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Gross motor skill performance in children with and without CHARGE syndrome: Research to practice



Pamela Haibach-Beach^{a,*}, Melanie Perreault^a, Elizabeth Foster^b, Lauren Lieberman^a

^a State University of New York, College at Brockport, United States

^b California State Polytechnic University, Pomona, United States

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ABSTRACT

Background: CHARGE syndrome is a multifaceted syndrome of complex birth defects. The heterogeneous nature of children with CHARGE syndrome brings unique issues and challenges affecting the overall motor development of the child, often resulting in developmental delays including motor delays.

Aims: The purpose of this research was to assess children with CHARGE Syndrome on locomotor and object control skills to better understand their motor development. This information is relevant to adapted physical education teachers, paraeducators, vision teachers, health care professionals (occupational therapists, physical therapists, and physicians) and parents and family members of children with CHARGE Syndrome.

Methods and Procedures: Thirty-seven children with CHARGE syndrome and thirty peers without disabilities participated in the study. Each participant was assessed on two object control and three locomotor skills with modifications, if necessary. In addition, the age of onset of independent walking was recorded for each participant.

Outcome and results: Children with CHARGE syndrome performed significantly behind their same age peers in most gross motor skills with the biggest deficits found in the run and kick. Age of onset of walking was associated with performance in jumping, running, and throwing.

Conclusions and implications: Early intervention services should focus on gross motor skills such as throwing, kicking, as well as walking at an early age.

What this paper adds

A typical child with CHARGE syndrome is significantly delayed in their gross motor skill performance. This research explores motor skills in higher functioning children with CHARGE syndrome, as this study only examined children who were able to ambulate without assistance. The results of this research provides component analyses on several locomotor and object control skills with a comparison to typically developing controls. The largest effect sizes were found in the run and throw, specifically component 3 (narrow foot placement) and 4 (nonsupport leg bent 90°). In addition, age of walking was examined in relationship to the motor skills assessed. Age of walking was an important indicator of performance with large negative correlations found in four out of five of the skills. The findings of this study reveal the critical need for early intervention services focusing on motor milestones leading to improved motor skill development in children with CHARGE syndrome.

* Corresponding author at: State University of New York College at Brockport, 350 New Campus Lane, Brockport, NY 14420, United States.
E-mail address: phaibach@brockport.edu (P. Haibach-Beach).

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1. Introduction

CHARGE syndrome is a multifaceted syndrome of complex birth defects. The heterogeneous nature of children with CHARGE syndrome cannot be overstated and the complexity of the syndrome brings unique issues and challenges. The original clinical criteria for diagnosing an individual consisted of identifying four of the six characteristics of CHARGE syndrome. The original six characteristics were based on the mnemonic C-H-A-R-G-E: coloboma of the eye, heart defects, atresia of the choanae, restriction of growth and/or development, genital and/or urinary abnormalities, and ear abnormalities and deafness (Pagon, Graham, Zonana, & Yong, 1981; Stratton & Hartshorne, 2018). Currently the diagnosis is based on clinical features, which can be extremely variable from one child to the next; however, the most common features include vision loss, hearing loss, and balance issues (Verloes, 2005). In 2004, the mutation of the chromodomain helicase DNA binding protein 7 (CHD7) gene was reported in approximately two-thirds of children with CHARGE syndrome (Vissers et al., 2004). With the discovery of the causative gene, it has been suggested to revise the current clinical diagnostic criteria to include broadening of the major characteristics and examining CHD7 variants (Hale, Niederriter, Green, & Martin, 2016).

The combined loss of vision, hearing, proprioception and vestibular function and the need of surgeries and hospital stays due to complication of CHARGE negatively affects the overall motor development of the child, often resulting in developmental delays including significant motor delays (Dammeyer, 2012; Haibach & Lieberman, 2013; Salem-Hartshorne & Jacob, 2004; Smith, Smith, & Blake, 2010). Currently, there is minimal research on the balance and age of walking in children with CHARGE Syndrome and no known research assessing fundamental motor skills. Therefore, it is important to determine the range of capabilities of children with CHARGE to better inform physical educators, adapted physical educators, and allied health professionals on the various characteristics that may influence motor skill development and performance in order to implement effective educational techniques for children with CHARGE syndrome.

Children with CHARGE syndrome are delayed in the development of motor milestones (i.e., crawling, standing, and walking) (Thelin, Curtis, Maddox, & Travis, 2011). The average age of independent walking (taking a minimum of five steps without assistance) in children with CHARGE syndrome is approximately 35–60 months, with some children never acquiring independent walking (Hartshorne, Nicholas, Grialou, & Russ, 2007). However, 80% of children with CHARGE syndrome will acquire independent walking by 6–7 years (Hartshorne et al., 2007). In a recent research study, the mean age of independent walking in children with CHARGE syndrome was 41.65 months (Haibach & Lieberman, 2013). This is in comparison to the onset of walking in typically developing children between the ages of 12 and 14.5 months (Karasik, Tamis-Lemonda, & Adolph, 2011; Størvold, Aarethun, & Bratberg, 2013). Independent walking requires the coordination, balance and strength of the muscular skeletal systems in addition to the child's ability to solve problems. Age of walking is also greatly affected by vision which is typically compromised in children with CHARGE Syndrome.

In addition, previous researchers have indicated a strong positive correlation between the earlier attainment age of independent walking in children with CHARGE syndrome in relation to the acquisition of communication and language (Dammeyer, 2012; Thelin & Fussner, 2005), adaptive behavior scores (Salem-Hartshorne & Jacob, 2004), executive function (i.e., self-monitored behavior and actions, Hartshorne et al., 2007), and intellectual outcome (Dammeyer, 2012, Raqbi et al., 2003). It is also likely to be strongly related to fundamental motor skill performance, which is the purpose of the present study. Therefore, motor development should be an early intervention priority and researched to examine potential variables that impact motor development and strategies to decrease the motor delay.

1.1. Purpose

The purpose of this research was to assess children with CHARGE syndrome on locomotor and object control skills to better understand their motor development. A component analyses of their motor skill performance can be used to develop curriculum to improve motor skills in this population. This information is relevant to adapted physical education teachers, paraeducators, vision teachers, health care professionals (occupational therapists, physical therapists, and physicians and parents and family members of children with CHARGE Syndrome.

2. Material and methods

2.1. Sample

Thirty-seven children with CHARGE syndrome who attended the 2017 International CHARGE Conference in Orlando, Florida participated in this study. Their mean age was 9.64 years (SD = 3.69 years), height 49.06 in. (SD = 7.16), and weight 60.97 lbs (23.14 lbs). Seventeen of the participants were boys and twenty participants were girls. Parents of the participants filled out a report that included demographic information and their age at walking. Parents of children with CHARGE also filled out information about their child's CHARGE characteristics. Based upon parental report, twenty of the participants had profound hearing loss, the other 17 participants had mild to moderate hearing loss in one or both ears and 20 participants were missing their semicircular canals and 17 had misformed semicircular canals. To participate in the study, participants had to have CHARGE syndrome, be between 4 and 14 years of age, and be able to ambulate independently (i.e., without any supportive devices such as walkers). Two participants dropped out after signing the consent form due to their inability to complete the study and four other participants were unable to complete the study due to either attention deficits, lack of interest, and/or inability to complete all of the tasks. The control group included 28

children between the ages of 4 and 14 years (Mean 10.1 years, SD 3.57 years), mean height 56.25 in. (SD = 8.99 in.), and mean weight 92.54 lbs (SD = 42.15 years). The control group were assessed at the lead investigator’s institution following the assessments in the children with CHARGE Syndrome to age and gender match the participants. Eighteen of the control participants were male (12 female).

This research was approved by the lead investigator’s institution and all participants and their parents signed informed consent forms approved by the Institutional Review Board committee. Participants were informed prior to beginning the tests that they could withdraw from participating in the study at any time during testing.

2.2. Instrument and procedures

Participants completed five motor skills: running, sliding, jumping, kicking, and overhand throwing. Each participant completed each of the five motor skills two times and all attempts were video recorded for later analyses to allow for independent assessment by two researchers trained in the TGMD-II. A videographer recorded each attempt with a view of the children’s entire body. The videos were downloaded within two days of recording and each were analyzed by the team within a month of data collection. The video analyses enabled the team to watch the videos multiple times or slower if necessary. If there was a disagreement, the lead researcher clarified any questions about scoring. Prior to data collection a graduate student was trained with the TGMD-II by two of the researchers by practicing assessments of other videos. During the training adequate interrater reliability was achieved across the two researchers, $\kappa = 0.60-0.86, p < 0.001$ (Landis & Koch, 1977). The two researchers with the highest interrater reliability scores of 0.86 conducted the assessments for the present study.

The criteria from the Test of Gross Motor Development II (TGMD II; Ulrich, 2000; for construct validity see Evaggelinou, Tsigilis, & Papa, 2002) was used to assess the participants’ gross motor performance of running, sliding, jumping, kicking, and overhand throwing. The TGMD II was chosen because it has been validated for children with visual impairments (Houwen, Hartman, Jonker, & Visscher, 2010) and in children with intellectual disabilities (Simons et al., 2008). There were several modifications used to help the children with sensory deficits such as a beeping ball for kicking, fluorescent tape, and a sound source for running. These modifications were validated as part of the Houwen et al. (2010) study for children with visual impairments. It should be noted that adapted equipment was found to not affect the performance of typically developing children (Lieberman, Haibach, & Wagner, 2014). Each criterion was evaluated as a one when present and a zero if not present. Each motor skill was completed two times. Motor testing was conducted and analyzed by the researchers of this study. Table 1 displays an example of the components and assessment for the run and Table 2 displays all of the components for each motor skill assessed.

2.3. Data analyses

Results on the protocol sheet were transferred to SPSS Version 24. The data of individual participants was stored anonymously and the protocol sheets were destroyed. The assessments were based upon observations and thus the motor skill scores and criterion scores are presented as medians and ranges. Analyses were non-parametric Mann-Whitney U tests (one-tailed). The statistical tests were set at a level of 0.05.

3. Results

3.1. Motor skills

The analysis revealed that the control group significantly performed better than the group of children with CHARGE syndrome on all motor skills: run ($U = 26, p < .001$), jump ($U = 299.5, p = .039$), slide ($U = 263.5, p = .003$), kick ($U = 198.5, p < .001$), and throw ($U = 128.5, p < .001$). See Table 3. Within the group of children with CHARGE, boys significantly performed better than girls only on the skill of kicking ($U = 60.5, p = .04$), all other skills there was no significant difference.

When the motor skills were broken down by component, the analysis revealed that the control group outperformed ($p > .05$) the group of children with CHARGE on all components except the following: jumping components 2 (arms extend forcefully forward and upward reaching full extension above head) and 3 (take off and land on both feet simultaneously) and kicking components 3 (nonkicking foot placed even with or slightly in back of the ball) and 4 (kicks ball with instep of preferred foot or toe). The participants with CHARGE struggled most with running components 3 (narrow foot placement landing on heel or toe) and 4 (nonsupport leg bent approximately 90 degrees), kicking component 2 (an elongated stride or leap immediately prior to ball contact), and all

Table 1
Example of scoring the run.

Skill	Performance criteria	Trial 1	Trial 2	Score
1	Arms in opposition to legs, elbow bent	1	1	2
2	Brief period where both feet are off the ground	0	1	1
3	Narrow foot placement landing on heel or toe	1	0	1
4	Nonsupport leg bent approximately 90 degrees	0	0	0
Skill Score		4 out of a possible total of 8		

Table 2
Components for each motor skill assessed.

Running Components

- 1 Arms in opposition to legs, elbow bent
- 2 Brief period where both feet are off the ground
- 3 Narrow foot placement landing on heel or toe (i.e., not flat footed)
- 4 Nonsupport leg bent approximately 90° (i.e., close to buttocks)

Jump Components

- 1 Preparatory movement includes flexion of both knees with arms extended behind the body
- 2 Arms extend forcefully forward and upward reaching full extension above the head
- 3 Take off and land on both feet simultaneously
- 4 Arms are thrust downward during landing

Slide Components

- 1 Body turned sideways so shoulder are aligned with the line on the floor
- 2 A step sideways with lead foot followed by a slide of the trailing foot to a point next to the lead foot
- 3 A minimum of four continuous step-slide cycles to the right
- 4 A minimum of four continuous step-slide cycles to the left

Kick Components

- 1 Rapid continuous toward the ball
- 2 An elongated stride or leap immediately prior to ball contact
- 3 Nonkicking foot placed even with or slightly in back of the ball
- 4 Kick the ball with the instep of preferred foot (shoelaces) or toe

Overhand Throw Components

- 1 Windup is initiated with downward movement of hand and arm
- 2 Rotates hip and shoulders to a point where the nonthrowing side faces the wall
- 3 Weight is transferred by stepping with the foot opposite the throwing hand
- 4 Follow-through beyond ball release diagonally across the body toward the nonpreferred side

Table 3
Comparison of skill scores between children with and without CHARGE syndrome.

	CHARGE (N = 31)		Controls (N = 27)		z	p	r
	Median	Mrank	Median	Mrank			
Run	4.00	16.84	8.00	44.04	6.48	< .000	.85
Jump	4.00	25.66	4.50	34.80	2.06	.039	.27
Slide	7.00	24.50	8.00	36.09	2.99	.003	.39
Kick	6.00	22.12	8.00	37.41	3.79	< .000	.50
Throw	1.00	19.78	8.00	39.91	4.73	< .000	.62

Table 4
Comparison of selected component scores between children with and without CHARGE syndrome.

	CHARGE (N = 31)		Controls (N = 27)		z	p	r
	Median	Mrank	Median	Mrank			
Run 1	2.00	24.23	2.00	36.39	3.58	< .000	.47
Run 2	2.00	25.94	2.00	34.50	3.06	.002	.40
Run 3	.00	19.23	2.00	41.93	5.86	< .000	.76
Run 4	.00	16.90	2.00	44.50	6.96	< .000	.91
Jump 1	2.00	25.98	2.00	34.45	2.21	.027	.29
Jump 4	.00	26.02	1.00	34.41	2.12	.034	.28
Slide 1	2.00	26.03	2.00	34.39	2.29	.022	.30
Slide 2	2.00	27.00	2.00	33.32	2.09	.037	.27
Slide 3	2.00	26.02	2.00	34.41	2.77	.006	.36
Slide 4	2.00	26.03	2.00	34.39	2.76	.006	.36
Kick 1	2.00	25.23	2.00	34.07	2.74	.006	.36
Kick 2	.00	21.48	2.00	38.09	4.29	< .000	.56
Throw 1	.00	20.40	2.00	39.25	4.73	< .000	.62
Throw 2	.00	20.97	2.00	38.64	4.52	< .000	.59
Throw 3	.00	21.50	2.00	38.07	4.24	< .000	.56
Throw 4	.00	21.33	2.00	38.25	4.24	< .000	.56

components of the throw (see Table 4).

3.2. Age at walking

The average age that the participants with CHARGE Syndrome were able to walk independently was 3.75 years ($SD = 1.57$ years). The average age the control participants walked independently was 1.05 years ($SD 0.10$ years). The analysis revealed a significant group difference, $F(59) = 18.93$, $p < .001$, wherein the control group ($M = 12.9$, $SD = 3.1$) walked significantly earlier than the participants with CHARGE ($M = 42.8$, $SD = 22.5$). Age at walking had a significant negative correlation ($p < .01$) with all motor skills except the jump. The age of walking had the strongest negative correlation to the skill of running ($r = -0.78$), followed by the throw ($r = -0.64$), kick ($r = -0.45$), and slide ($r = -0.34$).

4. Discussion

4.1. Summary and progress of knowledge

In this study, 31 children with CHARGE syndrome were assessed on their motor skills running, sliding, jumping, throwing, and kicking. Previous research has shown the need for physical education teachers and parents to focus on the specific area of motor skills in order to improve early motor development and motor skill development to enable the children to participate fully in sports and physical activity (Lieberman, Haibach, & Schedlin, 2012; Salem-Hartshorne & Jacob, 2004). The main finding in this study was that children with CHARGE syndrome were significantly behind their same age peers on all locomotor skills and object control skills tested, similar to findings assessing children who were blind with their sighted peers (Wagner, Haibach, & Lieberman, 2013). These results were determined even after the modifications of sound and color were provided to meet the need of the children's vision and hearing deficits.

In addition, this study examined the components for each motor skill assessed. Findings revealed that the children with CHARGE syndrome had specific deficits in running components 3 (narrow foot placement landing on heel or toe) and 4 (nonsupport leg bent approximately 90 degrees), kicking component 2 (an elongated stride or leap immediately prior to ball contact), and all components of the run, mirrors findings from a previous study by Haibach, Wagner, and Lieberman (2014) about children with visual impairments. These deficits in locomotor and object control skills may likely be due to the fact that children with CHARGE syndrome also have deficits in balance due to proprioceptive and visual impairments (Dammeyer, 2012; Haibach & Lieberman, 2013).

Consistent with literature, the children with CHARGE Syndrome walked independently significantly later than their typically developing peers, with a strong correlation to the age of independent walking with jumping, running, and throwing. Understanding the formation of motor skills is central to provide appropriate instruction, educational techniques, and interventions (Gallahue, Ozmun, & Goodway, 2012). Smith and Thelen (2003) stated that, "... developing organisms are complex systems composed of very many individual elements embedded within, and open to, a complex environment" (p. 343). The multicausality and influence of CHARGE syndrome will be different for each child. The degree of impairment can vary greatly between one child with CHARGE syndrome to the next and the loss can have varying impact as well. Doralp and Bartlett suggested "... a better understanding of infant characteristics in relation to motor development can have a significant impact on planning interventions & ensuring that a given environment is optimally suited to a child's disposition & needs" (2014, p. 327). Early intervention services should encourage balance and gross motor development to improve performance on these critical motor skills leading to more active and healthier lifestyles.

4.2. Practical implications

There are many practical implications as a result of these interesting findings. First and foremost, it is important to do all that we can to support children to walk as early as possible. This is by far one of the most memorable and proud moments for any parents, but one that parents of children with CHARGE syndrome cherish. Starting children early on with weight bearing activities and providing them opportunities to improve their balance and mobility through a variety of opportunities throughout the day is important to encourage independent walking.

The other practical implication taken from this study is the need for children with CHARGE syndrome to work on locomotor skills and object control skills at home and in school. The first step is to train support staff, caregivers, teachers, and parents, on the form of a throw, kick, slide, run, and a jump. These are critical skills that are used in many sports and recreational activities and can be infused into any backyard game, recess activity, or physical education class.

Many children may need modified equipment such as a beeper or sound device to run towards or a guidewire to run or perform locomotor skills along. These can be found through the American Printing House for the Blind at www.aph.org/pe/products. Other modified equipment may be beeping balls (also available through APH), bright lines or poly spots on the floor for jumping, bell whiffle balls, yarn balls, or bean bags for throwing. To learn the proper form to perform any of these skills, please see the Gross Motor Development video through the American Printing House for the Blind and Camp Abilities at either of these two links; <https://www.aph.org/physical-education/motor-video-feedback/> or <http://www.campabilities.org/instructional-materials.html>. Instructors can also read the Gross Motor Development Curriculum available as a free download from APH at <https://www.aph.org/files/manuals/GMDC/>. Early intervention on walking and intentional instruction, feedback, and support for gross motor skills will help all children including those with CHARGE syndrome gain valuable skills for successful transitions and potentially improve quality of life.

CHARGE Syndrome is the leading cause of child onset Deafblindness (National Center on Deaf-Blindness, 2017). This is important

to clarify as the current law identifies Deafblindness as one of the main 13 disability categories but not CHARGE itself. The definition of Deafblindness under the Individuals with Disabilities Education Improvement Act (IDEIA, 2004) states:

Deafblindness means concomitant hearing and visual impairments, the combination of which causes such severe communication and other developmental and educational needs that they cannot be accommodated in special education programs solely for children with deafness or children with blindness [34 C.F.R. §300.8(c)(2)].

Children with CHARGE Syndrome need unique additional assistance to meet their educational needs, including physical education. It is imperative that early interventions and individualized instruction is provided to these unique children to improve their overall development (Muller, 2006). Ninety percent of this population has one or more additional disabilities, with over 40% having four or more additional disabilities (National Center on Deaf-Blindness, 2017). Consequently, children with CHARGE syndrome are a vastly heterogeneous population and are considered a low incidence disability. The meaning of the term low incidence disability under IDEIA (2004) is as follows:

- a) a visual or hearing impairment, or simultaneous visual and hearing impairments;
- b) a significant cognitive impairment; or (c) any impairment for which a small number of personnel with highly specialized skills and knowledge are needed in order for children with that impairment to receive early intervention services or a free appropriate public education [34 C.F.R. §662(c)(3)].

The implication of this definition is that children with CHARGE syndrome need educators who are trained with specialized skills and interventions to meet the needs of children who are deafblind. As stated in IDEIA, children with deafblindness need specific interventions related to the dual sensory impairment that cannot be accommodated in special education programs solely for children with deafness or children with blindness (2004). Therefore, physical educators need to be cognizant of the motor skills and interventions needed to educate and develop a specially designed educational program for students who are deafblind in the least restrictive environment.

With increases in inclusion, physical educators and adapted physical educators must have the appropriate attitudes, knowledge, and skills needed to meet the educational and social needs for students with CHARGE syndrome. The Individualized Education Program (IEP) must address the impact of their dual sensory loss on motor skill acquisition. Educational techniques and interventions within special education are specifically designed to meet the unique needs of the students, which may include adaptations when appropriate in the content, methodology, or delivery of instruction (IDEIA, 2004). Furthermore, the earlier interventions and appropriate services can be provided, the sooner motor skill progressions can occur (Lieberman et al., 2012). However, many adapted physical educators have little or no knowledge and no previous training related to teaching students with CHARGE Syndrome (Lieberman et al., 2012). Many adapted physical educators also lack knowledge in how to develop appropriate programs and interventions for students with CHARGE Syndrome (Lieberman, Ponchillia, & Ponchillia, 2013; Lieberman & MacVicar, 2003). Since adapted physical educators infrequently encounter these students, they have little to no basis on where to start their assessment and what instructional strategies and interventions are needed to promote motor skill development.

4.3. Limitations and future directions

There were several limitations which should be taken into consideration with this study. Convenience sampling approach was used due to the very low incidence and large geographic dispersion of this population. Obtaining a random sample would simply not be feasible. Participants with CHARGE syndrome were recruited from a biannual international conference in which many parents of children with CHARGE syndrome bring their children. This is an opportunity for the parents to receive some of the latest research on CHARGE syndrome as well as for their children to meet other children with CHARGE syndrome, as many have not met another child with CHARGE outside of the conference. It is likely that the children with CHARGE syndrome who have the opportunity to attend this conference may be inadequate in representing the population and there is a possibility of a sampling error (Bornstein, Jager, & Putnick, 2013). The sampling size for children with CHARGE syndrome is large considering CHARGE is a very low incidence population. It should also be noted that to participate in this study, inclusion criteria included only children who were able to ambulate (i.e., walk) without assistance. It is important to note, that this criteria eliminated many other children with CHARGE syndrome and therefore cannot generalize to all children with CHARGE syndrome, rather children with CHARGE syndrome who are walking independently. As there is currently no known research conducted on the motor skill performance of children with CHARGE syndrome, the present study provides preliminary analyses. Subsequent analyses should examine more motor skills as well as the effects of age and sex.

5. Conclusion

Children with CHARGE syndrome will likely have deficits in some gross motor and object control skills. These deficits will be associated with late onset of walking ability. Early intervention services should focus on gross motor skills such as throwing, kicking, as well as walking at an early age. The use of modifications such as bright color balls, beeping balls, and bright tape can help children with their ability to perform certain gross motor skills. Teachers, parents, physical education or adapted physical education specialists, and allied health professionals should make every effort to universally design lessons to ensure a multisensory environment for the classes with children with CHARGE syndrome. With a proactive approach to movement, children with CHARGE syndrome can have active and health lives to the maximum extent possible.

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