peers do. She stated: I just want her to do anything that she can do. If any kid can do it, then I want her to be able to do it too. So, if she wants to run a marathon. So, she has a bike right now and she might bike, and I want her to be able to bike by herself someday, you know.

Gains confidence every time: Improvement

The second theme encompassed notions of improvement. All of the families wanted their child to improve on their motor skills, stamina, and balance.

Bradley's mother stated

Whatever you are doing, it is helping because he (Bradley) has picked up a lot of the gross motor skills that they had been working on for his goals and he kind of achieves everything they were working on in that.

The idea that the parents wanted the children to thrive in life and not allow the medical complications to hold them back was evident. Kiera's mother shared

I like the idea of an intervention to focus on something that we can do and that is something beyond medical because a lot of stuff the CHARGE kids deal with is very doctor orientated. I like that this was something that was more play. It was physical and it was important to do. It was fun and just a whole different ball game in my mind as far as something that we can work on together. Like we can sit down and work on your letters or eating, but this was like, hey, let's go outside and kick a ball. This will be huge. I don't know, it was kind of like a lightbulb, I guess.

The fact that gross motor development is not a major focus area for stakeholders within the esoteric world surrounding CHARGE syndrome was stated by several parents. They felt that this was an area that professionals and the community should focus on more often. Sienna's mother shared

It is something that they do not focus on a lot in CHARGE. It's hearing, vision, hearing, vision, oh yeah, they have low tone. It's just not focused on a lot, so it was kind of nice

and kind of refreshing to see a little study on that. I would say honestly, I have never seen an intervention other than walking. I thought it was kind of refreshing to enjoy and interesting something that you don't often see, and in terms of CHARGE Syndrome.

Clara's mother said "She gains confidence every time she is able to successfully do something. You build enough successes, then it's not such a battle or struggle."

The increase in motor skill performance was pronounced when two of the participants were released from physical therapy and one decreased his hours per week in physical therapy. Latisha's mother, shared "...it was interesting to see all of the gross motor work together to help every other skill that she needs." She and two other parents shared their relief that their children can now run on different surfaces more easily. Latisha's mother said "...she's comfortable on dynamic surfaces ... she'll go in grass and granted it's more of a slower run, but she does feel more confident moving on different surfaces and that was worth it in and of itself, just to see her be able to keep up with her peers"

Several parents said they were reluctant to work on catching due to difficulties with vision. However, parents were willing to try catching after they received the modified balls. Latisha's mother stated, "I would say that she went from catching every one out of five throws to fifty percent of the throws." Other areas that parents worked on were: running on different surfaces, catching (improvement from 20%-50% improvement on self-report), jumping, kicking (3), striking (2), rolling (2), sliding, throwing, hopping, and skipping. The least practiced skills were one hand striking, two hand striking, kicking, and hopping.

I don't know enough: Barriers

The major barriers to the motor skill intervention were the child's vision, attention span, time of year, being inside, time, balance issues, strength, and level of comfort of the child with some of the activities. Some of the parents said they did not know how to teach motor skills. Sienna's mother said, "Galloping, I have a hard time teaching how to do that. I just can't break that down well enough. I don't know enough of it." Not knowing how to teach some of the skills was a reason why some parents taught more of one skill or not another.

Some of the parents were very creative, such as Clara's mother who said, "...she is much more willing to try if she had something that made her feel safe, like a wall for hopping." Latisha's mother claimed that her daughter did not want any modifications and that was a barrier. "...the biggest thing for her is, she doesn't like modification, she likes to do what the big kids are doing." Not knowing how to teach the skills to their child and the child not wanting any modifications can be definite impediments to learning.

It is just play time: Overcoming challenges

This particular group of parents of children with CHARGE syndrome were resilient and very creative in overcoming challenges to instructing motor skills for their children. For example, the parents figured out positioning related to the child's muscle imbalances and vision. Parents also

created a variety of modifications that facilitated success with motor skills. For example, modifications ranged from larger equipment, to cueing attention, and changing distances of kicking and catching.

Some facilitators that worked for most of the children were making the skill development activities into games and making it fun. Latisha's mother stated, "If we could find a way to make it fun, she was down for it". Kiera's mother said "...we just did the hopping and sliding as like a dance move and we would have the music on, and she loves music." Kiera's mother also mentioned that they "...would set up goals in the backyard and be like okay let's see if you can kick it through here", that's what made activities more enjoyable.

Bradley's mother on recommending this program "I think it helps them (the child- Bradley) understand more why gross motor skills are so important. Also, learning the different activities that you could do and then realizing that it is just play time, it is not really a chore."

The parents were able to create positive reinforcers, such as music, stickers, siblings (for motivation), encouragement, and verbal cuing. Related to sibling motivation, Kiera's mother stated "The things she did the best would be the days that her sister was there with her and just to make it more fun.... She was like, this is cool and all, but it was way more fun when she had big sister helping her." Related to positive reinforcement, Kiera's mother stated "we made it a big deal if she could kick it through the goal".

All of the families said that repetition and practice were necessary to see improvement. Many of the parents had to give their children breaks due to issues with stamina and attention. Many also had to decrease the distance for kicking, throwing, and catching to help with vision issues and attention. As far as instructional strategies many parents used adapted balls, physical guidance, and verbal cues. The training materials that helped most were the training videos, the tip sheets, and the modified balls. Kiera's mother said "We weren't just modifying it for her to be able to do it, but modifying it to make it more interesting"

For the five families who implemented this gross motor development program, it appears that the resources were helpful in improving motor skills and they really enjoyed the process. Although parents gave recommendations for improving the program, the overall feeling was that the parents would recommend the intervention to other parents of children with CHARGE syndrome.

Intervention efficacy

Motor skill performance

Of the six participants, four participants completed both the pre- and post- motor skill assessments. Participants completed four pre-selected motor skill assessments chosen by the researchers based upon the frequency of the motor skills practiced according to the activity logs. Across these four participants, there were 15 pre- and post-intervention scores for eight motor skills from the TGMD-3: hop ($n = 1$), jump ($n = 1$), slide ($n = 1$), two-hand strike ($n = 1$), catch ($n = 1$), kick ($n = 4$), overhand throw ($n = 4$), and underhand toss ($n = 2$). More than half of the

scores increased from pre- to post-intervention ($n = 10$) while the others either remained stable ($n = 3$) or decreased ($n = 2$). See Table 4 and Figure 1. Improvement in skill scores ranged between 12.5% and 50% ($d = .89 - 2.29$). Across participants, the skill that each family practiced the most resulted in the greatest improvement (see table 3). For example, Latisha improved her kicking performance by 50% after practicing it during 58.3% of the practice sessions, but she only improved her overhand throwing score by 12.5% after practicing it during 16.7% of the practice sessions.

Table 4

Change in pre- and post-intervention scores for each participant

Motor Skill	Latisha			Kiera			Clara			Sienna		
	Skill Change	Rate Change	Practice Freq	Skill Change	Rate Change	Practice Freq	Skill Change	Rate Change	Practice Freq	Skill Change	Rate Change	Practice Freq
Hop							+25%	+40%	8/10 (80%)			
Jump							+12.5%	0%	5/10 (50%)			
Slide										+12.5%*	+20%	5/12 (41.7%)
Strike				+10%	0%	2/14 (14.3%)						
Catch	0%	0%	2/12 (16.7%)									
Kick	+50%	+40%	7/12 (58.3%)	-25%	0%	2/14 (14.3%)	+12.5%	+20%	2/10 (20%)	0%	+20%	4/12 (33.3%)
Overhand Throw	+12.5%*	+40%	2/12 (16.7%)	+25%	+20%	4/14 (28.6%)	+25%	+20%	3/10 (30%)	0%	0%	6/12 (50%)
Underhand Toss				+25%		0/14 (0%)				-25%		7/12 (58.3%)

Parent perceptions

Overall, parents' ratings of their children's motor skill ability remained stable or increased from pre- to post-intervention with the largest increases for two of the children, Latisha and Clara (see Table 4). The majority of parent ratings of their child's motor skill ability from pre- to post-intervention aligned with actual changes in skill performance across participants. Out of the 13 ratings (the underhand toss did not have an equivalent parent rating), nine parent ratings were consistent with the child's motor performance. For example, the parent rating for Clara's hopping ability increased by 40% and her actual hopping performance increased by 25%. However, parents' views on their child's ability to change his or her motor skills did not vary from pre- to post-intervention with the exception of Sienna whose parent rating dropped by over a point. There were no other noteworthy changes in parent ratings for the remaining questionnaire items.

Discussion

The purpose of this study was to determine the feasibility of a home-based gross motor skill intervention for children with CHARGE syndrome including intervention adherence, intervention acceptability, and intervention efficacy. All hypotheses were met including that most families would adhere to the intervention protocols, children would engage in the intervention, and children would show motor skill improvements and parents would change their perceptions of motor skill competence.

Intervention Adherence

Overall, the parents in this study followed the protocols for the motor skill intervention as directed by the research team. This current study has similar findings to the recent research by Scally et al., (2019). This study determined that a “First Steps” activity pack parent intervention led to increased physical activity and motor skills. Our intervention was similar to the intervention in this larger study in that we provided the parents with the equipment and curriculum resources to build skill acquisition. In the current study the parent intervention was for six weeks as opposed to the three weeks in the Scully et al., 2019 study. Both of these interventions led to improvements in motor skills with the equipment and guided lessons provided.

Intervention Acceptability

The parent interviews revealed intervention acceptance when discussing the motor skill activities, modifications, and movement interests of their child during the intervention. In the first theme “I just want her to do anything she can do: Personal goals”, parents shared that they wanted their children to be able to do what their peers do. This is similar to the findings by Lane et al. (2020) where the parents wanted their children to do what their peers were doing in physical education. The parents believed that the improvement of these fundamental movement skills would help them with their involvement in typical skills. The second theme “Gains confidence every time: Improvement” illuminated the idea that their child could improve their motor skills with practice. This is similar to the Hamilton et al. (1999) study where the children with developmental delay improved their skills with parental support.

Opportunities and parental support for improving competence in fundamental motor skills are vital for children with CHARGE syndrome; otherwise, they may struggle to participate in free time recreational activities in adolescence and into adulthood (Imel et al., 2020; Štěrbová et al., 2014). Gross motor skills have also been used as the foundation of placement decisions in physical education (Hilgenbrinck et al., 2020). The child’s gross motor skills helped drive the programming in physical education to ensure that they had the strong foundation of gross motor skills for their lifetime of involvement which may therefore increase opportunities for socialization and friendships. Appropriate motor development contributes to children’s physical, cognitive, and social development in addition to their health and well-being (Piek, Baynam, et al., 2006; Haga, 2008).

However, the third theme “I don’t know enough: Barriers” shared the very real issue of parents not knowing the process with some of the motor skills. This illuminates the need for parent training to help them understand how to teach their children with CHARGE Syndrome. To date there is no research to outline the training protocol of parents with children with CHARGE syndrome. The last theme “It is just play time: Overcoming challenges” described the parents as resilient and creative. They found ways to motivate and engage their child in some activities that they may not have wanted to do. This is similar to the findings of Lane et al. (2020) and Lieberman et al. (2012) where parents had to be creative and help the physical education teachers

because they did not know how to teach children with CHARGE syndrome in physical education.

Intervention Efficacy

Of four participants completing both the pre- and post- motor skill assessments, more than half of the scores increased from pre- to post-intervention ($n = 10$) while the others either remained stable ($n = 3$) or decreased ($n = 2$). Not surprisingly, it was consistently seen that the skill that each family practiced the most resulted in the greatest improvement. This is important to know as it has been found that running, kicking, leaping, and catching are skills where delays are seen for children with visual impairments (Wagner et al., 2013). Knowing the skills that are deficits for this population can help families target needed areas for improvement.

Related to the parents' ratings of their children's motor skill ability, it was found that the rating either remained stable or increased from pre- to post-intervention. As shared in the results, there was alignment with the parent ratings of their child's motor skill ability from pre- to post-intervention with actual changes in skill performance across participants. This is similar to the tripartite model where the belief of the parents in motor skills aligned with their actual performance with children with visual impairments (Lent et al., 2002). Knowing this, it is important for the parents to have high expectations for their children related to motor skill potential in order for the children to truly reach their potential.

In this study, 66% of the participants improved on the gross motor skills that they practiced during the intervention. The families that participated in this study were grateful that the simple auditory and bright balls they were given. They also appreciated the free resources of the curriculum, the videos, and tip sheets. In addition, the parents improved their perceptions of what their child could do related to the motor skills assessed. These results are similar to the findings of parent lead motor skill interventions of children with developmental delays (Altunsöz et al., 2016; Bremer et al., 2015; Hamilton et al., 1999). In the current study, the skills that were worked on the most were skills that the parents felt would be used in sports opportunities and skills that they knew how to do in order to teach them.

Early opportunities and promoting motor skill development is critical to leading a physically activity lifestyle also associated with language and social skill development (Stodden et al., 2008). Additionally, fundamental motor skills are the basis of recreational programming for all children including those who are deaf-blind (Lieberman et al., 2018). With the absence of fundamental motor skills, children with CHARGE syndrome will likely have difficulty participating in recreational programming when they transition into adolescence and adulthood (Imel et al., 2020; Štěrbová et al., 2014). This very issue was highlighted in the research on the transferability of fundamental motor skills to home and the community (An & Goodwin, 2007; Chaapel et al., 2013).

One major issue with children with CHARGE syndrome as discussed in the introduction is the medical complications and extended hospitalization stays. These hospitalizations can have long lasting effects on growth and development including fundamental motor skills (Hartsthorne et

al., 2011; Salem-Hartshorne & Jacob, 2005). These prolonged absences from physical education and skill development opportunities may greatly affect the gross motor development of children with CHARGE syndrome (Hilgenbrinck et al., 2020). Thus, interventions such as this home intervention study are paramount in order to lay the foundation of fundamental motor skills to prepare the child for a lifetime of physical activity and recreation.

The findings from the current study support the notion that motor skill interventions for children with CHARGE syndrome should include parents in the process (Imel, et al., 2020, Scully et al., 2019). According to previous research, parents may be a more critical component of their children's motor development if their child has a disability (Altunsöz et al., 2016; Bremer et al., 2015; Hamilton, et al., 1999). For example, it is known that parents of children with visual impairments often promote and encourage their children by supporting them to be active and providing transportation and financial assistance (Linsenbigler et al., 2018). Alternatively, overprotecting their children from getting hurt may be a barrier to motor skill development (Lieberman et al., 2006). Parents can help with motor skills as well as their child's academic and transition goals of their IEP. The more motor skills the child can engage in the more sports and activities will be available to them- thus increasing the number of people they can engage with. In addition, the more sports and activities they can do the more they have to talk about with family, peers, and community members therefore increasing socialization and self-determination (Lieberman, et al., 2021).

Similar to previous studies, this study supports the importance of parents of children who are deaf-blind in facilitating goals and objectives of the IEP with the many roles that they play (McKittrick, 2019). These important and impactful roles include case management, advocacy, and knowledge sharing with team members. These roles on the IEP team can transfer to the home to promote motor skills and then back to school and the IEP team to share benchmarks and goals learned. Clearly parents, and their perceptions related to motor skills, do play a critical role in the motor competence of their children with visual impairments as seen in the current study.

Limitations

This study provided an important first step toward developing a robust intervention to be implemented with children with CHARGE syndrome. The resources and equipment can be used by families who have children with visual impairments, deaf-blindness, or other sensory impairments and severe disabilities. However, this study was not without limitations. These results indicate that this study is feasible and should be conducted on a larger sample size. The very small sample size limits the generalizability of the results. In addition, it should be noted that CHARGE syndrome is a highly variable and unique disorder, which is why interventions should be individualized for each child. In addition, it should be noted that the pre-test was conducted at the International CHARGE Syndrome Conference with the post testing being conducted in the individual homes of the participants. Parents received training videos for recording their child's motor skill performances, however, parents did not have previous experience with conducting motor skills assessments. Reduced space or poor camera angles may have affected the assessments of the post testing. It is likely that these conditions would have only limited their scores, meaning it is more likely that participants would have performed even

better with trained researchers due to demonstrations, instructions, more space, and optimal camera angles.

Another limitation could have been the weather as the intervention occurred during the winter months of January and February. Even parents of children in warm climates indicated that the weather was a deterrent in some instances preventing them from working on a wider range of skills. This is similar to the findings of Scully et al., (2019) where weather had an impact on the intervention protocol. In addition, the intervention was conducted just before COVID hit, which caused delays and challenges with the post testing. It should also be cautioned that the implication of school and many event closings due to COVID may have negatively impacted the children's motor gains. Finally, future motor skill intervention research on children with CHARGE syndrome should include other practices such as physical therapy, occupational therapy, and/or orientation and mobility services that the child is receiving that could affect their motor skill development as well as include a control group which does not receive the treatments.

Conclusions

Children who have CHARGE syndrome have significant health issues early in their lives that often delay their motor development significantly. This delay in motor skills can have negative consequences for their future. This feasibility study showed that parents can implement a gross motor intervention at home with resources that can be accessed on-line and with equipment found in common stores or websites. The more practice and feedback children experience the more they can improve on these fundamental skills and the higher potential for a better quality of life in their future.

Corresponding Author:

Lauren J. Lieberman

State University of New York at Brockport

llieberman@brockport.edu

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