IMPACT OF RATE LIMITERS AND AFFORDANCES ON THE DEVELOPMENTAL MILESTONE OF WALKING IN CHILDREN WITH CHARGE SYNDROME

A DISSERTATION

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BY

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To the Dean of the Graduate School:

I am submitting herewith a dissertation written by Elizabeth A. Foster entitled "Impact of Rate Limiters and Affordances on the Developmental Milestone of Walking in Children with CHARGE Syndrome." I have examined this dissertation for form and content and recommend that it be accepted in partial fulfillment of the requirements for the degree of Doctor of Philosophy with a major in Kinesiology.

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Accepted:

Dean of the Graduate School

DEDICATION

I dedicate this dissertation to my Pop-Pop, James J. Houghton. Thank you for always providing intellectual challenges throughout my childhood and for always maintaining such high expectations on learning and experience. Your work ethic and desire to continue to learn and challenge your mind has instilled into my professional attitude and philosophy. I understand the importance and strength of human contact. I miss you so much and love you tons! Thank you for always being around when I need you.

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"Every story has an end, but in life every end is just a new beginning." M.S. Forbes

I am ready for ours. I love you.

ABSTRACT

ELIZABETH A. FOSTER

IMPACT OF RATE LIMITERS AND AFFORDANCES ON THE DEVELOPMENTAL MILESTONE OF WALKING IN CHILDREN WITH CHARGE SYNDROME

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Children with CHARGE syndrome have a significant delay in acquiring developmental motor milestones, including independent walking (Blake et al., 1998; Hartshorne & Cypher, 2004; Hartshorne, Nicholas, Grialou, & Russ, 2007; Travis & Thelin, 2007). CHARGE syndrome is a multifaceted syndrome of complex birth anomalies, which usually includes a degree of vision and hearing loss at birth. The purpose of this investigation was to examine the impact of individual rate limiters and affordances on acquiring the independent walking developmental milestone in children with CHARGE syndrome. The investigation was divided into two phases that were performed concurrently. The participants in Phase I were 62 children with CHARGE syndrome whose information was entered into the CHARGE Syndrome Clinical Database Project (CSCDP) by a parent or primary caregiver. The participants in Phase II were 23 parents or primary caregivers of a child with CHARGE syndrome who acquired independent walking.

In Phase I, the CSCDP was used to obtain the following information: left and right eye vision levels; left and right ear decibel loss; balance problems; occurrence of 5-point crawling; occurrence of back scooting; type of hearing loss; and small or absent

semicircular canals in the left and right ear. An analysis of covariance, analysis of variance, and multiple regressions were performed to analyze the data. Based on the results, an increase in the severity of vision loss and hearing loss significantly delayed the attainment age of independent walking in children with CHARGE syndrome. Furthermore, right eye acuity levels and left ear decibel loss were significant predictors on the attainment age of independent walking.

In Phase II, semi-structured interviews were used to acquire the parents' or primary caregivers' perspective on rate limiters or affordances related to the attainment of independent walking in their child with CHARGE syndrome. Interviews were transcribed and then analyzed using thematic analysis based on the research questions and the dynamic systems theory. Additional individual, environmental, and task constraints were reported that encouraged or hindered the attainment of independent walking. Within the limitations of this investigation, the conclusions were drawn that the findings supported the dynamic systems theory.

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INTRODUCTION

Movement is displayed in every aspect of a child's life, and understanding the formation of movement is central to the field motor development. The understanding of motor development provides guidelines for effective and appropriate educational techniques and interventions (Gallahue, Ozmun, & Goodway, 2012). One guideline that is often used to gauge children's development are developmental motor milestones. The use of developmental motor milestones provides information about a child's rate and extent of his or her overall developmental progress. Since the developmental milestones are relatively sequential and provide a progressive pattern any delay in the attainment of these developmental motor milestones may impede further development (Johnson & Blasco, 1997).

Specifically, the attainment of independent walking usually occurs subsequently after the developmental motor milestones of standing independently, crawling with hands and knees, and sitting independently (World Health Organization [WHO] Multicentre Growth Reference Study Group, 2006). A typical child with CHARGE (coloboma of the eye, heart defects, atresia of the choanae, retardation of growth and/or development, genital and/or urinary abnormalities, and ear abnormalities and deafness) syndrome is delayed significantly in gross motor development, particularly with the developmental milestone of walking independently (Hartshorne, Hefner, Davenport, & Thelin, 2011).

The attainment of developmental motor milestones, through the lens of the dynamic system theory (DST), can be understood by considering the interaction of the individual, environmental, and task constraints (i.e., rate limiters, affordances).

The DST is an ecological perspective of motor development, which emphasizes that movements and tasks are controlled and produced by interactions of the individual's body subsystems (i.e., vision, hearing, muscular, skeletal) and the environment (Kugler, Kelso, & Turvey, 1982; Smith & Thelen, 1993; Thelen & Ulrich, 1991). The process of motor development is then connected to changes within the individual, environment, and task (Newell, 1984, 1986; Thelen & Smith, 1996). The individual's body subsystems are in constant self-organization in order to adapt to change as new structures and different patterns of movement are acquired (Kamm, Thelen, & Jensen, 1990). As the process of motor development occurs, the subsystems of the body must reorganize due to new behavioral change, known as motor learning. Motor learning, ". . . is a relatively permanent change in motor behavior resulting from practice or past experience" (Gallahue et al., 2012, p.14).

A significant developmental behavioral change can be seen when infants attain the developmental motor milestone of walking. In order for an infant to attain this motor milestone, the body subsystems must be developed to support the body in an upright position through the use of posture, balance, and strength (Thelen, 1986a). In addition, the infant must have the ability to execute and control the body through the walking step pattern. Therefore, the attainment of walking is based on the organization of the body subsystems and the individual's unique characteristics. In addition, the environmental

characteristics (i.e., uneven surfaces) along with the task of walking can alter the skill of walking. These characteristics of the individual, the environment, and the task are viewed as constraints that can either encourage or discourage movement (Newell, 1984).

The dynamic system approach describes the concept of constraints as potential rate limiters. A rate limiter "is an individual constraint or system that hold[s] back or slows the emergence of a motor skill" (Haywood & Getchell, 2009, p. 23). In addition, different subsystems within the child (i.e., vision, hearing) may also act as rate limiters for specific motor skills (Thelen, 1995, 1998). Gibson (1977, 1979) stated the importance of the process of perception and action in regards to movement. Perceptual skills are based on an individual's ability to interpret and integrate sensory information to determine a movement outcome (Gabbard, 2004; Haywood & Getchell, 2009). Affordances are individual perceptions, a person's capabilities based on the movement possibility related to the task and the environment (Gibson, 1977, 1979). Affordances tend to promote and encourage developmental change. If an infant or child is unmotivated or unable to process perceptions of his or her environment or body capability, then the acquisition of movement will be negatively impacted or delayed (Levtzion-Korach, Tennenbaum, Schnitzer, & Ornoy, 2000; von Hofsten, 2004).

Based on the dynamic systems theory, each subsystem is interdependent with other subsystems, which contribute to the attainment or delay of developmental motor milestones. Smith and Thelen (2003) stated that, ". . . developing organisms are complex systems composed of very many individual elements [subsystems] embedded within, and open to, a complex environment" (p. 343). Therefore, multicausality and the influence of

the subsystems is one major aspect of the dynamic systems theory on motor development. Impairments to specific body subsystems (i.e., vision, hearing) can cause possible constraints to motor development and the attainment of developmental motor milestones (Kamm et al., 1990). Children who have dual sensory loss have impairments in the vision and hearing subsystems within their bodies, which may act as individual rate limiters. Although vision and hearing are usually not totally absent, the degree of loss can greatly impact other subsystems concurrently impacting his or her motor development.

The current leading identifiable cause of infants born with a dual sensory loss of vision and hearing is CHARGE syndrome (National Center on Deaf-Blindness [NCDB], 2014). CHARGE syndrome is a multifaceted syndrome of complex birth anomalies, which at many times includes a degree of vision and hearing loss at birth. Researchers have shown that individuals with CHARGE syndrome may not achieve normal developmental motor milestones (Hartshorne, Nicholas, Grialou, & Russ, 2007; Travis & Thelin, 2007). For instance, typically, children with CHARGE syndrome do not walk independently until 3 to 5 years of age (Hartshorne et al., 2011; Hartshorne et al., 2007). This age range exceeds the standard window of achievement for children without disabilities based on the results of the WHO Multicentre Growth Reference Study (2006); therefore, it is important to investigate what contributes to this delay of the independent walking motor milestone.

It is essential for researchers studying motor development to determine the potential effect of an infant's characteristics (i.e., weight, activity level, motivation) on motor development (Doralp & Barlett, 2014). Other researchers have indicated the importance

of infant characteristics; however, the majority of researchers have not addressed specific infant characteristics (Bradley, 1994; Shonkoff & Meisels, 2000). Doralp and Bartlett suggested " . . . a better understanding of infant characteristics in relation to motor development can have a significant impact on planning interventions and ensuring that a given environment is optimally suited to a child's disposition and needs" (2014, p. 327). Therefore, an understanding of the impact of various characteristics that influence motor development is necessary to develop effective and appropriate educational techniques and interventions that encourage motor development in infants and children with CHARGE syndrome.

Motor Development

Motor development is one field of study under motor behavior research (see Figure 1). The field of motor behavior began in the late 18th century and continues to be of interest and researched to this present day (Haibach, Reid, & Collier, 2011). Motor behavior refers, ". . . to changes in motor learning control and development that embody learning factors and maturational processes associated with movement performance" (Gallahue, Ozmun, & Goodway, 2012, p. 14). During the years of infancy and early childhood, developmental motor milestones provide specific ages in which normal functioning is established. In order for an individual to walk independently, most children will first attain the developmental milestones of sitting, creeping, crawling, and then standing. The average ages of developmental motor milestones achieved by children were originally researched by Bayley (1936, 1969) and Shirley (1931, 1963).

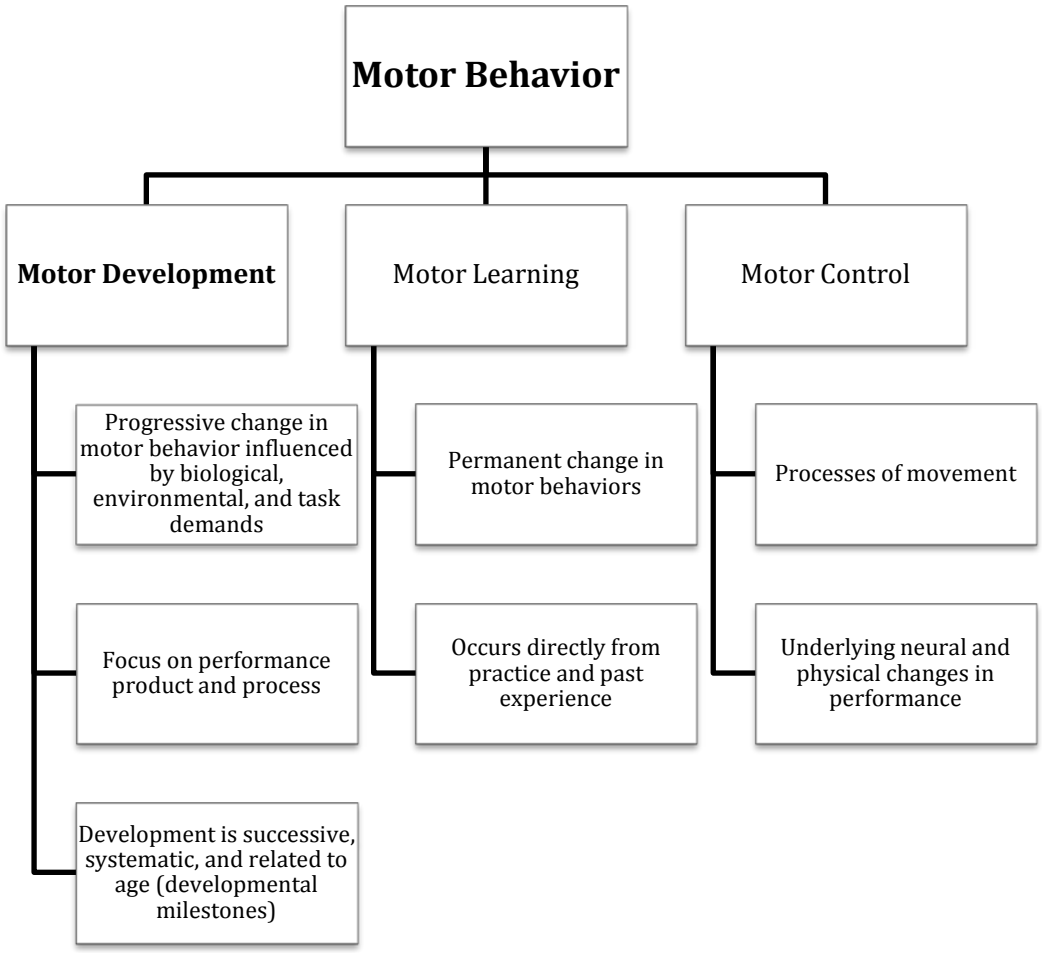

Figure 1. A flow model of the field of motor behavior, which includes the study of motor development, motor learning, and motor control. (Gallahue et al., 2012; Haibach et al., 2011).

In 2006, the WHO implemented a study, the Multicentre Growth Reference Study, to assess development and produce age of achievement standards of six developmental motor milestones. The WHO Multicentre Growth Reference Study (2006) followed 816 children from Ghana, India, Norway, Oman, and the United States from birth until 5 years of age. The motor development data were used to generate a standard window of achievement limited by the 1st and 99th percentiles of the study sample. The window of

achievement for each developmental milestone contains the normal variation in ages of achievement among healthy children. Based on this investigation, the window of achievement for walking alone, without assistance, was from 8.2 to 17.6 months with the mean age of achievement being 12.1 months (see Figure 2; WHO Multicentre Growth Reference Study, 2006). In addition, 90% of children achieved five of the developmental milestones (i.e., sitting without support, standing with assistance, walking with assistance, standing alone, walking alone) following a common sequence. Based on the results of the investigation, there was no significant difference of achievement of motor milestones between genders and countries.

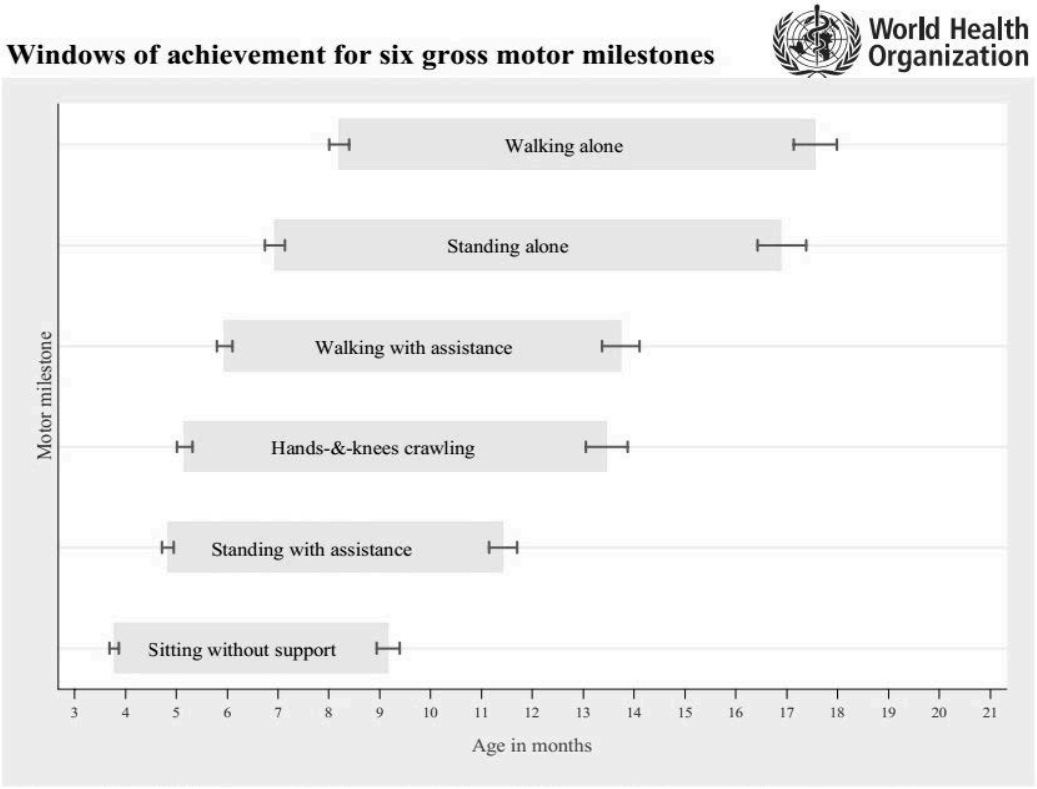

Figure 2. Windows of achievements for six gross motor milestones related to walking. From "*WHO Motor Development Study: Windows of achievement for six gross motor development milestones*" by WHO Multicentre Growth Reference Study Group, 2006 (no permission needed to reprint).

Deafblindness[1]

Since 1986, the National Center on Deaf-Blindness (NCDB), has been conducting an annual National Child Count of Children and Youth who are Deaf-Blind. In December 2013, a total of 9,454 individuals, birth through 21 years old, were included in the national child count. Many children who are deafblind now are provided early intervention and special educational services under Individuals with Disabilities Education Improvement Act (IDEIA, 2004). Under this Act:

> Deaf-blindness 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)].

Due to the concomitant degree of loss in hearing and vision, children need unique additional assistance to meet their educational needs. It is imperative that early interventions and individualized instruction is provided to these unique individuals 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 (NCDB, 2014). Consequently, children with deafblindness 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;

[1] The term deafblindness will be used throughout this paper without the hyphen to recognize the impact and unique condition of the combined sensory loss of both hearing and vision, which should not be investigated as one or the other but rather the impact of the combined loss.

> (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 deafblindness need educators who are trained with specialized skills and interventions to meet the needs of individuals 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, educators and service providers need to be cognizant of the skills and interventions needed to educate and develop a specially designed educational program for students who are deafblind in the least restrictive environment.

The WHO and the World Bank (2011) prepared a World Report on Disability, which included information on education. Based on the report, an inclusive environment for children with disabilities should be a priority of all countries and is an element of quality education within a child's least restrictive environment. Inclusive education should remove barriers and provide reasonable accommodations and support services required for students with disabilities. Inclusive education can be seen in physical education settings with more students with disabilities being included with their peers and a need for additional preparation and supports within this setting (Block, 2013).

Successful inclusion provides a significant need for information, resources, and research in the field of deafblindness pertaining to general and adapted physical

education (Lieberman, Ponchillia, & Ponchillia, 2013). The philosophy of inclusion is to educate students with disabilities in the same environment with their peers without disabilities by addressing their educational and social needs (Block, 2007, 2013). With the increase in inclusion, adapted physical educators must have the attitudes, knowledge, and skills needed to meet the educational and social needs for students who are deafblind.

Educational Techniques and Interventions

Understanding the impact of vision and hearing on motor development can provide valuable information to general and adapted physical educators when developing an Individualized Education Program (IEP) to improve motor development. The program must address the impact of the dual sensory loss on motor development. Furthermore, the earlier interventions and services can be provided, the more children who are deafblind will benefit (Hartshorne et al., 2011). One area related to motor development within the education system is general and adapted physical education. 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).

Early interventions and appropriate instructional strategies are critical components in the success and participation of a child who is deafblind (Hartshorne et al., 2011). Highly qualified adapted physical educators are professionals from standard-based physical education teacher education programs who can design, implement, and evaluate motor skills, levels of fitness, and athletic skills of students with disabilities (American Association for Physical Activity and Recreation & National Consortium for Physical

Education for Individuals with Disabilities, 2007); especially from low incidence populations (Anderson & Smith, 2013; Regan & McElwee, 2013). In addition, these professionals can increase positive lifelong health habits for students with low incidence disabilities attending culturally diverse and high poverty schools (*Federal Register*, 2014). Moreover, highly qualified adapted physical education teachers must possess a comprehensive content knowledge in disability studies, assessment methods, special education law, development of IEPs, adaptations and modification for physical education, behavior management, individual teaching and learning styles, inclusive practices, instructional design and planning, professional leadership, and assistive technology for physical education (Kelly, 2006).

However, most adapted physical educators have little or no knowledge and no previous training related to teaching students with deafblindness (Lieberman, Haibach, & Schedlin, 2012; Lieberman & Houston-Wilson, 1999). Many adapted physical educators also lack knowledge in how to develop appropriate programs and interventions for students who are deafblind (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 development.

Over 60% of the children with deafblindness in special education are receiving their education in local schools (NCDB, 2014). In addition, 65% of elementary school aged children with deafblindness are being educated a portion of their day in a regular classroom (NCDB, 2014). With a large portion of these individuals being instructed in

their regular classroom for some part of their day as their least restrictive environment, physical educators need to be prepared to include students with deafblindness in general physical education and adapted physical education. These children are unique in their degree of vision and hearing loss; however, the effects of both occurring together make a significantly greater impact on these individuals and their need for appropriate educational techniques.

Since children with deafblindness have limited exposure and ability to obtain information about the environment surrounding them they learn best by direct instruction through quality teaching. Qualified teachers " . . . provide equitable access and opportunities that build upon and extend what learners already know in facilitating the ability to acquire, construct, and create new knowledge" (Hollins, 2011, p. 395). Furthermore, qualified teachers demonstrate effective teaching strategies (Lytle, Lavay, & Rizzo, 2010).

Due to the dual sensory loss of children who are deafblind, the ability to obtain information and benefit from incidental learning is limited. Incidental learning advances knowledge through the automatic flow of sensory information. This continuous knowledge, exposure, and experience within one's environment are available to an individual through incidental learning (Alsop et al., 2012). However, children who are deafblind do not fully receive the same amount of knowledge and experience due to their vision and hearing loss. Therefore, quality teaching must also include the knowledge of learners, which comprises of individual characteristics, background experiences, and prior knowledge (Hollins, 2011). Since many times the incidental knowledge and

experience may be received incomplete or distorted due to the vision and hearing loss, educational interventions must be used to fill in those misconnections of knowledge.

In addition to limited access to incidental learning, children with deafblindness also have fewer opportunities to participate in physical activities, are overall less active than their peers, and have motor skill delays (Lieberman, Bryne, Mattern, Watt, & Fernandez-Vivo, 2010; Lieberman & Houston-Wilson, 1999; Sherrill, 2004). Specifically for children with deafblindness, movement provides a means to explore the world around them. Movement allows the child to explore new objects, people, and environments. This exploratory movement has been shown to associate with perceptual and cognitive development (Gibson, 1988).

A child's mobility is affected by his or her ability to process the environment, know when and how to move, and interact with the surrounding environment (Levtzion-Korach et al., 2000). A child with deafblindness not only has decreased sensory input but limited motivation needed to stimulate purposeful movement within the environment. Motivation is thought to trigger movement (von Hofsen, 2004). The exploration and movement performed throughout the environment broadens a child's perspectives of his or her world and knowledge. As stated earlier, movement is crucial for the overall development of children with deafblindness. Acquiring independent movement is a stepping-stone to gaining independence, knowledge, and experience about the world around them.

CHARGE Syndrome and Motor Development

Within the estimated 9,454 individuals with deafblindness documented in the National Child Count, 864 have CHARGE syndrome as their primary identified etiology

(NCDB, 2014). Therefore, this syndrome is currently the leading identified cause of childhood deafblindness. This syndrome is a multifaceted syndrome of complex birth defects. The original clinical criteria for diagnosing an individual consisted of identifying 4 of the 6 characteristics of CHARGE association, currently known as 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,

Retardation of growth and/or development,

Genital and/or urinary abnormalities, and

Ear abnormalities and deafness (Pagon, Graham, Zozana, & Yong, 1981).

However, Blake et al. revised the clinical criteria in 1998, identifying additional major and minor characteristics of CHARGE syndrome, which are now used to diagnose a child. The child must have three of the four major characteristics or two major plus three minor characteristics to be diagnosed with CHARGE syndrome (Blake et al., 1998). The major characteristics are more specific to the syndrome than the minor characteristics. However, the major and minor characteristics are rarely seen in combination with any other conditions or syndromes.

The major characteristics that are commonly related to CHARGE syndrome are: (a) coloboma, which is a cleft or hole in one of the structures in the eye (i.e., iris, retina, choroid); (b) choanal atresia, which is narrowed or completely blocked nasal pathways; (c) cranial nerve dysfunction, which can cause swallowing problems, facial paralysis,

diminished or absence sense of smell, and sensorineural hearing loss; and (d) characteristic ear abnormalities (Blake et al., 1998). The minor characteristics are: (a) genital hypoplasia; (b) developmental delay; (c) cardiovascular malformations; (d) growth deficiency; (e) orofacial cleft (i.e., cleft lip, palate); (d) tracheoesophageal fistula; and (e) distinctive face. In addition, there are some occasional findings of additional characteristics in individuals with CHARGE syndrome but at lower frequencies. 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 chromodomain helicase DNA binding protein 7 (CHD7) gene was reported in approximately two-thirds of children with CHARGE syndrome have this gene mutation (Vissers et al., 2004). The one-third of children with CHARGE syndrome without the identified mutation of the CHD7 gene were diagnosed based on the major and minor characteristics associated with the syndrome. Therefore, CHARGE syndrome is a congenital autosomal dominant genetic disorder (Vissers et al., 2004). With the discovery of the causative gene, revision to current clinical diagnostic criteria included broadening of the major characteristics and examining CHD7 variants (Hale, Niederriter, Green, & Martin, 2016).

The effect of a dual sensory loss in children with CHARGE syndrome cannot be examined as two separate entities of vision and hearing; it is the combined degree of loss in both vision and hearing that can potentially affect the overall motor development of the

individual. CHARGE syndrome often results in developmental delays including motor delays (Dammeyer, 2012; Smith, Smith, & Blake, 2010).

In addition, many children with CHARGE Syndrome spend much of their infancy and childhood in and out of the hospital for various surgeries (e.g., heart, choanal atresia, tracheostomy), which can cause motor (e.g., rolling, sitting, walking) delays from the restricted environment (Hartshorne et al., 2011; Salem-Hartshorne & Jacob, 2004). The loss of both distant senses of vision and hearing and time hospitalized greatly affects and presents challenges to overall human development and learning. For instance, balance and mobility depend on vision and vestibular function, which are affected by vision and hearing loss (Hartshorne et al., 2011; Pogrund & Fazzi, 2002).

Each particular movement task (e.g., sitting, crawling, walking) can be affected differently by the degree of hearing and vision loss. Aspects of the task should be examined when determining adaptation to instruction (Lieberman et al., 2013). Since vision and hearing are both considered distant senses, these senses provide information about the surrounding environment to the child (Hartshorne et al., 2011). The use of any residual hearing or vision can greatly affect their ability to gather additional information and perform a task. Many researchers have reported out of the five senses, vision and hearing are reported to be used approximately 90% for learning and gathering information about the environment (Chrétien, 1995; United States Department of Labor, n.d.). In support of this statement, Houwen, Visscher, Lemmink, and Hartman (2009)

stated the following are functions of vision related to motor development:

> (a) incentive function – to motivate children to move, (b) spatial function – to provide information about distance and direction of movements and objects, (c) protective function – to anticipate dangerous situations, and (d) controlling/feedback function – to detect errors and correct the ongoing movement by ongoing regulation of the movement (p. 465).

With a loss of vision and hearing, a child with CHARGE syndrome is prone to have a delay in acquiring developmental motor milestones (e.g., walking; Hartshorne et al., 2007).

For movement to occur, children must have a minimal level of balance. Characteristics present in children with CHARGE syndrome result in delays in balance and mobility development (Hartshorne et al., 2011; Pogrund & Fazzi, 2002; Thelin, Curtis, Maddox, & Travis, 2007). There are a variety of characteristics (i.e., vision, hearing, vestibular) in children with CHARGE syndrome that would further delay the development of balance and mobility, therefore, affecting motor development. Vestibular dysfunction (i.e., small or absent semi-circular canals) is considered to be the major characteristic causing this delay (Thelin et al., 2007). Vestibular abnormalities are present in 80 to 90% of the population with CHARGE syndrome (Hartshorne et al., 2011). However, few children with CHARGE syndrome have a formal vestibular evaluation to determine the effect of vestibular function on balance (Travis & Thelin, 2007). This is due to the nature of the test which requires individuals to maintain a particular position, or perform a task for a significant period of time (Travis & Thelin, 2007). Eye movement

tests for vestibular function may also be affected by the individual's colobomas, making the test invalid.

However, Travis and Thelin (2007) suggested that the cause to the delay in achieving independent walking could be related to vestibular abnormalities, visual deficit, or muscular hypotonicity. Travis and Thelin (2007) also reported that children with CHARGE syndrome could achieve gross motor developmental milestones, including walking; however, the milestones are acquired at a much slower rate compared to their peers without disabilities. Further, a concomitant loss of hearing and vision has a direct negative effect on an child's balance, motor development, postural development, and acquiring developmental milestones (e.g., walking; Hartshorne et al., 2011). In addition, previous research indicated a strong correlation between the attainment age of independent walking in children with CHARGE syndrome with the acquisition of symbolic communication (Thelin & Fussner, 2005), language (Petroff, 1999), adaptive behavior scores (Salem-Hartshorne, 2003; Salem-Hartshorne & Jacob, 2004), and executive function behaviors (Hartshorne et al., 2007). It is still unknown, what is the direct cause of delay in acquiring independent walking in children with CHARGE syndrome. Nevertheless, as illustrated in the previous investigations, there is a need for children who are deafblind to acquire independent mobility through walking.

Theoretical Framework

The DST was used in this investigation to gain an insight about the interactions of characteristics in children with CHARGE syndrome impact the developmental motor milestone of walking. Shirley (1931) stated that independent walking was the most

important and most impressive of the developmental milestones, as it allows the hands to be free to further explore the environment. This is particularly important to children who are deafblind, as their hands many times act as their eyes and ears by performing the exploration of their environment tactilely.

In the DST, the concept 'dynamic' can be defined as development that is nonlinear and discontinuous. Therefore, as a child develops, the development may not always be smooth and hierarchical. Developmental behavioral change (i.e., motor learning) is highly individualized. Kugler, Kelso, and Turvey (1982), in addition to a few others, first originated this dynamic systems approach to motor control. It then became a popular framework within developmental psychology and infant development (Thelen, Kelso, & Fogel, 1987).

In the DST, the term 'systems' conveys the concept that the human organism is composed of numerous body subsystems interacting to produce movement (Haibach et al., 2011). The environment also influences the continuous interactions of the subsystems within the individual. The human organism of subsystems self-organizes based on the ". . . specific conditions within the biology of the individual and the environment are met . . ." (Gallahue et al., 2012, p. 28). The body's ability to self-organize stimulates new and stable movement change to occur, ". . . only when all the components reach critical functioning and the context is appropriate does the system assemble a behavior" (Thelen, 1995, p. 82). Therefore, movement production emerges from a constant interaction of the individual, the environment, and the task (Newell, 1984, 1986).

Newell defined constraints as characteristics within the individual, environment, or task that either encourage or discourage movements (Newell, 1984). Within the DST, these constraints that promote and encourage development and behavioral change to occur are termed as affordances (Gibson, 1977, 1979). Whereas constraints that impede or delay developmental change are termed rate limiters (Thelen, 1995, 1998). The DST relies on the interconnectedness of perception, action, and cognition to perform behavior change, which is impacted by the individual, task, and environment. Various constraints must reach a critical level influenced by nonlinear interactions for motor development to occur (see Figure 3).

Movement will occur within specific contexts due to the self-organizing and interaction of various systems (i.e., sensory, motor, perceptual). In addition, the physical and social environment influences the development of new movement behaviors (Thelen & Fogel, 1989). All systems interact together for new movement patterns to emerge. New movement patterns only become preferred patterns under certain conditions. Affordances and rate limiters are both seen as constraints, which can encourage or discourage the development of new movement patterns.

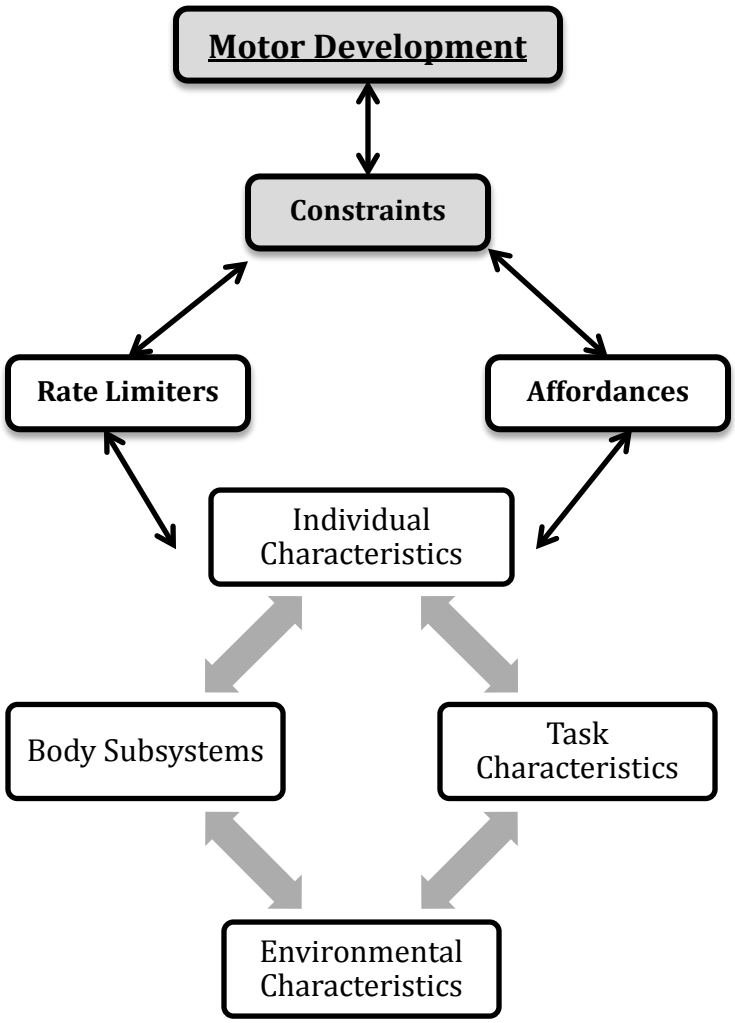

Figure 3. Dynamic Systems Theory model of the interaction and interrelation of constraints that influence motor development.

Unique constraints exist within the individual, task, and environment that lead to preferred patterns of movement development. These preferred patterns of movements typically use the least amount of energy and therefore, may lead to atypical or inefficient patterns (Thelen, 1989). The emergence of movement patterns is developed by individual constraints based on the maturation of the central nervous system, development of posture, balance, muscular strength and endurance, and the ability to process sensory

information (Haywood & Getchell, 2009). “Self-organization means that behavior emerges strictly as a cooperative function of the subsystems within particular environmental and task contexts” (Thelen & Ulrich, 1991, p. 24).

The subsystems will self-organize based on the variables from contributing constraints, which will result in movement behavioral changes in order to regain stability and organization of the movement being produced (Thelen, 1989; Thelen & Fogel, 1989). Bernstein (1967) referred to the ‘degrees of freedom’ as a limitless combination of movements that provides knowledge and control of each joint within a movement task to the individual. The ‘degrees of freedom’ is based on the individual’s ability to control and coordinate movement of neurons, muscles, and joints to perform a particular movement solution (Kugler et al., 1982).

The child’s ability to self-organize decreases the ‘degrees of freedom’ (e.g., the infinite amount of possible movement options) of the neural, anatomic, and energetic components related to the specific task or movement (Thelen, 1992). The body subsystems will always self-organize; however, self-organization may not always lead to the desired movement (e.g., independent walking) unless the rate limiters and affordances construct the most attractive option (i.e., attractor state). A child’s disability (i.e., vision, hearing loss) can alter the body’s self-organization due to numerous constraints being placed on the subsystems, which may reduce the limitless combinations of movements (i.e., degrees of freedom; Cowden & Torrey, 2007). The arrangement of a movement pattern is then the result of the constraints provided at a specific moment in time, within a specific situation or environment (Coker, 2013). If a child does not have the requisite

affordances to walk independently (i.e., balance, muscular strength), the body will self-organize to a movement pattern with a more stable attractor state (e.g., scooting).

Attractors are preferred states of stability and impact the self-organizing process (Coker, 2013). When a change in constraints is imposed on a system, its stability is endangered and a new form is established (Thelen & Ulrich, 1991). The attractor states occur in a similar pattern to basins; where deep attractor basins are much more stable and are difficult to change, compared to shallow attractor basins, which are more susceptible to change (Coker, 2013; Ennis, 1992; see Figure 4).

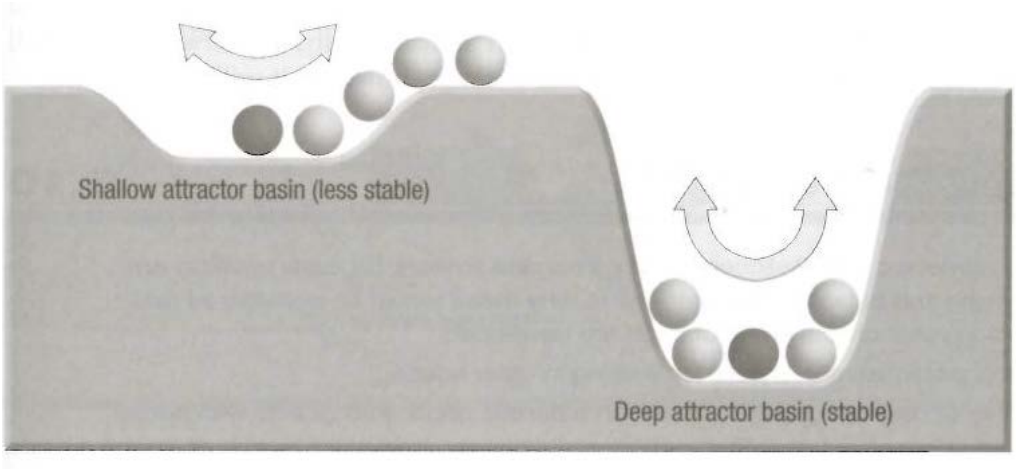

Figure 4. Shallow versus deep attractor basins. Shallow attractor states are more susceptible to change than deep attractors. From *Motor Learning and Control for Practitioners, With Online Labs,* 3rd ed., by C. A. Coker. Copyright ©2013 by Holcomb Hathaway, Publishers (Scottsdale, AZ). Used with permission.

Therefore, motor development delay can occur when a child is not able to shift into a new movement pattern and maintain stability in a desirable attractor state (Cowden & Torrey, 2007). The new movement pattern may be more or less stable due to the individual, task,

and environmental constraints. Attractor states can become more or less stable and shift into new movement patterns (Thelen, 1992).

Thelen and Ulrich stated that "… new forms [of movement] can arise only from perturbations that disrupt the stability of the old forms" (1991, p. 4). The systems in the body are 'softly assembled' in which component subsystems are able to reassemble in different ways based on the specific context (Thelen, 1989; Thelen & Fogel, 1989; Thelen & Ulrich, 1991). The coordinated structure of the individual's body subsystems lead to the use of the child's preferred pattern of movement. In addition, the interrelationship between the individual, task, and environmental constraints can also influence the stability of each attractor state, and leads to a subconscious choice of the most preferred movement for each performance (Thelen & Ulrich, 1991).

A phase shift is a transition from one movement pattern to the next (e.g., crawling to walking). During a phase shift, the first movement becomes unstable (e.g., a shallow attractor basin); therefore, encouraging a shift to another movement pattern until a more stable pattern (e.g., a deep attractor basin) is established. Movement patterns prefer to be in a state of stability rather than instability. The attractor state instability will increase the variability of the movement (Harbourne & Stergiou, 2009). As the movement pattern travels through the phase shift, it will develop into a combination of the old stable movement pattern and the new unstable movement pattern. Changes in behavior are therefore a result of a series of phase shifts and transitions between the systems' state of stability with the new movement pattern eventually developing into a more stable option that becomes the new norm (Coker, 2013).

This phase shift into a new attractor state comes from various variables and constraints applied to the systems that change the child's behavior or replace a patterned behavior with a new movement. The constraints within a task, the individual, and the environment interact and can potentially modify and influence the system's stability and the attractor state of a new movement pattern (Thelen, 1992). Control parameters (i.e., affordances) are constraints that move the system into new attractor states, such as increased muscular strength and balance. In contrast, rate limiters are constraints that function to hinder or hold back the ability of a system to change, such as fear.

Therefore, as new movements emerge during phase shifts, the attractor states stabilize and destabilize as a function of rate limiters and affordances. The task and the environment can be adapted to facilitate the phase shift to a new attractor state. Intervention strategies and practice may be needed to cause instability to an already deep attractor state (e.g., back scooting, 5-point crawl) in order for a change in movement to occur (Coker, 2013). This change in movement patterns is fluid and nonlinear; however, it is always seen as nonrandom (Thelen, 1989; Thelen & Ulrich, 1991). Eventually through practice and the body's positive feedback loop, a new attractor state is formed, and eventually with continued practice it will gain stability and develop into a deep attractor basin (Coker, 2013).

Researchers have reported that children with CHARGE syndrome have a significant delay in acquiring developmental motor milestones, including independent walking (Blake et al., 1998; Hartshorne & Cypher, 2004; Travis & Thelin, 2007). With the use of the DST, investigation about the impact of rate limiters and affordances on the

development of walking in children with CHARGE syndrome may provide some understanding about their developmental delay and offer potential suggestions for future interventions. Due to the complexity of the syndrome, it is difficult to understand the various constraints and the intricacy of each child's subsystems, which may impact the attainment and stability of independent walking as an attractor state.

Purpose of the Study

The purpose of this investigation was to examine the impact of individual rate limiters and affordances on acquiring the independent walking developmental milestone in children with CHARGE syndrome. The investigation was divided into two phases that were performed concurrently and both examined the constraints that delayed (i.e., rate limiters) or contributed (i.e., affordances) to the attainment of the independent walking developmental milestone in children with CHARGE syndrome.

Research Questions

For the purpose of the study, there are four research questions and two phases that are as follows:

Phase I: CSCDP: children with CHARGE syndrome rate limiters and affordances

1. What is the significant impact of vision and hearing loss as rate limiters in children with CHARGE syndrome on their attainment of independent walking while statistically controlling for the months in which the child with CHARGE syndrome was hospitalized?

2. What are additional significant rate limiters or affordance variables that are related to the age at which independent walking in children with CHARGE syndrome is attained?

Phase II: Parent or primary caregiver perspective of their child with CHARGE syndrome rate limiters and affordances

3. What do parents or primary caregivers propose are affordances that lead their child with CHARGE syndrome toward the ability to walk independently?
4. What do parents or primary caregivers propose are rate limiters causing a delay in walking for their child with CHARGE syndrome?

Limitations

This study is subject to the following limitations:

1. The effect of balance issues related to vision loss and hearing loss on motor development cannot be addressed quantitatively.
2. The amount of interventions and services provided to the individuals (i.e., early intervention, physical therapy) may affect the motor performance of the child with CHARGE syndrome and could not be controlled.
3. The heterogeneity of the group's characteristics of CHARGE syndrome could not be controlled entirely.

Delimitations

This study is subject to the following delimitations:

1. Parents or primary caregivers attended an international conference and were a part

of the CHARGE Syndrome Foundation; therefore, they may have been more proactive in their child's development.

2. Only children with CHARGE syndrome who have achieved independent walking were participants for this investigation.
3. Descriptive information used to analyze the developmental milestones was limited by self-reports from the participants' parents or primary caregivers, both in the database and interviews.
4. Participants were a convenience population, which potentially limits the ability to generalize the results.
5. Exclusive use of age of acquisition of independent walking as measured in months.
6. Exclusive use of parents or primary caregivers who expressively and receptively spoke and read English.

Definition of Terms

The definitions of the following terms are essential to the understanding and purposes of this investigation:

Balance (static and dynamic): refers to the ability to maintain equilibrium of the body whether in a static (i.e., stationary) or dynamic (i.e., moving) position (Haywood & Getchell, 2009; Payne & Isaacs, 2002). "Static balance involves maintaining one's equilibrium while the center of gravity remains stationary. Dynamic balance involves maintaining one's equilibrium while the center of gravity shifts" (Ozmun & Gallahue, 2011, p. 388). Stability and balance are at times interchangeably defined due to both

contributing to essential elements of movement. Stability is defined by inhibiting mobility by resisting movement (Haywood & Getchell, 2009).

Blind (blindness): ". . . an impairment in vision that, even with correction, adversely affects a child's educational performance. The term includes both partial sight and blindness" [34 C.F.R. §300.8 (c) (13), IDEIA, 2004]. The classification of blind according to the *World Health Organization, International Classification of Diseases* (ICD-10) is a visual acuity of 20/400 or less (WHO, 2010).

Deaf (deafness): " . . . a hearing impairment that is so severe that the child is impaired in processing linguistic information through hearing, with or without amplification that adversely affects a child's educational performance" [34 C.F.R. §300.8 (c) (3), IDEIA, 2004].

Deaf and Deafblind: Although in this investigation, the term deaf with a capital "D" and deafblind with a capital "D" will not utilized, the researcher acknowledges and respects those who are within the Deaf culture and community and the Deafblind culture and community, who have their own unique language, history, arts, and sports (Hodge, Lieberman, & Murata, 2012). Since the researcher is unaware of each participant's involvement in the Deaf culture and community and the Deafblind culture and community, the researcher will use person first language within this study when referring to individuals who are deaf, deafblind, or who have CHARGE syndrome, which is supported by the United States Centers for Disease Control and Prevention. If specific articles and references use the term Deaf pertaining to their research, then those terms will also be reflected throughout this study.

Deafblind (deafblindness): For the purpose of this investigation, deafblind and deafblindness will be used without the hyphen to recognize the impact and unique condition of the combined sensory loss of both hearing and vision. Deafblindness is " . . . 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), IDEIA, 2004]. Deafblindness is also referred as a dual sensory loss, which will be used interchangeably throughout this investigation.

Gross motor: The term gross motor involves movements and skills, which use large muscles of the body, such as legs or arms (Haywood & Getchell, 2009; Payne & Isaacs, 2002).

Hard of hearing: Although a person who is hard of hearing is defined in IDEIA, 2004, as a person with a hearing impairment, the term hard of hearing will be used instead in this investigation. The term hard of hearing is valued within the Deaf culture and the Deaf community instead of the term hearing impairment. In IDEIA (2004) hearing impairment is defined as, "an impairment in hearing, whether permanent or fluctuating, that adversely affects a child's educational performance but that is not included under the definition of deafness in this section" [34 C.F.R. §300.8 (c) (5)].

Hearing loss: Refers to those individuals who are hard of hearing or deaf in which the loss of hearing impacts an individual's learning and development (American

Speech-Language-Hearing Association, 2014). The WHO (2015) will be used for classification of hearing loss. WHO defines hearing loss in children 13 years and younger with a loss greater than 30 decibels.

Locomotor skills: Locomotion is the act of moving by transporting the body from one place to another location (Haywood & Getchell, 2009). Locomotor skills can include but are not limited to the following: walk, run, skip, slide, leap, jump, and hop (Ulrich, 2000).

Motor behavior: "Is an 'umbrella' term referring to changes in motor learning control and development that embody learning factors and maturational processes associated with movement performance" (Gallahue et al., 2012, p. 14).

Motor development: "Refers to the continuous, age related process of change in movement, as well the interacting constraints (or factor) in the individual, environment, and task that drive these changes" (Haywood & Getchell, 2009, p. 5).

Motor milestone: "A fundamental motor skill whose attainment is associated with the acquisition of later voluntary movements. The order in which an infant [or child] attains these milestones is relatively consistent, although the timing differs among individuals" (Haywood & Getchell, 2009, p. 101).

Motor performance: Performance of a voluntary movement task or action of one or more body parts that is observable and is based on the interaction of the individual, the environment, and the task characteristics (Gallahue et al., 2012; Hutzler, 2007; Newell, 1986).

Sensorineural hearing loss: This type of hearing loss is caused by damage to the sensory cells in the inner ear, including the cochlea and its receptors, and or the nerve

fibers from the inner ear to the brain (National Institute on Deafness and Other Communication Disorders, 2013).

Vestibular dysfunction: Damage to the vestibular system (i.e., utricle, saccule, semicircular canals, VII cranial nerve) may disrupt information about the body's motion, equilibrium, and spatial orientation and impact gaze and balance abilities (Hanes & McCollum, 2006; Rine, 2009; Shumway-Cook & Woolacott, 2001; Vestibular Disorders Association, 2008).

Visual impairment: Defined in IDEIA (2004) as, ". . . an impairment in vision that, even with correction, adversely affects a child's educational performance. The term includes both partial sight and blindness" [34 C.F.R. §300.8 (c) (13)]. Visual impairment is when vision loss is defined in terms of visual acuity loss or visual field loss within the organ functions (WHO, 2010). In addition, vision loss is a general term that encompasses a broad degree of visual impairment, including low vision and blindness. The ICD will be used for classification of low vision and blindness. ICD defines low vision with a range from 20/70 to 20/400 of visual acuity and blindness as 20/400 or less (WHO, 2010).

Walking: The conceptual definition of walking is a locomotor skill in which "weight shifts from the left to the right foot, with one foot in contact with the ground at all times" including a "support phase and a swing phase" of each foot to perform a gait cycle (Haibach et al., 2011, p. 101). In addition, the operational definition used in the present investigation of walking includes that the individual is able to upright bimodal locomote independently for three to five consecutive steps without falling or hesitating.

Yaguramaki and Kimura (2002) define the onset of independent walking, “as the moment at which infants can walk five steps independently” (p. 69).

CHAPTER II

REVIEW OF LITERATURE

As more children with deafblindness are being educated within their local school districts and in general education classrooms, it is crucial that educators (e.g., general and adapted physical educators) collaborate with other professionals to accelerate the motor development of children with CHARGE syndrome (coloboma of the eye, heart defects, atresia of the choanae, retardation of growth and/or development, genital and/or urinary abnormalities, and ear abnormalities and deafness). Improved motor development, especially independent walking, can promote an increase in academic achievement and independence. The purpose of this investigation was to examine the impact of individual rate limiters and affordances on acquiring the independent walking developmental milestone in children with CHARGE syndrome. The cause of developmental motor milestone delay in children with CHARGE syndrome is still unknown; however, some researchers have speculated some constraints that may impact development.

Numerous researchers have considered the walking developmental milestone as an indicator of the level of intelligence, language, behavior, and communication in individuals with deafblindness (Dunlap, 1985; Hartshorne & Cypher, 2004; Petroff, 1999; Salem-Hartshorne, 2003; Salem-Hartshorne & Jacob, 2004; Thelin & Fussner 2005). Previous researchers have indicated the effect vision and hearing loss have on

motor performance. However, there is limited research on the impact of the combined vision and hearing loss. Therefore, in this chapter, the investigator reviewed the literature related to the development of walking and the impact of vision loss and hearing loss on motor development. The dynamic systems theory (DST) was used as the conceptual filter to provide the reader both an explanation and a connection between the literature review and the methodology and results that are conducted in this investigation.

The chapter is organized into six sections: (a) Adapted Physical Activity Taxonomy, (b) Development of Walking, (c) Barriers to Physical Activity Participation, (d) Effect of Vision loss on Motor Development and Walking, (e) Effect of Hearing loss on Motor Development and Walking, and (f) CHARGE Syndrome and Motor Development. These six sections provide significant support to investigate and examine the potential impact of both vision and hearing loss on independently walking in children with CHARGE syndrome.

Adapted Physical Activity Taxonomy

Adapted Physical Activity Taxonomy (APAT; Carano, 2014) was used to evaluate the quality of strength within a field of study through research articles and to determine the level of recommendation for the organization of evidence in all the retrieved research that pertains to the impact of dual sensory loss and motor development. APAT involves a systematic review of literature by determining the quality of strength for each study into three levels and a level of recommendation for a sound physical activity practice. Taxonomy evaluation tables identified in this chapter are located in Appendix A. These tables within the appendix are identified throughout this chapter as A, which represents

Appendix A, and then the table number indicating the specific research article, for example: (Thelen & Fisher, 1982, Level 3, A 1).

Quality of Strength of the Study

The APAT is used to determine the quality of strength of a study. A research study can be placed into three different levels of strength: Level 1, strong; Level 2, moderate; and Level 3, weak. The quality of strength is based on five domains assessed within the original research article. Each domain has quality of study indicators that must be met. The domains that are evaluated in the original research article are the: (a) introductory section; (b) method section; (c) result section; (d) discussion section; and (e) other, which includes the references and appendices.

The APAT has four general types of research methodologies, each containing their own quality indicators for the study pertaining to each level of strength within the domains. The four research methodologies are: qualitative design, correlational design, single subject design, and experimental/quasi-experimental designs. The task force members of the division for research for the Council for Exceptional Children support the four research methodologies to provide evidence for effective practices in special education (Odom et al., 2004). This current body of literature included 1% (n = 4) of Level 1 studies, 18% (n = 9) of Level 2 studies, and 74% (n = 36) of Level 3 studies.

Level of Recommendation

The APAT provides three levels of recommendation for the organization of evidence pertaining to this research study that is incorporated in this chapter. The levels of recommendation are based on the grading system used in the Strength of

Recommendation Taxonomy (SORT) grading guidelines (Newman, Weyant, & Hujoel, 2007). Under the APAT, the three levels of recommendation are A, B, C, or not needed if the study recommendation is not relevant to adapted physical activity. Recommendation Level C is based on opinion, consensus, or field-based experiences. Improvements to individuals with disabilities without examining the effectiveness of individual outcomes or an intervention are also granted a Level C recommendation. Recommendation Level B consists of inconsistent or limited evidence of significant value pertaining to adapted physical activity and individuals with disabilities. Recommendation Level A consists of reliable and high quality significant research with a validated intervention in the field of adapted physical activity for individuals with disabilities. This current body of literature included 35% (n =17) of Level A studies, 43% (n =21) of Level B studies, and 22% (n =11) of Level C studies.

Development of Walking

The majority of an infant's motor development follows a predictable sequence of behaviors emerging from reflexes and postural control to the progress of developmental motor milestones. Shirley (1931), McGraw (1932, 1940, 1941, 1945), Bayley (1936), and Gesell (1928, 1939, 1946, 1949, 1954) were among the first to research motor development and traditional developmental theories. Shirley outlined five specific phases of motor development that can be observed through infant movement and play which are: "(a) development of passive postural control, (b) development of active postural control, (c) active efforts toward locomotion, (d) locomotion by creeping and walking with support, and (e) walking alone" (1931, p. 193). McGraw (1932, 1940, 1941, 1945) then

began to analyze motor development based on the premise of the growth and function of the nervous system. McGraw established motor developmental norms through recorded observations of infant behaviors. She observed reflexive movements that appeared before controlled self-initiated motor patterns (McGraw, 1932). She also developed, based on her observations, the seven phases of erect locomotion (McGraw, 1945).

However, during this time, Bayley (1936) developed a scale to quantify infant motor development, named the *Bayley Scales of Infant Development*, which is used as a standardized instrument to provide valuable information. This information provided a sequence of infancy motor development. Gesell (1946), a pediatrician, explained general patterns and principles of infant motor development. The researcher suggested that development was based on maturation and development moved from cephalocaudal to proximodistal and the development was not always linear but a spiraling hierarchy (Gesell, 1928, 1939, 1949, 1954). Development is dynamic and progression occurs through advancements and regression of various developmental domains (Gesell, 1954).

In addition to the maturation theories, neurodevelopmental theories further examined the impact of reflexes and central nervous system on motor development. Sherrington (1906) investigated the interaction of the central nervous system and reflexes on motor movement. Rood (1956) further examined the impact of sensory systems on motor delay and the inability to separate motor functions from sensory input. The neurobehavioral theory of sensory integration was then originated by Ayres (1972, 1974, 1980, 1989), which emphasized the interaction of sensory input, motor output, and sensory feedback on movement.

Subsequently, based on various theories, the researchers shifted their focus from the initial research in motor development to examine different, new aspects of motor development. Since then, Thelen and her colleagues (Thelen, 1986a, 1986b, 1994; Thelen & Cooke, 1987; Thelen & Ulrich, 1991) have formulated a dynamic systems theory (DST) of motor development. This theory contested the maturation and neurodevelopmental theories, by providing new principles of motor development of infants.

Infant Stepping, Kicking, and Treadmill Stepping

Thelen and Fisher (1982, Level 3, A 1) examined infant stepping reflex and spontaneous kicking while in the supine position which began to contest the maturation and neurodevelopmental theories. The purposes of this investigation were to identity the difference between infant stepping reflex and kicking and to show that biomechanical demands can affect infant stepping reflex and kicking. A movement analysis was performed with concurrent electromyography and videotape frame analysis on the infants (n = 8). Each session consisted of 5 minutes of recorded spontaneous kicking with no stimulation then followed by upright supported position by the observer with the infant's soles on a mat (1.5 - 2 minutes).

There was no significant difference or correlation between the rate of stepping or kicking in the eight infants (see A 1). The stepping reflex and kicking both displayed alternative left and right movements of the legs; however, rate, alternations, and laterality did show some individual variability. The investigators also observed and recorded that the infant stepping reflex and kicking both used the same muscle synergisms through

kinematic and electromyographic examination. The investigators proposed that the disappearance in stepping and increase in supine kicking as the infant aged was due to gravity and the increased mass of the legs, which altered the dynamics of the leg movements.

In 1984, Thelen, Fisher, and Ridley-Johnson tested the proposed hypothesis by examining the relationship of physical growth on the infant stepping reflex (Level 3, A 2). “The traditional explanation for the disappearance of stepping is that this ‘primitive’ reflex is suppressed with the maturation of inhibitory tracts from the cortex” (Thelen, Fisher, & Ridley-Johnson, p. 480). However, the DST focuses beyond the central nervous system and into the interaction between additional body subsystems (i.e., muscular system).

Forty infants were observed at 2, 4, and 6 weeks of age on the number of steps they took and body size characteristics (i.e., weight, crown-heel length, crown-rump length, circumference of thighs and calves, subscapular fat fold). In addition, an arousal 6-point scale was used to document the infants’ arousal state before and twice during each session. Repeated measures ANOVA showed a significant increase due to the effect of age for all variables except arousal state. In addition, an increase in arousal state was a significant predictor of an increase in the amount of steps the infant took at each age level. Furthermore, infants that gained weight rapidly between 2 and 4 weeks stepped less. Therefore, infants at 4 weeks old may be sensitive to changes in mass that impact the ability to produce steps.

In order to test that sensitivity, small weights were added to infant legs to simulate the average mass growth that happens between 4 and 6 weeks of age. The infants at 4 weeks of age averaged 14 steps in the no weight condition; however, in the weight added condition the infants' steps declined significantly to 9 steps. There were no arousal differences between the weight and no weight conditions. Therefore, the investigators reported that the infants displayed behavioral movement change based on leg mass.

The next phase of this investigation examined infant stepping rate while submerged in water. The average number of steps in the out of water condition was 10 which significantly increased to 20 steps during the in water test condition. Arousal scores between both conditions were not different. The explanation of the disappearing infant step reflex was therefore, contributed to changes in body build and muscular strength.

As examined in the previous investigation, individual characteristics (i.e., mass, strength) may be rate limiting constraints on the ability to achieve independent walking. In addition to individual characteristics, Thelen (1986b, Level 3, A 3) stated, ". . . new skills are a product not only of cognitive or central instructions, but also of perceptual, affective, attentional, motivational, postural, and anatomic elements interacting within a particular context" (p. 1498). Each component including the environmental influence may constrain movement.

Thelen (1986b, Level 3, A 3) examined the ability to generate alternating stepping in 7-month old infants during four different conditions: (a) supine kicking, (b) stepping with no treadmill, and (c) stepping with a treadmill set to two different speeds (0.10 meters per second and 0.19 meters per second) with the investigator allowing the infant to bear a

comfortable amount of weight on his or her feet. The infants were videotaped and the movements were compared on the frequency or rate, laterality, degree of alternating movements, and intralimb and interlimb coordination based on each condition.

Movements in both legs significantly increased on a moving treadmill when compared to supine position and stepping with no treadmill. In addition, an increase in treadmill speed displayed a significant increase in the frequency of steps. The treadmill also significantly increased the percentage of alternations and laterality preference (see A 3). However, the speed of the treadmill had no significant effect.

The interjoint coordination was analyzed based on the hip, knee, and ankle joints compared to a female adult. The infant steps were more synchronized, with the hip and knee joint flexing and extending together. Phase durations were significantly affected by treadmill speed with phase duration increasing as the speed increased. In addition, the swing and stance phase were significantly different based on the treadmill speed. The infant's steps were increased with the treadmill speed and phase adaptations were made similar to adults.

Therefore, when 7-month old infants were placed on a treadmill, complex coordination of alternating stepping was performed. However, when the infants were in supine position or an upright position with no treadmill conditions, the participants displayed fewer steps and movements, which were more asymmetrical, fluctuating, and uncoordinated. The infants also displayed an increase in stepping in no treadmill condition after experiencing the treadmill condition at two different speeds, but the pattern was more immature. Most all infants performed few or no steps without the

treadmill turned on even while in an upright position. Thelen (1986b) proposed that the treadmill provided mechanical leg stretch to generate alternating steps until the infant is strong enough and has enough balance to replicate these movements independently.

Thelen and Ulrich (1991, Level 3, A 4) continued to examine the acquisition of independent walking through a longitudinal study through the use of treadmill stepping in seven infants from the age of 1 month until 10 months of age, which introduced the DST to understand the processes related to motor development. Each infant was tested twice each month, which were 2 to 3 days apart from each other within a week of the infant's month anniversary. Infrared emitting diodes were used to analyze the infants' movements. An opticoelectronic motion analysis system was use to examine right and left foot movements during treadmill testing. Foot movement was coded as alternating step, parallel step, double step, or a single step.

During each testing, the infant had 5 to 10 minutes to assimilate to the testing environment. Then, eleven 20-second treadmill trials were performed, while the investigator assisted the infant in an upright position and allowed the infant to support as much body weight as possible. Of the 11 Trials, Trials 1 and 9 were baseline trials in which the treadmill belt was stationary. In trials 2 through 8, the treadmill moved at the same speed but gradually increased for each succeeding trial. Trials 10 and 11 were mixed speed, in which the right belt would move faster than the left (Trial 10) or the left belt would move faster than the right belt (Trial 11).

During the baseline trials (i.e., 1 and 9), the infants took very few steps; however, with the treadmill on, they produced steps similar to research conducted by Thelen and

Ulrich (1991). Furthermore, there was a significant increase in the number of steps with age and increased treadmill speed. Descriptive information of alternating steps in the infants displayed an initial phase of limited steps, followed by a short period in which the amount of steps declined then followed by a steady increase.

The treadmill stepping results were impacted by the overall rate of general motor maturation (i.e., *Bayley Scales*); developmental changes in the proportion and composition of the infant's legs; difference in arousal and mood; and changes in the predominant postures and movements of the legs (Thelen & Ulrich, 1991, Level 3, A 4). Therefore, it was concluded that the development of independent walking is an emergence of specific factors and is not a prescribed process (Thelen et al., 1987; Thelen, Ulrich, & Jensen, 1989). "Each movement for balance or traveling is associated not only with its functional consequences but also with the multimodal feedback from the visual, vestibular, tactile, and proprioceptive receptors that are concurrently activated" (Thelen & Ulrich, p. 91). With repeated trials and practice of the components of locomotion, this preferred attractor state forms into independent walking.

Balance and Stability

The ability to maintain balance and postural stability requires the integration of vestibular, proprioceptive, and visual information into appropriate motor responses; therefore Palm, Strobel, Achatz, von Luebken, and Friemert (2009, Level 3, A 5) investigated the influence of visual or auditory information on postural control. Twenty-three young, healthy, active participants who had no history of vestibular, vision, or auditory disease were used in this investigation. Postural stability was measured with a

Biodex Medical Systems *Balance System*™. The system measures medial-lateral stability, anterior-posterior stability, and the overall stability. The participants participated in three visual conditions: eyes closed, sway-feedback condition (i.e., visual feedback from the computer screen), and eyes open with no visual feedback from the screen. Within these three conditions, the participants either had auditory input (i.e., music at 75-80 decibels) or no auditory input. The six test conditions were randomized for each participant.

Auditory input (i.e., music) did not result in significant differences in the overall stability scores during the three different visual conditions. Regardless of the auditory input, there were significant differences between each visual condition based on overall stability. The overall postural stability was significantly worse in the eyes-closed than in the eyes-open condition and balance control was significantly improved with visual feedback provided in the sway feedback condition (see A 5). Therefore, in this investigation an increase in visual information lead to an increase in postural stability while distracting auditory information did not impact postural stability.

In addition to auditory and visual information to maintain postural stability, the somatosensory system, which includes proprioception, also provides information to assist in postural stability. The somatosensory systems provide information of touch, pain, temperature, and limb position through cutaneous receptors (i.e., skin) and proprioceptors (i.e., muscles, tendons, joints; Rose & Christina, 2006). The impact of somatosensory information used during the acquisition of independent upright stance was investigated longitudinally through four developmental periods: pull to stand, stand alone, walking

onset, and postwalking (i.e., 1.5 months of walking) by Barela, Jeka, and Clark (1999, Level 3, A 6). Five infants were tested within 1 week of each of the four developmental periods. The infants had time to acclimate themselves to the laboratory setting, which was a small room surrounded by black curtains to reduce distractions.

Within the room was a wood cube affixed to a force platform and a small pedestal on which the infant would stand to discourage foot movement. The infant would stand upright and touch the side of the cube, which had a polyvinyl chloride (PVC) tube attached that recorded the magnitude of the force and the position. Body sway was recorded during half the trials by a video system and the other half by a tracking positional system. Each trial lasted for 1 minute.

Applied force in the medial-lateral direction decreased from pull to stand to stand alone, and to walking onset. The infants' applied force in the vertical direction decreased throughout each of the four developmental periods. In addition, medial-lateral and anterior-posterior body sway significantly decreased at the postwalking period compared to walking alone and pull to stand. Applied force and body sway were positively related in the anterior-posterior position. The investigators reported that body sway controlled the applied force present in the pull to stand, stand alone and walk alone developmental phases. However, in the postwalking phase, applied force controlled body sway movements. Based on the results, infants used surface contact for mechanical support rather than sensory information to assist in the attainment of developmental motor milestones.

Metcalfe and Clark (2000, Level 2, A 7) continued this investigation by examining the role of sensory information in postural control within newly walking infants. In order to coordinate postural orientation, the body's muscles and joints must work together based on the variability of the task and ever-changing environment providing multiple degrees of freedom (i.e., limitless possibilities of movement). To organize this coordination, at times infants will decrease the degrees of freedom by focusing on body segmental movement instead of moving the entire body (Thelen & Smith, 1996). However, postural coordination must be flexible and move together in order to adapt actions as needed, which establishes the importance of a functional perception-action relationship.

When the infants used surface contact there was a significant decrease in head and shoulder sway. Overall, center of mass sway was also reduced with the use of surface contact (see A 7). Within the three walking age groups (i.e., 1-4; 5-8; and 9+ months of walking), the use of surface touch increased the postural coordination variability. This implies that infants use somatosensory cues to explore different patterns and degrees of freedom of postural coordination. Motor actions are used to explore and gain insight based on perception and action of the specific task.

Since vision, hearing, and the somatosensory systems provide information for balance and postural control then individuals with hearing or vision loss would have delays based on the limited information being provided about his or her body in space (Russel & Nagaishi, 2005). Therefore, Uysal, Erden, Akbayrak, and Demirtürk, (2010, Level 3, A 8) examined the effect of congenital loss of hearing and sight on gait and

balance in children. The standing balance subtests of the *Southern California Sensory Integration Tests* (Ayres, 1976) were used to measure balance on one foot with eyes open and eyes closed. Gait analysis was performed on a powered surface, which measured the individual's step length, stride length, step width and step angle. The number of steps per minute was also documented.

In all balance and gait analysis, the group with hearing loss performed significantly better than the group with vision loss. Furthermore, the control group performed significantly better in all the balance and gait tests when compared to the vision loss group and all but the gait speed when compared to the hearing loss group (see A 8). The body mass index of the participants did not affect the children's balance or gait. Therefore, children with vision loss had more difficulties balancing then the group with hearing loss and the control group. The children with vision loss also had shorter step and stride length, a wider support surface and foot angle, and a slower gait speed when compared to the group with hearing loss and the control group. The investigators reported that vision loss had a greater impact on balance and gait than hearing loss.

Prediction of Motor Development Level

Throughout each developmental motor milestone achieved, infants learn about their bodies and gain neuromuscular strategies based on the relationship of perception and action. Developing postural control is critical to the development of locomotor skills (Looper, Talbot, Link, & Chandler, 2015, Level 3, A 9). Movements from one postural or developmental milestone to another would provide infants with opportunities to explore and learn body control. Looper et al. (2015) examined if transitional skills (i.e., rolling,

rotating to sitting, pulling to stand, squatting to stand, squatting to the floor) and sitting correlated to locomotor skills (i.e., hands and knees crawling, cruising, walking).

Movement Assessment Battery of Children (Henderson, Sugden, & Barnett, 2007) was used to observe motor skills in eight children from 6 months old until the children reached 5 months of walking experience. Monthly visits were conducted with standardized equipment available for the child to play and interact with at each visit. Pearson product moment correlation was used to determine the correlation between transitional skills and locomotor skills and between sitting and locomotor skills. Four of the five transitional skills (i.e., rotating to sitting, pulling to stand, squatting to stand, squatting to the floor) displayed strong significant correlations to all three locomotor skills. Independent rolling and sitting were not significantly correlated to locomotor skills (see A 9). Therefore, majority of transitional skills are largely connected to the ability to locomote. The investigators stated the control of pelvis and trunk may be the factor that links transitional skills to be highly correlated to locomotor skills.

The ability to examine characteristics that promote and predict motor development are essential for early identification of motor delays or difficulties and can provide valuable information for interventions (Gallahue & Ozmun, 1997). Charitou, Asonitou, and Koutsouki (2010, Level 3, A 10) investigated the ability to predict motor development based on early motor performance indicators in infancy. The infants (n = 46) were tested with the *Alberta Infant Motor Scale* (*AIMS*; Piper & Darrah, 1994), which is an observational norm-referenced assessment scale, constructed to measure gross motor maturation in infants from birth through independent walking. The *AIMS* is

divided into four subscales positions – prone, supine, sit, and stand – containing a total of 58 items. Each item describes three aspects of motor performance: weight bearing, posture, and antigravity movements. The participants in this investigation were divided into two groups. One group was reassessed at 2 months and the other was reassessed at 6 months. Stepwise regression was used to predict the infant's motor development.

The four subscales of the *AIMS* were significantly different from the total *AIMS* score in both the 2 months and 6 months reassessment groups. Supine position significantly predicted infant development within the group that was reassessed after 6 months. In the group that was assessed after 2 months, supine and prone positions were significant early motor performance predicting factors of motor development (see A 10).

Transition from Crawling to Independent Walking

"The transition from traveling on all fours to moving upright entails different body postures, patterns of coordination between limbs, and relations between the body and the environment" **(**Adolph & Tamis-LeMonda, 2014, p. 187). Children must move from crawling, a less effective strategy, to a new attractor state of walking. This new skill of walking will initially be less functional but will eventually become a more effective movement pattern.

To examine this transition from crawling to independent walking, Adolph et al., (2012, Level 3, A 11) examined natural infant locomotion observed through active free play. The crawling and walking age was determined when the infant could travel 10 feet (3.048 meters) without stopping. Twenty crawlers and 114 walkers were observed and videotaped (15 to 30 minutes) in a laboratory playroom filled with furniture, different

ground surfaces, and toys, in which the infants freely could move throughout the environment. Fifteen additional walkers were observed at their home setting (60 minutes) in which the parents interacted normally with their infants. At the end of the laboratory testing, each infant's walking skills were analyzed over a pressure sensitive mat to examine step length and step width. In addition, accumulated time walking or crawling, number of steps, and number of falls were used to measure each infant's natural locomotion.

The result of functional skills of 12-month old crawlers and walkers were that novice walkers fell significantly more times per hour than expert crawlers. However, walkers spent significantly more time in motion, accumulated more steps, and travelled greater distances than crawlers (see A 11). Falling rate decreased in walkers when the increase in activity and movement was also examined. Walking skills (i.e., step length, step width) were significantly correlated to test age and walking age. Older infants who had more walking experience also took significantly longer and narrower steps. The older infants also spent more time walking, took more steps, and fell less. Expert crawlers were not more skilled than novice walkers, allowing walking to be seen as a more appealing attractor state than crawling.

Walking allows infants to move faster and further without increasing the risk of falling (Adolph et al., 2012, Level 3, A 11). Crawlers do increase speed over weeks of experience but speed continues to increase when infants begin walking (Adolph, 2008). Infants are also able to see more when they transition from crawling to walking. When crawling, initially an infant's neck and eyes are pointed downward, allowing his or her

vision to primarily be focused on his or her hands and the floor. However in contrast, walking permits infants to see distant objects and people (Franchak, Kretch, Soska, & Adolph, 2011, Level 3, A 12; Kretch, Franchak, & Adolph, 2014, Level 3, A 13). Franchak et al. (2011, A 12) attached a head mounted eye tracker to 14-month-old infants to examine their visual behavior during natural interactions. The infants' prospective fixations of objects and obstacles were significantly different for reaching, crawling, and walking, primarily due to the different physical constraints (i.e., hand, head positions) involved with each of the three actions.

Kretch et al. (2014, Level 3, A 13) examined visual input difference between crawling and walking. Infants 13 months old in Study 1 crawled or walked down a straight path wearing a head mounted eye tracker that recorded gaze direction and head-centered field of view. On 25.7% of steps taken, the crawler's view contained only the floor for the entire videotape of the scene. Therefore, while crawling, infants may miss visual input from distal parts of the room around them. The highest point of visibility was significantly greater for walking infants than those who crawled. The closest point of visibility for crawlers were closer to the crawler's hands (M = 8.22 inches/20.89 centimeters) than to the walker's feet (M = 32.68 inches/83.00 centimeters). Also, 9 of the 15 infants who were crawling switched to sitting during mid-trial. Sitting significantly increased the infants' point of visibility and the infants were more likely to be able to view their caregivers' face and toy. In Study 2, crawlers displayed significantly more variability in head position than did the walkers. In addition, crawlers also

displayed significantly larger deviations in head pitch angle between consecutive steps than walkers (see A 13).

Furthermore, in addition to greater visual access, walking frees both hands to be used to grab and transport objects. Karasik, Tamis-LeMonda, and Adolph (2011, Level 3, A 14) reported that 13-month old experienced crawlers travelled to distance objects only 4 times per hour compared to beginner 13-month old walkers who travelled 12 times per hour. At 13 months, walkers were three times more likely to travel to objects to interact with them compared to crawlers. In both groups, infants significantly increased in the frequency of carrying objects; however the increase was significantly larger in the crawler-walker group. There was no difference in using objects for social exchange with mothers based on the two groups (i.e., crawlers-crawlers, crawlers-walkers). However, the crawler-walker group significantly increased moving object bids from 0.54 (*SD* = 1.47) at 11 months to 7.13 (*SD* = 7.26) at 13 months while stationary bids remained unchanged (see A 14). Therefore, infants who walk explore their environment, objects, and people differently than crawlers and are provided new opportunities for actions and interactions (Adolph & Tamis–LeMonda, 2014).

Summary

Based on the DST and the literature review pertaining to the development of walking, the following possible constraints are reported (see Table 1).

Table 1

Possible Constraints Related to the Development of Walking

	Constraints	References
Individual	Leg mass, muscular strength	Thelen & Fisher, 1982; Thelen, Fisher, & Ridley-Johnston, 1984
	Visual, vestibular, tactile, proprioceptive systems	Thelen & Ulrich, 1991
	Visual, auditory, vestibular, postural control	Palm, Strobel, Achatz, von Luebken, & Friemert, 2009
	Somatosensory system	Barela, Jeka, & Clark, 1999
	Sensory information and perception	Metcalfe & Clark, 2000
	Hearing loss, vision loss, body mass index	Uysal, Erden, Akbayrak, & Demirtürk, 2010
	Postural, body, pelvis, trunk control	Looper, Talbot, Link, & Chandler, 2015
	Supine, prone position	Charitou, Asonitou, & Koutsouki, 2010
	Visual access	Karasik, Tamis-LeMonda, & Adolph, 2014
Environment	Gravity	Thelen & Fisher, 1982; Thelen, Fisher, & Ridley-Johnston, 1984
	Mechanical support through surface contact	Barela, Jeka, & Clark, 1999
	Surface touch	Metcalfe & Clark, 2000
	Social exchange	Karasik, Tamis-LeMonda, & Adolph, 2014
Task	Treadmill	Thelen, 1986; Thelen & Ulrich, 1991
	Crawling (hand, head positions)	Franchak, Kretch, Soska, & Adolph, 2011; Kretch, Franchak, & Adolph, 2014

Barriers to Physical Activity Participation

Education of students with visual impairments and deafblindness may pose many barriers that affect appropriate educational experiences. Students with visual impairments who participate in successful physical activities can obtain greater independence and an overall improvement in the quality of life (Lieberman, 2011). Lieberman and Houston-Wilson (1999, Level 3, A 15) conducted a survey of physical education teachers

throughout New York State to identify barriers teachers experience when educating students with visual impairments and deafblindness in general physical education class. The barriers were separated into three groups: (a) teacher barriers; (b) student barriers; and (c) administrative barriers.

Teacher barriers consisted of physical educators feeling inadequately prepared to educate students with visual impairments and deafblindness due to the lack of professional preparation. Lieberman and Houston-Wilson (1999, Level 3, A 15) suggested the need to improve and provide additional professional and in service training for teachers that covers inclusion and instructional strategies, adaptations, and activity resources. Physical educators who participated in this investigation reported that curriculum units and activities that required visual-motor coordination were difficult to teach to students with visual impairments and deafblindness. An additional difficulty the physical educators expressed was trying to keep an appropriate pace of the lesson.

Further, due to the limited exposure and knowledge of students with visual impairments and deafblindness, Lieberman and Houston-Wilson (1999, Level 3, A 15) reported that educators might limit the student's expectations and be unaware of his or her potential. However, the use of trained peer tutors and varied teaching styles may foster a desired performance and quicker understanding (Houston-Wilson, Lieberman, Horton, & Kasser, 1997; Lieberman & Houston-Wilson, 1999; Wiskochil, Lieberman, Houston-Wilson, & Petersen, 2007). For student barriers, lack of opportunities and resources in physical activities for students with visual impairments and deafblindness were reported in addition to the lack of confidence and fear that some students displayed.

In addition, parental overprotection can be a barrier to participation and inclusion in physical activities.

Administrative barriers, including time, lack of appropriate equipment, and medical excuses, all affect success of inclusion and participation of students with visual impairments and deafblindness in physical education. However, based on the barriers reported by Lieberman and Houston-Wilson (1999, Level 3, A 15) it is imperative that educators, parents, students, and administrators address these barriers to enhance opportunities and participation in physical activities.

Similar barriers were reported by Lieberman, Houston-Wilson, and Kozub (2002, Level 3, A 16), about the inclusion of students with visual impairments in general physical education class. In this investigation, the perceived barriers recorded by physical educators prior to a professional development workshop were professional preparation, equipment, programming, and time. The participants identified professional preparation as the most prevalent barrier. Due to the unique needs and instruction of students with visual impairments, barriers perceived by physical educators impact their instruction and program. This information is important and crucial to the motor development of individuals with deafblindness because previous researchers have stated that in addition to barriers faced by instructors, students with visual impairments have delays in motor development, particularly locomotor activities, including walking, due to limited opportunities for physical activities (Brambring, 2006; Conroy, 2012; Gosch, Brambring, Gennett, & Rohlmann, 1997; Haibach, Wagner, Lieberman, 2014; Jan, Sykanda, &

Groenveld, 1990; O'Mara-Maida & McCune, 1996; Pereira, 1990; Wagner, Haibach, & Lieberman, 2013).

In addition, Conroy (2012, Level 2, A 17) performed a qualitative investigation to examine how physical education teachers support the inclusion of students with visual impairments and identify instructional needs and strategies. Successes, challenges, and needs were the three main themes, which emerged from the interviews of physical educators. The physical educators felt they did not have adequate background information about the developmental level of the student with the visual impairment, which lead to difficulties in selecting the amount of support and type of instruction to provide. In addition, the infrequent collaboration and communication with teachers of the student or the IEP team was seen as a challenge in this investigation. All of the participants in this investigation reported the need for more training, which is seen in the previous investigations. The participants acknowledged the need of differentiated instruction as an inclusion strategy for students with visual impairments.

Inclusion of children who are deafblind in physical education and physical recreation activities is important for obvious health benefits as well as an increase in movement and social opportunities (Lieberman, 2002a, 2002b; Smith, 2002). Lieberman and MacVicar (2003, Level 3, A 18) conducted a study to examine the play and recreational habits of youth who are deafblind to determine parental satisfaction of physical education and physical recreation activities, and examine barriers families faced. The top five recreational activities selected by the youth who were deafblind in this investigation

(n = 51) were swimming (57%), swinging or rocking (33%), walking (31%), climbing equipment (24%), and biking (24%).

Based on the results from the parents' questionnaire, the researchers also reported that children with deafblindness had approximately 20 hours during the week and 24.6 hours during the weekend of free time. However, even with that amount of time, parents were often not satisfied with the opportunity and ability for their child to participate in recreational activities. The parents' satisfaction for physical education and physical recreation experiences fell between "neutral" and "not very satisfied" on a 5-point Likert scale. Many parents expressed frustration of barriers they faced for their child with deafblindness to participate and access recreational activities.

The most common barrier (33%) expressed by the parents was related to the disability of deafblindness. The investigators believed the additional barriers were present due to the previous most common barriers. These barriers included lack of knowledge about specific adaptation (31%), appropriate programming ideas (25%), adequate staff that were knowledgeable and willing to work with children who were deafblind (25%), adequate communication (16%), time (12%), money (8%), accessibility (4%), and transportation (4%).

In summary, the researchers indicated the need for additional professional preparation and appropriate support for physical educators and physical activity recreational specialists as stated in the previous articles pertaining to barriers that children who are deafblind do require more support and assistance. However, physical recreation opportunities within a community should be available based on the accessibility needs of

being deafblind. An increase in knowledge about deafblindness and the unique needs of these children and youth would assist in minimizing the barriers faced by the families in this study and increase the opportunities for these children with deafblindness to be active members of their community, participate in physical education and physical recreational activities, and increase social opportunities with peers.

Summary

Based on the DST and the literature review pertaining to the barriers associated with physical activity, the following possible constraints are reported (see Table 2).

Table 2

Possible Constraints Related to the Barriers to Physical Activity

	Constraints	References
Individual	Teacher preparation	Lieberman & Houston-Wilson, 1999
	Professional development	Lieberman, Houston-Wilson, & Kozub, 2002
	Background knowledge, communication	Conroy, 2012
	Vision, hearing, knowledge communication	Lieberman & MacVicar, 2003
Environment	Opportunity, resources, parental overprotection, appropriate equipment	Lieberman & Houston-Wilson, 1999
	Equipment, program, time	Lieberman, Houston-Wilson, & Kozub, 2002
	Collaboration, support, instruction	Conroy, 2012
	Accessibility, transportation	Lieberman & MacVicar, 2003
Task	Training	Conroy, 2012

Effect of Vision Loss on Motor Development and Walking

There is limited research on the motor skill performance in children with visual impairments and even less research on the effect of visual impairment severity on motor skill performance. Visual impairment is estimated to occur in 80 to 90% of children with CHARGE syndrome (Blake et al., 1998). The effect of the visual impairment for children with CHARGE syndrome may cause developmental delays in acquiring independent walking. Vision loss has been researched and examined as a variable which may affect or impede motor development (i.e., walking).

Interaction Between the Individual and Task

Wagner, Haibach, and Lieberman (2013, Level 1, A 19) examined the gross motor performance of children with visual impairments compared to similar age and gender characteristics of peers with vision. The participants with visual impairments had significant deficits in performing locomotor and object control skills compared to their peers with vision. Running and leaping were most affected by the children with vision loss in the locomotor section, while kicking and catching were most affected in the object control section when compared to their peers with vision. This information is important in the field of adapted physical education and has practical implication for the need to increase opportunities and time to practice gross motor skills in children with visual impairments. Development of fundamental motor skills is the foundation to motor proficiency, fitness, and overall health.

In 2014, the same investigators (Haibach et al., Level 1, A 20) conducted a follow up study to examine determinants of gross motor performance in children with visual

impairments. In this study, the investigators examined the effect of age, gender, and the severity of the visual impairment on gross motor performance. Two age groups were used to compare the effect of age on gross motor performance (6-9 years and 10-12 years). The severity of visual impairment was separated into three levels pertaining to the United States Association for Blind Athletes vision classification system. Based on the results of this investigation, age and gender did not play a significant role in determining gross motor skill performance. However, the severity of the visual impairment was a significant factor in all assessed motor skills when B1 (totally blind/light perception) participants were compared to B2 (20/600, travel vision), and B3 (20/200, legally blind). Participants in the B2 (n = 25) and B3 (n = 52) group performed similar in the motor skills, except for the run, catch, and throw.

Overall there was not a significant difference between the B2 (n = 25) and B3 (n = 52) groups on gross motor performance (see A 20). Therefore, the investigators concluded that children with more severe vision loss perform worse on gross motor performance than their peers with more vision. There also was a common level of performance for children who had functional level of vision and those who were legally blind. For children with CHARGE syndrome, the findings of this investigation may apply in that children with more severe vision loss will not acquire independent walking until later months than those with more vision.

In addition to gross motor performance, researchers have reported that congenital visual impairment or blindness causes a delay in the development of gross motor milestones when compared to children with vision (Adelson & Fraiberg, 1974; Bouchard

& Tétreault, 2000, Brambring, 2006, 2007; Celeste, 2002; Ferrell, 2000; Fraiberg, 1977; Hatton, Bailey, Burchinal, & Ferrell, 1997; Jan, Freeman, & Scott, 1977; Kastein, Spaulding, & Scharf, 1980; Norris, Spaulding, & Brodie, 1957; Pereira, 1990; Tröester & Brambring,1993; Tröester, Hecker, & Brambring, 1994). Bouchard and Tétreault examined the difference in 30 children with moderate low vision compared to gender and age matched children with vision (2000, Level 3, A 21). The *Bruininks-Oseretsky Test of Motor Proficiency* (*BOTMP*; Bruininks, 1978) was used to assess the motor proficiency of the two groups of participants. In addition, the age of acquisition of five developmental motor milestones (i.e., sitting, crawling, creeping, standing, walking) were obtained through a parent questionnaire.

In all five developmental motor milestones the participants with vision loss were delayed when compared to their peers with vision. Participants with low vision obtained independent siting, standing, and walking significantly later than the participants with vision. The participants with low vision also acquired the developmental milestone of walking at an average of 14.5 months, while the group with vision acquired walking at an average of 12.2 months. In addition, children with vision loss obtained significantly lower scores in six of the eight subtests in the *BOTMP* (see A 21). The balance subtest score performance was significantly lower (children with low vision, age equivalence of 80.5 months, *SD* = 28.7; children with vision, age equivalence of 162.4 months, *SD* = 32.4 months) than all other subtests when the children with vision loss were compared to the children with vision.

Therefore, balance can be seen as a major contributing factor to development of motor performance in children with vision loss. In addition, the investigators stated the following possible factors could impact motor development and performance: limited experience, lack of interest, fear of space, socially isolated, communication gap, parents' overprotection attitude, poor physical fitness, and difficulty imitating.

Based on the results of this study, it was recommended that early intervention, including motor activities, was essential in the development of gross motor skills in children with visual impairments. In addition, increasing the child's confidence in performing motor activities is equally as important. It is also important to recognize a delay in motor development as soon as possible in order to provide appropriate early interventions and services. Recognizing the age at which children should be attaining developmental motor milestones and additional motor skills can provide valuable information about a child's development.

Further research by Brambring (2006, Level 2, A 22) examined the age at which children who were blind acquired 29 gross motor skills over 4 years and compared them to the age norms of children with vision. The information was gathered through observations during home visits. Developmental data for children with vision were obtained from four standardized developmental tests: *Bayley Scales of Infant Development* (Bayley, 1969), *Denver Developmental Screening Test* (German version; Flehmig, Schloon, Uhde, & von Bernuth, 1973), *Griffiths Developmental Scales* (German version; Brandt, 1983) and *Entwicklungskontrolle für Krippenkinder* (Zwiener & Schmidt-Kolmer, 1982) which is a German language developmental test. From these

developmental tests, 29 items were selected to compare gross motor skills of the four participants who were blind and the age norms of children with vision.

Children who were blind had an average developmental delay of approximately 11.9 months at the age of 30 months when compared to children with vision. The median age of walking three steps was 16.5 months in children who were blind compared to 12.4 months for children with vision. Children who were blind compared to the children with vision revealed significant developmental delays in the total comparison, dynamic balance, attainment of locomotion, and refinement (i.e., performance level) of locomotion (see A 22).

A regression analysis of the acquisition ages of children who were blind and children with vision displayed a high correlation, meaning the sequence of developmental skills were acquired sequentially. The investigator stated that children who were blind learned and applied alternative compensatory strategies, such as the use of other sensory information, to compensate for the loss of vision. Children who are blind may develop motor development and walking later in order to be able to process and understand the physical or verbal guidance needed to assist in the acquisition of motor skill development and performance, while children with vision can imitate skills they are able to see (Brambring, 2006, Level 2, A 22).

Vision also provides motivation to move and engage with possible incentives in an environment and provides feedback of an individual's position in open space. For children with vision loss, the fear of space (Cratty, 1970) may inhibit movement, due to their inability to know where they are in the environment. Brambring (2006) reported that

major developmental divergences in skills that required movement through space without physical contact with the environment (e.g., walking without holding onto furniture, without assistance). However, motor skills can be presented early to children with vision loss through a differentiated approach using various senses within instructional strategies.

In addition to the effect of vision on motor development, Tröester, Hecker, and Brambring (1994, Level 3, A 23) reported that prematurity of infants who were blind exhibited a greater delay in motor development when compared to full term infants who were blind and full term infants with vision based on the *Bayley Scales of Infant Development* (Bayley, 1969). Tröester, Hecker, and Brambring stated that, "prematurity is one of the main risk factors for congenital blindness" (1994, p. 63). Therefore, these investigators examined the impact of prematurity on motor development in infants who were blind.

The full term infants who were blind walked three steps unassisted at a median age of 15.5 months compared to preterm infants who were blind that performed the skill at a median age of 26.5 months. Differences between full term and preterm infants who were blind were larger than difference between full term infants who were blind and full term infants with vision, identifying that motor development may be significantly impacted by prematurity combined with vision loss.

However in contrast, Tröester and Brambring (1993, Level 2, A 24) reported that infants who were blind and preterm, with age adjusted for prematurity, when compared to infants who were blind and born full term did not significantly differ in blind-specific motor skills of locomotion on the *Bielefeld Developmental Test for Blind Infants and*

Preschoolers (Brambring, Dobslaw, Klee, Obermann, & Tröester, 1987). Blind-specific motor skills are skills that require visuomotor coordination skills. Therefore, those with vision loss would have delayed acquisition and refinement due to limited use of vision. The reported delay in locomotor skill is, "predominantly a consequence of their blindness" (Tröester & Brambring, 1993, p. 99). Infants who were blind in both age groups, 9 months and 12 months old, when compared to infants with vision were significantly delayed in locomotion. Nevertheless, variances of the two age groups in infants with vision did not differ significantly in locomotion. In contrast when compared to infants with vision, both age groups of infants who were blind were significantly delayed in locomotion.

However, infants who were blind had a significant variability between the mean of the entire locomotion scale for 9 months being 4.0 and for 12 months being 11.6. Therefore, it was concluded that age might have an integral impact on motor development based on the need to have conceptual, perceptual, and cognitive capabilities to enable, "audiomotor coordination in place of visuomotor coordination" in infants who are blind (Tröester & Brambring, 1993, p. 103). An infant's ability to adapt to his or her lack of visual stimulation and feedback within an environment is crucial in developing motor skills and as previous shown. This may require a longer time in infants who are blind. In addition, visual stimulation activates motor stimulation early on in an infant's life, which negatively impacts the motor activity of infants who are blind since they are less simulated than those with more vision. Therefore, the motor delay in children who

were blind was due to their inability to replace visuomotor coordination with audioproprioceptive coordination and control of movement.

The impact of the degree of vision loss was further examined on gross motor development using the *Peabody Developmental Motor Scales* (Folio & Fewell, 1983) in children with visual impairments (Celeste, 2002, Level 2, A 25). Eighty-four families of children with visual impairments from age 4 months to 4 years were surveyed about their children's gross motor development. Based on the results of this investigation, children with the least amount of vision had the poorest motor outcomes. However, the sample subgroup that had additional disabilities (n = 59) performed worse than the subgroup with a visual function of no light perception or only light perception (n = 31). In addition, those who were born premature (n = 37) had the poorest motor development outcome of the other two subgroups. All the participants (N = 84) in the investigation were most delayed in the mobility developmental motor milestones (e.g., walking). The *Peabody Developmental Motor Scales* (Folio & Fewell, 1983) scores indicated that the developmental motor milestone of independent walking should be performed between 9 to 17 months. However, 50% (n = 12) acquired the developmental milestone of independent walking between 37 to 48 months with an additional 31.3% (n = 5) did not achieve this milestone until later than 48 months old. Therefore, the children with the least vision had the poorest gross motor outcomes.

Researchers have reported that a visual impairment can impact motor development, especially pertaining to independent walking. In addition to the delay of attaining the developmental milestone of walking, Hallemans, Ortibus, Truijen, and Meire (2011,

Level 3, A 26) investigated the age-related changes in the walking gait of individuals with loss vision, blindness, and a control group with vision. The participants with visual impairments (n = 31; low vision n = 22; blind n = 9) were between the age range of 1 to 44 years (children n = 21; adults n = 10) and all had congenital peripheral visual system disorders that were compared to an age-related (children; adult) control group of individuals with vision.

The following information on the walking gait was obtained: walking speed, heading angle, stance phase duration, double support phase, dimensionless stride length, step frequency, and step width. Children with a visual impairment displayed a slower walking speed, a shorter stride length, a prolonged duration of stance phase duration, and double support during the gait pattern. In addition those participants who were blind walked at a slower speed, displayed a shorter stride length, and had a prolonged duration of stance when compared to both the control and group with low vision. Therefore, as vision loss increases there is a greater display of a more pronounced gait pattern.

Part of the gait pattern in walking is a double support, when both feet are in contact with the ground. This part of the gait pattern or cycle is used as a period of recovery in balance (Hallemans, et al., 2011, Level 3, A 26). Therefore, an increase in the double support phase for children with vision loss may indicate a result of balance difficulties (Hallemans et al., 2011; Hallemans, Ortibus, Meire, & Aerts, 2010; Sutherland, 1997). In addition, the prolonged double support stance allows the foot to gain sensory feedback about the ground. Patla, Davies, and Niechwiej (2004) expressed this movement as haptic exploration. This adaptation to the gait pattern has been investigated to provide sensory

feedback to compensate for a loss of visual feedback of the environment (Buckley, MacLellan, Tucker, Scally, & Bennett, 2008; Hallemans et al., 2009a, 2009b; Hallemans et al., 2010, Hallemans et al., 2011). Therefore, these gait adaptations may be due to balance difficulties or are used as a strategy to gain more environmental input. However, within this investigation (Hallemans et al., 2011) an increase in age in children with visual impairments or blindness displayed overall improvements to their walking gait patterns.

Interaction Between the Environment and Task

Both the Dynamic Systems Theory (Shumway-Cook & Woollacott, 2001; Thelen & Smith, 1996) and the World Health Organization (WHO, 2001) support the importance of parents being vital proponents of motor development. Motor functioning occurs through the interaction of the individual and environmental factors, which parents and others can manipulate (Thelen & Smith, 1996). Parents of children with vision loss should concentrate on auditory-tactile association by providing a tactile stimulus associated with a sound to replace the visual input (Levtzion-Korach et al., 2000, Level 3, A 27). Improvements in motor development have been reported when parents alter the child's environment by providing adequate stimulation based on his or her visual function through the use of language and physical contact (Fraiberg & Adelson, 1977; Norris et al., 1957).

Levtzion-Korach et al. (2000, Level 3, A 27) examined the motor development of 40 children who were blind in Israel on 10 different fine and gross motor skills based on the *Bayley Developmental Scale* (Bayley, 1969) and the *Revised Denver Developmental*

Screening Test (Frankenburg, Fandel, Sciarillo, & Burgess, 1981). The children who were blind were significantly more delayed in all motor skills based on the standard milestone age for children with vision. Levtzion-Korach et al. (2000) stated that,

> . . . a child, whether blind or sighted, has basic needs in order to progress in its motor development, such as neuromuscular maturation, a good body awareness and image, acquirement of the concept of permanence and the ability of localizing a sound together with the active urge of exploring (p. 229).

These individual and environmental characteristics, along with the involvement of parents (Levtzion-Korach et al., 2000, Level 3, A 27) and early educational interventions (Bouchard & Tétreault, 2000, Level 3, A 21) can assist in the motor development of children with vision loss.

Regardless of the attainment of developmental gross motor milestones, Pereira (1990, Level 3, A 28) further examined positional spatial concepts (i.e., the ability to move in relation to objects and the environment and the ability to form object-to-object relationships) and balance performance in children who were visually impaired in relation to their motor development. The *Hill Performance test of Selected Positional Concepts* (Hill, 1981) and an adaptation of Leonard's (1969) test were used. Pereira concluded that differences in motor performance in children with visual impairments when compared to children with vision were reported due to poor balance performance (1990, Level 3, A 28). Therefore, poor balance performance was related to low visual acuity, a reduction in visual clarity, and alterations of the vestibular system. However,

better results of positional concepts were independent of visual acuity and visual field loss, which is the total area in which objects can be seen.

Pereira also examined two pedagogical approaches to improve motor development, one was with more cognitive activity than motor and the other was with more motor activity and less cognitive activity. The pedagogical approach that focused on stronger cognitive demands demonstrated to be more effective in improving motor development. Improvements of motor development were dependent on pedagogical strategies, which included: (a) descriptive positive reinforcement from the teacher; (b) variety of teaching cues; and (c) child's engagement and time on task.

Summary

Based on the DST and the literature review pertaining to effect of vision loss on motor development and walking, the following possible constraints are reported (see Table 3).

Table 3

Possible Constraints Related to the Effect of Vision Loss on Motor Development and Walking

	Constraints	References
Individual	Severity of vision loss	Haibach, Wagner, & Lieberman, 2014
	Vision, balance, experience, interest, fear, communication, attitude, fitness level, imitation	Bouchard &Tétreault, 2000
	Vision	Brambring, 2006
	Prematurity	Tröester, Hecker, & Brambring, 1994
	Visuomotor coordination, audiomotor coordination, blindness, age, conceptual capabilities, perceptual capabilities, cognitive capabilities, adaption	Tröester & Brambring, 1993
	Vision, prematurity	Celeste, 2002
	Engagement, time on task	Pereira, 1990
	Neuromuscular maturation, body awareness, concept of permanence, localize sound	Levtzion-Korach, Tennenbaum, Schnitzer, & Ornoy, 2000
	Vision, age, balance, sensory feedback	Hallemans, Ortibus, Truijen, & Meire, 2011
Environment	Space, isolation	Bouchard &Tétreault, 2000
	Contact with surfaces	Brambring, 2006
	Visual stimulation	Tröester & Brambring, 1993
	Positional spatial concepts, positive reinforcement, teaching cues	Pereira, 1990
	Parental involvement, audio-tactile stimulus	Levtzion-Korach, Tennenbaum, Schnitzer, & Ornoy, 2000
Task	Locomotor-running, leaping, object control-kicking, catching	Wagner, Haibach, & Lieberman, 2013
	Early intervention	Bouchard &Tétreault, 2000
	Cognitive activities	Pereira, 1990
	Gait patterns	Hallemans, Ortibus, Truijen, & Meire, 2011

Effect of Hearing Loss on Motor Development and Walking

Ear abnormalities are present within an estimated 90% of individuals with CHARGE syndrome (Blake et al., 1998). The ear abnormalities can cause either a conductive or a sensorineural hearing loss. For children with a sensorineural hearing loss, there may be vestibular problems, which may affect their abilities to balance. The development of balance and ability to walk independently are dependent on vestibular, proprioceptive, motor, and visual systems (Kaga, 1999; Suarez et al., 2007). Children largely rely on their vision to maintain balance during their early stages of development (Inoue et al., 2013). However, an impairment to the vestibular functions may lead to developmental delays in motor function and achieving gross motor milestones such as walking (Kaga, 1999; Kaga, Shinjo, Jin, & Takegoshi, 2008).

Interaction Between the Individual and Task

In order for children to produce upright bimodal independent walking, both balance and motor coordination must occur. However, children with hearing loss, especially sensorineural hearing loss, may have vestibular dysfunctions. This can pose difficulties to attain independent walking due to balance issues. Many researchers have investigated the effect or relationship of vestibular function and motor development in children with profound hearing loss (Crowe & Horak, 1988; Cushing, Chia, James, Papsin, & Gordon, 2008; De Kegel, Maes, Baetens, Dhooge, & Van Waelvelde, 2012; Inoue et al., 2013; Kaga, Suzuki, Marsh, & Tanaka, 1981; Kaga et al., 2008; Maes, De Kegel, Van Waelvelde, Dhooge, 2014; Masuda & Kaga, 2014; Potter & Silverman, 1984; Rapin, 1974; Rine, Cornwall et al., 2000; Suarez et al., 2007). The vestibular system can produce

three motor reflexes to assist the body with movement: (a) vestibular-ocular reflex assist with visual stabilization; (b) vestibular-collic reflex assist with neck stabilization; and (c) vestibular-spinal reflex assists with posture and the orientation of one's body in space which is needed to attain motor developmental milestones (De Kegel et al., 2012; Nandi & Luxon, 2008; Suarez et al., 2007; Tribukait, Brantberg, & Bergenius, 2004). The vestibular system, which includes the utricle, saccule (also called the otolithic organs), and three semicircular canals in each ear, provides information about the body's motion, equilibrium, and spatial orientation (Hanes & McCollum, 2006; Shumway-Cook & Woolacott, 2001; Vestibular Disorders Association, 2008). The utricle and saccule detect gravity, vertical orientation, and linear movement or acceleration while the semicircular canals detect rotational movement (Vestibular Disorders Association, 2008).

Inoue et al. (2013, Level 3, A 29) investigated the influence of vestibular nerve dysfunction on the development of gross motor function in children with profound sensorineural hearing loss. The gross motor function was evaluated by the age of acquiring head control and independent walking. Function of the superior and inferior vestibular nerve systems was evaluated for dysfunction. The damped rotation test and caloric test were used to determine the function of the superior nerve systems. The vestibular evoked myogenic potential (VEMP) test was used to evaluate the function of the inferior nerve systems.

The participants consisted of 89 children with profound sensorineural hearing loss between the ages of 20 to 97 months. Based on the results, it was indicated that the age of independent walking was significantly delayed in the group of participants with

combined superior and inferior dysfunction and in the group of participants with inferior dysfunction when compared to children with normal vestibular function. However, the investigators reported that there was no significant difference between the group with normal function and the superior dysfunction group or between the inferior dysfunction group and the combined dysfunction group.

Of the participants used in this study, only 20% had a dysfunction in both the superior and inferior vestibular nerve systems and 40% had a dysfunction in the inferior nerve system. Twenty participants (26%) in the study did not walk until later than 18 months of age, which was considered to be delayed by the modified Japanese version of the *Denver II* (Frankenburg, Dodds, & Archer, 1990). However, all children were able to walk independently within 30 months. Therefore, hearing loss may be a contributing factor of motor delay in children with CHARGE syndrome due to sensorineural hearing loss and vestibular dysfunction.

De Kegel et al. (2012, Level 1, A 30) examined the predictive ability of vestibular function on motor performance of children with hearing loss. Children in the study who had an absence of the VEMP response performed significantly worse on static balance and had a greater postural instability than children with hearing loss who had a VEMP response. However, children with unilateral loss or moderate hearing loss performed equally to those with bilateral, profound hearing loss in both motor and balance tasks. Therefore, although children with unilateral loss or moderate hearing loss may not have the same risk for communication and language delay as children with bilateral;

individuals with profound hearing loss, may still encompass the same risk for delay in developing motor and balance tasks (De Kegel et al., 2012).

In addition to the vestibular system, vision, proprioceptive, somatosensory, and the central nervous system all contribute to an individual's ability to maintain posture and walk independently (Horak, Shumway-Cook, Crowe, & Black, 1988; Kaga, 1999; Suarez et al., 2007), which supports the dynamic systems theory that all systems are working together to establish development and movement. In order for an individual to walk, vertical postural control must be obtained. Therefore, Suarez et al. (2007, Level 3, A 31) examined the balance sensory organization and sensory information used in children with profound hearing loss and cochlear implants to maintain vertical postural control. The participants in this investigation were assessed on their vertical postural control using a force platform while measuring sway velocity and center of pressure distribution area based on two conditions. In Condition 1, the participant stood on the force platform with eyes open while in Condition 2, the participant stood on a 7.87 inch/20 centimeter thick foam placed on the force platform with their eyes covered.

Thirty-six children with sensorineural hearing loss (28 with normal vestibular function, 8 with hypoactive vestibular function) along with 22 children with no hearing loss as controls participated in both conditions. The sway velocity and center of pressure distribution were higher in Condition 2 compared to Condition 1 in all participants. However, those with a hypoactive vestibular system (n = 8) had significantly higher value in sway velocity and center of pressure distribution in Condition 2 than those with a profound sensorineural hearing loss with normal vestibular function and those with no

hearing loss. In addition, there was no significant difference in postural control with the cochlear implant turned on or off in Condition 2.

In Condition 2, participants had to rely solely on vestibular information due to the lack of visual information and an altered somatosensory input due to the thick foam. Therefore, the investigators reported that children with profound sensorineural hearing loss with hypoactive vestibular function rely on visual and somatosensory information to maintain postural control. In addition, children with vestibular dysfunction should be taught to use substitutive strategies such as vision, proprioceptive, and somatosensory input to assist in postural control (Suarez et al., 2007, Level 3).

Damage to a child's vestibular structures due to sensorineural hearing loss can cause motor development delay (Cushing et al., 2008; Gayle & Pohlman, 1990; Horak et al., 1988; Jackler & de la Cruz, 1989; Jacot, Van Den Abbeele, Debre, & Wiever-Vacher, 2009; Kaga et al., 2008; Nadol & Hsu, 1991; Shinjo, Jin, & Kaga, 2007; Shumway-Cook & Woolacott, 2001). Sensorineural hearing loss is damage to the sensory cells in the inner ear, including the cochlea and its receptors, and/or the nerve fibers from the inner ear to the brain (National Institute on Deafness and Other Communication Disorders, 2013). Rine et al. (2000, Level 2, A 32) examined the motor proficiency of children with sensorineural hearing loss and concurrent vestibular dysfunction using the *Gross Motor Scale of the Peabody Developmental Motor Scales* (Folio & Fewell, 1983).

Regardless of age, children with sensorineural hearing loss scored significantly lower on the Gross Motor Scale (e.g., locomotor category). However, there was no correlation between vestibular function and *Z* scores on the *Peabody Developmental Motor Scales* or

skill categories. Nonetheless, a hypofunction vestibular score identified 91% of participants with balance delays and delays in the Gross Motor Scale were lower after repeated testing. In children with sensorineural hearing loss, the motor delay was persistent across various repeated measures as the child aged, which was reported contrarily to research findings by Siegel, Marchetti, and Tecklin (1991, Level 3, A 33) and Dummer, Haubenstricker, and Stewart (1996, Level 3, A 34).

Siegel et al. (1991, Level 3) compared scores on the balance subtest of the *Bruininks-Oseretsky Test of Motor Proficiency* (*BOTMP*), in three age groups (i.e., Group 1: 4.5 to 6.5 years; Group 2: 8 to 10 years; Group 3: 12.5 to 14.5 years) of children (n = 28) who were deaf due to sensorineural hearing loss, compared to children with hearing. The mean balance subtest score of the *BOTMP* was significantly lower in all three age groups when compared to the normative value. Based on the results both older groups (Group 2 compared to Group 1; Group 3 compared to Group 1) had a significant higher score on the mean balance scores compared to the youngest groups. However, no significant differences were based on gender. Furthermore, an increase in age indicated an increase on balance scores for the youngest age group, however, no significant increase from Group 2 to Group 3.

In another study, Dummer et al. (1996, Level 3), reported that age was a significant factor in gross motor skill performance. Motor development of children who were deaf was investigated with the use of the *Test of Gross Motor Development* (*TGMD*; Ulrich, 1985). The participants were videotaped as they performed three trials of each skill, in addition distances for the throw, kick, and jump, and times for the 15-yard dash were

recorded. Based on the results, age (in months), which was the covariate, was the most significant variable in increasing the performance of both the locomotor skills and the object-control skills. Therefore, the mean scores on the *TGMD* increased as chronological age increased. Age also accounted for the most variance on the throw for distance, kick for distance, jump for distance, and 15-yard dash (see A 34). Based on the norms of the *TGMD,* it was reported that children who are deaf demonstrated delays of 1 to 3 years in the acquisition of object-control skills. In the locomotor skills section, children who are deaf acquired the skill of skipping and leaping at later ages than those without hearing loss when compared to the norms of the test.

To further evaluate the effect of sensorineural hearing loss on walking Melo, Silva, Tassitano, Macky, and Silva (2012, Level 3, A 35) examined the balance and gait of students with sensorineural hearing loss and students with hearing. The participants were assessed using the *Tinetti's Balance and Mobility Scale* (Tinetti, 1986) and the timed *"Up and Go" Test* (Podsiadlo & Richardson, 1991). In the *Tinetti's Scale* results for balance the children who were deaf had no significant difference in performance compared to the children with hearing. However, in the *Tinetti's* gait characteristics portion, the children with hearing loss performed significantly lower than the children with hearing, even after that data were separated by gender. The children with hearing loss in this study had two distinctive gait characteristics that differed from the group with hearing (see A 35). During the gait analysis, the majority of the students with hearing loss exhibited trunk flexion during locomotion. In addition, the majority of the students did not display a

swing phase and heel strike. Instead the students dragged their feet and continuously maintained a phase of double support.

In the *"Up and Go" Test,* only the 7 to 10 year old group of children with hearing loss performed significantly worse than the children with hearing. When compared by gender, both male groups did not differ but there was a significant difference with female children with hearing performing better than female children with hearing loss. Within the *"Up and Go" Test,* 17 of the 44 students with hearing loss were classified as having walking dependencies which increased the risk of falling. This may explain why the students with hearing loss dragged their feet, walked at a slower pace, and had shorter step lengths to assist in maintaining balance. However, children with hearing loss decreased the degree of walking dependency as their age increased.

With the advancement in technology, many individuals with hearing loss are receiving cochlear implants. However, individuals with additional disabilities are usually not routinely precluded for cochlear implants (Amirsalari et al., 2012) but more are currently being provided a choice. Amirsalari et al., (2012, Level 3, A 36) examined the effects of motor developmental delay on the outcome of cochlear implants. Children with sensorineural hearing loss and normal motor development were compared to children with sensorineural hearing loss and delayed motor development. Speech perception tests were performed before surgery and 24 months after cochlear implantation. The results of the speech intelligibility ratings and the auditory perception scales were not significantly different between the group with normal motor development and the group with delayed motor development.

Gheysen, Loots, and Waelvelde (2008, Level 3, A 37) examined the impact a cochlear implant has on motor development of children who were deaf. Motor development was measured by the *Movement Assessment Battery for Children* Dutch validated version (*M-ABC*; Smits-Engelsman & Niemeijer, 1998), one-leg standing test, and the *Körperkoordinationstest für Kinder (KTK*; Schilling & Kiphard, 1974), which measures gross motor coordination. Motor performances of children with hearing were significantly better for the total score of the *M-ABC* and all scales of the one-leg standing test. Children with hearing had better manual dexterity, ball skills, and dynamic and static balance on the scales of the *M-ABC* than children who were deaf. Children with hearing also demonstrated significantly higher balance scores when standing on one leg, with eyes open and closed, compared to children who were deaf (see A 37).

The children with hearing when compared to children who were deaf with cochlear implants and without cochlear implants performed significantly better than only the children who were deaf with cochlear implants on the *M-ABC* balance subtest, the *KTK* walking backward subtest, and all of the one-leg stand scores. In addition, children with hearing performed significantly better than the children who were deaf without cochlear implants on only the *KTK* jumping on one leg subtest. Only one significant difference was reported in the *KTK* test in which children without cochlear implants walked backwards on the beam significantly better than children with cochlear implants. Therefore, the results of this investigation, the researchers concluded that there was no significant impact of cochlear implants on the motor development of children who are deaf.

Interaction Between the Environment and Task

The majority of children with hearing loss or who are deaf have hearing parents; researchers have shown that Deaf individuals who have Deaf parents function better academically (Ritter-Brinton & Stewart, 1992), linguistically (Courtin, 2000; Harris, 2001; Vaccari & Marschark, 1997), and socially (Hadadian & Rose, 1991; Harris, 2001). Children with hearing who were born to Deaf parents and learned sign language as their primary language did not slow down their rate of language acquisition after attaining a developmental motor milestone (Bonvillian, Orlansky, & Novack, 1983). These results are contrary to children with hearing who use spoken English as their primary language, in which the rate of language acquisition is slowed down after attaining a developmental motor milestone (McCarthy, 1954; Shirley 1933a, 1933b). To further investigate the impact of parents hearing level in children who are deaf, Lieberman, Volding, and Winnick (2004, Level 3, A 38) examined the difference in motor development of Deaf children to Deaf parents to Deaf children to hearing parents.

The *TGMD* (Ulrich, 1985) was used to measure the children's locomotor and object control gross motor development. Each testing session was videotaped and then analyzed and scored. There was no significant difference between the two groups on the *TGMD* locomotor and object control subtest. Based on the results of a factorial ANOVA there was a significant main effect on the difference in locomotor and object control subtests scores but no significant main effect of the hearing status of parents. Therefore, the hearing statuses of the parents were not a factor in Deaf children's motor performance.

To further assist the influence of the environment, Majlesi, Farahpour, Azadian, and Amini (2014, Level 2, A 39) designed a proprioceptive training intervention, which assessed the gait, and balance of children who were deaf (> 75 decibel loss) compared to children with hearing. The 12-session exercise program consisted of 20 minutes of stability and balance training and 20 minutes of dynamic balance activity, which focused on somatosensory awareness. Gait measurement was performed by the use of a camera and reflective markers positioned on the participants lower limbs. Balance measurements were taken in two conditions while on a force platform, in tandem standing (i.e., feet heel to toe) and tandem standing on a 7.87 inch/10 centimeter thick foam block. Sway anterior-posterior and medial-lateral were recorded while on the force platform.

There were no significant differences between the two groups in age, height, weight, or body mass index prior to the intervention. After the intervention, children who were deaf had a significant decrease in postural sway. The postural sway in all participants was significantly greater in anterior-posterior than medial-lateral directions. In addition, after the intervention, both groups average sway while standing on the foam block significantly decreased. Gait velocity was not significantly affected after the intervention in the group with hearing loss (see A 39). The proprioceptive training intervention that enhanced somatosensory ability displayed improvement in postural control and decreased sway in children with hearing loss.

Another intervention to improve postural control and motor development in children with sensorineural hearing loss and vestibular dysfunction was implemented by Rine et al., (2004, Level 2, A 40). Children with sensorineural hearing loss and vestibular

dysfunction were assigned to either a placebo or exercise intervention, which lasted 12 weeks consisting of 30-minute sessions taken place three times a week. Both groups were similar in age. Postural control was measured by posturography sensory conditions testing (SCT) and motor development was measured by the *Peabody Developmental Motor Scales* (*PDMS*; Folio & Fewell, 1983). The placebo group participated in language development training while the exercise intervention group participated in eye hand coordination and general coordination activities, and balance training. All the activities were designed to focus on visual and somatosensory awareness.

All children had significantly lower *PDMS* scores than the testing norms and postural control scores on the SCT-3 (i.e., sway-references vision, fixed support vision, somatosensory ratios) were significantly lower than norms during preintervention testing. In the postintervention testing, those in the exercise intervention group had significant improvements in the *PDMS* raw scores and intervention quotient compared to preintervention. The postintervention *PDMS* intervention quotients of the exercise intervention group were significantly higher than those of the placebo group. The placebo group actually had a .78 to .55 decline in the intervention motor quotient score after postintervention; however, once they participated in the exercise intervention the group had a significant increase in both the raw scores and motor quotient scores (see A 40).

In the postural control test, the postintervention exercise group, not the placebo group, had significant improvements in the SCT-3 scores but no significant difference in the vision and somatosensory ratio scores. Although those scores did increase in the postintervention exercise group and were within the normative sample. Similar

improvements were demonstrated when the placebo group participated in the exercise intervention after postintervention of the language development. When both group's post exercise intervention scores were merged, significant improvements were seen in the vision and somatosensory ratio scores. Therefore, exercise focused on coordination and the use of visual and somatosensory information improved postural control and motor development in children with sensorineural hearing loss.

Based on the review of literature, hearing loss may be a contributing factor of motor delay in children with CHARGE syndrome. Children with hearing loss, especially sensorineural hearing loss, need appropriate identification of motor developmental delay and function of vestibular system to implement effective intervention strategies to improve motor development. Children who are deaf need to receive early intervention and services if there is a suspected motor delay or vestibular dysfunction, in addition to communication and language development (Rine et al., 2004; Rine et al., 2000).

Summary

Based on the DST and the literature review pertaining to effect of hearing loss on motor development and walking, the following possible constraints are reported (see Table 4).

Table 4

Possible Constraints Related to the Effect of Hearing Loss on Motor Development and Walking

	Constraints	References
Individual	Vestibular nerve function	Inoue et al., 2013
	Vestibular function, balance, hearing loss	De Kegel et al., 2012
	Balance, hearing loss, cochlear implants, postural control, vision, proprioceptive, somatosensory	Suarez et al., 2007
	Sensorineural hearing loss, vestibular function	Rine et al., 2000
	Age, deafness, balance	Siegel, Marchetti, & Tecklin, 1991
	Age, deafness	Dummer, Haubenstricker, & Stewart, 1996
	Balance	Melo et al., 2012
	Cochlear implant	Amirsalari et al., 2012; Gheysen, Loots, & Waelvelde, 2008
	Eye hand coordination, coordination, balance, visual and somatosensory awareness	Rine et al., 2004
Environment	Sensory information	Suarez et al., 2007
	Parental hearing status	Lieberman, Volding, & Winnick, 2004
Task	Object control, locomotor	Dummer, Haubenstricker, & Stewart, 1996
	Gait	Melo et al., 2012
	Proprioceptive training	Majlesi, Farahpour, Azadian, & Amini, 2014
	Exercise intervention	Rine et al., 2004

CHARGE Syndrome and Motor Development

Motor milestones are usually delayed in children with CHARGE syndrome, with the average age for walking independently being between 35 to 57 months (Blake et al., 1998). Comparatively, the normal achievement of independent walking is 12 months with

the acceptable range being from 8.2 to 17.6 months (WHO Multicentre Growth Reference Study Group, 2006). Adapted physical education provides appropriate educational interventions, which can support children with CHARGE syndrome to reach their full potential of motor development and attainment of motor milestones. It is important that educators are aware of the range of outcomes (e.g., independent walking, communication) possible for children with CHARGE syndrome to achieve (Salem-Hartshorne & Jacob, 2004).

In addition to educational intervention, children with CHARGE syndrome also need equal access and opportunities as their peers have in physical education. Due to various characteristics of CHARGE syndrome, instruction in general and adapted physical education is crucial for the development and enhancement of their motor development and skills. Lieberman, Haibach, and Schedlin gained information about the current status of physical education for children with CHARGE syndrome by a parent questionnaire (2012, Level 3, A 41).

The questionnaire was developed to gather information about the status of physical education settings, modes of communication, modifications, and successful and difficult curriculum units. A total of 26 parents of children with CHARGE syndrome completed the questionnaire. The age range of the children with CHARGE syndrome was from 6 to 19 years old. The parents reported a range of physical education settings for their child with CHARGE syndrome being inclusive (42%) to segregated (23%) and a combination of both inclusive and segregated settings (19%). In addition, parents reported that the placement affects their child's success in physical education and more success was seen

in physical education when support staff (i.e., paraeducator, intervener, or teacher assistant) attended the class with the child. Communication used during physical education class was primarily American Sign Language (17%) with a combination of communication methods being used 17% throughout the class. Equipment modification was used most frequent (n = 6) to generate more access and success in physical education. However, 5 out of the 26 parents indicated that no modifications were used and another five parents were unaware if any modifications were made in the physical education lesson.

Nevertheless, children with CHARGE syndrome were reported by their parents to be most successful in individual motor skills (i.e., jumping, kicking, throwing) as opposed to sport units that involved balls, were fast-paced, and/or incorporated a team. Parents reported that their children had difficulty most with fundamental motor skills. The more information that is known about each child the better the educational team can make appropriate changes to the child's physical education program. Training teachers and support personnel on strategies, communication mode, and modifications to implement in the physical education settings will also increase the child's success (Lieberman, 2007). Modifications and appropriate support and placement are essential for children with CHARGE syndrome to have successful opportunities in physical education class.

Importance of Walking

Dunlap (1985, Level 3, A 42) suggested that a functional classification system for individuals with deafblindness could potentially be meaningful for educational purposes. Instead of using the degree of the disability, Dunlap developed the classification based on

the abilities of the individual. Two hundred and fifty-one individuals with deafblindness were assessed on 199 activity or skill statements using the *Severely Handicapped Progress Inventory* (Dunlap, Evelsizer, McQuiddy, & Powers, 1982). The instrument was grouped in 11 categories: gross motor, fine motor, perceptual, self-care, language, Braille, cognition, socialization, independent level, work activities, and leisure/recreation. Cluster analysis was used to group individuals based on the 11 instrumental subscores categories through hierarchical single linkage algorithm. Clusters were then tested for significant differences using an analysis of variance. Three groups emerged from the data, by stepwise regression to predict group membership. The four variables retained in the stepwise process were gross motor, language, leisure, and socialization, which were used for classification functions. Those four classification functions correctly grouped 83.7% of all the participants into three different groups (Group 1: 90%; Group 2: 84.9%; Group 3: 76%).

In addition to the four variables, the descriptive data on the degree of vision and hearing for each participant were used to understand the difference among the groups. Participants in Group 1 had the most vision loss but the least hearing loss. Group 1 participants were also the lowest functioning in all activities including both physical and cognitive activities. The differences between Groups 1 and 2 were physical activity performance and between Groups 2 and 3 were cognitive activity performance. Group 2 functioned higher on the physical activities than Group 1 but lower on the cognitive activities than Group 3. Group 2 also had less vision loss but more hearing loss than

Group 1. Group 3 was the highest functioning group, which had the least amount of vision loss but the most amount of hearing loss.

The most significant variables were gross motor and language between all three of the groups. With the differences among the three groups considered using just those two variables, the results seemed clearer; with the greatest difference between Groups 1 and 2 being gross motor and the greatest difference between Groups 2 and 3 being language. Dunlap (1985) indicated that gross motor activity skills may predict the cognitive abilities of individuals who are deafblind. The investigator further suggested the need to determine the relationships and influences of combined hearing and vision loss on activities, including gross motor performance.

Furthermore, Petroff (1999, Level 1, A 43) surveyed the characteristics and experiences of teens who were deafblind and who also finished high school, across the nation. Based on the results of the survey completed by parents (N = 102), the majority of participants who were deafblind were divided into two very different categories. The largest group of the investigation was classified under the following category: participants who did not communicate with language (i.e., signed or spoken) and did not walk independently. The other category included participants who communicated effectively using spoken and/or sign language and walked independently. Of the participants who were deafblind, 36.6% used a wheelchair and depended on others to be mobile (highest percentage) with the next highest percentage of youth, 30.1%, who walked independently without assistance or supervision. Therefore, walking independently was an important characteristic of youth with deafblindness.

Gross motor (i.e., age of walking) was once again used as a predictor of performance of children with deafblindness. In the investigation by Salem-Hartshorne and Jacob (2004, Level 3, A 44), gross motor, along with medical involvement (i.e., length of hospitalization at birth, frequency of surgeries, number of hospitalizations after birth), degree of deafblindness, degree of hearing loss, and degree of vision loss were used as factors associated with the developmental progress in children with CHARGE syndrome measured by the *Adaptive Behavior Evaluation Scale* (*ABES*; Carney, 1995).

Salem-Hartshorne and Jacob (2004) were interested in the developmental outcomes in children with CHARGE syndrome. A correlation analysis was used to determine the relationship of the factors. The *ABES* score correlated negatively with later age of walking, degree of hearing loss, degree of deafblindness, and medical involvement. The participant's earlier age of walking had the strongest positive relationship to the *ABES* score. In addition, the increase in the degree of deafblindness and medical involvement were positively correlated with an increase in delay of the age of acquired walking.

Other investigators examined the executive functions in children with CHARGE syndrome and how the functions may be related to behavior (Hartshorne et al., 2007, Level 2, A 45). The ability to regulate, control, and monitor thoughts, behaviors, and actions is defined as executive functions (Carlson, Davis, & Leach, 2005). Ninety-eight parents of children with CHARGE syndrome completed a history questionnaire (Salem-Hartshorne & Jacob, 2004), a *Behavior Rating Inventory of Executive Function* (*BRIEF*; Gioia, Isquith, Guy, & Kenworthy, 2000), and an *Autism Behavior Checklist* (Krug, Arick, & Almond, 1993).

All participants (*N* = 98) reported that their child with CHARGE syndrome had a delay in reaching motor milestones. The average age of walking was 3.08 years and the average age of crawling was 1.64 years. Correlation between age of walking, age of crawling, and walking ability with the three indexes (i.e., Behavioral Regulation Index, Metacognition Index, Global Executive Composite) on the *BRIEF* was performed. Age of walking was significantly associated with all three indexes on the *BRIEF*. As age of walking was more delayed (i.e., age increases), scores on the *BRIEF* indexes increased and were more clinically significant. The age of walking in children with CHARGE syndrome has also been related to some additional characteristics. A delay in walking has also been associated with the need for psychotropic medication (Wachtel, Hartshorne, & Dailor, 2009), an increase in behavior similar to children with autism (Hartshorne, Grialou, & Parker, 2005), and sleeping difficulties (Hartshorne et al., 2009).

CHARGE Syndrome

Accomplishing independent walking takes a great deal of balance, coordination, and postural stability, all which can be affected by dysfunction systems within the body. For children with CHARGE syndrome, the impact of vision loss and colobomas affect the visual system of the body (Russell-Eggitt, Blake, Taylor, & Wyse, 1990) while hearing loss and ear abnormalities (i.e., semicircular canals) effect the hearing and vestibular systems (Thelin, Mitchell, Hefner, & Davenport, 1986).

Due to the high occurrence rate of vision loss and hearing loss in children with CHARGE syndrome balance issues may be present. Haibach and Leiberman (2013, Level 3, A46) examined the balance and self-efficacy of balance in children with

CHARGE syndrome. Of the 22 children with CHARGE syndrome in the investigation, 14 had fallen in the past year and 14 parents stated that their child with CHARGE syndrome had a fear of falling. The average age of attaining independent walking was 41.65 months with a majority (n =18) using some type of assisted walking devices at times while walking. Balance was measured by a modified version of the *Berg Balance Scale* (Franjoine, Gunther, & Taylor, 2003), which consisted of 14 tasks. Self-efficacy was measured by a modified *Activities-Specific Balance Confidence Scale* (*ABC*; Powell & Myers, 1995) survey with their parents.

The participants with CHARGE syndrome scored significantly lower on all of balance tasks within the *ABC*. Based on the results from the balance scale scores, 9 of 22 the participants with CHARGE syndrome were at a low risk of falling with an additional 8 at a medium fall risk, and 4 at a high fall risk. None of the children without CHARGE syndrome were at a fall risk. In addition, the participants with CHARGE syndrome scored significantly lower on the *ABC* scale when compared to children without CHARGE syndrome. On a scale of 0 to 10 for balance confidence, with a 10 being completely confident, 19 of the children with CHARGE syndrome indicated a balance confidence score of 0. Scores on the *ABC* were correlated, moderately positive to the balance scale scores (see A 46). However, the balance scores were not significantly correlated by gender or age. Therefore based on the results of this investigation, children with CHARGE syndrome may be at risk for falling and may lack balance confidence.

For children with CHARGE syndrome difficulties in balance may be related to vestibular abnormalities or dysfunctions (Murofushi et al., 1997, Level 3, A 47).

Murofushi et al., (1997) examined five individuals with CHARGE syndrome between the ages of 3 to 23 years old. Of those five, all had vestibular abnormalities (i.e., absence of semicircular canals) and only three obtained independently walking. The other two were unable to independently walk at approximately 6 years old. The three who were able to walk independently performed the milestone at 18 months, 3 years, and 5 years. However, all three performed a walking gait with a wide base and stiff legged and had difficulties walking and balancing on uneven surfaces.

An additional investigation was conducted to examine if motor development was delayed due to vestibular areflexia, particularly if it is combined with vision difficulties in six individuals (age range 3 to 26 years old) with CHARGE syndrome (Admiraal & Huygen, 1997, Level 3, A 48). Electronystagmography was used for eye movements and audiograms, behavior audiometry, and brain stem auditory evoked potentials were performed. Of the six individuals with CHARGE syndrome, ages 3 to 26 years old, all had vestibular areflexia and had some degree of bilateral hearing loss. Only one individual had horizontal pendular nystagmus. The age range of independent walking was between 2 to 4 years of age; however, one individual's age was not reported. The investigators stated that intact saccular function may be important for upright stance and gait to occur. Adequate assessments and correct diagnosis are also essential in development of educational support for individuals with CHARGE syndrome.

In addition to balance, vestibular dysfunctions can also impact postural development and the development of walking in children with CHARGE syndrome (Abadie et al., 2000). To examine the vestibular abnormalities impact on postural development in

children with CHARGE syndrome (*N* = 17; age range 3 to 9 years old) Abadie et al., (2000, Level 3, A 49) documented the attainment of postural and developmental milestones through physical examinations and interviews with the parents using the *Brunet-Lésine scale* (Brunet & Lésine, 1983). Furthermore, a computerized tomography scan was performed on the individuals to examine the inner ear semicircular canals, vestibule, vestibular aqueduct, and cochlea. A vestibular function evaluation was also performed using otolith vestibular-ocular system responses (VOR). Throughout the investigation, it was noted qualitatively unusual patterns of development that were displayed.

Some of the unusual postural behavior exhibited by 17 children with CHARGE syndrome (Abadie et al., 2000) included:

1. deep hypotonia during the first months of life contrasting with normal tone after the acquisition of walking;
2. poor use of hands and arms support to compensate for lack of frontal or lateral balance that delays sitting, followed by a poor use of hands;
3. avoidance of the prone position with poor tone for raising on forearms;
4. predilection for the supine position resulting in the first displacement skill being crawling on the back;
5. frequent inability to crawl on all fours without resting the head on the floor;
6. easy straddling a toy car with feet pushing on the ground and hands resting on the wheel;
7. prolonged duration of the standing with support stage,

8. walking only possible in a familiar place, on a flat and regular floor, with walking outdoors not being acquired until about 1 year later;

9. when walking, changes of direction often resulting in falls and performed without turning the head, with no suppleness of the neck, as if head and trunk were frozen; and

10. inability to ride a bicycle without [stabilizers]. (p. 571)

Furthermore, the participants' developmental milestone mean ± SD and range were: stable sitting 14 ± 3.3 months (8 to 18 months), standing with support 19.8 ± 4.9 months (12 to 27 months), standing without support 23.9 ± 6.1 months (17 to 33 months), and walking indoors 29.8 ± 8.7 months (18 to 52 months). Ten of the 17 participants had bilateral complete absence of all three semicircular canals and vestibular function abnormities were present in all participants. Of the 10 with absent semicircular canals, none were able to walk before 18 months of age. On average, the developmental milestones were reached 50% later than in children with normal motor development. However, those participants with residual otolith function had the closest range of normal developmental milestone ages. It was concluded that any residual use of vision and proprioception systems could assist and compensate for vestibular abnormities that most children with CHARGE syndrome face.

Summary

Based on the DST and the literature review pertaining to CHARGE syndrome and motor development, the following possible constraints are reported (see Table 5).

Table 5

Possible Constraints Related to CHARGE Syndrome and Motor Development

	Constraints	References
Individual	Communication	Lieberman, Haibach, & Schedlin, 2012
	Language, communication	Petroff, 1999
	Hearing loss, vision loss, deafblindness, adaptive behaviors	Salem-Hartshorne & Jacob, 2004
	Regulate, control, and monitor thoughts, behaviors, and actions	Hartshorne, Nicholas, Grialou, & Russ, 2007
	Psychotropic medication	Wachtel, Hartshorne, Dailor, 2009
	Behavior	Hartshorne, Grialou, & Parker, 2005
	Sleep	Hartshorne et al., 2009
	Colobomas, vision	Russell-Eggitt, Blake, Taylor, & Wyse, 1990
	Hearing loss, ear abnormalities	Thelin, Mitchell, Hefner, & Davenport, 1986
	Balance, fear, confidence	Haibach & Leiberman, 2013
	Vestibular dysfunction, bilateral hearing loss, saccular function	Murofushi et al., 1997
	Vestibular areflexia	Admiraal & Huygen, 1997
	Vestibular abnormalities, hypotonia, balance, falls	Abadie et al., 2000
Environment	Education placements, support staff, modifications	Lieberman, Haibach, & Schedlin, 2012
	Hospitalization, surgeries, birth	Salem-Hartshorne & Jacob, 2004
	Familiar place, flat floor, outdoors	Abadie et al., 2000
Task	Wheelchair	Petroff, 1991
	Assistive devices	Haibach & Leiberman, 2013
	Toy car, 5-point crawl, supine position, stand with support	Abadie et al., 2000

Conclusion

In conclusion, the effect of vision, hearing, somatosensory, and vestibular systems all impact the ability of a child to balance, maintain postural control, and perform motor actions (e.g., walking). In children with CHARGE syndrome, it is common that all three of those systems are impacted based on the syndrome. In order to increase motor development, it is essential to know which of the systems and subsystems impact movement and how the other systems function to allow movement to occur in children with CHARGE syndrome.

In Chapter II, a literature review was conducted that focused on constraints that built a foundation for the reader and provided support for the rationale of this investigation. In the following Chapter III, a detailed explanation of the methodology is provided, which includes a description of data collection procedures and analysis that have been completed. The method section has been separated into Phases I and II, to ensure clarification for the reader.

CHAPTER III

METHOD

The purpose of this investigation was to examine the impact of individual rate limiters and affordances on acquiring the independent walking developmental milestone in children with CHARGE syndrome (coloboma of the eye, heart defects, atresia of the choanae, retardation of growth and/or development, genital and/or urinary abnormalities, and ear abnormalities and deafness). The CHARGE Syndrome Clinical Database Project (CSCDP) was used in Phase I of this investigation. Interviews with parents or primary caregivers of a child with CHARGE syndrome were used in Phase II. Both phases were performed simultaneously to examine factors and characteristics that delayed (i.e., rate limiters) or contributed (i.e., affordances) to the attainment of the independent walking developmental milestone in children with CHARGE syndrome.

The information gathered was guided by the research questions. The variables examined in each investigation phase were based on the dynamic system theory (DST). The data were analyzed separately for Phase I and Phase II and then through triangulation the results of Phase I and Phase II were compared (see Table 6). Parents or primary caregivers provided the data and information obtained in both Phase I and Phase II about their child with CHARGE syndrome.

This investigation involved a mixed method approach to generate a more complete and meaningful understanding of the results gathered from this investigation. The use of a

mixed method approach included several measurement approaches with varying designs to achieve triangulation.

Table 6

Study Description of Phase I and II

Phase	Explanation
Phase I (CSCDP Children with CHARGE syndrome rate limiters and affordances)	• Reviewed CSCDP for qualifying participants • Examined data to establish grouping based on degree of vision and hearing loss. • Conducted an ANCOVA analysis; controlling for hospitalization time. • Conducted a multiple regression analysis to examine the impact of additional rate limiters/affordances on the age acquiring the independent walking developmental milestone.
Phase II (Parent or primary caregiver perspective of their child with CHARGE syndrome; including rate limiters and affordances)	• Selected primary caregivers or parents of the participant (i.e., person with CHARGE syndrome) from Phase I or through recruitment to participate in the Phase II interview. • Scheduled and performed semi-structured interviews through the use of telephone or Skype voice call. • Transcribed recorded interviews. • Sent transcripts to participants to perform a member-check, to read through transcripts for validity and reliability of content. • Analyzed transcripts (i.e., coding) to determine major themes. • Themes reviewed by professor in the field of deafblindness. • Performed triangulation comparison from results of Phase I.

Phase I: CSCDP Children with CHARGE Syndrome Rate Limiters and Affordances

Phase I data were examined for impact on walking and the relationship of selected variables (i.e., rate limiters or affordances) on age of walking in children with CHARGE syndrome. The clinical data of individual characteristic information (i.e., vision loss, hearing loss, hospitalizations) were obtained from participant files in the CSCDP. This

project is located at Saint Louis University and has been endorsed by the CHARGE Syndrome Foundation. The methods that were used in Phase I of the investigation are described under the following headings: (a) Participants, (b) Development of the CSCDP, (c) Procedures, (d) Research Design, and (e) Data Analysis.

Participants

Participants were children with CHARGE syndrome (N = 62) who were currently in the CSCDP database and met the following inclusion criteria: (a) identified with CHARGE syndrome as his or her etiology, either through genetic testing or through current CHARGE syndrome characteristic evaluation and (b) achieved independent walking. The exclusion criteria were: (a) parents or primary caregivers who had children who are deafblind but did not have CHARGE syndrome, (b) parents or primary caregivers who were not receptively and expressively fluent in English, and (c) parents or primary caregivers who did not have access to the internet or email. The parents or primary caregivers of the child with CHARGE syndrome inputted the data and information into the CSCDP.

Development of the CHARGE Syndrome Clinical Database Project

The CSCDP database was used to obtain information pertaining to participants with CHARGE syndrome. The CSCDP is an Internet web-based database hosted by Saint Louis University (SLU), Missouri (IRB #22507; approved 6-11-13: Board #3; amended 6-2-15: Board #1) and includes responses from questionnaires related to CHARGE syndrome. Specifically, the CSCDP was designed to collect clinical data on individuals with CHARGE syndrome across a longitudinal time scale reported by their parent or

primary caregiver. A data transfer agreement from the CSCDP was obtained and completed. The data were shared unidentified (i.e., anonymous). Initially, it took a parent or primary caregiver two to three hours to complete the entire database questionnaire. However, the information could have been inputted throughout multiple sittings; therefore, a parent or caregiver could have logged in to the database multiple times to complete the questions and update information as needed. Further, parents were sent annual reminders to update information yearly.

The CSCDP questionnaire is accessible through Qualtrics, which is a survey research company contracted with SLU. Qualtrics provides data security for Protected Health Information. Qualtrics meets the rigorous privacy standards imposed on health care records by the Health Insurance Portability and Accountability Act. All Qualtrics accounts are hidden behind passwords and all data are protected with real-time data replication.

Parents or primary caregivers needed an email address and internet access to be able to enter data into the CSCDP database. Parents or primary caregivers were allowed to access the link to the database at a location of their choice with a computer as long as there was internet or Wi-Fi available. Parents or primary caregivers had the option to choose to complete only some sections of the CSCDP questionnaire.

The CSCDP consists of the following 12 numbered sections: (1) demographics; (2) birth history; (3) major diagnostic characteristics; (4) CHARGE syndrome minor diagnostic characteristics; (5) genetic testing; (6) surgeries and hospitalizations;

(7) vision; (8) hearing; (9) milestones and growth; (10) neurology; (11) medication, bone health, and sleep; and (12) photographs and comments.

The focus of this present investigation was on the developmental milestones of walking in addition to the usual precursor developmental milestones including sitting independently, crawling, and then standing (World Health Organization [WHO] Multicentre Growth Reference Study, 2006) from the CSCDP. The parents or primary caregivers were instructed to access information from medical documents (i.e., audiograms, eye examinations, growth charts) to ensure accurate data input of the information. It should be noted, Bodnarchuk and Eaton (2004) reported that parental reports of gross motor milestone attainments are valid when compared to a home visitor's observations.

Procedures

Institutional Review Board (IRB) approval from Texas Woman's University was obtained (IRB protocol #18216, see Appendix B) for this investigation, prior to the start of any data collection. A letter of support for this investigation was also obtained from the CHARGE Syndrome Foundation (see Appendix C). In addition, a data use transfer agreement form was completed by the primary investigator of this investigation and with the CSCDP administrator (see Appendix D) in order to access data within the database. The following information from CSCDP was gathered: vision acuity; type of hearing loss (i.e., conductive, sensorineural, or mixed); decibel loss; developmental milestone ages; and months hospitalized (i.e., surgery, illness) prior to the attainment of independent walking. Some additional variables gathered were: CHARGE syndrome characteristics,

height and weight, birth history (i.e., prematurity), and known vestibular or semicircular canal dysfunction.

The administrator of the CSCDP from SLU sent an email containing the questions that are asked in each of the 12 sections of the database questionnaire to the primary investigator of this investigation. After reviewing all 12 sections of multiple pages of questions, the primary investigator and two faculty members, one in the field of adapted physical education and the other in the field of motor learning, analyzed and unanimously agreed on specific questions that should be examined that related to the purpose of this investigation. The specific questions selected were then examined by an additional two faculty members, one in the field of exercise physiology and the other in the field of deafblindness, to determine if all questions were relevant to the purpose of this investigation.

Once the section questions were finalized, the primary investigator sent an email with the attached section questions (see Appendix E) to the administrator of the CSCDP. The administrator then accessed the CSCDP and selected the specific information from the participants' files within the database. An Excel spreadsheet with all the information pertaining to each individual participant based on the section question requested was sent to the primary investigator. Once the Excel spreadsheet of data were retrieved, demographic information about the individuals with CHARGE syndrome was organized and analyzed.

Research Design

The data provided by the CSCDP contained information pertaining to the child's vision and hearing levels. The following tables (see Tables 7 & 8) indicate the information obtained about the vision and hearing levels of individuals with CHARGE syndrome.

Table 7

The CSCDP Questions Pertaining to the Levels of Vision Loss in the Left and the Right Eye

	Normal (1)	Mild loss (2)	Moderate loss (finger counting) (3)	Light/dark shadows only (5)	Completely blind (6)	Unknown (7)
Left eye (1)	❍	❍	❍	❍	❍	❍
Right eye (2)	❍	❍	❍	❍	❍	❍

Table 8

The CSCDP Questions Pertaining to the Levels of Hearing Loss in the Left and the Right Ear

	Normal (0-15 dB) (1)	Slight (15-25 dB) (2)	Mild (26-30 dB) (3)	Moderate (30-50 dB) (4)	Moderately severe (50-70 dB) (5)	Severe (71-90 dB) (6)	Profound (91+ dB) (7)	Unknown (8)
Left ear (1)	❍	❍	❍	❍	❍	❍	❍	❍
Right ear (2)	❍	❍	❍	❍	❍	❍	❍	❍

Note. dB = decibel

As a result of the procedures from Phase I, the following research question guided the development of the current investigation.

Research question one (RQ1). What is the significant impact of vision and hearing loss as rate limiters in children with CHARGE syndrome on their attainment of independent walking while statistically controlling for the months in which the child with CHARGE syndrome was hospitalized? In order to examine the impact of vision and hearing loss on the attainment of independent walking, four independent groups were established. The four groups were based on the reported levels of vision and hearing of the participants with CHARGE syndrome. The four levels were based on the child's best corrected vision and aided hearing level. Within the current education system in the United States, the WHO, and also stated in the United States Association of Blind Athletes (USABA), the best corrected level of vision in the best eye is used for identification and classification (Individuals with Disabilities Education Improvement Act, 2004; USABA, 2016; WHO, 2010). The Deaflympics also classifies athletes based off of the hearing loss in the better ear (International Committee of Sports for the Deaf, 2016).

Therefore, the four levels of vision and hearing were based on the best eye and best ear and were as follows: (a) near normal vision and near normal hearing (n = 10), (b) near normal vision and hearing loss (n = 31), (c) vision loss and near normal hearing (n = 5), and (d) vision loss and hearing loss (n = 16). The criteria for the levels of vision loss and hearing loss were based on the *World Health Organization, International Classification of Diseases, 10th Revision* (ICD-10; WHO, 2010). Near normal vision included visual acuity levels from 20/10 to 20/70, which included the normal and mild loss categories within the CSCDP. ICD-10 stated that low vision or a visual impairment through

blindness included the visual acuity of 20/70 to 20/1200 or less (WHO, 2010). Therefore, those participants with CHARGE syndrome who were reported as having moderate vision loss, light and dark shadows only, or completely blind in their best eye were identified as having vision loss.

Children with near normal hearing were within the range of -10 to 25 decibels. Children with hearing loss above 30 decibels are defined as having moderate to profound hearing loss (WHO, 2015, n.d.). Therefore, normal, mild, or moderate hearing loss categories within the CSCDP were identified as individuals with near normal hearing levels. Children with a hearing loss in their best performing ear at a level of moderate loss or higher were considered to have a significant hearing loss. The effect of assistive devices (e.g., glasses, hearing aid, cochlea implant) on vision and hearing were obtained if parents or caregivers knew the effect and if the child with CHARGE syndrome used them on a daily basis.

Consequently, for RQ1, the research design consisted of examining the effect of the independent variable, which was the combined vision and hearing loss, on the dependent variable of age in months of acquired independent walking. Furthermore, since research performed by Salem-Hartshorne (2003) and Salem-Hartshorne and Jacob (2004) on children with CHARGE syndrome indicated that hospitalization had a negative impact on development. Therefore, months the children were hospitalized was used as a covariate within the research design.

Since the data provided by the CSCDP contained information on the left and right eye and ear, the primary investigator determined a deafblind score for each participant.

The deafblind scores were based on the level of both their left and right eye and their left and right ear hearing levels. This deafblind scale ranged from 4 (i.e., near normal vision, hearing) to 24 (i.e., profound vision, hearing loss). The deafblind score has been used in previous research (Salem-Hartshorne, 2003; Salem-Hartshorne & Jacob, 2004) as a quantifiable way of examining the dual sensory loss of individuals with CHARGE syndrome. Three groups were established based on the deafblind scale. Those participants that scored between 4 through 10 were grouped together (i.e., mild deafblind loss; $n = 5$) then those who scored between 11 through 17 (i.e., moderate deafblind loss; $n = 32$) and then 18 through 24 (i.e., severe deafblind loss; $n = 25$) were grouped together. Therefore, the effect of the independent variable, which consisted of three deafblind severity groups, on the dependent variable of age (in months) of acquired independent walking was also examined.

Within the field of deafblindness, particularly related to CHARGE syndrome, many complex variables may impact motor development besides vision and hearing loss (Hartshorne et al., 2011). Therefore, an additional research design was developed based on the second research question in this phase of the investigation.

Research question two (RQ2). What are additional significant rate limiters or affordance variables that are related to the age at which independent walking in children with CHARGE syndrome is attained? To examine this research question, variables that were presented and discussed in the literature review (see Chapter II) as possible significant rate limiters and affordances were examined based on information that was available in the CSCDP. The following variables available within the CSCDP were: left

and right eye vision levels; left and right ear hearing levels; deafblind score; balance problems; occurrence of 5-point crawling; occurrence of back scooting; type of hearing loss; and small or absent semicircular canals in the left and right ear confirmed by a computerized tomography (CT) scan or magnetic resonance imaging (MRI) scan.

Data Analysis

In Phase 1, pertaining to RQ1, vision and hearing loss were analyzed as individual rate limiters to examine the impact of varying degrees of combined loss on the acquired independent walking developmental milestone in children with CHARGE syndrome. The impact of vision and hearing loss was analyzed through a between subjects ANCOVA, controlling for months in which participants have been hospitalized. The covariate statistically controlled for the impact of being hospitalized within the four different groups. The dependent variable was the developmental milestone of independent walking, which was measured in months. Descriptive statistics were conducted to provide additional information about the participants' demographic information and precursor developmental milestones (i.e., sitting, crawling, standing) leading to independent walking. The alpha level was set to .05. All statistical analyses were conducted using SPSS Version 19.0 (SPSS Inc.).

This data file was examined for the assumptions needed to analyze the data through the use of an ANCOVA. Therefore, the data were examined for normal distribution of the data, outliers, Levene's test for homogeneity of variances, and homoscedasticity (Portney & Watkins, 2009). The covariate was also examined for a linear relationship to the age of walking in each of the four groups of vision and hearing loss and for homogeneity of

regression slope to assure there was no interaction between the covariate and the loss of vision and hearing.

Since the ANCOVA only reported the overall significant differences between the groups, due to it being an omnibus test statistic, if a significant main effect ($p < .05$) was established then a post hoc test was performed. A planned last comparison post hoc test was used to analyze the comparisons of the groups, which determined any significant difference among the four groups (Portney & Watkins, 2009).

Another data analysis was performed to examine the impact of the three deafblind severity groups on the age in months of acquired independent walking (i.e., dependent variable). This data file was also examined for the needed assumptions (i.e., distribution of the data and outliers, Levene's test for homogeneity of variances, and homoscedasticity) to analyze the data through the use of an ANOVA (Portney & Watkins, 2009). The covariate, months hospitalized, did not meet the assumptions needed to analyze the data through an ANCOVA.

Based on RQ2, a subsequent multiple regression (standard enter method) data analysis was performed. Multiple regression was used to examine the impact of additional rate limiters and affordances on the age of independent walking. The data were examined for independence of errors, a linear relationship, normal distribution, homoscedasticity, multicollinearity, and any significant outliers. The data were then examined for a linear relationship between the ages of independent walking with each independent variable. Once it was determined that the data met the assumptions, a

multiple regression analysis was performed to examine the overall fit of the model and the relative contribution of each independent variable on the total variance explained.

Multiple regression analysis was applied to examine whether the age of independent walking in months could be predicted based on the following variables: left and right eye vision levels, left and right ear hearing levels, deafblind score, balance problems, occurrence of 5-point crawling, occurrence of back scooting, type of hearing loss, and small or absent semicircular canals in the left and right ear. Since this was the first multiple regression model to be examined pertaining to the age of walking in children with CHARGE syndrome, block multiple regression model was used with all variables entered at the same time. Due to the low sample size and number of variables used in this regression model, a subsequent multiple regression analysis was used from the first model.

Phase II: Parent or Primary Caregiver Perspective of Their Child with CHARGE Syndrome Rate Limiters and Affordances

During Phase II, a semi-structured interview was conducted with the parents or primary caregivers of a child with CHARGE syndrome. The information gained was used to gain further information regarding key variables (i.e., rate limiters or affordances) that impacted the age of walking. The qualitative analysis results were used in conjunction with the results from Phase I (i.e., quantitative data analysis) to better understand and validate impressions regarding the impact of rate limiters and affordances associated with achieving the developmental milestone of independent walking in children with CHARGE syndrome. The purpose of using multiple data sources was to enhance the

level of reliability within the investigation (Denzin, 1970, 1978; Johnson, Onwuegbuzie, & Turner, 2007). The following reflects the components of Phase II under the succeeding headings: (a) Participants, (b) Instrument, (c) Procedures, and (d) Data Analysis.

Participants

Phase II participants (*N*= 23) of the investigation meet the following inclusion criteria: (a) parent or primary caregiver of a child identified with CHARGE syndrome, (b) their child with CHARGE syndrome independently walked three to five independent consecutive steps, (c) parent or primary caregiver was receptively and expressively fluent in English, and (d) parents or primary caregivers whose son or daughter with CHARGE syndrome acquired the independent walking (i.e., three to five independent, consecutive steps) developmental milestone within the past 5 years.

Potential participants were recruited (see Appendix H) through: (a) flyers that included the research purpose and the primary investigator's contact information, were placed at the designated research information table at the International CHARGE Syndrome Conference; (b) recruitment scripts were read to potential families during the meet and greet conference session and throughout the International CHARGE Syndrome Conference from July 30th to August 2nd, 2015; and (c) emails were sent to participants within the CSCDP database and CHARGE Syndrome Foundation explaining the research purpose and intent, including the informed consent form. An individual's consent form (see Appendix I) had to be signed by the parent or primary caregiver and obtained by the primary investigator prior to the start of the interview process.

Twenty-eight participants were recruited from the International CHARGE Syndrome Conference in July 2015. Of those 28, 20 were randomly selected to participate in the interview; an email was sent on August 17th, 2015 to each participant to set up a date and time to conduct a telephone interview. Another email was sent (September 17th, 2015) to the 20 participants recruited from the International CHARGE Syndrome Conference reminding those who had yet to request a date and time for the interview and thanking those participants who had already completed the interview. Eight of those 20 participants who were contacted and informed about this phase of the investigation completed the interview.

In addition, the administrator of the CSCDP sent an email on September 1st, 2015 to potential participants within the CSCDP (i.e., Phase I) who met the inclusion criteria to be interviewed. Furthermore, an email was sent on behalf of the CHARGE Syndrome Foundation on September 26th, 2015 to all participating members and families, which included information about the research investigation and an email address to contact the primary investigator if interested. Of those contacted through the CSCDP and CHARGE Syndrome Foundation, 15 showed an interest in the study and completed the interview. An additional seven potential participants showed interest; however, they did not meet the inclusion criteria to participate (i.e., son or daughter was not walking independently yet or did not acquire walking within the past 5 years). Therefore, a total of 23 participants took part in the semi-structured interviews.

Instrument

The use of qualitative research within this mixed methods research investigation was performed to enhance the collection of information of variables that were not available within the database and to provide a more in-depth understanding and validation of the complex concepts involved in the study **(**Hesse-Biber, 2010). "Mixed methods research is an intellectual and practical synthesis based on qualitative and quantitative research" (Johnson et al., 2007, p. 129). A semi-structured interview was used to gain insight from the parents or primary caregivers pertaining to their experience and perspectives regarding their child achieving independent walking in response to the following research questions: (a) what contributing factors (i.e., rate limiters) caused a delay in walking for their child with CHARGE syndrome and (b) what contributing factors (i.e., affordances) lead their child with CHARGE syndrome to achieve the developmental milestone of walking independently.

The semi-structured interview protocol (see Appendix F) entailed a specific set of questions based on the theoretical framework of the dynamic systems theory (DST) and the purpose of this investigation. Interview questions focused on gaining insight about the parents' perspectives and experiences of their child with CHARGE syndrome development pertaining directly to the child's ability to achieve independent walking. The main interview questions were followed by probing questions to seek and gain more information and insight on the parent's perspectives (Roulston, 2010), therefore, enriching the data obtained from the parents and primary caregivers. The probing

questions were developed based on the DST of interacting systems and the impact of the individual and environment on a specific task.

The interview protocol was validated by the use of two experts in the field of qualitative research, with one who has conducted qualitative research in the field of deafblindness. The two experts edited the interview questions until both agreed on content validity. The primary investigator also conducted two pilot phone interviews with parents of children without disabilities. At the end of the interview, a discussion occurred based on the comprehension of questions asked and the specific information that was provided.

Each semi-structured interview was approximately 10 to 25 minutes in length. All interviews were conducted through the telephone (n = 21) or through a Skype call (n = 2), which is an online telephone service. In addition, a list of referral resources was also provided to each participant as a resource to obtain more information about CHARGE syndrome and to network with other families in the event he or she experienced any feelings of discomfort (see Appendix G). Information obtained from each interview was compared to the data provided in the CSCDP questionnaire for accuracy. The DST framework was used as a lens to associate the findings of Phase I to the results of Phase II.

Procedures

Prior to the interview, the primary investigator emailed each participant with the interview protocol and a document which included figures of developmental motor milestones (see Appendix J). Each interview was audio recorded and then transcribed by

the primary investigator. During the interview, the primary investigator wrote down additional anecdotal notes that were analyzed in conjunction with the transcripts. The name of each participant and any additional identifiable information used throughout the interviews were replaced with pseudonyms to maintain confidentiality. Transcriptions were emailed back to each participant to ensure the validity of the information. Participants had the opportunity to read the information, make edits, and/or add additional information to the interview transcriptions prior to the completion of data analysis. This process of member checks increased the probability of trustworthiness and validity within the investigation by reflecting the participants' responses more accurately (Schutt, 2012).

Data Analysis

After the interviews were completed, the raw data obtained from the audio recordings were transcribed and the analysis of themes and codes began (Miles, Huberman, & Saldaña, 2014). The primary investigator transcribed each participant's recorded interview in order to provide a comprehensive qualitative analysis. The main experiences and perspectives stated by the parents or primary caregivers during the interviews were diagrammed to examine relationships among the variety of responses (Maxwell, 1996). The qualitative analysis was driven by the research questions and the DST. The following are the research questions for this phase of the investigation:

Research question three (RQ3). What do parents or primary caregivers propose are affordances that led their child with CHARGE syndrome toward the ability to walk independently?

Research question four (RQ4). What do parents or primary caregivers propose are rate limiters causing a delay in walking for their child with CHARGE syndrome?

In order to answer the research questions, each interview transcription was analyzed to build on the development of a set of codes, which led to the identification of categories or themes. “Codes are labels that assign symbolic meaning to the descriptive or inferential information compiled during a study” (Miles et al., 2014, p. 71). Coding also refers to a systematic process of organizing qualitative data into specific categories and themes (Bogdan & Biklen, 2006).

The coding was the beginning of analyzing the information, which then was used for data condensation and to detect reoccurring categories and themes. Categories and themes were defined, in part, by the quantitative data and by the DST. The categories symbolized clusters of related information. The themes represented understandings that reflected the most evidence and presented impressions related to the research inquiries within a particular category.

The qualitative data were examined for incidents and comments that either supported or raised questions about results that emerged from the quantitative methods (i.e., Phase I) and the DST. The DST pertains to constraints related to the individual, task, and environment. Therefore, the categories of the coding incidents and comments in the data were based on the following; Individual (I), Environmental (E), or Task (T) which related to the DST and captured the essence of what each individual stated.

As themes began to emerge, major themes were highlighted according to relevance based on the research questions and DST. Within the DST, there are two separating

categories, which are affordances and rate limiters. With further examination, the initial codes (i.e., I, E, T) were then separated into two subcategories, which related directly to each research question. Affordances were comments made from the parents or primary caregivers of children with CHARGE syndrome that assisted and lead to the development of walking (i.e., RQ3). Rate limiters were comments that pertained to information that impacted or delayed the child's ability to acquire independent walking (i.e., RQ4).

As the data accumulated, the coding of transcriptions was further examined for information related to the individual, environment, and task constraints through the use of NVivo. NVivo is a computer assisted qualitative data analysis software program for the analysis of text data (QSR International Pty Ltd, 2012). A qualitative comparative analysis was performed to systematically identify concepts, which resulted in additional coded themes that were present across participants (Schutt, 2012). The qualitative research process also involved grouping or clustering categories at increasingly complex levels. NVivo facilitated comparison of categories across data files to detect patterns both within and across cases. The coded themes assisted in the development of specific summary outcomes, in addition to the primary investigator's categories and themes. Additional independent coders also examined the reliability of the codes and themes. The disagreements between coders were examined and additional discussions and revisions were made to the major themes. This process validated the codes and established the themes. The interrelationships of the codes and categories led to the development of higher-level analytic meanings, providing information about the DST (Kugler et al.,

1982; Smith & Thelen, 1993; Thelen & Ulrich, 1991) pertaining to the achievement of independent walking in children with CHARGE syndrome.

Integration of Phase I quantitative results were examined with the results of Phase II qualitative analysis in order to gain more in depth results about the research questions. Johnson, Onwuegbuzie, and Turner (2007) stated that based on the research questions mixed design research can, " . . . provide the most informative, complete, balanced, and useful research results" (p. 129). In this mixed methods investigation, triangulation was used to compare the results and data from Phase I with the information obtained from Phase II.

Denzin (1970, 1978) and Patton (1999) identify four types of triangulation: methodological triangulation, which is used in this investigation; investigator triangulation; data triangulation; and theoretical triangulation. Methodological triangulation is the use of more than one method for gathering data to examine the consistency of findings generated by the different data collection methods (Denzin, 1978). In this investigation, methodological triangulation included the combined use of quantitative and qualitative research methods to provide a more complete set of findings than could be arrived by the use of one research method (see Figure 5).

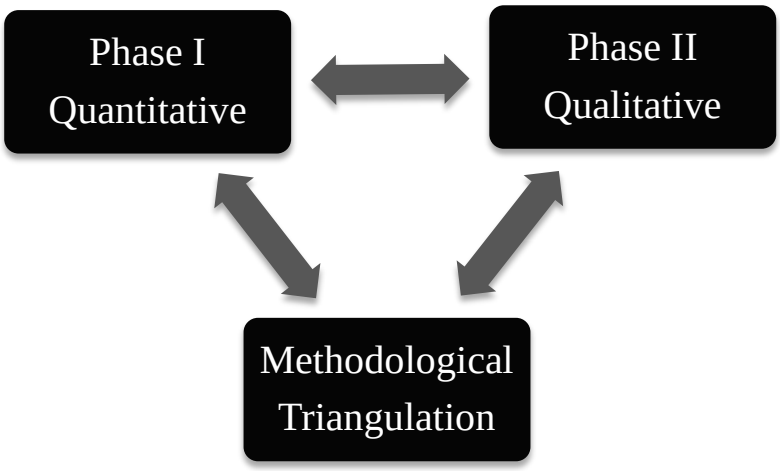

Figure 5. Visual representation of methodological triangulation analysis of data obtained from this mixed methods approach investigation.

Triangulation was used to enhance the credibility and convergent validity of this research investigation (Denzin, 1970). The purpose of the qualitative research was to provide an enriched understanding of the human experience within individual, environmental, and task constraints impacting the child with CHARGE syndrome. In addition, the use of methodological triangulation (i.e., quantitative, qualitative data) enhanced the confirmation or disconfirmation of conclusions and authenticated or supported the findings (Silverman, 2011). In addition, research bias related to the results has been lessened with the use of the dynamic systems theoretical filter.

Chapter III provided a detailed explanation of the method used in both Phase I and Phase II of this investigation. In the following Chapter IV, both the quantitative and qualitative data, which have been analyzed, are presented. The results section has been separated into Phase I and Phase II, to ensure clarification and understanding of the findings.

CHAPTER IV

RESULTS

The purposes of this investigation were to: (a) determine the significant impact of vision and hearing loss as rate limiters in children with CHARGE (coloboma of the eye, heart defects, atresia of the choanae, retardation of growth and/or development, genital and/or urinary abnormalities, and ear abnormalities and deafness) syndrome on their attainment of independent walking; (b) explore additional significant rate limiters or affordance variables that may be related to the age of independent walking; (c) determine what parents or primary caregivers propose are affordances that lead their child with CHARGE syndrome toward the ability to walk independently; and (d) determine what parents or primary caregivers propose are rate limiters causing a delay in walking for their child with CHARGE syndrome. According to the purposes of this investigation, two investigational phases were developed and implemented: Phase I (CHARGE Syndrome Clinical Database Project [CSCDP], Quantitative) and Phase II (Semi-structured interviews, Qualitative). The conceptual framework and theory applied throughout this investigation was the dynamic systems theory. In this section, the results of each phase are presented in sequence.

Phase I: CSCDP Children with CHARGE Syndrome; Rate Limiters and Affordances

The purpose of this phase was to examine the impact of vision and hearing loss on the acquired age of independent walking in children with CHARGE syndrome. The purpose was also to explore additional rate limiters or affordances and examine the relationship additional variables have on the development of walking. Based on the purpose of the study, the following research questions were derived:

Research Question One (RQ1). What is the significant impact of vision and hearing loss as rate limiters in children with CHARGE syndrome on their attainment of independent walking while statistically controlling for the months in which the child with CHARGE syndrome was hospitalized?

Research Question Two (RQ2). What are additional significant rate limiters or affordance variables that are related to the age at which independent walking in children with CHARGE syndrome is attained?

The following are the results of Phase I under the following headings: (a) Participant Demographics; (b) RQ1: Impact of Vision and Hearing Loss; and (c) RQ2: Additional Rate Limiters and Affordances Related to the Age of Walking.

Participant Demographics

Participants' data were obtained through the CSCDP. A parent or primary caregiver completed the information in the CSCDP based on behalf of their son or daughter with CHARGE syndrome. Approximately 300 families requested a user identification to complete the CSCDP. Of those 300, approximately 240 signed the CSCDP consent form

and entered data. Of those, approximately 190 participants signed the amended CSCDP consent form, which granted shared data access to other researchers. A total of approximately 125 participants completed Section 9, Milestones and Growth. Therefore, a total of 110 participants that had information entered in Section 9 and also consented to share data were obtained; however, many were missing information throughout Sections 2 through 9. Consequently, a data cleaning process examined the 110 records in the CSCDP databank. As a result, data were obtained from 62 (58.2%) participants, children identified with CHARGE syndrome, who had all the necessary data (i.e., Sections 2 through 9; see Appendix E) in the CSCDP were used for this present investigation.

Once parents or primary caregivers were provided a user identification to access the CSCDP, they were able to move throughout the sections and select information that they would like to share and complete. Subsequently, many of the parents or primary caregivers did not complete all sections of the CSCDP, which impacted the total amount of participants available for this investigation. In addition, the primary investigator of the CSCDP had to resubmit for signatures on the consent forms from CSCDP participants due to an amendment to the Institutional Review Board consent form. The amendment to the consent form permitted other researchers access to the information provided in the CSCDP without obtaining additional consent form signatures. Therefore, if participants of the CSCDP did not re-sign the consent form, their data were not available for this investigation. Shown in Table 9 are the descriptive statistics of developmental motor milestone information available through the CSCDP of the 62 participants from Phase I.

Table 9

Descriptive Statistics (Age in Months) of Participants with CHARGE Syndrome from Phase I Selected Developmental Motor Milestones Achievement

Motor Milestone	*n*	Range	Min.	Max.	Mean	*SD*
Raise head	34	23	1	24	6.1	5.2
Sit with support	48	28	2	30	9.4	5.0
Sit independently	50	29	7	36	14.5	5.8
Crawl on stomach	39	41	7	48	16.8	10.3
Crawl on Back	31	42	2	44	14.5	8.0
Pull to stand	41	28	8	36	17.9	6.8
Stand holding on	45	29	8	37	17.8	6.8
Cruise holding on	39	31	11	42	20.4	7.5
Stand independently	42	63	9	72	27.5	12.5
Walk independently	62	71	13	84	36.6	15.7
Run	25	102	18	120	45.4	26.3
Jump	24	94	20	114	57.7	26.4
Climb stairs with alternating feet	28	108	24	132	71.6	30.8

Note. *n* = number of participants with information available in the CSCDP, *SD* = standard deviation.

Of the 62 participants with CHARGE syndrome, 42 were males and 20 were females. Based on previous research pertaining to motor development, gender was not a significant factor. Furthermore, many children with CHARGE syndrome are hospitalized throughout their infancy. The average months hospitalized of the 62 participants with CHARGE syndrome was 4.6 months (*SD* = 4.7). In addition, the 62 participants had on average 11.2 surgeries (*SD* = 10.6). The type of surgeries, which most required hospitalization, undergone by the participants is presented in Figure 6.

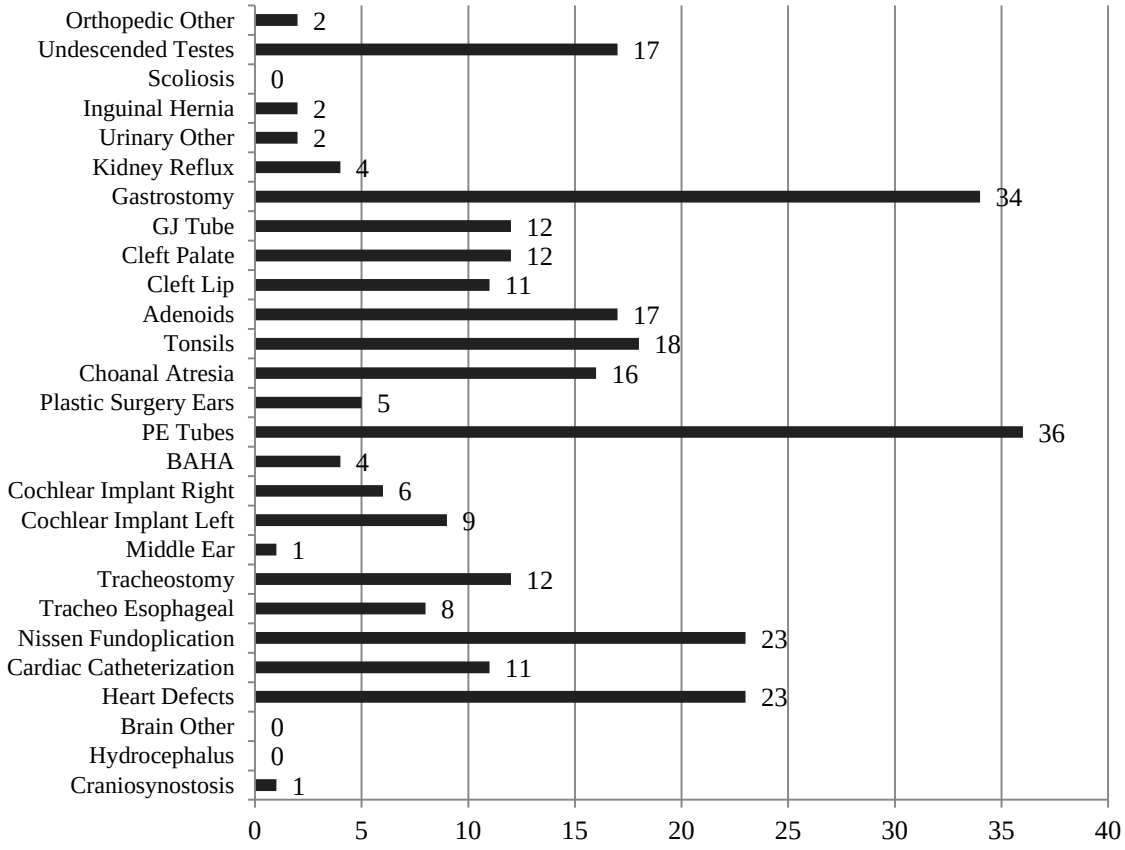

Figure 6. Type of Surgeries Experienced by the Participants with CHARGE Syndrome from Phase I. GJ tube = gastrostomy jejunostomy tube; BAHA = bone anchored hearing aid; PE tubes = pressured equalized tube.

In addition, the vision and hearing level severity for the 62 participants with CHARGE syndrome is provided (see Figure 7, Vision; Figure 8, Hearing). Vision severity was split into five levels based on the child's visual acuity loss which ranged from: normal (1), mild loss (2), moderate loss (3), light/dark shadows (4), to completely blind (5). The hearing severity was split into seven levels based on the child's decibel loss which ranged from: normal (1), slight loss (2), mild loss (3), moderate loss (4), moderately severe loss (5), severe loss (6), to profound (7).

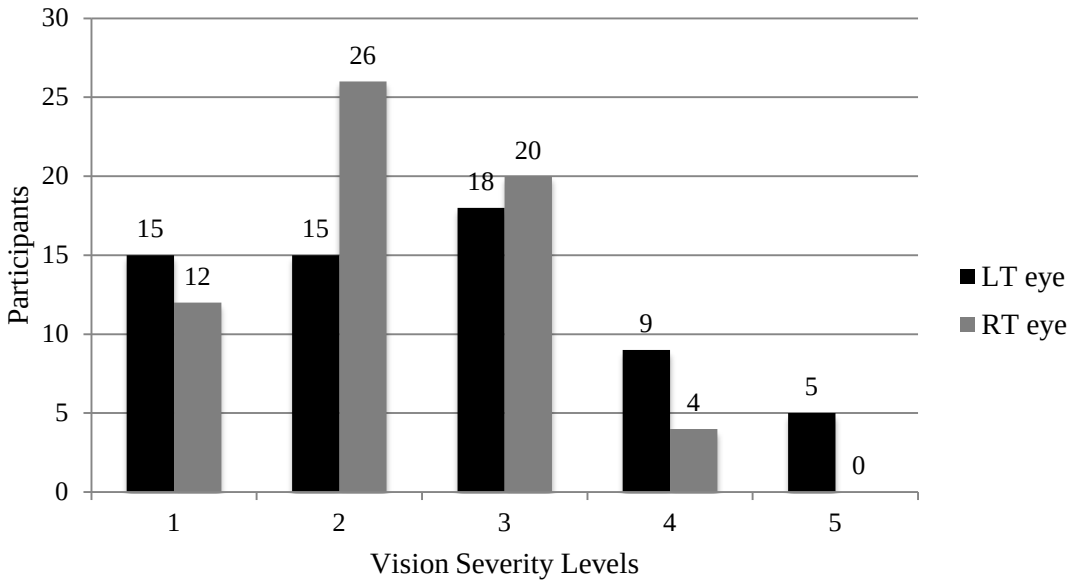

Figure 7. Vision Severity of the Left and Right eye of the Participants with CHARGE Syndrome from Phase I. LT = Left; RT= Right

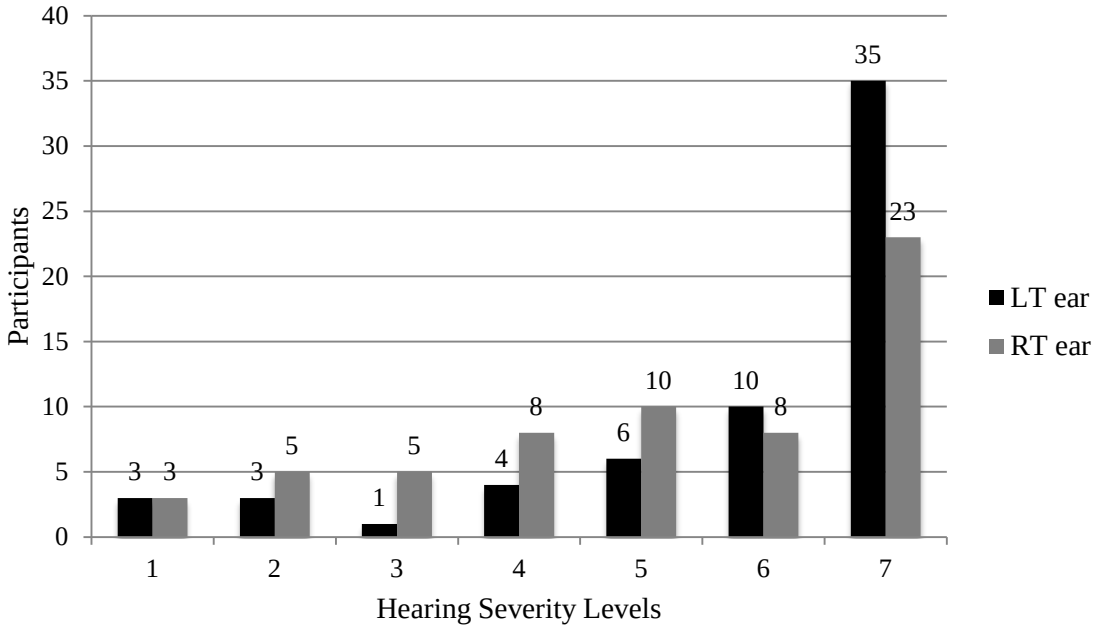

Figure 8. Hearing Severity of the Left and Right ear of the Participants with CHARGE Syndrome from Phase I. LT = Left; RT= Right

Furthermore, the ear structure and type of hearing loss present in the participants with CHARGE syndrome from Phase I are shown in Figure 9.

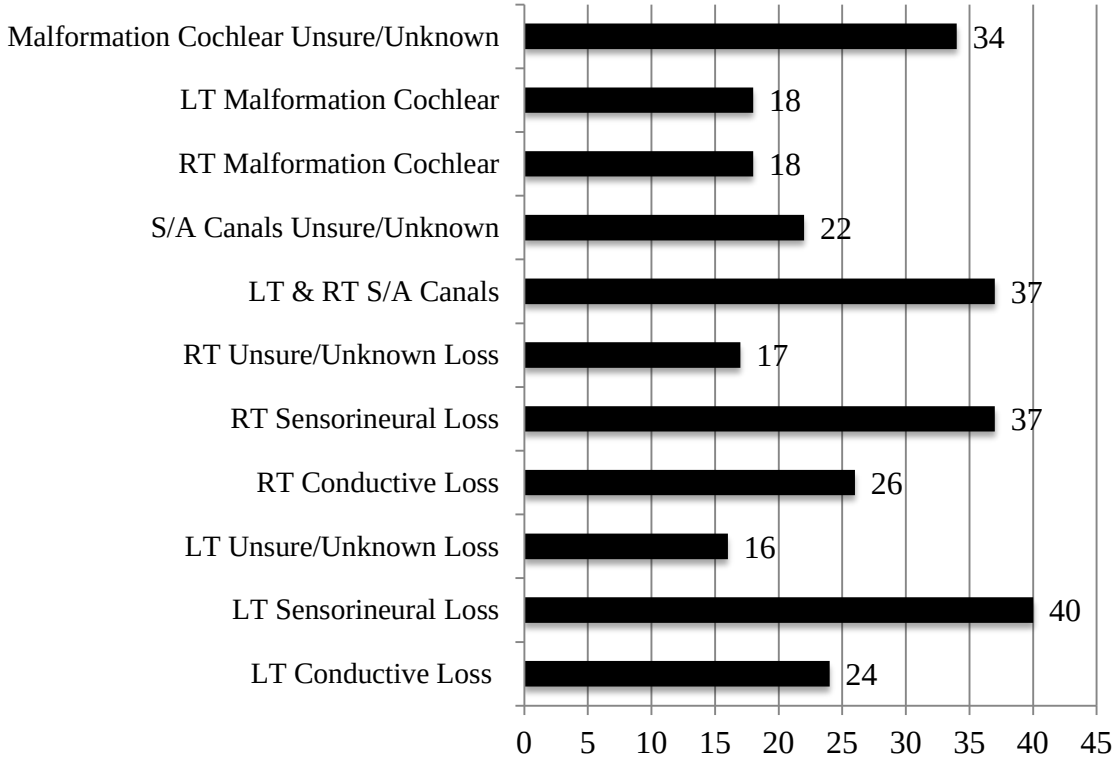

Figure 9. Inner ear Structure and type of Hearing loss of the Participants with CHARGE Syndrome from Phase I. LT = Left; RT= Right; S/A = Small or absent inner ear canals.

RQ1: Impact of Vision and Hearing Loss on age of Walking

To examine the impact of vision and hearing loss on the age of walking of children with CHARGE syndrome, the following procedures were followed:

Data Cleaning. Once the raw data were obtained from the primary investigator of the CSCDP, the data were opened in a large Excel document. The raw data (N = 62) were then reviewed to assure usable, reliable, and valid information for data analyses (Portney & Watkin, 2009). First the data were analyzed for duplicate data; two sets of duplicate

data were located and one duplicate in each set was deleted from the file. Second, the data were also compared to the 23 participants who participated in Phase II of this investigation to establish internal (intra-rater) reliability in both phases. If there was a difference of information reported in the interview that was presented in the database, an email was sent to the parent or primary caregiver (n = 4) for clarity and confirmation of the correct information presented. Data from a total of 62 participants were used for the following statistical test, based on the availability of the data needed for each variable.

Assumption tests. Numerous assumptions must be met in order to generalize the results (Field, 2013). After the data cleaning, the data were examined to determine if the following assumptions were met. In this phase of the investigation, the assumptions for the statistical ANCOVA method were examined prior to data analysis.

The data were first examined for outliers, which may have an impact on data analysis. Visual inspection of boxplots, Z scores, and Cook distance values were used to examine the data for outliers. A case was considered an outlier if: (a) value was greater than 1.5 box lengths of a box plot; (b) Z score was ± 3 standard deviations; or (c) Cook distance value was greater than 1 (Cook & Weisberg, 1982). Based on the outlier examination results, one case was identified as an outlier, as assessed by the inspection of a boxplot for values greater than 1.5 box-lengths from the edge of the box and Z score of 3.0345. That case was part of Group 2. Instead of excluding that case for further data analysis, the value was adjusted to the next highest score within that dataset for Group 2.

The models of errors (i.e., differences between the model and the observed data) were determined to be a normal distribution. Normal probability plots, generated by

SPSS 19.0, were visually examined and the values of the plots were laid over a straight diagonal line; therefore, concluding the assumption was met. A scatterplot (i.e., standardized predicted values of the dependent variable by standardized residuals) was visually examined for the assumption of homoscedasticity. Based on the illustration of the plots, the values were evenly spread out and therefore the assumption of homoscedasticity was met. The homogeneity of variances assumption was met, as assessed by Levene's test of homogeneity of variance (p = .406).

In addition, the covariate variable (i.e., months hospitalized) was visually examined for the assumption of linearity through a grouped scatterplot. Although all the groups did not have a linear relationship, the covariate was also examined for homogeneity of regression slope to assure there was no interaction between the covariate and the loss of vision and hearing. The homogeneity of regression slopes was not statistically significant, $F(3,54) = 0.967$, p = .415. This assumption assured that there was no interaction between the covariate and the loss of vision and hearing. All the assumptions for the ANCOVA were met and the data analysis was performed.

RQ1 results. After adjustment for months being hospitalized, there was a statistical difference in age of walking between the vision and hearing groups, $F(3,57) = 3.10$, p = .034, partial $n^2 = 0.14$. The covariate (i.e., months in hospital) was also significant (p = 0.045). The adjusted means for each vision and hearing group were: Group 1 (near normal vision and hearing) M = 32.7, SD = 13.2; Group 2 (near normal vision, significant hearing loss) M = 33.3, SD = 12.9; Group 3 (significant vision loss, near normal hearing)

M = 29.5, *SD* = 16.2; and Group 4 (significant vision and hearing loss) *M* = 45.1, *SD* = 16.8.

Analysis was performed with the use of a simple (last) planned comparison. A significant ($p < .05$) difference between Groups 1, 2, and 3 when compared to Group 4 was reported. Group 4 was significantly ($p < .05$) more delayed in the age of independent walking when compared to Groups 1, 2, and 3 (see Table 10).

Table 10

Results of Simple (last) Planned Comparison Between Group 4

Vision Hearing Group	Simple Contrast*		DV: Walk (months)
Group 1 vs. Group 4	Contrast Estimate		-12.410
	Std. Error		5.636
	Sig.		.032
	95% Confidence Interval for Difference	Lower Bound	-23.695
		Upper Bound	-1.126
Group 2 vs. Group 4	Contrast Estimate		-11.798
	Std. Error		4.394
	Sig.		.009
	95% Confidence Interval for Difference	Lower Bound	-20.597
		Upper Bound	-2.999
Group 3 vs. Group 4	Contrast Estimate		-15.626
	Std. Error		7.179
	Sig.		.034
	95% Confidence Interval for Difference	Lower Bound	-30.002
		Upper Bound	-1.250

Note. * = Reference group 4, DV = Dependent variable

To further examine the impact of vision and hearing loss on the age of walking in children with CHARGE syndrome, the additional procedures were followed:

Data cleaning. In the following analysis, the level of vision loss and the level of hearing loss presented by each participant were used to obtain a combined deafblind

score. The scores of vision and hearing loss were combined to generate the deafblind score (i.e., 4 near normal hearing and vision through 24 profound hearing and vision loss). The deafblind scores for each participant were divided into three severity groups of deafblind loss. Group 1 was mild deafblind loss (scores 4-10), Group 2 was moderate deafblind loss (scores 11-17), and Group 3 was severe deafblind loss (scores 18-24). After the data modifications were determined, the following assumptions were performed.

Assumption tests. Based on the outlier examination results, there were two outliers in the data. Both cases values were greater than 1.5 box lengths of a box plot. One participant's data point was from Group 2 (Z = 3.04) and the other was from Group 3 (Z = 3.04); both were adjusted to the next highest score within the dataset for each group. Therefore, Participant Number 29 from Group 2, age of walking was adjusted from 84 months to 60 months; for Participant Number 59 from Group 3, age of walking was adjusted from 84 to 60 months.

Normal distribution for each level of the independent variable was visually examined by normal probability plots generated by SPSS 19.0. All data points were placed on the diagonal straight line, indicating normal distribution assumption was met. Through Levene's test for homogeneity of variances, equal variances were assumed (p = .781).

The covariate was visually examined for the assumption of linearity through a grouped scatterplot, in which this assumption was violated. In addition, the homogeneity of regression slope was also significant, F (2,56) = 3.998, p = .024. Therefore, this

assumption was also violated. Since assumptions were not met to perform an ANCOVA, an ANOVA was used in the data analysis.

RQ1 results. A between subjects ANOVA was conducted to determine if the age of acquired independent walking was different for groups with different deafblind severity levels. Participants were classified into three groups: mild deafblind (n = 5), moderate deafblind (n = 32), and severe deafblind (n = 25). The age of acquired independent walking in months increased from mild deafblind (Group 1: M =25.0, SD = 11.2), to moderate deafblind (Group 2: M =33.5, SD = 13.3), to severe deafblind (Group 3: M = 40.4, SD = 13.4). There was a significant difference between the three deafblind severity groups and the age of independent walking, $F(2, 59) = 3.632$, $p = .033$, partial $n^2 = 0.110$. A simple (last) planned comparison analysis was used. A significant ($p < .05$) difference between the mild deafblind severity group (Group 1) and the severe deafblind severity group (Group 3) was reported. The severe deafblind severity group (Group 3) was significantly ($p = .021$) more delayed in the age of independent walking than the mild deafblind severity group (Group 1; see Table 11). However, the severe deafblind severity group (Group 3) was not significantly ($p = .057$) more delayed than the moderate deafblind severity group (Group 2).

Table 11

Results of Simple (last) Planned Comparison Between Group 3

Deafblind Level Simple Contrast*			DV: Walk (months)
Group 1 vs. Group 3	Contrast Estimate		-15.360
	Std. Error		6.476
	Sig.		.021
	95% Confidence Interval for Difference	Lower Bound	-28.318
		Upper Bound	-2.402
Group 2 vs. Group 3	Contrast Estimate		-6.860
	Std. Error		3.529
	Sig.		.057
	95% Confidence Interval for Difference	Lower Bound	-13.921
		Upper Bound	.201

Note. * = Reference group 3, DV = Dependent variable

RQ2: Additional Rate Limiters and Affordances Related to the Age of Walking

To examine additional rate limiters and affordances the following procedures were performed:

Data cleaning. Specific acuity levels for the left and right eyes were obtained from the CSCDP. Of the 62 participants, 18 did not have specific acuity levels for the left eye and 26 did not have specific acuity levels for the right eye. It has been reported that it is difficult to obtain an accurate and exact visual acuity levels for children with CHARGE syndrome due to colobomas and difficult testing procedures (Salem-Hartshorne & Jacob, 2004). Therefore, the missing specific acuity level data were obtained by the use of the vision loss scale. The acuity data was established by *World Health Organization, International Classification of Diseases, 10th Revision* (ICD-10; WHO, 2010) based off of the vision loss scale. All visual acuity levels from the United States notation were

converted into the decimal notation for the vision acuity data to be continuous variable. Normal vision, 20/20, was then provided with the decimal notion 1.0. Total blindness visual acuity decimal notion was 0.0. A range between 1.0 and 0.0 was used based on each child's vision acuity or vision level. Left and right decibel loss was derived from the hearing loss scales provided in the CSCDP (i.e., normal hearing = 0 decibel loss, slight = 15, moderate = 30, moderately severe = 50, severe = 71, profound = 91).

Data obtained from the CSCDP that indicated either a left or right small or absent inner ear canal for a participant were provided with a 1. If the response listed in the data was unsure or no, the participant received a 0. This procedure (i.e., yes =1, unsure/no = 0) remained consistent for the following variables: balance problems, 5-point crawl, and scooted on back. Furthermore, if a participant had sensorineural hearing loss listed in either his or her left or right ear, the data point was a 1. If conductive or unsure was reported then the participant received a 0. This procedure was performed the same for small or absent inner ear canals. Once all the data were modified to meet the corresponding statistical test then assumption tests were performed.

Assumption tests. The assumption of independent errors was met because Durbin-Watson statistics indicated 1.424, which also was close to 2. The assumption would have been violated if Durbin-Watson statistic was less than 1 or more than 3. Based on the illustration of the scatterplots, values were evenly spread out, which indicated the homoscedasticity assumption and the assumption of a linear relationship were met. In addition, partial regression plots were examined and assumptions of linearity were met based on each independent variable and the dependent variable

(excluding categorical variables; e.g., balance problems, 5-point crawl, scooted on back, hearing type, small or absent inner ear canals).

Based on the collinearity statistics, the assumption of multicollinearity has not been violated. There were no significant outliers detected in the dataset, based on the casewise diagnostic analysis using SPSS 19.0. There were no Cook's distance values above 1 (Cook & Weisberg, 1982) and no Mahalanobis distance values greater than 31.26. The models of errors (i.e., differences between the model and the observed data) were determined to be normally distributed. In the histogram and the normal probability plot generated by SPSS 19.0, the curves were in a normal distribution shape and the values of the plots were laid straight over a diagonal line.

RQ2 results. The exploratory enter method multiple linear regression was used first to examine the independent variables (i.e., left acuity level; right acuity level; left decibel loss; right decibel loss; months hospitalized; number of surgeries; and the occurrence of balance problems, 5-point crawl, back scooting, small or absent semi-circular canals, sensorineural hearing loss) prediction of the dependent variable (i.e., age of independent walking). Based on the following model, $R = 0.548$; $R^2 = 0.300$, the variables in this model explained 30% of the variability of the age of walking. *Adjusted* R^2 is an estimate effect size, $R^2 = 0.146$ is indicative of a medium effect size, according to Cohen's (1988) classification. The results of the overall regression model was not significant, $F(11, 50) = 210.414$, $p = .054$. Based on the exploratory regression analyses the following variables were selected to be analyzed in a second regression model: right acuity level ($\beta = -.360$, $p = .065$), left decibel loss ($\beta = .277$, $p = .058$), months

hospitalized (β = .093, p = .584), and number of surgeries (β = .090, p = .586). The selection of these variables was based on the highest correlation beta value and significant value (i.e., closest to p = .05) in relation to the age of walking.

A second multiple regression was performed to predict the age of independent walking in children with CHARGE syndrome from right acuity level, left decibel loss, months hospitalized, and number of surgeries. The assumptions of linearity, independence of errors, homoscedasticity, no outliers, and normal distribution were met. Based on the following model R = .537, R^2 = .288, therefore, the variables included in this model explained 28.8% of the variability of the age of walking. The *Adjusted* R^2 is 0.238, indicative of a large effect size according to Cohen's (1988) classification. The overall prediction model was significant, $F(4, 57) = 5.766$, p = .001. The acuity level of the right eye had a large negative correlation with the age of walking (r = -.406). Within the regression model, the right eye visual acuity level (p = .004) and left ear decibel loss (p = .011) were significant predictor variables (see Table 12). Based on the results, the right eye acuity level had the most contribution toward predicting the age of independent walking (β = -.353). Based on the unstandardized coefficients, a decrease in the acuity of the right eye equals an increase in the age of walking by 17.453 months. Furthermore, an increase loss in the left ear equates to a .162 month increase in the age of walking.

Table 12

Summary of Multiple Regression Coefficients

	Unstandardized Coefficients		Standardized Coefficients			Correlations		
Model	B	Std. Error	Beta	t	Sig.	Zero-order	Partial	Part
(Constant)	27.628	5.706		4.842	.000			
Acuity Right*	-17.453	5.738	-.353	-3.041	.004	-.406	-.374	-.340
Left dB Loss*	.162	.061	.294	2.628	.011	.295	.329	.294
Hospitalization	.370	.477	.111	.775	.442	.254	.102	.087
Surgeries	.160	.215	.108	.747	.458	.274	.098	.083

Note. Dependent Variable = Achieved milestone: Walk alone (months), dB = decibel, * = $p < .05$.

Phase II: Parent or Primary Caregiver Perspective of Their Child with CHARGE Syndrome Rate Limiters and Affordances

The purpose of this phase was to gain further information regarding key variables (i.e., rate limiters, affordances) that impacted on the age of walking in children with CHARGE syndrome. Through the use of semi-structured interviews, the participants (i.e., parents or primary caregivers of children with CHARGE syndrome) shared their insights on the impact of the various systems on motor development. The participants also discussed the impact of the individual, task, and environmental constraints on the development of walking in their son or daughter.

Although there is some support in the literature for the impact of various systems (i.e., vision, hearing, vestibular, muscular) on motor development (see Chapter II), the research indicates there may be some mixed results in concluding the impact within children with a dual sensory loss and additional factors caused by CHARGE syndrome (Hartshorne et al., 2011; Hartshorne et al., 2007; Hefner, 2008). These concerns have

stimulated the primary investigator to examine the impact of rate limiters and affordances on the development of walking in children with CHARGE syndrome. The focus of this research component was to engage the parents and primary caregivers of individuals with CHARGE syndrome in a qualitative interview protocol to identify critical information regarding how his or her child obtained independent walking. The following research questions were central to this research investigation. **RQ3:** What do parents or primary caregivers propose are affordances that lead their child with CHARGE syndrome toward the ability to walk independently? **RQ4:** What do parents or primary caregivers propose are rate limiters causing a delay in walking for their child with CHARGE syndrome?

A variety of themes that have significance to the research investigation questions emerged from the 23 interviews. The following reflects the results of Phase II under the succeeding headings: (a) Participant Demographics, (b) Data Cleaning, (c) RQ3: Cross Case Analysis of Affordance Themes, (c) RQ4: Cross Case Analysis of Rate Limiter Themes, and (d) Triangulation.

Participant Demographics

Participants (N = 23) for Phase II of this investigation were parents or primary caregivers of children with CHARGE syndrome who also had their child's information entered into the CSCDP. All 23 participants reported to be a parent and primary caregiver. In addition, 6 of the 23 included their spouse (i.e., husband or wife) as also being the parent and primary caregiver of their child with CHARGE syndrome. The participants' ages ranged from 27 to 64 years old, with a mean age of 38 years old. Nineteen of the participants were mothers, which included two mothers who adopted

their son or daughter who had CHARGE syndrome. Of the 23 participants, three were fathers who participated in the interview. In one interview, both the mother and father were present and the telephone was placed on speakerphone to allow both participants to respond. One participant preferred to answer the interview questions through email. A telephone interview was then used as a follow up clarification to the emailed responses. Additional descriptive information about the participants is provided in Table 13.

Data cleaning. After the completion of the interviews, the primary investigator transcribed each interview. After the transcriptions were complete, the primary investigator reviewed the anecdotal notes that were taken during the interviews with the transcriptions to make sure all relevant information was present. Transcriptions were then sent back to each participant (i.e., member-checking) for them to reread and check for accurate information. At that time the participants could edit and add information to the transcription if needed. If no edits or corrects were needed, then the participant did not need to respond or take any actions. Of the 23 participants, one participant responded through email with additional information to clarify and provide more in depth information pertaining to the questions asked during the interview. Transcriptions were edited of any filler vocalizations (i.e., umm, ah).

Table 13

Descriptive Information of the Participates in Phase II

	Region	Relationship to Child	Age	Child's Gender	Child's Age	Type and Age of Diagnosis	Interview Duration and Type
R. 1	U.S.	Mother	35	Female	2y 4m	Genetic +; 5m	15m 34s; telephone
R. 2	U.S.	(A) Mother	64	Female	6y 5m	Clinical; 6m	9m 50s; email/telephone
R. 3	U.S.	Mother	27	Male	3y 6m	Clinical, 2 weeks; Genetic +, 9m	19m 35s; telephone
R. 4	U.S.	Father	45	Female	2y 6m	Clinical, 2 weeks; Genetic +, 3 weeks	17m 45s; telephone
R. 5	U.S.	Father	45	Male	3y 7m	Genetic +; 3 weeks	20m 21s; telephone
R. 6	U.S.	Father	50	Female	8y	Genetic +; 2 weeks	17m 31s; telephone
R. 7	U.S.	Mother	42	Male	7y	Clinical, birth; Genetic +; 4y	12m 27s; telephone
R. 8	U.S.	Mother	31	Male	4y	Clinical, birth; Genetic +; 2m	10m 20s; telephone
R. 9	U.S.	Mother	30	Male	3y	Clinical, 6m; Genetic +, 11m	17m 25s; telephone
R. 10	U.S.	Mother	34	Female	5y	Clinical, 16m; Genetic +, 16m	22m 33s; telephone
R. 11	U.S.	Mother	37	Male	4y 6m	Clinical, 1 week; Genetic +, 4m	24m 44s; telephone
R. 12	U.S.	(A) Mother	29	Male	4y	Clinical, 2m; Genetic -, 3m	21m 40s; telephone
R. 13	U.S	Mother	38	Male	5y	Clinical, 2 weeks; Genetic +, 2m	22m 31s; telephone
R. 14	Canada	Mother	50	Male	8y	Clinical, 1 week	14m 35s; telephone
R. 15	U.S.	Mother	44	Female	5y 6m	Clinical, 1 week; Genetic +, 1m	12m 5s; telephone
R. 16	U.S.	Mother	32	Female	4y	Clinical, 4m; Genetic +, 14m	10m 31s; telephone
R. 17	U.S. Israel	Mother Father	40	Male	10y	Clinical, 3 weeks	15m 20s; telephone
R. 18	Saudi Arabia	Mother	36	Male	5y	Genetic +, 4m	13m 13s; Skype call
R. 19	Australia	Mother	35	Male	2y 8m	Clinical, 1 week; Genetic +, 6m	26m 10s; Skype call
R. 20	U.S.	Mother	33	Male	3y	Genetic +, 11m	13m 24s; telephone
R. 21	U.S.	Mother	39	Female	7y 10m	Genetic +, 2y	17m 2s; telephone
R. 22	U.S.	Mother	29	Female	4y 6m	Clinical, 1 week; Genetic -, 1m	17m 2s; telephone
R. 23	U.S.	Mother	33	Female	3y 6m	Clinical, 1m; Genetic +, 1m	21m 17s; telephone

Note. R = respondent; A = adopted; y = years; m = months; s = seconds; Clinical testing is based on CHARGE syndrome characteristics; Genetic DNA testing is used to identify the mutation of chromodomain helicase DNA binding protein 7 (CHD7) gene.

RQ3: Cross Case Analysis of Affordance Themes

Based on the responses from participants a variety of themes emerged based on variables which assisted their child in developing the independent walking milestone. The following are identified themes associated with this research question and DST. The themes are divided into three sections: individual, environmental, and task. Nvivo was used to develop a word frequency figure based on the affordance responses of the parents and primary caregivers (see Figure 10). The themes are supported by a few examples of participant transcriptions from the data to support the results. Any names provided in the interviews have been replaced with pseudonyms to maintain confidentiality of the participants. The verbatim transcriptions from the respondents are labeled in terms of respondent number (R.); whether it was a mother, adopted mother, father, or both (M, AM, F, M/F); and the respondent's age. For an example: R.1, M, 35.

Figure 10. Affordance word frequency of the 100 most frequent, 4 letter minimum, words from participant transcriptions.

Individual

After examining and coding the transcriptions, some themes pertaining to the child with CHARGE syndrome emerged as contributing factors in assisting with the attainment of independent walking. The following are major themes, supported by quotes from the participants.

Determined personality trait. Most of the parents and primary caregivers described their children with CHARGE syndrome as being a fighter and determined. These personality traits were described to assist in the attainment of independent walking. In the following quotations, participants described their child's personality traits and the impact they had specifically on the child's walking development.

"He would hold your hand [when walking] but it was like when he started getting stable enough to where he didn't need your hand, he would push you away. He started doing that very early, when he was independent. I would say determination, independent are probably his two factors that helped him." This respondent continued to discuss her son's independence. "I feel like he was very independent when he wants to do things. He doesn't want help a lot of times, but we are dealing right now with frustration because he doesn't want help but he wants to be able to tell us what he wants. I feel like with walking it was almost the same way." (R. 9, M, 30)

"She is very determined, she practiced a ton, just very determined she was going to walk herself so I think that was a big factor in all of the progress." (R. 1, M, 35)

"Trish has always been very happy and very determined! She never gives up!" Later in the interview, when asked if there was any additional information about her child achieving independent walking that she would like to share, the mother stated, "she is a fighter and she did her part by doing her best to do what her therapists asked of her." (R. 2, AM, 64)

"I think for himself, personally, he has been described this way by a number of therapist that he has a very strong agenda. If he wants to do something, he will do it. When that sort of mechanism clicked in his head, that yes, I want to get moving [independent walking], that's really what has helped him learn so quickly. That has been quite a big thing for him." (R. 19, M, 35)

"He is very determined, I mean he can't do something, he is going to keep doing it until he can get it right. He was putting his own shoe and socks on before he could even sit up on his own. So, when he puts his mind to something there is no stopping him from doing it." (R. 3, M, 27)

"She is pretty determined when she wants to do something, [pause] yea, she's you know, she is pretty strong headed about it, she works on things til you know until she can get it or find a way to get around it." (R. 4, F, 45)

"He has that personality and he always has. If you give him that goal and are like come on buddy, and tell him, you got to do this. He always over achieves . . ." (R. 11, M, 37)

Feeling of comfort and safety. As the interview discussions continued, many respondents mentioned the importance of their child feeling comfortable and safe in order for motor development to improve. Independent walking is a risk that children with CHARGE syndrome need to feel ready to take. With an increase in feelings of comfort and safety, as per the parents' perspectives this positively impacted their attainment in independent walking.

One respondent was explaining variables associated with CHARGE syndrome (i.e., lack of vestibular input, vision, hearing) which caused her daughter difficulty when trying to achieve independent walking, " . . . until she finally got comfortable [with walking] in the works and wanted to get here to there in a faster way." (R. 6, F, 50)

"I feel like in our situation, therapy didn't really make him walk. I feel like he walked, he started walking when he was comfortable and felt safe to walk, actually this October he will have been walking a year, 29 months is when he started walking." Then, after talking about her son's ability to cruise using furniture she stated, "Just until he got comfortable with that [cruising furniture] and started feeling safer, he would start walking a little bit." (R. 9, M, 30)

The feeling of comfort, for this respondent's child consisted of understanding how to manipulate and move her body, on her own terms. "I think it was her getting use to how her own body worked. I hate to say but a lot of therapies, as much therapy we did, I know it is good for them but until she has figured out her own lens, she is not going to do it, no matter how long she practices. I am sure some fundamentals must be there. It was really her coming to turns with how does this part of my body work." (R. 10, M, 34)

“I think just working on it [walking] in her comfort zone really helped.” (R. 15, M, 44)

Confidence. With the use of assistive devices (i.e., walker, push toy), many parents responded that their child gained a sense of confidence. However, time was also a perceived factor the impacted the use of a walker and the timetable to achieve walking independently.

“Maybe she felt ready at 3 years of age [to walk independently].” (R. 23, M, 33)

“. . . his legs have always been strong, stronger than his upper body he just doesn’t know how to get them going forward and his confidence. His confidence came gradually and so that is why I think it took so long for him to actually walk. He wanted to but he was just scared.” (R. 20, M, 33)

“And also, I think he [son with CHARGE syndrome] walked when he was ready, when his balance system was ready.” (R. 7, M, 42)

One mother stated that she practiced with her son, walked with him, and taught him how to fall. “That [practice] gave him the confidence where he wasn’t just going to fall and crash and hit his head. I think he started learning that oh, if I fall it is not so bad if I am walking by myself. I think motivation in just cruising using the coffee table, he just needed that confidence with something to hold on. That’s why that summer when he finally let go and moved he was just, I can remember his face, he was just so excited that he finally made it without falling. So I think a lot of it was the confidence and the ability to do it without crashing.” (R. 13, M, 38)

“It was a long road. She didn’t end up getting the confidence to walk independently until 28 months, which is still actually a little bit early. She started using a gait trainer, a walker, at about, around when she turned 2.” The same respondent continued to state that the walker enhanced her level of confidence. “I think having to wait so long to get the official gait trainer probably was a delay. Because once we got her in it and use to it, there were levels of confidence that we didn’t see with the pushcart. I wish we would had been able to get that sooner.” (R. 10, M, 34)

“She could pretty much walk holding onto our pinky fingers by the time she was 22 months. She didn’t have the confidence to let go. At 24 months she took her first steps on her own and then she was off from there.” The mother then explained, “the walker, gait trainer, gave her a big boost in confidence . . .” (R. 16, M, 32)

Summary. Evident from the quotations, the respondents expressed individual characteristics that their child exhibited or presented that improved the child's ability to independently walk. A majority of parents described their child as independent, determined, and confident, which fostered the achievement of independent walking. Furthermore, parents expressed the need for their children to feel safe and comfortable in their own bodies and within their surroundings to be able to produce new movement patterns.

Environmental

As with normally developing children, the environment plays a key role in motor development in children with CHARGE syndrome (Gabbard, Caçola, & Rodrigues, 2008; Thelen, 1995). The participants in this investigation shared what they perceived as environmental factors that contributed to their child with CHARGE syndrome independently walking.

Use of furniture. Many participants mentioned the use of furniture to assist in the development of walking. Since cruising is usually seen as a precursor to independent walking, many parents rearranged their houses and manipulated toys to encourage cruising and standing. Parents mentioned these various strategies that they implemented for the purpose of encouraging a child's motor development. For instance:

"I think, you know we did do some of the [pause] try to move stuff close enough together where she had a bridge between the furniture and those type of things." (R. 6, F, 50)

"Well, we have a lot of furniture, which was helpful for him to be able to hold on to the furniture and take steps without being independently walking. So he could go from couch to couch and chair and walking around the full living room without letting go of anything." (R. 3, M, 27)

"We would position furniture at certain distances, his PT [physical therapist] helps us set it up so he could, as he would slowly take a couple of steps after a week when he was comfortable with that we would move furniture a little bit more away, and when I say move furniture I am talking about 6-10 inches, it wasn't a lot." (R. 9, M, 30)

"We would put a padded ottoman a foot away from the couch, trying to get her to take that step." The mother continued to explain the progression of walking with the use of furniture manipulation as a motivator to encourage her daughter to walk further. "We got a play kitchen, we set it up 5 feet away from the couch, where she had been cruising for months. Eventually, it took about 2 months; she would then walk from the couch over to the kitchen. That was one of the first things that got her motivated to get over to where she wanted too. Then one day she just literally stood up in the middle of the living room and took a couple of steps and turned around and looked at me. I was like what did you just do?!? She has been walking ever since." (R. 10, M, 34)

"We would put him at the coffee table and he would watch TV [television] there and he would stand for an hour and a half. He would stand forever as long as he was holding something." The mother then stated, " . . . he started going from the coffee table to the couch or something like that and he would get really excited when he finally made it without falling and that started it [walking independently]." (R.13, M, 38)

"One suggestion that they [physical therapist] made which was very good, was to position some toys so many spaces apart so then she had to take one step so get to the next toy so we would do that with coffee tables, we would push them back to she would have to take that one step and then eventually two steps on her own and then further and further. That was one good suggestion that they gave." The parent then continued to state that, "she [her daughter with CHARGE syndrome] had a kitchen that we did that with, so she had to take one step to get to the kitchen. And she had a little doll station that we would position that one or two steps away from the couch as well as the coffee table so she had to move to get to it." (R. 16, M, 32)

"Around 18 months, she took her first step. We pushed two couches together and then we would try to have her go in between and then kind of move the couches further apart, it is more like an ottoman." (R. 21, M, 39)

"We also had her crib in the living room and so she actually loved practicing standing while holding onto the crib, grabbing the crib and walking by herself. She used to practice by herself, she liked to practice by herself. She pulled herself up to stand around 2 years; I am not exactly sure, but roughly 2 years." (R. 23, M, 33)

"A lot of holding onto the wall and moving around and then slowly letting go of furniture to go a couple steps . . ." (R. 1, M, 35)

New areas. In addition to the parents manipulating the furniture and toys within their household to encourage independent walking, many parents stated that they would travel to new areas within their community which motivated their child with CHARGE syndrome to move. Many parents responded that exploration and curiosity of the new environment stimulated movement. However, parents and primary caregivers stated the importance of the new environment having open areas for exploration. The following excerpts provide support:

". . . we would take her to the stores where it was bigger space and 'kinda' let her run [with the assistance of a walker] around. Our house, we have limited space for her to run and kind of work on her [pause] endurance. So we would go places to let her run. So that was at least 5 months of the gait trainer so I would say probably 8 months assisted walking." After being asked what were some things that were most helpful or contributed to your daughter walking, the mother responded with, "I think letting her just have a lot of freedom to run and explore on her own and we just 'kinda' followed her around a lot, that helped a lot." (R.1, M, 35)

"We [the parents] would take him to the mall and he would walk around with it [walker]." (R. 9, M, 30)

"His legs were very strong but he had no upper body strength. We would go to the playground and I would make him hang on the bar and it was play but he didn't realize I was making him work. I would make him do some pull ups or I would make him try to sit and I would put something underneath him." (R. 13, M, 38)

"We moved to Georgia into a bigger house, which gave her more inspiration to walk further, it [motor development] really started blossoming and that is when she became an independent walker." (R. 15 M, 44)

"I mean, we lived in a city so we would walk to different places holding hands with him. So, I mean, he was in an environment which, goes like anywhere in society in which walking is a basic mode of transportation . . ." The father continued to add, "when we do go for a lengthy walk or we walk somewhere in a destination we have to hold his hand… But if we take him to a playground, he will motivate himself to walk anywhere. He likes the climbing wall at the playground, he will climb up that. I guess that is not walking but it is connected to the family of movement." (R. 5, F, 45)

This father at the end of the interview, when asked if there is anything else that he would like the primary investigator to know, responds with, “you know I think that’s the main thing that you missed here, where just you know sometimes is just more trying to connect or explore [pause] you know if it is just taking her in a wagon or doing something else I think contributed to getting her desire up, wanting to risk it, finally finding out there are places they [children with CHARGE syndrome] want to get in the world and other people are walking there and if they I can walk there, there has some pluses for me.” (R. 6, F, 50)

“We live in Saudi Arabia so it is nice outside all year round and there is no snow so I think having access to the grass all the time has helped. We also have big, huge trampoline in the backyard and when he was crawling and starting to stand up we were on the trampoline daily. And his brother was on there to, jumping and getting him to feel the vibrations of the up and down movement. Other times it was me [mother] on with him, where he was, knowing he could stand up and move without falling down and getting hurt, I think that helped him a lot.” (R. 18, M, 36)

Peer modeling. There seemed to be a consensus between the parents of children with CHARGE syndrome that observations of a child walking as a role model had a positive impact on their child’s motor development. Parents and primary caregivers were particularly aware of role models who walked and ran in their child’s surrounding environment. For example:

As one mother explained, “. . . I noticed that kids that have siblings, they seem to walk faster than kids that don’t and she [her child with CHARGE syndrome] doesn’t have any siblings. I think that would have helped promote her walking quicker if she did. If she had a model in the house, her age.” (R. 15, M, 44)

“. . . it was just holding, holding hands forever, two hands and then one hand and [pause] finally we were just at the doctor’s office one day and she saw this little boy running around, grabbing books off a shelf and she [her child with CHARGE syndrome] just decided she was going to walk and chase after him and she just decided she was going to walk around the office and [pause] she would fall and get right back up chasing this little boy.” Then later on in the interview, the mother stated that, “I think being around other kids was a big factor for her, I guess, learning to walk, being at school, wanting to chase after kids and wanting to do things they were doing was a pretty big motivator for her. We saw a lot of progress in a lot of areas when she started school and being around other children.” (R. 1, M, 35)

“Just the fact that she was in school with other children who did walk alone may have been an encouragement to her to keep trying.” (R. 2, AM, 64)

“Then he had brothers and he just wanted to do everything that they do. So, he is being push because of them, he just wants to keep up with them.” (R. 3, M, 27)

“What motivated her was seeing other kids, I think even to this day she wants to be like everyone else, so being around other friends and seeing them walk was motivating for her.” (R. 22, M, 29)

“He does have a big brother who loves to like run around, well Phil would be in the walker, his brother would be behind it and push it really fast. To make Phil run with it [walker] he would, they would just be laughing. He loved it so.” (R. 5, F, 45)

“I think the one thing that was helpful was that he had a twin brother that was always running around so you know just to have him messing around, to see your brother walking and running around.” (R. 7, M, 42)

“We often have a household full of kids who are around 5 and 8, who are wrestling and are boisterous, he is right in there. I shouldn't say he completely keeps up endurance wise he does but he is not as fast.” (R. 18, M, 36)

This respondent provides insight into the impact of the use of video modeling, which was beneficial and contributed to her son acquiring independent walking. “Other things, I would definitely say watching other kids learning to walk. An interesting thing was the first time he actually got up off the floor by himself, standing up unassisted, was immediately after I showed him a YouTube video of a baby learning to walk and get up off the floor. There [are] a lot of points during time where I am like, oh my God that’s amazing and that was one of them. It was an immediate sort of reaction he had to that. He literally got up off the floor and he didn’t do it again for quite a few weeks, at least a month after that, he didn’t really try to do it again. He watches something, he will attempt it and then he will bank it and come back to it. That is what he’ll do with that.” (R. 19, M, 35)

“One time in point, when we took her to the nursery, she saw other children her age walking much more than the usual and so I think this encouraged her a lot. She was conscious enough to leave the walker and walk towards us.” (R. 23, M, 33)

Encouragement. The last theme associated with environmental constraints that emerged from the interviews with the participants in this investigation revolved around an encouraging environment. Evident from the following quotations, encouragement was

needed and provided to their child with CHARGE syndrome to assist in independent walking development. The encouraging environment included an increase in opportunities to practice and execute independent walking.

"We gave him lots of different opportunities to try it out. We would hold his hand and take walks on different terrains and would go up and down hills holding his hands. We did a lot of practicing until he was independent." (R. 18, M, 36)

"What things were most helpful, for him, honestly like the push walkers we had a few different types of those and just encouragement really. He had PT [physical therapy] once every other week so not a ton. So just really continuously working with him." (R. 8, M, 31)

"It is just constant encouragement to just take one step and then two steps, rather than it takes 6 weeks, it takes a year but we got there!" (R. 10, M, 34)

"We have told him you need to walk and trying to encourage him." The mother continued to state, "we work really hard with him and he still has PT at pre-school and at home once a month. We work with him daily." Furthermore, the mother provides input about being able to provide the time needed to work with their child. "I think, we [parents] were really consistent with [pause] he was our first child, so we had more time then if he would have been our second or third to be quite honest. We had all our focus on him. With every therapy we really worked hard, all the time, constantly with him to do stuff. I think that probably helped a little bit too." (R. 11, M, 37)

"I think what helped him the most was that we just kept encouraging him and working with a physical therapist and an occupational therapist. We pushed him. It was my goal, even on all his IEPs [individualized education programs] and everything is walking. With the help with the different strategies and support, he started. I can't say really one thing, just a good team effort. We have a great team. I have the dream team." (R. 14, M, 50)

"There were many factors that were playing [assisting in the attainment of independent walking] but which one was the most contributing one. I know we did all of this [physical therapy, walker, and going to the nursery to watch other children] to encourage her." (R. 23, M, 33)

"You know we had we had a good strong support system between Rachel's and my parents . . ." (R. 4, F, 45)

Summary. The parents and primary caregivers of children with CHARGE syndrome manipulated the environment to engage their children and provide additional opportunities to practice and feel a sense of success in regards to independent walking. The participants provided specific examples to increase the likelihood of walking to be a preferred mode of movement.

Task

The task of walking comprises the production and interaction of multiple body systems to attain the task (Adolph, 2008; Piek, 2002; Thelen, 1995; Thelen & Fisher, 1982). Walking requires the use of the muscular and skeletal systems to biomechanically move the body to produce movement (Shumway-Cook & Wollocott, 2001; Yaguramaki & Kimura, 2002). Parents and primary caregivers shared task constraints (i.e., affordances) that supported and aided the development of walking.

Practice. Throughout the interviews, various parents and primary caregivers explained the use of practice as an affordance. This beneficial constraint provided children with CHARGE syndrome multiple opportunities to engage in motor development, learn about their body and the surroundings, and strengthen their muscles. The following excerpts provide evidence:

When asked about how your daughter with CHARGE syndrome achieved independent walking, this mother responded with, " . . . she practiced a ton!" (R.1, M, 35)

"We have seen this in so many things in her life, it's really practice makes perfect. It is just practice takes a long time." This mother then spoke about how she would add gallon jugs of water to a push cart, to provide proprioceptive feedback back to her daughter with CHARGE syndrome so she could continue to practice walking. "As she got more practice and got more comfortable, I took some of the weight off [weighted push cart]." (R. 10, M, 34)

When asked about what were some things that were most helpful or contributed to your son acquiring independent walking, the mother responded with, “I think just practice, every day we practiced.” (R. 13, M, 38)

“So instead of doing physio-muscle [physical therapy] time once a month and [pause] we just did a lot of practice at home. Here she’s more comfortable and it was more like a play, play therapy, rather than in an office, where she didn’t really perform very well so this was a better fit for her. I think it was more the practice.” The mother explained that the aid worker would attend physical therapy twice a month and then implement those exercises and techniques with the child with CHARGE syndrome at the house. “She [daughter with CHARGE syndrome] had a developmental aid worker which came to the house every day for two hours, 5 times a week, who would hold her hand, walk around, walk outside, lots of practice with walking holding her hands. We [mother and daughter with CHARGE syndrome] did a bunch of cruising, she loved cruising, and hanging onto things. I guess the practice and the walker would be my two things.” (R. 16, M, 32)

“A lot of homework and practice and that sort of thing.” (R. 19, M, 35)

“It took a while for her to take sub sequential steps, so we practiced doing that a lot with the physical therapist and at home.” (R. 21, M, 39)

Therapy support. Respondents described various supports that promoted independent walking in their child with CHARGE syndrome. Most parents and primary caregivers spoke about the benefit of their child participating in early childhood interventions and therapies. Several participants described the benefits of physical therapy, while others did not see as large of an influence on motor development. One mother (R. 9, M, 30) stated, “I feel like in our situation, therapy didn’t really make him walk, I feel like he walked, he started walking when he was comfortable and felt safe to walk . . .” However, the majority of the parents believed that the therapy that was provided assisted their child with CHARGE syndrome in the development of walking independently. Here are some examples:

“We had an amazing physical therapist that did his [son with CHARGE syndrome] therapy for the first 3 years of his life. She was wonderful and really pushed him and tried

different chairs and she did not baby him at all. She would make him do a lot more than any other therapist would do." (R. 12, AM, 29)

"I would say, first of all early intervention. The earlier the better, even though we didn't see a lot of improvement for the first 7 months that he was doing that but it was one of the main things, getting that training into him. That was the main thing but also physio [physical] therapy and occupational therapy." (R. 19, M, 35)

"I think the physical therapy obviously played a 'humongous' role, I don't think we could have done it without that. The physical therapist was absolutely amazing and I think one really important thing with just CHARGE kids in general or my kid in general, is that you have to push them harder than you think you can because they can do it." (R. 11, M, 37)

" . . . we [the parents] got her into physical therapy, both land and warm water therapy, and she started walking I think, she was about, 18-month marker. Initially it was assisted you know we use the little straps to hold her then she eventually migrated to a walker and then to independent walking." The father then continued to speak more about physical therapy, stating, "I think the physical therapy played a big role in it, you know getting her out and having them work with her." Then later in the interview, he stated " . . . the county has a program for early childhood development, they would come in once a week and they made sure everything was going well. They would bring an occupational and physical therapist that would work with her also at our home once a week on those days." (R. 4, F, 45)

"You know I honestly think that [pause] we did hippotherapy, you know physical therapist worked with him on horseback, and I feel that was the one thing that got his trunk stronger, but honestly I just think, you know I think [pause], I think that that was a big contributing factor because he did that for all summer and then into the fall and it was a new therapy at that time and that was when he walked." (R. 7, M, 42)

"I think hippotherapy really helped a lot, if nothing else it got her over some of her phobia, in her fear of walking or losing her balance. I think that helped a lot." (R. 15, M, 44)

"Occupational therapy was able to assist her with crawling and encouraged us and taught us to do a "hand under hand" technique." (R. 2, AM, 64)

" . . . we were constantly going to OT [occupational therapy] and PT [physical therapy], three times a week to help him get to where he is now." (R. 3, M, 27)

Many of these children with CHARGE syndrome attended multiple therapies as this mother stated, "We had occupational therapy and physical therapy and orientation and mobility, we had all that." She then continued to converse on additional therapies, "we

have first steps so we have a wonderful program for early intervention. We had lots of therapist. He got physical therapy, 2 hours a week and occupational therapy was another 2 hours. . .” (R. 13, M, 38)

“Hippotherapy was probably one of the biggest factors, and the early intervention helped a lot with the physical therapy, like guiding us through the beginning and practicing.” (R. 21 M, 39)

“Oh, another thing that helped him was that he started therapeutic riding, we truly believe working on that core and sitting up on the horse really helped his balance because he wasn’t walking when we started riding and then after he started riding he started walking. …that [therapeutic riding] was a big factor, I can’t believe I forgot that. I definitely recommend the therapeutic riding.” (R. 14, M, 50)

Assistive equipment. The participants described various pieces of assistive or supportive equipment that was used to encourage movement, particularly related to walking, throughout the interviews. For instance, one mother (R. 9, M, 30) described the benefit of her son with CHARGE syndrome using a walker, “. . . I honestly feel like the walker he was in, actually got him walking sooner, even though it is against what the normal is for physical therapy.” Many participants mentioned the benefit of walkers, gait trainers, and/or the use of push toys. The parents stated that the use of assistive equipment contributed to the development of independent walking in their child with CHARGE syndrome. However, one mother (R. 20, M, 33) described her child’s struggle with the use of a walker. “He wouldn’t take a walker or any other type of aid that we were trying to give him to help him but he just started walking by himself.” She then stated that her son with CHARGE syndrome, “. . . had a normal toy walker you see at Target, he liked to walk with that. His physical therapist got him a medical walker but he would not use that at all.” It is important to note that the parents had to make adjustments to the assistive equipment to fit their child’s needs in order to provide increased

opportunities to practice walking. The following are some excerpts from parents and primary caregivers of the benefits they shared of using assistive equipment:

"I will tell you we had two different walkers. The first walker that we had was a Restin and a therapist recommended it but it was a little too safe for him, cause he just leaned on it and didn't do anything. The minute he moved two months later after we got the walker, the therapist was like, I don't think this is really working for him cause he just leaned on it and he walks with just a radio flyer wagon, better. So she switched him to a different walker, Nimbo, which doesn't have any contractions to hold him in and he loved it." The mother than provided advice for other parents of children with CHARGE syndrome, "I think that he wouldn't have walked if I would have kept him in that [Restin walker], I don't know how long it would have taken him to walk. I think that is something parents need to be very conscience of, I think they need to question and ask. The other walker was called the Nimbo. That worked great for my son but they may not work great for another kid. They [parents of children with CHARGE syndrome] just have to find what works best for their kid, but if they are not walking, maybe they actually need something with less contractions because they are like 'well this has a seat so I am just going to sit down.' Some of them will need it, it is a great walker, when you take that all off, it is really big and its really heavy, so that is why the Nimbo worked best for us because it was smaller and he could move faster. (R. 11, M, 37)

"I really feel like the walker helped him a lot. I know a lot of PTs [physical therapists] are against it because they feel like it delays walking because they become dependent on it but for him it wasn't so much dependent, like he just sat in the sling and it allowed his muscles to atrophy, he mainly just used it as a little bit of support to keep his body upright, it wasn't so much for him to sit on and rest. But I mean he did a lot of PT, I know PT strengthened his core and his hips. But I honestly feel like the walker he was using actually got him walking sooner, even though it is against what the normal is for PT." The mother provided the following additional information about the walker, "he started using a walker at 18 months, right when he just started to crawl. He was not really good at it at 18 months. He had to have a sling to hold him up because he would fall and he was weak. We would let him do it for little trial periods at the beginning. I would say by 20 months, 19, 20 months he was better. He would only do it for short periods of time. But from 21 months up until he started walking, he would fly around the house with his walker. It was just like his normal walking, he would go outside with it. We didn't have to have the sling on it anymore or any support, it was really there for his glance so he didn't fall down." (R. 9, M, 30)

"Also, the walker was helping him and all these years he had a male nurse who was really good with him and was pushing him and challenging him. He wasn't afraid to try to make him try to do it on his own, he would push him in the right way. We weren't afraid, he always said no let him do it, let him do it." (R. 17, M/F, 40)

“When Sheri first started walking, we used Walking Wings [straps] to assist her. This was really helpful because of her lack of balance and low muscle tone.” (R. 4, F, 45)

“. . . that [walker] transitioned into a weighted shopping cart which was easier for her to navigate at school.” (R. 15, M 44)

“We [parents] did get her a push toy and she used that a lot. She didn’t have a walker.” (R. 21, M, 39)

“Her first very few steps were with assistance walking behind a push toy at 23 months old.” (R. 2, AM, 64)

“We did gait training. We did a gait trainer when she started school, last December and we probably used it for about 4 months.” The mother further discussed the use of the walker, “I do think the gait trainer gave her some independence [pause] but I think also the timing we introduced it was good, cause if we would have introduced it earlier it would have become a different ‘kinda’ crutch for her versus a, you know, she was following us along trying to do things on her own at that point, it was just [pause] ‘kinda’ a step in the process then something that hindered her. I know lots of physical therapist worried that it would slow down the progress. But in her case it really helped her stand more upright and ‘kinda’ get the right muscles going . . .” (R. 1, M, 35)

“He did really well in it [gait trainer], and then the insurance decided not to pay for the gait trainer so we just got a walker. And he did amazing with that.” (R. 3, M, 27)

“Finally, what helped a lot was he got a walker. It was the walker, the one where you saw a lot at the conference, where it is open in the front and he has his hands on the sides and he walks forward [pause] so the wheels are ‘kinda’ on the sides and behind him. He loved it! He got really good at it and ran around everywhere with it.” (R. 5, F, 45)

“Walking started in a standard [walker] and then eventually she progressed to using a walker some and she ended up using that for kind of awhile and then she would go between things until eventually around a little past age 4 of when she started to walk independently.” The father also stated that “ . . . the walker felt safe . . .” for his daughter to walk through big, open areas. (R. 6, F, 50)

“. . . push walkers, we had a few different types of those . . .” (R. 8 M, 31)

“The walker, gait trainer, gave her a big boost in confidence, which she was lacking and she fell a lot so she was scared to go out on her own. So it [walker] was really good for her confidence and for building strength in her legs and endurance to walk.” (R. 16, M, 32)

". . . we [parents] bought him a walker, a Kaye walker. Bought that for him around 17 months of age, about 2 months before he started taking his first unassisted steps. He started using a walker and I do believe that was a really big part of him wanting to walk for himself." (R. 19, M, 35)

"We also used a push cart that we weighted down because she needed that feedback; push back from it so we would put a gallon jug of water in her push cart. She would love it. We had a lightweight one that a typical kid would use and that was not sturdy enough for her. That was very important for her. Even when we would play shopping cart and I taped hand and ankle weights to the bottom of it. To give it enough weight and to give her enough stability and push back for her to feel comfortable pushing it." (R. 10, M, 34)

"I would say a walker, that helped a ton. She used one of those jumpers to get strength in her legs. As well as, just any sort of walking tool that you hold onto [pause] toys with wheels around but with support, that helped. She couldn't just do it on her own or she would fall so you have to hold the toy so she could roll it." (R.22, M, 29)

Fall safely. Ultimately the level of safety was a concern for many of these parents and primary caregivers of children with CHARGE syndrome. The participants spoke about teaching their son or daughter with CHARGE syndrome how to fall safely to the ground when trying to stand and walk. This instruction that parents provided on how to fall safely coincided with an increase in confidence and a decrease in fear and being afraid to attempt to walk. The quotations below describe the parents' experience teaching their son or daughter how to fall and the benefits their child gained:

"And that is probably how she is different from another kid cause when she would fall at first, she would fall like a rock because she couldn't brace herself or handle it. And her ability to fall better probably coincided with pretty closely of when she started walking. Because she could do a controlled fall compared to just a crash." (R. 6, F, 50)

" . . . he [son with CHARGE syndrome] had a lack of saving mechanism, responses when you put your hands out when you are falling; saving reflexes. He didn't have them automatically; we had to teach him that." (R. 19, M, 35)

"… I taught him how to fall. Every day I would come home from work and I would do 20 reps [repetitions] of making him fall so I would hold his hands and then I would make

him fall forwards so he learns to put his hands out or make him fall to his bum, left, right, backwards, forewords, until every day I did that at least 20 times." (R. 13, M, 38)

"I think as far as the independent walking, one of the good things we did was to teach her [daughter with CHARGE syndrome] how to catch herself when she fell. Because by the time she was doing assisted walking she was old enough to build up a pretty healthy fear of falling down and getting hurt which was holding her up from walking independently. So we worked on teaching her how to catch herself with her hands when she fell down so she wouldn't be quite as scared." (R. 10, M, 34)

" . . . I [mother] don't blame the therapist at all but she liked to play it safe. She was like I don't want him to fall over or hit his head. We talked about even getting a helmet because he's falling and crashing and hit[ting] his head. When we switched therapist, she was like I am going to teach him how to fall and how to catch himself . . ." (R. 11, M, 37)

Summary. Throughout the interviews, parents and primary caregivers of children with CHARGE syndrome shared experiences and perceived factors that increased their child's ability to attain independent walking. Through the qualitative analysis, themes emerged from individual, environmental, and task affordances. The excerpts provided by the parents and primary caregivers revealed the overlap and influence the individual, environmental, and task affordances had on their child's attainment of walking. Many participants provided insight about their trial and error approaches with assisting their son or daughter to achieve independent walking. The parents or caregivers manipulated the environment and provided assistive equipment to support the development of independent walking.

RQ4: Cross Case Analysis of Rate Limiter Themes

Based on the responses from participants a variety of themes emerged, which seemed related to causing a delay in their child developing the independent walking motor milestone. The following are identified themes associated with the research question and

dynamic systems theory (DST). The themes are divided into three sections: individual, environmental, and task. Nvivo was used to determine a word frequency based on the responses of the parents and primary caregivers on potential rate limiters (see Figure 11). Excerpts from the participants' transcriptions are provided, which support the themes and results.

Figure 11. Rate limiters word frequency of the 100 most frequent, 4 letter minimum, words from participant transcriptions.

Individual

There was a consensus between all participants that the complexity of CHARGE syndrome had an impact on motor development. This overall theme related directly with the DST. Most participants spoke about the characteristics associated with CHARGE syndrome as possible contributing factors that caused their child to be delayed in motor milestones, especially the motor milestone of walking. When asked, "What were some roadblocks or difficulties that delayed your son in walking independently?" one mother

(R. 9, M, 30) answered, “definitely [pause] he is missing his semicircular canals. His balance and his ears [vestibular system], I would say would be the biggest thing that caused him a delay in walking, sitting, and crawling. Probably after that low muscle tone. With the balance comes fear so that kind of covered a lot of the stuff; the fear of falling. I would say balance and fear for sure.” Many of the parents and primary caregivers responded with multiple rate limiters associated with characteristics of CHARGE syndrome, which will be presented throughout the major themes.

Another father (R. 6, F, 50) answered, “I think general weakness, the lack of vestibular system, lack of vision, and lack of hearing, you know just, physically took a toll . . .” Therefore, some of the responses contained multiple individual rate limiters pertaining to their child with CHARGE syndrome. The major themes that emerged from the participant’s responses are as follows:

Fear and feelings of being afraid. The respondents expressed that their child with CHARGE syndrome felt fearful or afraid to walk independently. This feeling acted as a rate limiter, which decreased a child’s motivation to attempt to walk independently. The following quotes provide support and evidence:

> “We [parents] tried to encourage her to leave the walker and come but she was very afraid.” The mother continued to explain how fear was the most impactful variable. “Mainly her fear, because at first she was very afraid. I think she was afraid because of her vestibular system because things weren’t really stable for her. We knew at a certain stage that her muscles were strong enough to let her walk but she was afraid to come toward us without holding on. The reason, I think it may have been because her vestibular systems wasn’t well developed and she was always afraid to fall down.” (R. 23, M, 33)

"Because by the time she was doing assisted walking, she was old enough to build up a pretty healthy fear of falling down and getting hurt which was holding her up from walking independently." (R. 10, M, 34)

"Essentially his balance. He would fall, he would hit his head multiple times a day. He was falling backward, he was falling forwards. He just didn't know how to brace himself so he would completely fall on his head. He wouldn't walk because he was afraid that he was going to fall." (R. 3, M, 27)

". . . she hasn't quite a bit of vision to use as well, so I think it is a combination of fear and the walker felt safe as well." (R. 6, F, 50)

"I feel like he could have started walking well before 29 months but I just don't think he felt safe doing it. I think he was afraid of getting hurt if he fell, he was very cautious." (R. 9, M, 30)

Low muscle tone. One characteristic associated with CHARGE syndrome is low muscle tone. The muscular system must be at a strong enough point to support the body to stand and walk. The respondents explained the impact of low muscle tone on walking in the following excerpts:

" . . . her low muscle tone could be a factor . . ." The mother then provided additional information, "she [her daughter with CHARGE syndrome] has low muscle tone, even now, though it has improved over time." (R. 2, AM, 64)

"Her muscle tone. She has really poor muscle tone, especially core muscle tone. I think that was probably the biggest setback." The father stated "initially she had really low muscle tone, so we got her into physical therapy . . ." (R. 4, F, 45)

"She has some low muscle tone but it was more of a coordination slash bravery thing for her." (R. 10, M, 34)

"He has low muscle tone, he has a problem with his hips too." (R. 17, M/F, 40)

"Alright, so basically it was very difficult for her [to attain independent walking], low muscle tone, low very dysfunctional vestibular system, low balance." (R. 6, F, 50)

"He [has] low muscle tone, so he didn't do the normal progression . . ." (R. 14, M, 50)

“She is getting better every day and stronger but she still has low muscle tone and lacks her balance and is exhausted by the end of the day.” (R. 15, M, 44)

Ear and vestibular malformations. The ‘E’ in CHARGE syndrome represents ear abnormalities and deafness (Pagon et al., 1981). Ear malformations have the potential to impact the vestibular system (i.e., utricle, saccule, semicircular canals, VII cranial nerve), which can act as a rate limiter. The following excerpts provide evidence on the impact of ear malformation on walking:

“We don’t know what kind of vestibular input she is getting, it’s not much of any.” The mother also reported “she was born with no inner circular canals to give her balance.” (R. 1, M, 35)

“We use[d] to think he was low tone when he was younger, in infancy, as he sort of developed, I think it is more to do with his lack of coordination than tone. It was sort of like you could tell the difference for a long time. I don’t think it was low tone at all when I look back at now, I just think it was that he didn’t have that vestibular input and didn’t know how to push himself.” The mother responded during the interview with more information about the impact of his vestibular input; “he doesn’t have any semi-circular canals, they are completely absent so he has no vestibular input I suppose. In his ability to walk, for him, he had to rely on vision and proprioceptive alone.” (R. 19, M, 35)

“. . . he definitely [has] serious malformations with them [ears] in his cochlea and vestibular function. So he, I think, so his result [is] he has terrible sense of balance. That’s the primary thing here . . .” (R. 5, F, 45)

“I think his semicircular canals because they are small, that give him the balance issues and he has to figure that out.” (R. 11, M, 37)

“Oh, definitely, the balance issues, when you don’t have the equipment [semicircular canals] to have balance, I think it was a cause for delay [of walking].” The mother stated that, “he has absence of semicircular canals in both ears and only 1 and 1/2 turns of the cochlea compared to the usual 2 and 1/2.” She then made a reflective statement “the fact that he has [absent] semi-circular canals and balance issues, the fact that he is walking is quite a miracle.” (R. 14, M, 50)

“His semicircular canals aren’t formed appropriately so how his doctor explained it to me was it was like being dizzy, like you are on a merry go round like 20 hours a day.” (R. 7, M, 42)

Low vision. Although vision loss caused by colobomas is present in many children with CHARGE syndrome, the majority of parents and primary caregivers did not reference vision as major rate limiter throughout the interview. Rather, vision loss was mentioned in combination with other factors associated with CHARGE syndrome. Below are two responses from mothers, who explained in more depth the impact vision had on motor development.

"I think mostly it's been, she doesn't see, she is usually looking up or down, it is hard to get her to notice things on the ground when she is looking forward and so forth so the vision is one of the big factors." The mother explained that her daughter, " . . . tracts slowly with her vision so if something was coming at her or moving to her it throws her off her balance." The mother further explained the impact of her vision loss, " . . . with the limited vision, without the environmental hearing, she was kind of stuck in a little bubble and only curious about what was going on left and right with her vision" (R. 1, M, 35)

" . . . for vision perception he has to touch the ground to know where he is, he has to put his hands down. Some of the things, he doesn't have terrible vision, but he can't see a lot and I think we take for granted that some of the things he does is actually a vision issue." (R. 12, AM, 29)

Difficulty balancing. Coinciding with the DST, various systems (i.e., vision, vestibular, muscular), which have already been mentioned by the participants, would have an influence and impact on balance. Therefore, many of the parents or primary caregivers of children with CHARGE syndrome found it difficult to acknowledge one factor as a rate limiter, rather they suggested the combination of rate limiting factors impact on the attainment of independent walking. Evidence is provided in the following quotations:

"She was just about to turn 4 when she started walking, she has CHARGE syndrome obviously and severe balance issues which made it really hard for her to achieve that

milestone." The mother stated her daughter with CHARGE syndrome, ". . . knew she was unsteady and I think that delayed her unease with her balance, maybe kind of kept her from walking sooner." (R. 15, M, 44)

"The balance, the semi-circular canals we were told were not formed correctly so I think his balance is off and still even now when he walks, he digs his toes in as if he is still needing that extra little support." (R. 18, M, 36)

"He has [pause] he was born with very low muscle tone. How much of a role that played compared to how much the lack of balance and vestibular system played a role, I am not sure. I am sure it is all connected but our sense was that it was more about the sense of balance than the strength and muscle tone." (R. 5, F, 45)

"His balance and coordination is horrible, it's gotten better and he has been more independently walking but it is still pretty bad." (R. 3, M, 27)

"Her balance is somewhat off and she took some bad falls so after that her confidence went down. She ended up with a concussion at one point. So she stopped walking for a couple of weeks after that." (R. 16, M, 32)

"I think balance was his number one challenge." The mother stated the timing of walking was based on his ability to balance. "And also, I think he walked when he was ready, when his balance system was ready. You know, he definitely, he was still pretty physically strong, his muscle were strong, but balance was still very challenged." To conclude the mother reiterated the point " . . . just balance and you know using your balance system to kind of coordinate his limbs and all that was, I think of, was his biggest challenge." (R. 7, M, 42)

"Balance is a huge issue, [pause] even for him now at age 4. So just trying to stand independently fighting the whole balance issue was a challenge. That was pretty much the main only one [factor which caused a delay in walking], I think." The mother then responded with, "other than the balance, I think that was really the main issue for him, once he kind of let go of the push walkers it was just trying to navigate while being stable." (R. 8, M, 31)

"Probably about 6 months [of assisted walking], because I kept thinking oh, she is going to be walking anytime but then it never happened. It was the longest waiting period. But I think she had to get her balance and figure that out before she was able to really learn to walk." (R. 22, M, 29)

Tactile aversion. Although not a major theme, it is important to recognize that individuals working on motor development with children with CHARGE syndrome may

need to understand the child's level of comfort with touch. Two parents provided input and identified the sensitivity of touch as a potential rate limiter. One mother (R. 2, AM, 64) explained that her daughter with CHARGE syndrome, " . . . did not like having anyone touch her hands, making it more difficult to help her in her journey . . ." Another mother (R. 19, M, 35) stated " . . . for about the first 10 months of his life he didn't move at all. He didn't roll over, he didn't want to be picked up even, it was, he was very sensitive about being touched, he didn't want to move." The identification of tactile aversion as a rate limiter may impact how service providers as well as parents are able to interact and instruct children with CHARGE syndrome related to motor development.

Summary. Prevalent throughout the interviews with the parents and primary caregivers was the interconnectedness of the body's subsystems, impact on motor development. A mother (R. 13, M, 38) provided this example of the impact of various rate limiters on walking. "The balance, his ability to hold his head up, being able to look down at anything that was in front of him was hard because anytime he would put his head down he would fall or it was called the drunk walk where then he couldn't look back up because then he would lose his balance and get frustrated. I think that is basically it, the vision, the hearing, and the balance all put together took away from his ability to start walking." A different mother (R. 22, M, 39) spoke during the interview about the impact of various rate limiters on learning. "My personal opinion is that with the hearing, the vision loss, and all those things, I kind of learned that kids with multiple disabilities they kind of focus on one thing as a time, so for her maybe walking was not important for her at a certain time because maybe she was working on speech or that kind of stuff.

Once she accomplished, I can't think of what she was working on before that, but once she accomplished that, then she focused on walking." It is the combined impact of various rate limiters that caused a delay in independent walking for children with CHARGE syndrome.

Environmental

Changes to the environment can alter the performance of a particular movement task. Parents and primary caregivers provided insight on environmental variables, which caused a developmental delay of the task of independent walking in their child with CHARGE syndrome. The following were emerging themes perceived by the respondents, which included the impact of uneven surfaces, inadequate space, hospitalizations, and surgeries. Each theme is supported by direct quotes from parents and primary caregivers of children with CHARGE syndrome.

Difficulty walking on uneven surfaces. The skill and performance level of walking was altered based on variations of surfaces for children with CHARGE syndrome. Parents identified uneven surfaces as a rate limiter, which hindered the child's ability to walk independently. Difficulties walking on uneven or non-level surfaces remained, even after some of the children with CHARGE syndrome acquired the ability to walk independently. A confirmation is presented in the following excerpts:

"He is still very unstable at times, when the surface changes he may fall or if the sun gets in his eyes when we are outside he doesn't do well in those type of environments, if the ground is un[even] he doesn't do well with that either." (R. 9, M, 30)

"She doesn't do well on any uneven ground. She still has some stability issues because she has low vision and is blind [pause] so that's a factor." (R. 1, M, 35)

“She still has a hard time on uneven surfaces anywhere with a threshold until she gets comfortable with it and then she is totally fine.” (R. 10, M, 34)

“… around 3 years old she was still doing a lot of face plants, especially walking on uneven surfaces.” (R. 21, M, 39)

“Outside, uneven grass and ground even after he started walking that was always a challenge to get him to walk outside and anywhere that wasn't completely flat.” (R. 8, M, 31)

Inadequate space. Parents and primary caregivers described space as a potential rate limiter. Inadequate space hindered the development of walking by restricting the child’s incentive to move and ability to explore through movement. The following are supportive participant responses:

“Just kind of space to walk around in, she didn’t have a whole lot of area that was a flat surface for her to get around so she was just stuck to one little spot. And outside it was very difficult for her to get up by herself so she didn’t really attempt it unless someone was holding her hand.” (R. 16, M, 32)

“From an exploring space thing and or even [pause] she is really, if she goes into a big open area she will sit down because it is too scary from a navigation stand point.” The father then indicated when his daughter with CHARGE syndrome, “. . . gets to wide open spaces she would get nervous and [sit] down.” He continued to explain, “. . . I think there was a point where the world was still that scary that she didn’t have much desire to leave her immediate bubble.”(R. 6, F, 50)

“I think at first, he was quite scared of walking outside, being in the house was different, but when he was outside and walking [on] grass, he had all sort of sensory issues that was a challenge for him.” The mother spoke about her new house which had an “ . . . open area without a lot of contrast between the wall color and the floor color that may not have helped him.” (R. 19, M, 35)

“The main room was very small and so for her [daughter with CHARGE syndrome] it was like 10 steps across the floor is all she could get so there was room limitations.” (R. 15, M, 44)

Hospitalization and surgeries. Research by Salem-Hartshorne (2003) and Salem-Hartshorne and Jacob (2004) on individuals with CHARGE syndrome indicated that

hospitalization had a negative impact on development. Participants in this investigation also shared the negative influence of hospitalization and surgeries on walking. In addition, some parents identified specific medical equipment as a rate limiter, due to the restriction it caused on his or her child with CHARGE syndrome ability to move freely within a specific environment. In the following quotations, participants describe the impact of hospitalizations and surgeries on the development of walking.

"Hospitalization and surgeries were huge. We spent nearly the first year in the hospital." Hospitalization varies for each child with CHARGE syndrome; this child, ". . . spent the first 10 months in the hospital, and he had 15 surgeries within the first 10 months, so we were, yeah, he spent the first 11 weeks in the NICU, and then another 2 months after his trach [tracheostomy] surgery in the hospital and then we spent another 2 months in the fall and another 2 months after that." The mother indicated her son with CHARGE syndrome had to be fed by a feeding pump in which nutrients are transferred through either a button or a tube (e.g., percutaneous endoscopic gastrostomy, gastrostomy, nasogastric tube) into the child's stomach. "The fact that he is attached to his food for so many hours [22 hours] a day, that's a big obstacle for him . . . just being in the hospital and going through as much as he did for the first two years really held him back." The mother continued to state, "I think he would have been walking sooner if he hadn't been connected to his GI [gastrointestinal] pump for you know, for 22 hours a day. So that is a big issue for him because if he only has so much line for him to be able to walk so one of the hard things for him was to figure out how to walk around with his cords and IV [intravenous therapy]." (R. 3, M, 27)

"I just think health concerns for the first year of life for all kids with CHARGE really pushes them behind." The mother provided information related to the impact of surgeries and hospitalizations on her daughter with CHARGE syndrome. "She was born, in the NICU [Neonatal Intensive Care Unit] for 6 months for 4 major surgeries, heart surgery, she has a trach [tracheostomy], feeding tube, [and] swallowing difficulties. So when she came home at 6 months, she was basically a newborn and the doctors had kind of said when we do any evaluations to subtract 6 months off of her life because she came home as a newborn, she was going to be delayed. We knew that. It all started, she is on a ventilator, she is on a ventilator for 24 hours a day, until she was about a year and a half and then slowly started to wean her off. When she started to get weaned off and completely weaned off, that was when she started crawling, she wouldn't even do tummy time when she was on the vent. I think it was just too much equipment." (R. 22, M, 29)

“From birth to the time Trish walked alone, she had 19 surgeries including three open heart surgeries, pacemaker surgery, two cochlear implants, cleft lip and cleft palate repairs, tonsils and adenoids removed, and other less serious surgeries.” (R. 2, AM, 64)

“He was in the hospital for a while just lying down so he didn’t have a chance to move around like other children. He was hospitalized so he wasn’t moving and he was connected to many machines. He was also very fragile and he didn’t have a lot of physical contact. At that point when he was very little, he was stuck in the hospital and he was taking a lot of medication. He was also under a lot of sedation and the doctor was saying to us that it would take a long time to get all the medicine out of his body. I think the recovery from the hospitalization; he was so behind cause of that.” (R. 17, M/F, 40)

“I think definitely some of her feeding issues gave her a roadblock because she was on a feeding tube so for some time she couldn't really be mobile, or as mobile as she wanted to be. Since she was connect or she would wear a backpack which would weigh her down because she was so small when she was doing the feeding, that was a little bit I think of a roadblock. And also she basically threw up daily for like 3 1/2 years so I am sure, I am sure that delayed her globally. That pulled her attention and energy.” (R. 10, M, 34)

“I think because he has CHARGE, I think one of the biggest things was that he was in the hospital for the first 7 months of his life. There wasn’t a lot we could do leg exercises but there wasn’t a lot of the rolling over either because of all the stuff he was hooked up to. He has had four heart surgeries and you wouldn’t want to lie on your chest after that. I think that was one of the biggest things.” (R.11, M, 37)

Summary. Understanding the impact of environmental constraints can provide valuable information in regards to adaptations needed to increase practice opportunities and feelings of success in children with CHARGE syndrome. Since many children with CHARGE syndrome spend a majority of their first year in hospitals, unable to move freely, it is imperative that environments are designed and manipulated to promote movement, not postpone the development any longer. Providing adequate space in an area with level surfaces can increase the potential for motor development to occur. In addition, task performance (i.e., walking) was impacted due to equipment (i.e., ventilator, feeding machine) constraints.

Task

The demands of the task of walking interact with the individual and the environmental factors present at a specific point in time. Constraints within the task "not only interact but have the potential for modifying and being modified" (Gallahue et al., 2012, p. 29). The parents and primary caregivers of children indicated changes in movement patterns with CHARGE syndrome as perceived rate limiters. The goal of walking is to be in an upright position where hands are free and movement is performed at a quicker rate than crawling (Adolph et al., 2012; Adolph & Tamis-LeMonda, 2014). However, for children with CHARGE syndrome, walking may not always be the task solution based on the interaction and influence of other constraints.

Head down and 5-point crawl. The attainment of certain movements themselves can act as rate limiters (Haywood & Getchell, 2009). In children with CHARGE syndrome it is common for them to exhibit back scooting and a 5-point crawl position with their head on the ground as they are developing (Hefner, 2008; Hefner & Davenport, 2002). One mother (R. 11, M, 37) during the interview actually stated, "I had heard at the conference [International CHARGE Syndrome Conference] that if they scooted on their back that they wouldn't walk until later. But he didn't [delay walking]." A different parent mentioned that since her son performed the 5-point crawl that he would be delayed (see R. 14, M, 50 quote). Therefore, the parents of children with CHARGE syndrome acknowledged that certain movements acted as rate limiters.

". . . he [son with CHARGE syndrome] was slow in rolling over but he sat up at a really early age and then he started to roll to get places [pause] then he would scoot on his butt then he turned to 'bulldozing', like the 5-point crawl, and head down and crawling that

way everywhere. We knew he would be delayed walking since he was a bulldozer with his head down." (R. 14, M, 50)

"He [son with CHARGE syndrome] would be in the 5-point crawl position but he didn't move in that position. He liked to be in that position, he liked to get that feedback on his head, deep pressure on his head, looking like he was about to do a tumble. He liked to do that a lot." (R. 19, M, 35)

Difficulty standing independently. Gallahue, Ozmun, & Goodway stated, "the infant must first be able to control the body in a standing position before tackling the dynamic postural shifts required of upright locomotion" (2012, p. 144). However, a theme that was present in the transcriptions of parents and primary caregivers of children with CHARGE syndrome was the inability to stand still, even after achieving independent walking.

"He learned to stand still after he learned to walk." (R. 9, M, 30)

"He can't stand up by himself; he can only pull himself up. If he stops walking he just falls gently to the ground. When we are out and about he never stops walking." (R. 20, M, 33)

"Another big thing that we had to work on, when he first learned how to walk, he would walk but he couldn't stop and stand still. I didn't realize that stopping and standing still was actually really hard work. People use to think he was so 'hyper' because he was going back and forth but I was like he can't stop, cause if he stops he is going to fall unless he can grab onto something, he is going to fall. And he can't just stand still. Now he can stand still and he is fine with that but that is something we had to work really, really hard on." The mother later in the interview restated, "no, he walked first and he could ran and walk before he could stand still. It seems opposite right, we were always working on it but he just took off. He could stop with his walker but he had handles. He really navigated that thing. We worked on it, when he got going he could stop if he held onto something but if it was in the middle of space he had a hard time. We worked on it and now it isn't even a problem." (R. 11, M, 37)

Wide and wobbly gait. In the context of the DST, optimal task patterns and movement capabilities of an individual are determined by the interaction of constraints

(Caldwell & Clark, 1990; Clark, 1995). If children with CHARGE syndrome are instable in the movement task of walking, then the pattern of the task will be adjusted to reach a more stable attractor state, such as a wide stance. The following are excerpts from parents and primary caregivers of children with CHARGE syndrome.

"He is still rather wobbly and has a wide gait. I don't know how many times he has been stitched up for running into things or just losing his balance. You can tell when he is overly tired, because he is more wobbly and falls down more." (R. 18, M, 36)

"He definitely still has a wide gait to his walk but he can be very independent with his walk. If he gets tired or gets going really fast he will still drop to his knees and do a hand and knee crawl." (R. 12, AM, 29)

"Very wide stance at first and wobbling from side to side." (R. 19, M, 35)

"She was very wobbly and unconfident but she was definitely standing and cruising a lot earlier than she was walking." (R. 15, M, 44)

"And she walks with a wide stance and pretty wobbly, kind of a drunken sailor walk a little bit." (R. 6, F, 50)

"When he did start walking, he was very wobbly of course, walking independently. He did walk with a walker before. He was determined but really wobbling." (R. 9, M, 30)

Summary. Movement solutions and behaviors are ever changing and evolve over time. In addition, movement variability is part of the learning process to gain stability in functional movement patterns (Newell, Liu, & Mayer-Kress, 2001). When rate limiters impose on an individual the stability of the movement will change. Rate limiters increase the individual's subsystems stability. For children with CHARGE syndrome, within the context of the DST, walking occurs as a functional movement pattern when the movement pattern gains increased stability and less variability of the movement pattern. However, as the individual gains experience, an awareness of his or her body, and

perceptual cues; the optimal movement pattern to accomplish the goal will be performed (Shumway-Cook & Woollacott, 2007). Affordances promote and encourage developmental change to occur from one movement to a new movement.

Conclusion

Based on the present investigation, major themes pertaining to affordances and rate limiters in children with CHARGE syndrome are a result of parents' perspectives (see Table 14). Parents offered valuable information about variables that hindered and enabled their child with CHARGE syndrome to acquire independent walking. Many of the parents' concerns were based on interacting constraints having an impact on their child's motor development. This evidence supports the DST that both the individual and the environment can impact the task of walking. In addition, task constraints may also hinder or enable the child's abilities to attain independent walking.

Table 14

Qualitative Evidence of Affordances and Rate Limiters

Constraints	Individual	Environmental	Task
Affordances	Determined personality trait Comfort and safety Confidence	Furniture Open and flat areas Peer modeling Encouragement	Practice Therapy support Assistive equipment Fall safely
Rate Limiters	Fear or afraid Low muscle tone Ear or vestibular malformations Low vision Balance Tactile aversion	Uneven surfaces Inadequate space Surgeries and hospitalizations	5-point crawl Stand independently Wide and wobbly gait

Methodological Triangulation

Methodological triangulation was used to gain rich and comprehensive analysis of the data (Denzin 1970, 1978). The results from Phase I (i.e., quantitative) and Phase II (i.e., qualitative) were used to formulate additional information of the impact of rate limiters and affordances on the attainment of independent walking in children with CHARGE syndrome. The deepening and widening of one's understanding is the results of the use of methodological triangulation. The nature of the DST and human interaction, particularly within the context of CHARGE syndrome, is complex due to the multitude of characteristics, which influence the individual's intention and interaction with the environment and task. Therefore, quantitative and qualitative methods were used in this investigation.

The complexity of CHARGE syndrome dictates the use of a mixed method research design to seek in depth understanding about the potential of significant variables. Based on the results of the quantitative analysis, it can be concluded that the loss of vision and hearing has a negative impact on the age of independent walking in children with CHARGE syndrome. Furthermore, the impact of hospitalization also can be seen as a potential rate limiter to the development of walking. However, many additional variables may also be significant rate limiters and affordances pertaining to children with CHARGE syndrome and motor development.

With the results of the qualitative analysis, participants have provided a deeper understanding of the impact of vision and hearing loss and provided valuable additional information that was not obtained with the quantitative analysis. Parents of children with

CHARGE syndrome provided support and validation of the results presented in Phase I. One mother (R. 1, M, 35) indicated the lack of motivation to walk " . . . with the limited vision, without the environmental hearing, she was kind of stuck in a little bubble and only curious about what was going on left and right with her vision." This information provides additional insight about the impact of vision and hearing loss on the task of walking.

In addition, throughout the interviews, the parents reported on additional rate limiters that provide more information affecting motor development. Parents reported on the lack of vestibular input causing their child with CHARGE syndrome to have balance issues and walk with a wide and wobbly gait. In addition, children with CHARGE may be afraid or have a fear of walking due to the chance of falling and getting hurt. The interaction of the individual and the environment impact the task of walking, which is supported by the DST and by responses of the parents. Additional affordances and rate limiters were revealed with the use of methodological triangulation, which improved the current knowledge, and understanding of the results of this investigation.

Summary

To summarize, quantitative and qualitative mixed methods were analyzed within this chapter to closely examine factors and characteristics that contributed (i.e., affordances) or delayed (i.e., rate limiters) the attainment of the independent walking developmental milestone in children with CHARGE syndrome.

Chapter IV provided the results of this mixed method investigation. In the subsequent chapter, a synthesized discussion of the results will be presented in

comparison to related literature from previous research. This may provide additional insights into the impact of affordances and rate limiters on the attainment of independent walking in children with CHARGE syndrome. The discussion will also focus on the concepts and context of the DST based on the results provided. The implications of the findings, research limitations, and conclusions will also be included in Chapter V.

CHAPTER V

DISCUSSION

The impact of individual rate limiters and affordances on acquiring the independent walking developmental milestone in children with CHARGE (coloboma of the eye, heart defects, atresia of the choanae, retardation of growth and/or development, genital and/or urinary abnormalities, and ear abnormalities and deafness) syndrome was examined in the present investigation. Based on the dynamic systems theory (DST; Kugler et al., 1982; Thelen, 1995, 1998; Thelen et al., 1987), movements occur in response to a dynamic interaction of the individual, environmental, and task constraints. Rather than movements always being produced by a central command center (i.e., central nervous system, brain), a movement can emerge based on the self-organization of the body's systems and subsystems based on ever changing constraints. Constraints are defined as boundaries that impact movement capabilities of an individual (Clark, 1995; Newell, 1986).

Constraints consist of the individual, environmental, or task variables that can be either limiting or enabling (Thelen et al.,1989). Individual constraints comprise of biological and functional characteristics (i.e., body attributes, personality characteristics, perceptual skills). Environmental constraints include both the physical environment (i.e., surface, light, gravity) and social environmental factors (i.e., expectations, family). Newell (1986) proposed three categories of task constraints: (a) goal, related to the

product or outcome of the movement task; (b) rules, governed by a specific sport skill or optimal pattern of coordination; and (c) implements or machines.

Currently, it is unknown what specific constraints impact the delay in motor development in children with CHARGE syndrome. Vision, hearing, somatosensory, and vestibular systems all impact the ability of a child to balance, maintain postural control, and perform motor actions (e.g., walking; Shumway-Cook & Woolacott, 2001). It is common that all these systems are impacted in some combination in children with CHARGE syndrome (Hartshorne et al., 2011; Salem-Hartshorne & Jacob, 2004).

In order to support motor development, it is essential to know which systems impact movement and how the other systems function to allow movement to occur in children with CHARGE syndrome. There is some evidence in the literature that vision, hearing (e.g., specifically vestibular dysfunction), muscle weakness, and long hospitalization stays impact motor development, particularly independent walking, in children with CHARGE syndrome (Abadie et al., 2000, Admiraal & Huygen, 1997; Haibach & Lieberman, 2013; Murofushi et al., 1997; Salem-Hartshorne & Jacob, 2004).

The present investigation was designed and conducted to examine the impact of rate limiters and affordances on acquiring the independent walking developmental milestone in children with CHARGE syndrome. Children with CHARGE syndrome have a multiple number of anomalies and the severity of involvement varies greatly, leading to a heterogeneous group, which is difficult to measure due to the combination of impairments.

Therefore, methodological triangulation was conducted in the current investigation, based on quantitative and qualitative methods. Data from this present investigation were collected and analyzed using a mixed method approach. Further, a discussion of the results was presented in two phases. The data were synthesized and the following discussion is presented.

Provided in this chapter is a discussion related to the findings based on the research questions of this investigation:

Phase I: CSCDP children with CHARGE syndrome; rate limiters and affordances

1. What is the significant impact of vision and hearing loss as rate limiters in children with CHARGE syndrome on their attainment of independent walking while statistically controlling for the months in which the child with CHARGE syndrome was hospitalized?
2. What are additional significant rate limiters or affordance variables that are related to the age at which independent walking in children with CHARGE syndrome is attained?

Phase II: Parent or primary caregiver perspective of their child with CHARGE syndrome; rate limiters and affordances

3. What do parents or primary caregivers propose are affordances that lead their child with CHARGE syndrome toward the ability to walk independently?
4. What do parents or primary caregivers propose are rate limiters causing a delay in walking for their child with CHARGE syndrome?

This chapter consists of five sections: (a) Discussion, (b) Implications of the Findings, (c) Limitations, (d) Conclusion, and (e) Recommendations.

Discussion

The following is a discussion related to the findings based on the purposes of this investigation. The discussion consists of three sections: (a) Interaction Between the Individual and Task; (b) Interaction Between the Environment and Task; and (c) Task of Walking Independently. Throughout the discussion, elements of the DST are infused relative to the findings.

Interaction Between the Individual and Task

The interaction concept of the DST implies that individual constraints impact the task of independent walking (Kugler et al., 1982; Smith & Thelen, 1993; Thelen & Ulrich, 1991). In this investigation, the process of attaining independent walking was in part negatively impacted by the child's vision and hearing which acted as rate limiters (Thelen, 1995, 1998).

Impact of vision and hearing loss. Based on the quantitative findings of this investigation, there were significant differences between the attainment age of independent walking and the severity of vision and hearing loss in the children with CHARGE syndrome. Qualitative data were used to support these findings; parents of children with CHARGE syndrome indicated that vision and hearing loss hindered their development of independent walking. Based on the qualitative findings, parents or primary caregivers were aware of the impact their child's vision and hearing loss had on his or her motor development, particularly related to attainment of independent walking.

In addition, the acuity level of the right eye and the decibel loss of the left ear were significant predictor variables on the age of independent walking. Therefore, a decrease in visual acuity in the right eye or an increase in the decibel loss in the left ear would significantly increase the attainment age of independent walking in children with CHARGE syndrome, further delaying them from being able to walk independently. This is a very unique finding, since there are no known researchers who have indicated the negative impact of vision or hearing loss in either the left or right side on the developmental motor milestone of independent walking.

Furthermore, another interesting finding of this present investigation was that when the vision and hearing level groups were compared on the attainment age of independent walking, the group that had the lowest mean value of attainment age of independent walking (e.g., began walking at the youngest age) was the group with significant vision loss and near normal hearing. This is most likely due to the low sample size in this group (*n* = 5*)* compared to the other three groups. It was postulated that the group with near normal vision and hearing levels would have had the lowest attainment age of independent walking. However, there was no significant difference between the group with significant vision loss and near normal hearing when compared to the group with near normal vision and hearing or the group with near normal vision and significant hearing loss. Nonetheless, the group with significant vision and hearing loss was significantly more delayed then the other three groups. These results reiterate the impact of vision and hearing as rate limiting constraints on the attainment of independent walking in children with CHARGE syndrome (Thelen, 1995, 1998).

These findings support the literature and results of previous research related to the impact of vision and hearing loss on motor development and gross motor performance. Researchers, through empirical studies, have supported in theory that vision loss delays in the acquisition of gross motor skills, including independent walking (Adelson & Fraiberg, 1974; Bouchard & Tétreault, 2000, Brambring, 2006, 2007; Celeste, 2002; Ferrell, 2000; Fraiberg, 1977; Hatton et al., 1997; Jan et al.,1977; Kastein et al., 1980; Norris et al., 1957; Pereira, 1990; Tröester & Brambring, 1993; Tröester et al., 1994). Furthermore, the findings of the present investigation support the results of Haibach et al., (2014), who reported that the increase in severity level of vision loss negatively impacted the gross motor performance of children with vision loss. Similar to the results of this investigation, the group with the most vision loss performed significantly lower in the gross motor skills assessed when compared to the two other groups who had less vision loss. Additionally, Celeste (2002) reported similar results that indicated children with the least vision had the poorest gross motor outcomes related to walking independently.

Furthermore, the findings of Bouchard and Tétreault (2000) and Brambring (2006), who examined the motor development (i.e., attainment of walking) of children with vision loss compared to children with vision, supported the results of this present investigation. The results indicated that participants with vision loss attained independent walking significantly later than participants with vision. Within this investigation, the average attainment age of independent walking for children with CHARGE syndrome was 36.6 months, which when compared to the World Health Organization (WHO)

Multicentre Growth Reference Study Group, exceeds the window of achievement for walking independently (8.2 to 17.6 months; WHO Multicentre Growth Reference Study Group, 2006).

In addition to the delay in walking, a delay in posture control (i.e., prone, supine, sitting, standing) in infants who were blind were reported and considered to result from the lower level of motor stimulation due to the lack of sight (Tröester & Brambring, 1993). Based on the results of this investigation, a mother's perspective of her child with CHARGE syndrome supports the previous findings. The mother stated, " . . . with the limited vision, without the environmental hearing, she [daughter with CHARGE syndrome] was kind of stuck in a little bubble and only curious about what was going on left and right of her vision." The mother's response indicated her child's lack of motor stimulation to move due to her vision and hearing loss. Brambring (2006) further reported that vision related constraints "prevents or restricts the ability to engage in adequate learning experiences while acquiring motor skills" (p. 621). Therefore in this investigation, supported by previous research, vision was indicated as a rate limiting constraint on the attainment of independent walking in children with CHARGE syndrome.

In addition to vision loss, the majority children with CHARGE syndrome (90%; Blake et al., 1998) exhibit hearing loss and vestibular dysfunctions, which has a negative impact on the attainment of independent walking. Although vestibular dysfunction (i.e., small or absent inner ear canals) and sensorineural hearing loss were not significant predictors of the age of walking in this present investigation; the impact of hearing loss

combined with vision loss did significantly delay the attainment of independent walking in children with CHARGE syndrome. Previous researchers supported the negative impact of vestibular dysfunction and sensorineural hearing loss on the task and attainment of walking (Crowe & Horak, 1988; Cushing et al., 2008; De Kegel et al., 2012; Inoue et al., 2013; Kaga et al., 2008; Kaga, Suzuki et al., 1981; Potter & Silverman, 1984; Rapin, 1974; Rine et al., 2000; Suarez et al., 2007).

Impairment to the vestibular functions (i.e., hearing system) has the potential to lead to developmental delays in motor function and achieving gross motor milestones such as walking (Kaga, 1999; Kaga et al., 2008; Rine et al., 2000), specifically in children with CHARGE syndrome (Abadie et al., 2000; Admiraal & Huygen, 1997; Hartshorne et al., 2011). Ear structure malformations may impact the vestibular system (i.e., utricle, saccule, semicircular canals, VII cranial nerve, cochlear), which can disrupt information about the body's motion, equilibrium, spatial orientation, and balance abilities (Hanes & McCollum, 2006; Rine, 2009; Shumway-Cook & Woolacott, 2001; Vestibular Disorders Association, 2008). Upright, bimodal independent walking requires postural control and balance, which the vestibular system (i.e., utricle, saccule, semicircular canals) provides. The vestibular system can produce three motor reflexes to assist the body with movement: (a) vestibular-ocular reflex assist with visual stabilization, (b) vestibular-collic reflex assist with neck stabilization, and (c) vestibular-spinal reflex assists with posture and the orientation of one's body in space (De Kegel et al., 2012; Nandi & Luxon, 2008; Suarez et al., 2007; Tribukait et al., 2004). All three motor

reflexes contribute to postural tone and are necessary for the acquisition of motor developmental milestones (Nandi & Luxon, 2008; Tribukait et al., 2004).

In contrast to the results of this investigation, Inoue et al. (2013) reported that the age of independent walking was significantly delayed in the group of participants with profound sensorineural hearing loss and combined superior and inferior vestibular dysfunction and with inferior vestibular dysfunction when compared to children with normal vestibular function and no hearing loss. However, to explain the inconsistency with the results of this investigation, the investigators reported that there were no statistically significant differences between the group with normal function and the group with superior vestibular dysfunction, the group with inferior vestibular dysfunction, or the group with combined vestibular dysfunction.

Furthermore, in a case study investigation of six children with CHARGE syndrome, the researchers reported that intact saccular function might be an important factor for upright stance and proper gait pattern (Admiraal & Huygen, 1997). The age range of independent walking was between 2 to 4 years of age for those children with CHARGE syndrome (one of the six individuals' age was not reported; Admiraal & Huygen, 1997). Residual otolith function may also enhance the development of motor skills and walking in children with CHARGE syndrome (Abadie et al., 2000). Therefore, depending on the type of vestibular dysfunction, varied results may be reported related to the impact vestibular dysfunctions has on the attainment age of independent walking. No specific information related to the type of vestibular function was provided in the databank or analyses within this investigation.

Potter and Silverman (1984) reported that children with hearing loss might use other organs and body systems to compensate for vestibular dysfunction. However, the task of walking (i.e., gait patterns) differed between the children with sensorineural hearing loss compared to the children with hearing (Melo et al., 2012) indicating that hearing loss causes modification to the task of walking. The results align with the DST and the results of this present investigation, in that constraints may provide specific modifications in movement patterns to remain in a state of stability (Coker, 2013; Thelen & Ulrich, 1991).

In order for an infant to attain the independent walking developmental motor milestone, the body subsystems must be developed to support the body in an upright position through the use of posture, balance, and strength (Thelen, 1986a). Parents and primary caregivers of children with CHARGE syndrome within this present investigation provided information consistent with this assertion. When asked by the primary investigator to explain any difficulties or roadblocks of their son or daughter with CHARGE syndrome had acquiring independent walking, one mother responded with, “Oh, definitely, the balance issues, when you don't have the equipment [semicircular canals] to have balance, I think it was a cause for delay [of walking]. Hartshorne et al. (2011) stated, “. . . vision impairment combined with vestibular dysfunction leads to delayed motor milestones” in children with CHARGE syndrome (p. 5).

Abadie et al. (2000), reported that there was no significant difference in the attainment age of independent walking indoors in children with good or poor otolith function compared to children with CHARGE syndrome who had also good or poor otolith function. Those findings support the findings of the present investigation.

Furthermore, Abadie et al. (2000) reported that of all the parameters needed for balance acquisition, most of them are defective in children with CHARGE syndrome, which contributes to their developmental delay.

The development of balance and ability to walk independently are therefore dependent on vestibular (i.e., hearing system), proprioceptive, motor, and visual systems (Kaga, 1999; Nakajima, Kaga, Takekoshi, & Sakuraba, 2012; Suarez et al., 2007). As previously stated, vision has a role in postural control; in addition, postural stability is also directly associated with the correct functioning of the hearing organs (Walicka-Cupryś et al., 2014). The type of hearing loss and structure of a child's inner ear provides information about the function of a child's vestibular system. Sensorineural hearing loss is caused by damage to the inner ear (i.e., vestibular structures) that may impact vestibular system function (National Institute on Deafness and Other Communication Disorders, 2013). Suarez et al. (2007) reported that children with sensorineural hearing loss (i.e., dysfunction of the hearing organs) primarily used visual and somatosensory information for postural control. Furthermore, Rine et al. (2000) reported that children with sensorineural hearing loss had a deficit in balance development and a delay in motor development. Within this present investigation, 43 of the 62 (69.4%) participants had a known sensorineural hearing loss. In addition, children largely rely on their vision to maintain balance during their early stages of development (Inoue et al., 2013). This information could explain the walking gait pattern (i.e., wide and wobbly) adaptations that children with CHARGE syndrome perform to assist with

posture control, balance, and additional sensory information that was reported in this investigation by the parents' or primary caregivers' perspectives.

Therefore, if vision and hearing are both affected, then the development of postural control, balance, and motor development will also be delayed. The findings of this investigation provide evidence that vision loss and hearing loss are rate limiting constraints that delay the attainment of independent walking in children with CHARGE syndrome.

Low muscle tone. Based on the qualitative findings of this investigation, low muscle tone was a significant rate limiter in the attainment of walking in children with CHARGE syndrome. Muscular strength and gravity have been previously determined as potential constraints to the production of movement (Thelen & Fisher, 1982; Thelen et al., 1984). Abadie et al. (2000) reported that hypotonia (i.e., low muscle tone) was prevalent in children with CHARGE syndrome within their first few months after birth; however, muscle tone increased to a normal level after the attainment of walking.

Furthermore, Brown (2005) reported that low muscle tone is very persistent in children with CHARGE syndrome. Due to additional anomalies, the issue of low muscle tone is compounded by the lack of stimulation and motivation to move (Brown, 2005). Brown (2005) further reported that tactile defensiveness may be present in children with CHARGE syndrome due to "poorly modulated tactile and proprioceptive senses" (p. 269). Similar results were reported in this present investigation.

Parents and primary caregivers provided the following evidence to support the impact of both low muscle tone and tactile aversion in children with CHARGE syndrome on the attainment of independent walking (below are three different parent perspectives):

“Her muscle tone. She has really poor muscle tone, especially core muscle tone.”

“Alright, so basically it was very difficult for her [to attain independent walking], low muscle tone, low very dysfunctional vestibular system, low balance.”

“He [has] low muscle tone, so he didn’t do the normal progression [of developmental motor milestones] . . .”

The following is evidence provided by the qualitative results pertaining to tactile aversion,

“. . . for about the first 10 months of his life he didn’t move at all. He didn't roll over, he didn’t want to be picked up even, it was, he was very sensitive about being touched, he didn’t want to move.”

Both of these variables (i.e., low muscle tone, tactile aversion) can constrain the ability to acquire independent walking in children with CHARGE syndrome and should be acknowledged by instructors (i.e., teachers, therapists).

Impact of fear. Fear is identified within the DST as an individual constraint (Newell, 1986) impacting the production of movement. The present investigator reported that the impact of fear decreases an individual’s confidence, comfort, and safety (i.e., affordances) and increases their delay in the attainment of independent walking in children with CHARGE syndrome. The support and evidence of fear being a rate limiter in acquiring independent walking was perceived by the parents and primary caregivers of children with CHARGE syndrome and supported by previous researchers.

Haibach and Leiberman (2013) examined the balance and self-efficacy of balance in 22 children with CHARGE syndrome and reported that fear was a factor in balance performance. In this previous study, it was reported that 14 children had fallen in the past year and 14 of the 22 parents stated that their child with CHARGE syndrome had a fear of falling. Based on the results of a balance scale, 9 of the 22 participants with CHARGE syndrome were at a low risk of falling, with an additional 8 at a medium fall risk, and 4 at a high fall risk, compared to children without CHARGE syndrome who were not at a fall risk (Haibach & Leiberman, 2013). The qualitative findings within this present investigation were similar to the findings of Haibach and Leiberman (2013). One participant within this investigation shared a similar experience as reported by Haibach and Leiberman (2013),

> "That [practice] gave him the confidence where he wasn't just going to fall and crash and hit his head. I think he started learning that oh, if I fall it is not so bad if I am walking by myself. I think motivation in just cruising using the coffee table, he just needed that confidence with something to hold on. That's why that summer when he finally let go and moved he was just, I can remember his face, he was just so excited that he finally made it without falling. So I think a lot of it was the confidence and the ability to do it without crashing."

Based on the results of the previous research findings (Haibach & Leiberman, 2013), on a scale of 0 to 10 for balance confidence (with a 10 being completely confident), 19 of the 22 children with CHARGE syndrome indicated a balance confidence score of 0. Therefore, fear appears to be correlated with balance. As balance increased, the child's confidence also increased, which supported motor development. Other investigators reported that a lack of confidence and fear displayed by students who had visual

impairments or were deafblind was a barrier (i.e., rate limiter) to participation in physical activities (Lieberman & Houston-Wilson, 1999). One mother in this investigation stated,

> "By the time she [her daughter with CHARGE syndrome] was doing assisted walking, she was old enough to build up a pretty healthy fear of falling down and getting hurt which was holding her up from walking independently."

An investigation on 17 children with CHARGE syndrome reported, when walking, changes in direction often resulted in falls (Abadie et al., 2000).

Contrary to the majority of the findings related to fear in this current investigation, Bouchard and Tétreault (2000) reported a fear of space in children with vision loss. The parents and primary caregivers of this present investigation reported that fear was associated with falling and therefore delaying the acquisition of independent walking. However, due to the child's limited vision and hearing, the fear to explore space was noted by a few parents within this investigation. The mother stated,

> "I think it is a combination of fear and the walker felt safe as well. From an exploring space thing and or even now, if she goes into a big open area she will sit down because it is too scary from a navigation stand point but if you give her a walker, she would go right through that because she feels safe. . ."

Another mother stated, ". . . when she [daughter with CHARGE syndrome] gets to wide open spaces she would get nervous and [sit] down." Bouchard and Tétreault (2000) reported that fear of space was an additional possible rate limiter that could delay motor development and gross motor performance in children with vision loss, which was present in two interviews with parents of children with CHARGE syndrome. For children with vision loss, the fear of space (Cratty, 1970) may inhibit movement, due to their inability to know where they are in the environment. In addition, as reported by previous

studies and in this present investigation, Bouchard and Tétreault (2000) reported that an increase in a child's confidence in performing motor activities is essential.

Therefore, the impact of fear (i.e., of falling, of space) was another significant rate limiter in the acquisition of independent walking in children with CHARGE syndrome. In addition, confidence was a significant affordance, which assisted in the attainment of independent walking in children with CHARGE syndrome.

Interaction Between the Environment and Task

Vision and hearing loss impacts the perception of a child's environment. The environment is a constraint which may impact the production of preferred movement tasks as related to the DST (Newell, 1984, 1986; Thelen & Smith, 1996). Environmental constraints can act as both rate limiters and affordances on gross motor performance and motor development. Therefore, individual constraints (e.g., body systems) are influenced by environmental constraints. The body systems interact together as a functional self-organizing unit to enable the preferred form of movement to be performed based on the task and environmental constraints. An example of this was reported by Bouchard and Tétreault (2000), in which infants with vision loss were not stimulated to raise their heads and therefore, typically preferred to be in a prone position to seek light sources within their environment. Accordingly, in the present study the impact of the environment played a role in the development of independent walking in children with CHARGE syndrome.

Environmental impact. An area (i.e., physical space) in which an individual is performing gross motor skills has the potential to influence his or her performance and

the development of additional gross motor skills (Abadie et al., 2000). Within this present study, multiple constraints were related to the area in which an individual with CHARGE syndrome was performing movement. Results from parent perspectives illustrated that the environment and the surface (i.e., flat, open, uneven) and surrounding objects (i.e., furniture) impacted the development of independent walking in children with CHARGE syndrome.

When asked what were some difficulties or roadblocks that her daughter with CHARGE syndrome experienced while developing to walk, two different mothers responded:

"Just kind of space to walk around in, she didn't have a whole lot of area that was a flat surface for her to get around so she was just stuck to one little spot. And outside it was very difficult for her to get up by herself so she didn't really attempt it unless someone was holding her hand."

"Outside, uneven grass and ground, even after he started walking, that was always a challenge to get him to walk outside and anywhere that wasn't completely flat."

Similar findings were reported in Abadie et al. (2000), in which all of the 17 children with CHARGE syndrome within the investigation had difficulty walking outdoors on uneven surfaces. Furthermore, Adolph (2002) stated that walking is only possible if the floor is "appropriately stable, flat, rigid, wide and with sufficient traction to support the movements required for balance and propulsion" (p. 88).

Abadie et al. (2000) further reported that for children with CHARGE syndrome walking was only possible in a familiar place that is on flat and regular floors. The findings of this present study agree with the previous research. One mother stated,

"She [her daughter with CHARGE syndrome] still has a hard time on uneven surfaces anywhere with a threshold until she gets comfortable with it and then she is totally fine."

This statement provides evidence that a familiar place that has even floors supports independent walking in children with CHARGE syndrome.

In addition, Brambring (2006) reported major developmental divergences in skills that required movement through space without physical contact with the environment (e.g., walking without holding on to furniture or without assistance) in children with vision loss. The findings of this present investigation support the use of physical contact with furniture to assist in the development of walking in children with CHARGE syndrome. In this present study, it was reported that environmental constraints were altered in a way to promote independent walking in children with CHARGE syndrome. One mother stated,

"We would position furniture at certain distances his PT [physical therapist] helps us set it up so he could, as he would slowly take a couple of steps after a week when he was comfortable with that we would move furniture a little bit more away, and when I say move furniture I am talking about 6-10 inches, it wasn't a lot."

The use of furniture as an affordance encouraged the development of independent walking in children with CHARGE syndrome. Another mother stated how the use of furniture and toys assisted in the development of independent walking in her daughter with CHARGE syndrome:

". . . position some toys so many spaces apart so then she had to take one step so get to the next toy so we would do that with coffee tables, we would push them back so she would have to take that one step and then eventually two steps on her own and then further and further."

During the acquisition of walking, previous research (Barela et al., 1999) reported that surface touch was used initially for mechanical support and then after having walking experience (1.5 months) for a source of sensory information. This may be the reason why many children with CHARGE syndrome use surface touch as an affordance to assist in the development of independent walking. Metcalfe and Clark (2000) reported that the use of surface contact also significantly decreased head and shoulder sway in newly walking infants and stabilized posture. Therefore, both infants with CHARGE syndrome and those without use somatosensory information to aid in upright postural control.

Impact of hospitalization and surgeries. In this present investigation, further analysis results in additional constraints on the task of independent walking in children with CHARGE syndrome when being in a hospital. The amount of months children with CHARGE syndrome were hospitalized was a significant covariate in this study. CHARGE syndrome is medically complex, with many children with CHARGE syndrome fighting to survive within the first year of their life. Many children with CHARGE syndrome have multiple lifesaving surgeries within their first year of life due to the numerous factors present at birth (Admiraal & Huygen, 1997; Harvey, Leaper, & Bankier, 1991; Tellier et al., 1998; Wiznitzer, Rapin, & Van de Water, 1987). One mother shared that her daughter with CHARGE syndrome was,

". . . on a ventilator for 24 hours a day, until she was about a year and a half and then slowly started to wean her off. When she started to get weaned off and completely weaned off, that was when she started crawling, she wouldn't even do tummy time when she was on the vent."

Another mother stated her son with CHARGE syndrome,

". . . had four heart surgeries and you wouldn't want to lie on your chest after that. I think that was one of the biggest things."

The findings from the qualitative results continued to support and provide evidence in the relationship of increased months hospitalized and the number of surgeries with an increase in the attainment age of independent walking.

As reported by parent perspectives, hospitalization and surgeries had a significant impact on delaying motor development in their child with CHARGE syndrome. One father indicated that,

"He [his son with CHARGE syndrome] was in the hospital for a while just laying down so he didn't have a chance to move around like other children. He was hospitalized so he wasn't moving and he was connected to many machines. He was also very fragile and he didn't have a lot of physical contact."

Another mother stated,

"hospitalization and surgeries were huge. We [the mother and child with CHARGE syndrome] spent nearly the first year in the hospital."

Furthermore, another mother provided information related to the impact of hospitalization and surgeries and hospitalizations on her daughter with CHARGE syndrome.

"She was born, in the NICU [Neonatal Intensive Care Unit] for 6 months for 4 major surgeries, heart surgery, she has a trach [tracheostomy], feeding tube, swallowing difficulties. So when she came home at 6 months, she was basically a newborn and the doctors had kind of said when we do any evaluations to subtract 6 months off of her life because she came home as a newborn, she was going to be delayed. We knew that."

The result of the impact of hospitalizations and surgeries on children with CHARGE syndrome is also supported by the limited research available on CHARGE syndrome.

Research performed by Salem-Hartshorne (2003), Salem-Hartshorne and Jacob (2004), and Abadie et al. (2000), indicated that hospitalization and surgeries had a negative impact on motor development in children with CHARGE syndrome, which supports the findings of this investigation.

Impact of the social environment. Thelen and Fogel (1989) reported that the social environment influences the development of new movement behaviors. This generalization was also reported in this present investigation. The social environment included the use of encouraging parents, therapists, and walking peer models, who provided support in the development of independent walking in children with CHARGE syndrome.

Similar to the findings of this investigation, Levtzion-Korach, Tennenbaum, Schnitzer, and Ornoy (2000) reported that involvement of parents could assist in the motor development of children with vision loss. Improvements in motor development were seen when parents altered the child's environment by providing adequate social stimulation based on his or her visual function (Letzion-Korach et al., 2000) through the use of language and physical contact (Freiberg & Adelson, 1977; Norris et al., 1957). Infants who walk also explore their environment, objects, and people differently and are provided new opportunities for action and interactions (Adolph & Tamis–LeMonda, 2014). Within this present investigation, one father stated the impact of a social environment,

". . . sometimes it's just more trying to connect or explore [pause] you know if it is just taking her in a wagon or doing something else I think contributed to getting her desire up, wanting to risk it, finally finding out there are places they [children with CHARGE

syndrome] want to get in the world and other people are walking there and if they I can walk there, there has some pluses for me."

Previous researchers have supported that interactions with people increased as a result of obtaining the skill of walking (Karasik et al., 2014).

Furthermore the encouraging social environment also impacted the development of children with CHARGE syndrome on their execution of independent walking. One mother in this study stated,

"I think what helped him the most was that we just kept encouraging him and working with a physical therapist and an occupational therapist. We pushed him. It was my goal, even on all his IEPs [individualized education programs] and everything is walking. With the help with the different strategies and support, he started. I can't say really one thing, just a good team effort. We have a great team. I have the dream team."

The presence and characteristics of the parents and family can be an environmental social constraint impacting motor development (Haywood & Getchell, 2005). Pereira (1990) reported that social positive reinforcement improved the motor development of children with vision loss. Furthermore, Gibson (1987, 1997) reported that infants explore multisensory experiences when information is afforded by environmental interactions. Only after infants can locomote can infants better understand the environment around them and are more involved in the interpretation and exploration of the environment (Metcalfe & Clark, 2000). Independent movement changes the way infants experience their environment around them (Clearfield, 2011).

Similar to the results of this present investigation, previous researchers report that the social surrounding environment provides support for motor development (Barela et al., 1999). The results of this investigation indicated the importance of a peer model in the

development of independent walking in children with CHARGE syndrome. As two mothers in this investigation stated,

". . . one time in point, when we took her to the nursery, she saw other children her age walking much more than the usual and so I think this encouraged her a lot. She was conscious enough to leave the walker and walk towards us."

"What motivated her [her daughter with CHARGE syndrome] was seeing other kids, I think even to this day she wants to be like everyone else, so being around other friends and seeing them walk was motivating for her."

Modeling refers to behavioral change that comes directly from observing others (Berger, 1977) within a conducive environment. Bandura (1969) reported that modeling is observational learning that reflects acquisitions of new behavioral patterns. However, Bandura further stated that the ability to learn from a model depends on developmental factors (1986). However, within this present study, parents of children with CHARGE syndrome reported the motor developmental benefits their child gained from observing other children walking. The use of peer modeling and peer tutors have been previously reported by researchers as a beneficial strategy to increase motor performance in children with vision and hearing loss (Block, 2007; Cervantes, Lieberman, Magnesio, & Wood, 2013; Hodge et al., 2012; Houston-Wilson, Dunn, Hans van der Mars, McCubbin, 1997; Lieberman, Dunn, Hans van der Mars, & McCubbin, 2000; Schultz, Lieberman, Ellis, Hilgenbrinck, 2013; Wiskochil et al., 2007). In this present study, the results indicated that peer modeling of walking was a beneficial social environmental constraint for children with CHARGE syndrome.

Task of Independent Walking

The theoretical framework that guided this research was based on the DST. The task of independent walking is therefore, influenced by the interaction of the individual and environmental constraints (Newell, 1986; Thelen, 1995, 1998). The performance of independent walking is modified to obtain a functional movement pattern based on the movement stability and the body's preferred attractor state (influenced by constraints; Thelen & Ulrich, 1991). In the present investigation, parents reported that the task of walking in children with CHARGE syndrome was performed independently with a wide, wobbly gait, due to individual constraints. This type of movement likely emerged as the most attractive option based on the individual constraints present in those with CHARGE syndrome. In Phase II, parents and primary caregivers were emailed a document with figures of developmental milestones prior to the interview (see Appendix J). This provided the parents and primary caregivers with a visual of when typically developing children attain certain motor milestones and how the milestone is performed.

Similar to the results of this investigation, Murofushi et al. (1997) reported that three children (out of five participants) with CHARGE syndrome that acquired independent walking, performed a walking gait with a wide base and stiff legs, and had difficulties walking and balancing on uneven surfaces. Abadie et al. (2000) also reported that, "balance acquisition in human development is the result of visual, proprioceptive, vestibular, and cerebellar maturation" (p. 573). This statement supports the findings of this present study where balance difficulties and walking modifications in children with CHARGE syndrome were reported. The parents or primary caregivers in this

investigation provided supportive evidence that the task of walking was affected by vision and vestibular dysfunction. One mother reported that her child with CHARGE syndrome had a “very wide stance at first and [was] wobbling from side to side.” Another father stated that, “she walks with a wide stance and pretty wobbly, kind of a ‘drunken sailor’ walk a little bit.” Yaguramaki and Kimura (2002) reported that lateral stability was the most important factor in gait development in infants. Therefore, if children with CHARGE syndrome have difficulties with balance and stability then a wide stance would be needed to assist with the dynamic balance of the task of walking. In addition, movement variability is a part of the learning process to gain stability in functional movement patterns (Newell et al., 2001).

Adaptation to the gait pattern has been investigated and it has been suggested to provide sensory feedback to compensate for a loss of visual feedback of the environment (Buckley et al., 2008; Hallemans et al., 2009a, 2009b; Hallemans et al., 2010, Hallemans et al., 2011). Hallemans et al. (2011) reported that individuals with visual impairments demonstrated a slower walking speed, a shorter stride length, a prolonged duration of stance phase duration, and double support phase during the walking gait pattern compared to children with vision. Underlying balance difficulties may alter the prolonged stance and double support phase during walking in individuals with vision loss (Hallemans, et al., 2011; Hallemans et al., 2010; Sutherland, 1997). The prolonged double support phase allows the foot to gain sensory feedback about the ground (Patla et al., 2004).

In addition to those with vision loss, Melo et al. (2012) reported gait adaptations in children with hearing loss. During the gait analysis, the children with hearing loss exhibited trunk flexion during locomotion and did not display a swing phase or heel strike. Instead the children with hearing loss dragged their feet and continuously maintained a phase of double support (Melo et al.). The researchers indicated once again this gait adaptation was the result of balance difficulties.

Motivation. For children with CHARGE syndrome, motivation is needed for movement to be performed. Due to their vision and hearing loss, an environment must be stimulating to motivate the child to want to move. Adolph et al. (2012) reported that children may be motivated to walk because they are able to move farther faster than crawling without an increased risk of falling. In addition, after the onset of walking independently, as the child becomes a more experienced walker, the child takes more steps, travels further, and falls less (Adolph et al., 2012). However, skilled independent walking takes months to develop in children without any additional disabilities or anomalies (Adolph, Vereijken, & Shrout, 2003; Hallemans, De Clercq, Van Dongen, & Aerts, 2006). Hallemans et al. (2006) also reported that as a child was gaining walking experience immature walking patterns were frequently observed until motor control improved. Therefore, the instability of the task of walking as they are learning to walk will alter a child's gait pattern and they will continue to crawl at time, as they develop the ability to walk.

It is important for parents and primary caregivers of children with CHARGE syndrome to learn what is motivating in order to provide an environment that stimulates

the child to move. The results of this investigation revealed multiple sources of motivation. One mother stated,

> "What motivated her [her daughter with CHARGE syndrome] was seeing other kids, I think even to this day she wants to be like everyone else, so being around other friends and seeing them walk was motivating for her."

A different father stated that his daughter with CHARGE syndrome, ". . . always wanted to walk. . . She was motivated to get out and go." However, at times motivation is difficult. As one mother in this present investigation stated, ". . . motivation has always been a challenge. Whether it was getting him walking, scooting, [or] crawling, anything. It was a very [big] challenge, he's better now but you know he's 7 now." Motivation can be a rate limiter or an affordance on the attainment of independent walking in children with CHARGE syndrome.

Consequently, it is important that parents and service providers discover motivational factors which can increase a child's desire to move. One mother stated, ". . . I [mother] just motivated him more with visual aids to get him up and going." Another father stated, ". . . if we take him to a playground, he will motivate himself to walk anywhere." Even though walking may not be as functional at the time for children with CHARGE syndrome, it is important for them to continue to experience the task of walking to continue to gain confidence and experience. Furthermore, Adolph (2008) reported that exploration plays a critical role in learning by acquiring visual, haptic, vestibular, and perceptual information, and gaining relevant information about their environment relevant to the locomotor task. As one mother in this present investigation stated,

"We got a play kitchen, we set it up 5 feet away from the couch, where she had been cruising for months. Eventually, it took about 2 months, she would then walk from the couch over to the kitchen. That was one of the first things that got her motivated to get over to where she wanted to [go]."

Assistive equipment. Previous researchers have supported the need for surface contact to assist in the motor development of children with vision or hearing loss (Barela et al., 1999; Brambring, 2006; Metcalfe & Clark, 2000). Furthermore, researchers have reported the use of assistive devices by individuals who are deafblind (Haibach & Leiberman, 2013; Petroff, 1999). Haibach and Leiberman (2013) reported that 18 out of 22 children with CHARGE syndrome used some type of assisted walking devices at times while walking.

The literature supports the findings of this present investigation and the use of assistive equipment in the development of walking in children with CHARGE syndrome. Assistive devices provide an increase in confidence and safety, which decreases their fear of falling compared to walking independently. The use of assistive equipment is a task constraint to the performance of walking (Newell, 1986). Based on the individual and environmental constraints, the use of assistive equipment can act as either a rate limiter or affordance in the development of independent walking. As one father in the present investigation stressed, it is essential to get the correct walker to benefit the child with CHARGE syndrome based on their unique individual needs. Furthermore, one mother in this present investigation stated,

"I think the walker definitely helped a lot because she was able to explore with [the walker]. And then I think 'kinda' the other piece to this is to [experience] the outside world enough to kind of have a really strong motivation to want to get out there, enough

to overcome some of the walking. And then it is just making it accessible for her, making sure we have the walker around."

5-point crawl. In the quantitative section of the present investigation, the 5-point crawl was not a significant predictor in the attainment of independent walking; qualitative results supported the impact on developing the task of walking independently. Previous researchers and the qualitative results indicate possible task constraints associated with the movement. The DST components can provide support and understanding of the preferred movement pattern of the 5-point crawl which is also frequently seen in children with CHARGE syndrome.

When crawling, an infant's neck and eyes are pointed downward, allowing him or her to primarily see the floor in front of his or her hands. However, walking permits infants to see distant objects and people (Franchak et al., 2011; Kretch et al., 2014). Individuals with vision loss, however, are not motivated by visual input to keep their head up and therefore, result in a 5-point crawl where their head is on the ground receiving proprioceptive feedback. Abadie et al. (2009) reported a frequent inability to crawl on all fours without resting the head on the floor in individuals with CHARGE syndrome. Further support in this investigation indicated early effect of poor balance in children with CHARGE syndrome included: (a) poor head control and poor ability to resist against gravity; (b) a preferred preference for lying flat on back for most activities, including locomotion; and (c) unique movement patterns including back scooting and 5-point crawl (Brown, 2005).

The current findings of the qualitative portion of this investigation supported previous studies. Two different mothers stated,

"[son with CHARGE syndrome] turned to bulldozing, like the 5-point crawl, and head down and crawling that way everywhere. We knew he would be delayed walking since he was a 'bulldozer' with his head down."

"He [son with CHARGE syndrome] would be in the 5-point crawl position but he didn't move in that position. He liked to be in that position, he liked to get that feedback on his head, deep pressure on his head, looking like he was about to do a tumble. He liked to do that a lot."

The attainment of the 5-point crawl may act as a rate limiter due to the stability of this preferred movement in children with CHARGE syndrome (Ennis, 1992; Haywood & Getchell, 2009; Thelin & Ulrich, 1991). However, within specific control parameters and through self-organization of the body's systems and constraints, independent walking can become the preferred movement pattern.

Implications of the Findings

The findings from this present investigation provided implications and numerous recommendations for professionals and practitioners who work with and instruct children with CHARGE syndrome related to independent walking. Since the severity of vision and hearing loss negatively impacts the delay in attaining independent walking, those professionals who provide programs and interventions should emphasize the use of an individual's residual hearing and vision.

It is important that within the school environment, educational techniques and interventions are developed to improve motor performance, gait patterns, and prevent falls in children with sensory loss (Melo et al., 2012). Furthermore, De Kegel, Maes,

Baetens, Dhooge, and Van Waelvelde (2012) reported that although communication and language are the main problems faced by children with hearing loss, they also have a higher risk for motor and balance deficits, which should not be overlooked.

Since previous researchers have indicated a strong positive correlation between the attainment age of independent walking in children with CHARGE syndrome in relation to the acquisition of symbolic communication and language (Petroff, 1999; Thelin & Fussner, 2005), adaptive behavior scores (Salem-Hartshorne, 2003; Salem-Hartshorne & Jacob, 2004), and executive function behaviors (Hartshorne et el., 2007) motor development should be an early intervention priority. Furthermore, communication development during infancy is part of an interdependent system, which includes motor development (Bates, Benigni, Bretherton, Camaioni, & Volterra, 1979). As development occurs in one system it concurrently facilitates the development of other systems. The inability to walk may limit an individual from communicating since most communication occurs between two or more individuals. The 'communication bubble' (i.e., another person) is necessary for communication (Hefner & Davenport, 2002). The 'communication bubble' is the area around an individual where a talker or signer can be heard or seen optimally, which may differ depending on the degree of hearing and vision loss (Hefner & Davenport, 2002). This may partially explain why there is a correlation between walking and communication levels of individuals who are deafblind (King, 2009; Thelin & Fussner, 2005). Furthermore, the attainment of walking is associated with an increase to normal levels of muscular tone in children with CHARGE syndrome (Abadie et al., 2000). Therefore, no part of children with CHARGE syndrome's

development should be ignored; particularly within early intervention. Programs should emphasize the importance of motor development (i.e., walking) for children with CHARGE syndrome.

Because of the numerous individual constraints pertaining to children with CHARGE syndrome, it is important that environments are recognized as potential constraints that can be manipulated to promote motor development and success. The experience of movement success in children with CHARGE syndrome can strengthen their motivation to continue to move and increase confidence, which will alleviate or reduce their fear. Brown (2005) stated that any input that assists in the development or use of the vision, balance, and proprioceptive systems within the body of an individual with CHARGE syndrome could assist in the development of walking. Further, Newell (1991) reported that basic actions of posture, locomotion, and manipulation allow an individual the ability to perform a variety of motor skills based on task constraints (e.g., self-help skills, vocational skills); therefore, leading to a more independent individual.

Additionally, Bouchard and Tétreault (2000) reported the need for early intervention to minimize the consequences of vision and hearing loss, which could contribute to motor development, specifically the development of gross motor skills (e.g., independent walking). In addition, providing an environment which is stimulating through visual, auditory, and tactile senses is important to increase the motivation levels of children with CHARGE syndrome to move (Levtzion-Korach et al., 2000). Furthermore, individuals who work with and assist children with CHARGE syndrome should situate them in a prone position and place items in their visual field (Bouchard & Tétreault, 2000).

Teachers and therapists should also acknowledge the awareness of touch, pain, and temperature that may fluctuate for children with CHARGE syndrome (Brown, 2005), which may have the potential to impact motor performance (i.e., rate limiter).

The theoretical framework that guided this research was the DST. The DST was used in this present investigation to gain an insight about the interrelations and interactions of characteristics in children with CHARGE syndrome that could have a positive (i.e., affordances) or negative (i.e., rate limiter) impact on the developmental motor milestone of walking. Within this ecological perspective, discovery learning and a 'hands-off' approach is emphasized, to lead the learner into actively developing the correct movement response (Coker, 2009; Davids, Button, & Bennett, 2008; Handford, Davids, Bennett, & Button, 1997; Newell, 1991). Therefore, the teacher or instructor is placed in a facilitator role to manipulate and identify constraints to lead the child to the optimal movement patterns.

From the results of this present investigation, the following are identified individual constraints that influence motor development in children with CHARGE syndrome: individual personality, comfort and safety level, confidence level, feeling of fear or being afraid, muscle tone, hearing loss, vision loss, vestibular function, balance, and tactile preference. Furthermore, from the results of the study, environmental constraints such as surface touch (i.e., furniture), adequate open areas, surface, and encouragement can impact the attainment of walking in children with CHARGE syndrome.

Specifically, general and adapted physical educators can benefit from the knowledge of individual and environmental constraints that impact the motor development and

attainment of independent walking in children with CHARGE syndrome. The facilitation of the program and educational interventions should be based on the unique needs of each individual. Since children with a dual sensory loss are a low incidence disability, challenges arise related to providing highly qualified instructors and evidence based instructional strategies (No Child Left Behind; Amended 2002). However, the results of this investigation can contribute to the literature in CHARGE syndrome and can increase the knowledge of the impact of vision and hearing loss on motor development and walking in children with deafblindness.

Limitations

The limitations of this investigation must be considered when generalizing and evaluating the results. First, the impact of balance was not quantitatively measured as a factor in the achievement of independent walking in children with CHARGE syndrome. Second, the amount of interventions and therapy services (i.e., early intervention, physical therapy, hippotherapy) provided to the children with CHARGE syndrome may have impacted the motor development of walking and was not evaluated or measured. Therefore, the interpretation of the results of this investigation are limited based on the duration and amount of interventions and therapies provided to the children with CHARGE syndrome. Third, the heterogeneity of the group and the impact of additional anomalies of CHARGE syndrome could not entirely be controlled. However, the uniqueness of the participants identified specific characteristics and factors related to motor development and children with CHARGE syndrome.

Fourth, parents or primary caregivers attended an international conference and were a part of the CHARGE Syndrome Foundation; therefore, they may have been more proactive in their child's development. Fifth, descriptive information used to analyze the developmental milestones related to independent walking was limited by self-reports from the participants' parents or primary caregivers, both in the database and interviews. Self-report questionnaires have been consistently used to obtain health-related information and developmental motor milestone measures have been deemed valid and reliable from parental reports (Bodnarchuk & Eaton, 2004). In addition, an electronic database was used to obtain data during the quantitative portion and no identifying personal information was available. Furthermore, interview questions were sent prior to the commencement of interviews and participants were reminded that responses would be documented anonymously.

Conclusion

In conclusion, based on the present findings, vision loss and hearing loss were rate limiters that significantly impacted the attainment of independent walking in children with CHARGE syndrome. The DST was used to further understand and explain the impact of various constraints on the attainment of independent walking in children with CHARGE syndrome. Similar to previous findings, the number of hospitalizations also was related to the attainment age of the developmental motor milestone of walking independently. The increase in severity of vision and hearing loss significantly increases the age of attaining the independent walking motor milestone in children with CHARGE syndrome.

Furthermore, parents and primary caregivers shared their perspectives and beliefs of the individual, environmental, and task rate limiters and affordances that impacted their child's independent walking. These findings support the impact of vision loss, hearing loss, and hospitalization has on delaying the attainment of independent walking. From these findings, the DST supports the dynamic interaction of the individual and environment influence on the performance of independent walking.

Recommendations

Based on the results from this investigation, there are numerous areas recommended for future researchers to pursue related to CHARGE syndrome and walking:

1. Future researchers should replicate the study and expand the scope of this investigation to include a larger sample size and possibly increase the use of additional measurable variables (e.g., balance test score, therapies provided, length of assisted walking) that may impact independent walking. In this study, the severity of vision and hearing loss negatively affected the attainment age of independent walking in children with CHARGE syndrome. Future investigations are needed to develop a concrete body of evidence in motor development impacted by sensory loss, specifically CHARGE syndrome.
2. Future researchers should explore the walking gait characteristics of children with CHARGE syndrome and the impact of various constraints on acquisition of independent walking gait. Based on the findings of this investigation, majority of children with CHARGE syndrome walked with a wide and wobbly gait pattern as

reported by parent perspectives, with many never achieving a mature form of independent walking.

3. Future researchers should examine the possible predictive ability of vestibular function test results on motor performance in children with CHARGE syndrome within independent walking. Vestibular test abnormalities are higher in individuals with sensorineural hearing loss. Within this present investigation, 43 of the 62 (69.4%) participants had sensorineural hearing loss.

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APPENDIX A

Adapted Physical Activity Taxonomy Evaluations

Table 1

Newborn Stepping: An Explanation for a "Disappearing" Reflex (Thelen & Fisher, 1982).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation B	Quantitative Correlation	Eight infants Age: 2 weeks Gender: 4 females, 4 males	To identity the difference between infant stepping and kicking and to show those biomechanical demands can affect infant stepping and kicking. Specific focus: N/A*	There was no difference between the rate of stepping or kicking in the 8 infants and there was no significant correlation (r = .639) between stepping and kicking rates. The stepping reflex and kicking both displayed alternative left and right movements of the legs; however, rate, alternations, and laterality did show some individual variability. The investigators concluded that the disappearance in stepping and increase in supine kicking as the infant ages was due to gravity and the increase mass of the legs which altered the dynamics of the leg movements.

Note. * = Not applicable (i.e., no keywords or specific focus was provided in the article)

Table 2

The Relationship Between Physical Growth and a Newborn Reflex (Thelen, Fisher, & Ridley-Johnson, 1984).

Strength Level & Recommendation Level	**Research Method**	**Population**	**Purpose**	**Summary of Results**
Level 3 Recommendation B	Quantitative Repeated Measures ANOVA Regression Dependent t-test	Study 1: 40 infants Age: 2 weeks Gender: 20 females, 20 males Study 2: 12 infants Age: 4 weeks Gender: 9 females, 3 males Study 3: 12 infants Age: 4 weeks Gender: 6 female, 6 male	To examine the relationship between growth and the stepping response (Study 1). To examine of manipulation of the leg mass would affect infant stepping (Study 2 & 3). Specific focus: Stepping reflex, somatic growth, newborn reflex, and body build.	Study 1: Repeated measures ANOVA reported a significant age effect for all variables except arousal state. A multiple regression analysis was performed at each of the three age levels (i.e., 2, 4, and 6 weeks). Arousal state was a significant predictor of the amount of steps the infant took at each level (2 weeks: F [1, 35] = 4.65, $p < .05$; 4 weeks: F [1, 38] = 11.8, $p < .01$; 6 weeks: F [1, 38] = 18.4, $p < .001$). In addition, infants that gained weight rapidly between 2 and 4 weeks stepped less (r = -.334, $p < .01$). Study 2: The infants at 4 weeks of age averaged 14.17 steps in the no weight condition; however in the weight added condition the infants steps declined significantly ($p < .01$) to 9.58 steps. Study 3: The average number of steps in the out of water condition was 10.41 which significantly ($p < .05$) increased to 20.27 steps during the in water test condition. Arousal scores between both conditions were not different. The explanation of the disappearing infant step reflex was therefore caused by changes in body build and muscular strength.

Table 3

Treadmill-Elicited Stepping in Seven Month-old Infants (Thelen, 1986).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation A	Quantitative Repeated Measures ANOVA	Six infants, one control female adult Age: 7 months Gender: 3 females, 3 males	To examine the ability to generate alternating stepping in seven month old infants. Specific focus: Infants, human, and locomotion.	Repeated measures ANOVA reported a significant effect of condition on kick or step rate ($p < .03$). Movements in both legs significantly increased ($p < .001$) on a moving treadmill when compared to supine position and stepping with no treadmill. In addition, an increase in treadmill speed displayed a significantly ($p < .02$) increase in the frequency of steps. The treadmill also significantly ($p < .001$) increased the percentage of alternations and laterality preference. However, the speed of the treadmill had no effect. Therefore, when 7-month old infants were placed on a treadmill; complex coordination of alternating stepping was performed. But, when the infants were in supine position or an upright position with no treadmill conditions, the participants displayed fewer steps and movements, which were asymmetrical, fluctuating, and uncoordinated. Therefore, the underlying mechanism of the stepping reflex did not disappear.

Table 4

Hidden Skills: A Dynamic Systems Analysis of Treadmill Stepping During the First Year (Thelen & Ulrich, 1991).

Strength Level & Recommendation Level	**Research Method**	**Population**	**Purpose**	**Summary of Results**
Level 3 Recommendation A	Quantitative *Bayley Scales of Infant Development* Longitudinal Repeated Measures ANOVA	Nine infants, full term without any disabilities Age: 1 month to 10 months	To examine the acquisition of independent walking through treadmill stepping. Specific focus: N/A	Repeated measures ANOVA reported a significant increase in the number of steps with age and increased treadmill speed. Descriptive information of alternating steps in the nine infants displayed an initial phase of limited steps, followed by a short period in which the amount of steps declined then followed by a steady increase. The overall rate of general motor maturation (i.e., *Bayley Scales*); developmental changes in the proportion and composition of the infant's legs; difference in arousal and mood; and changes in the predominant postures and movements of the legs all contributed to the treadmill stepping results.

Note. * = Not applicable (i.e., no keywords or specific focus was provided in the article)

Table 5

The Role and Interaction of Visual and Auditory Afferents in Postural Stability (Palm, Strobel, Achatz, von Luebken, & Friemert, 2009).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation B	Quantitative ANOVA	23 physically active soldiers Age: average 24 years of age Gender: 12 female, 11 male	To analyze the role of the visual and auditory systems in the maintenance of postural stability and to assess potential interaction between the two sensory systems. Specific focus: Postural control, visual system, auditory system, feedback, and posturography.	Auditory input (i.e., music) did not result in significant differences in the overall stability scores during the three different visual conditions: eyes closed: $p = .429$, sway feedback: $p = .797$, and eyes open no feedback: $p = .601$. Regardless of the auditory input, there were significant differences between each visual condition based on overall stability; the sway-feedback and eyes-open tests ($p < .05$), between the eyes-open and eyes-closed tests ($p < .001$), and between the sway-feedback and eyes-closed tests ($p < .001$). The overall postural stability was significantly worst in the eyes-closed than in the eyes-open condition and balance control was significantly improved with visual feedback provided in the sway feedback condition. Therefore, postural stability depended on vision and visual feedback.

Table 6

The use of Somatosensory Information During the Acquisition of Independent Upright Stance (Barela, Jeka, & Clark, 1999).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation B	Quantitative MANOVA Correlation	5 infants Age: Approximately 42 weeks old Gender: 1 female, 4 male Setting: Laboratory	To analyze the role of the visual and auditory systems in the maintenance of postural stability and to assess potential interaction between the two sensory systems. Specific focus: Postural, development, somatosensory, prospective, and infancy.	Multivariate analysis of variance resulted in a significant ($p < .001$) difference between the mean force applied throughout the four developmental periods. However, univariate analysis of variance only reported significant differences for medial-lateral force ($p < .01$) and vertical force ($p < .0001$). Applied force in the medial-lateral direction decreased from pull to stand to stand alone, and to walking onset. Applied force in the vertical direction decreased throughout each of the four developmental periods. In addition, medial-lateral and anterior-posterior body sway significantly decreased at the post walking period compared to walking alone and pull to stand. Therefore, initially infants use surface contact for mechanical purposes then later for orientation information to assist posture control.

Table 7

Sensory Information Affords Exploration of Posture in Newly Walking Infants and Toddlers (Metcalfe & Clark, 2000).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 2 Recommendation A	Quantitative MANOVA Correlation	13 infants Age: 13 to 24 months Gender: 7 female, 6 male Setting: Laboratory	To examine the role of sensory information in postural control within newly walking infants. Specific focus: Posture, development, perception action, exploration, and infancy.	When the infants used surface contact there was a significant decrease in head ($p < .01$) and shoulder ($p < .01$) sway with an effect size of 1.5. Overall, center of mass sway was also reduced ($p < .05$). Within the three walking age groups (i.e., 1-4; 5-8; and 9+ months of walking), the use of surface touch increased the postural coordination variability. Therefore, infants use somatosensory information to aid in upright postural control.

Table 8

Comparison of Balance and Gait in Visually or Hearing Impaired Students (Uysal, Erden, Akbayrak, & Demirtürk, (2010).

Strength Level & Recommendation Level	**Research Method**	**Population**	**Purpose**	**Summary of Results**
Level 3 Recommendation C	Quantitative ANOVA Mann-Whitney *U* Test	60 children (20 with hearing loss, 20 with vision loss, and 20 with hearing and vision) Age: average 10.2 years old Gender: 28 female, 32 male Setting: School	To examine the effect of congenital loss of hearing and sight on gait and balance in children. Specific focus: N/A	In all balance and gait analysis, the group with hearing loss performed significantly ($p < .05$) better than the group with vision loss. The control group performed significantly ($p < .05$) better in all the balance and gait tests when compared to the vision loss group and all but the gait speed when compared to the hearing loss group. Therefore, the children with vision loss had more difficulties with balance and gait than the children with hearing loss and the controls.

Note. * = Not applicable (i.e., no keywords or specific focus was provided in the article)

Table 9

The Relationship Between Transitional Motor Skills and Locomotion (Looper, Talbot, Link, & Chandler, 2015).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation C	Quantitative Correlation	8 children Age: 6 months Gender: 3 female, 5 male Setting: Home	To determine if transitional movements are correlated with locomotion. Specific focus: Transitional skills, walking, crawling, cruising, and sitting.	Pearson product moment correlation was used to determine the correlation between transitional skills and locomotor skills and between sitting and locomotor skills. Four of the five transitional skills (i.e., rotating to sitting, pulling to stand, squatting to stand, and squatting to the floor) displayed strong significant ($p < .05$) correlations to all three locomotor skills. Independent rolling and sitting were not significantly correlated to locomotor skills. Therefore, transitional skills assist infant's pelvic and trunk control needed for upright locomotion.

Table 10

Prediction of Infant's Motor Development (Charitou, Asonitou, & Koutsouki, 2010).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation B	Quantitative *AIMS* Regression	46 infants Age: approximately 10 months Setting: Greece	To predict motor development using indications from infant's early motor performance. Specific focus: Prediction, infants, motor development, *AIMS*, and motor skill.	The four subscales of the *AIMS** were significantly different ($p < .05$) from the total *AIMS* score in both the 6 month and 2 month reassessment groups. Stepwise regression showed that supine position ($r = .295$) could predict infant development within the group that was reassessed after 6 months. In the group that was assessed after 2 months, supine and prone positions ($r = .929$) were significant predicting factors of motor development.

Note. ***** = Alberta Infant Motor Scale

Table 11

How do you learn to walk? Thousands of steps and dozens of falls per day (Adolph et al., 2012).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation C	Quantitative ANOVA Correlation	151 infants Age: 11.8 to 19.3 months Gender: 72 females, 79 males Setting: Laboratory playroom and home	To examine natural infant locomotion. Specific focus: Infant development, learning, motor processes, and perceptual motor coordination.	The result of functional skills of 12 month old crawlers and walkers were that novice walkers feel significantly ($p < .02$) more times per hour than expert crawlers. However, walkers spent significantly more time in motion ($p < .01$), accumulated more steps ($p < .01$), and travelled greater distances ($p < .01$) than crawlers. Fall rate also decreased in walkers when the increase in activity and movement was examined. Walking skills (i.e., step length and step width) were significantly ($p < .01$) correlated to test age and walking age. In that, older infants who had more walking experience took significantly ($p < .01$) longer and narrower steps. Older infants also spent more time walking, took more steps, and fell less (all $p < .01$). Therefore, the benefits of walking outweigh the benefits of crawling in infants.

Table 12

Head-Mounted eye Tracking: A new Method to Describe Infant Looking (Franchak, Kretch, Soska, & Adolph, 2011).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation C	Quantitative ANOVA	6 infants Age: 14 months Gender: 3 females, 3 males Setting: Laboratory playroom	To examine visual behavior during natural interactions in mobile infants. Specific focus: N/A	Infants shifted gaze to their mother 53.9% after mothers' utterances. Infant response fixations to mothers were, 50% directed at mothers' body, 33.8% at their hands, and only 16.2% at the mothers' face. Repeated-measures ANOVA on the duration of fixations to the three parts of mothers' bodies did not reveal an effect of gaze location on fixation duration (p = .61). Nor did objects in mothers' hands increase duration of fixation to their hands (p = .48). Infants' prospective fixations of objects and obstacles were significantly (p = .001) different for reaching, crawling, and walking. Differences in fixation rates between walking, crawling, and reaching were based on physical constraints (i.e., hand and head positions). Therefore, visual exploration is dependent on the availability of information and the constraint of the infant's body.

Note. * = Not applicable (i.e., no keywords or specific focus was provided in the article)

Table 13

Crawling and Walking Infants see the World Differently (Kretch, Franchak, & Adolph, 2014).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation C	Quantitative GEEs Wald Chi-squared test	Study 1: 30 infants (15 crawlers, 15 walkers) Age: 13 months Gender: 15 females, 15 males Study 2: 13 infants (9 crawlers, 4 walkers) Age: 13 months Setting: Laboratory	To investigate whether there are differences in visual input between crawling and walking and which physical factors contribute to these potential differences. Specific focus: N/A	In study 1, 25.7% of steps, crawlers view contained only the floor for the entire scene video. Therefore, while crawling, infants may miss visual input from distal parts of the room around them. The highest point of visibility was significantly ($p < .01$) greater for walking infants than those who crawled. The closest point of visibility for crawlers were closer to the crawler's hands ($M = 20.89$ cm) than to the walker's feet ($M = 83.00$ cm). However, sitting significantly ($p < .01$) increased the infant's point of visibility and was more likely to be able to view their caregiver's face and toy. In study 2, crawlers displayed significantly ($p < .01$) more variability in head position and larger deviations in head pitch angle between consecutive steps than did the walkers. Therefore, visual input is impacted by the infant's posture.

Note. * = Not applicable (i.e., no keywords or specific focus was provided in the article)

Table 14

Transition from Crawling to Walking and Infants' Actions with Objects and People (Karasik, Tamis-LeMonda, & Adolph, 2011).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation C	Quantitative Factorial ANOVA Correlation	50 infants Age: 11 months Gender: 24 females, 26 males Setting: Home with natural parental interactions	To examine possible functional relations between the transition from crawling to walking and infants' object interactions. Specific focus: N/A	There were no significant differences between the crawler-crawlers and crawler-walkers on the amount of time spent with objects during either session (11 months or 13 months). At 11 months, the two groups significantly ($p < .01$) differed in engagement with distal objects. By 13 months, walkers were three times more likely to travel to objects to interact with them compared to crawlers ($p < .001$). In both groups, infants significantly ($p < .01$) increased in the frequency of carry objects; however the increase was significantly ($p < .01$) larger in the crawler-walker group. The amount of distal accessed objects and amount of carry objects at 11 months significantly ($p < .01$) predicted walking status at 13 months. Therefore, earlier object activities predicted walking status of infants.

Note. * = Not applicable (i.e., no keywords or specific focus was provided in the article)

Table 15

Overcoming the Barriers to Including Students with Visual Impairments and Deaf-blindness in Physical Education (Lieberman & Houston-Wilson, 1999).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation A	Quantitative Repeated Measures ANOVA	170 general physical education teachers Setting: Inclusion Physical Education	To examine major barriers that impede the inclusion of students with visual impairments in physical education and provide strategies based on those barriers. Specific focus: Physical education, people with visual disabilities, and training.	The following barriers were reported: lack of professional preparation; curriculum and activity instruction and inclusion; pace of lesson; limited expectations; parent overprotection; lack of opportunities and confidence; time; and lack of appropriate equipment.

Table 16

Barriers Perceived to Including Students with Visual Impairments in General Physical Education (Lieberman, Houston Wilson, & Kozub, 2002).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation A	Quantitative Correlation Logistic Regression Survey	148 physical education teachers Gender: 96 females, 52 males Setting: New York, Inclusion Physical Education	To examine barriers perceived by general physical education teachers when including students with visual impairments. Specific focus: N/A	In this investigation, the perceived barriers recorded by physical educators prior to a professional development workshop were professional preparation, equipment, programming, and time. Logistical regression did not indicate any significant predictors of professional preparation as a barrier ($p > .05$).

Table 17

Supporting Students with Visual Impairments in Physical Education: Needs of Physical Educators (Conroy, 2012).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 2 Recommendation A	Qualitative Interview	25 physical education teachers Age: 24 to 62 years old Experience: 3 to 26 years Setting: Inclusion Physical Education	To examine how physical education teachers support the inclusion of students with visual impairments and identify instructional needs and strategies. Specific focus: Inclusion, physical education, and visual impairments.	Themes emerged from the data that included successes, challenges, and needs. The physical educators felt they did not have adequate background information about the developmental level of the student with the visual impairment, which lead to difficulties in selecting the amount of support and type of instruction to provide. All participants in this investigation reported the need for more training.

Table 18

Play and Recreational Habits of Youths who are Deaf-Blind (Liberman & MacVicar, 2003).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation A	Mixed Design Descriptive Information Qualitative Themes Questionnaire	52 families of youth who were deafblind Age: 3 to 22 years old Gender: 20 female, 34 male	To examine the play and recreational habits of children who were deafblind. Specific focus: In infancy and childhood: blindness, deafness, and recreation.	The top five recreational activities selected by the youth who were deafblind in this investigation (n = 51) were swimming (57%), swinging or rocking (33%), walking (31 %), climbing equipment (24%), and biking (24%). The parent's satisfaction for physical education and recreational experiences were between "neutral and "not very satisfied on a 5-point Likert scale. The most common barrier (33%) expressed by the parents was related to the disability of deafblindness. The investigators believed the additional barriers were present due to the previous most common barrier. These barriers included lack of knowledge about specific adaptation (31%), appropriate programming ideas (25%), adequate staff that were knowledgeable and willing to work with children who were deafblind (25%), adequate communication (16%), time (12%), money (8%), accessibility (4%), and transportation (4%).

Table 19

Gross Motor Skill Performance in Children with and Without Visual Impairments – Research to Practice (Wagner, Haibach, & Lieberman, 2013).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 1 Recommendation A	Quantitative *TGMD-II** Quasi-Experimental Mann-Whitney *U* Test	23 children with visual impairments and 28 children with vision as control participants Age: 6 through 12 years old Gender: 9 female and 14 male children who were blind and 13 female and 15 male children with vision	To determine if there are differences in gross motor skill performance between children with and without visual impairments. Specific focus: Gross motor skill performance, visual impairments, children who are blind, *TGMD-II*, and whole-part-whole method.	Children with visual impairments had significant ($p < .05$) deficits in performing locomotor and object control skills when compared to sighted peers. Running, leaping, kicking, and catching were the most affected skills.

*Note. * = Test of Gross Motor Development: Second Edition*

Table 20

Determinants of Gross Motor Skill Performance in Children with Visual Impairments (Haibach, Wagner, & Lieberman, 2014).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 1 Recommendation A	Quantitative *TGMD-II** Quasi-Experimental Generalized linear model Pairwise Comparisons	100 individuals with visual impairments from seven states Age: 6 through 12 years old Gender: 39 female, 61 male	Assess gross motor skills performance in children with VI based on their age, gender, and level of visual impairment. Specific focus: Gross motor skills, children, visual impairment, blind, age, and gender.	Severity of visual impairment was a significant ($p < .05$) factor in all assessed motor skills, including locomotion and object control skills. B1 group performed significantly ($p < .05$) lower than B2 or B3 group in nearly all skills. Age and gender were not significant factors ($p > .05$).

*Note. * = Test of Gross Motor Development: Second Edition*

Table 21

The Motor Development of Sighted Children and Children with Moderate Low Vision aged 8-13 (Bouchard & Tétreault, 2000).

Strength Level & Recommendation Level	**Research Method**	**Population**	**Purpose**	**Summary of Results**
Level 3 Recommendation A	Quantitative *BOTMP** Pearson correlation T-tests Regression	30 children with low vision and 30 children with vision Age: 8 to 13 years of age Setting: Quebec	To examine the motor development of children with low vision compared to children with vision and identify factors that influence motor development. Specific focus: Vision and motor abilities in children.	Participants with low vision obtained independent siting (p = .001), standing (p = .001) and walking (p = .01) significantly later than participants with vision. Children with vision loss obtained significantly ($p < .0001$) lower scores in six of the eight subtests in the *BOTMP*. The balance subtest score performance was significantly lower (children with low vision, age equivalence of 80.5 months, SD = 28.7; children with vision, age equivalence of 162.4 months, SD = 32.4 months) than all other subtests when the children with vision loss were compared to the children with vision.

*Note. * = Bruininks-Oseretsky Test of Motor Proficiency*

Table 22

Divergent Development of Gross Motor Skills in Children who are Blind or Sighted (Brambring, 2006).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 2 Recommendation B	Quantitative Regression analysis	4 children who were congenitally blind Gender: 2 female, 2 male Setting: Germany	A longitudinal assessment of the age at which children who were blind acquire 29 motor skills over 4 years compared to children with vision. Specific focus: N/A	The mean developmental delay in children who were blind was 11.9 months at 30 months when compared to children with vision. A high degree in variability in relative developmental differences was reported. The median age of walking three steps was 16.5 months in children who were blind compared to 12.4 months for children with vision. Children who were blind compared to the children with vision revealed significant developmental delays in the total comparison ($Z = -5.199$, $p < .001$), dynamic balance ($p < .05$), attainment of locomotion ($p < .05$), and refinement of locomotion ($p < .01$). A regression analysis of the acquisition ages of children who were blind and children with vision displayed a high correlation ($r = .89$), meaning the sequence of developmental skills were acquired sequentially. Results indicated distinct developmental motor delays in children who were blind.

Table 23

Longitudinal Study of Gross-Motor Development in Blind Infants and Preschoolers (Tröester, Hecker, & Brambring, 1994).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation C	Quantitative Descriptive Information	10 infants who are blind Subgroup: 5 born full term and 5 born preterm Age: 7.5 to 16 months Gender: full term: 2 female and 3 male; preterm: 3 female and 2 male Setting: Germany	To examine the impact of blindness and prematurity on motor development in infants who are blind. Specific focus: N/A	Differences between full term and preterm infants who were blind were larger than differences between full term infants who were blind and infants with vision. The full term infants who were blind walked three steps unassisted at a median age of 15.5 months compared to preterm infants who were blind which performed the skill at a median age of 26.5 months. The five preterm infants who were blind demonstrated only slight delays in postural development, but greater delays in locomotor development (i.e., walking unassisted 3 steps).

Table 24

Early Motor Development in Blind Infants (Tröester & Brambring, 1993).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 2 Recommendation B	Quantitative *Bielefeld Developmental Test for Blind Infants and Preschoolers* Descriptive Information Mann-Whitney *U* Test	21 infants who were blind and control group of 47 infants with vision Age: 9 months or 12 months Gender: infants who were blind aged 9 months: 2 females and 3 males; 12 months: 9 females and 7 males; infants with vision aged 9 months: 11 females and 12 males; 12 months: 12 females and 12 males	To gain information on the effects of blindness on particular areas of motor development compared to age and gender matched infants with vision. Specific focus: Blindness, congenital, psychomotor development, oculomotor coordination, age, posture, locomotion, manual aptitude, vision disorder, eye disease, infant, motor control, and human.	Infants who were blind and preterm, with age adjusted for prematurity, did not differ significantly in blind-specific motor skills of locomotion ($p > 0.10$) when compared to infants who were blind and full term. In contrast when compared to infants with vision, both age groups of infants who were blind were significantly delayed in locomotion. The delay in posture control (i.e., prone, supine, sitting, standing) in infants that were blind was considered to result from the lower level of motor stimulation due to the lack of sight. Therefore, the delay in locomotor skills in children who were blind was due to their inability to replace visuomotor coordination with audioproprioceptive coordination and control of movement.

Table 25

A Survey of Motor Development for Infants and Young Children with Visual Impairments (Celeste, 2002).

Strength Level & Recommendation Level	**Research Method**	**Population**	**Purpose**	**Summary of Results**
Level 2 Recommendation A	Quantitative Descriptive Information Parent Survey	84 families of children with visual impairments Subgroups: premature birth, additional disabilities, and visual functioning Age: 4 months to 4 years old Gender: 45 female, 39 male Setting: Germany	To gather descriptive information about demographics, birth history, etiology and visual functioning, early intervention services, and gross motor development of children with visual impairments. Specific focus: Infant development; child development motor skills - in infancy and childhood; blindness - in infancy and childhood; vision, and subnormal - in infancy and childhood.	Based on the results, 50% (n =12) walked independently between 37 to 48 months with an additional 31.3% (n = 5) not reaching this milestone until later than 48 months old. Therefore, the children with the least vision had the poorest gross motor outcomes related to walking independently.

Table 26

Development of Independent Locomotion in Children with a Severe Visual Impairment (Hallemans, Ortibus, Truijen, & Meire, 2011).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation B	Quantitative Mixed Design ANOVA Regression	31 children and adults who had a visual impairment and 60 age-related individuals with vision Age: 1 to 44 years old Setting: Belgium	To describe age-related changes in gait in individuals with a visual impairment. Specific focus: Gait, child, adult, development, step-time parameters, low vision, and blind.	An increase in age resulted in overall improvements to gait parameters. Individuals with a visual impairment displayed a slower walking speed ($p < .002$), a shorter stride length ($p < .001$), a prolonged duration of stance phase duration ($p < .001$), and double support during the gait pattern ($p < .001$). Participants who were blind walked at a slower speed ($p < .001$), displayed a shorter stride length ($p < .001$), and had a prolonged duration of stance when compared to both the control and group with low vision. These gait adaptations may be due to balance difficulties or are used as a strategy to gain more environmental input.

Table 27

Early Motor Development of Blind Children (Levtzion-Korach, Tennenbaum, Schnitzer, & Ornoy, 2000).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation B	Quantitative One-sample *T*-test Two-sample Unpaired *T*-test	40 children who were blind and 24 children with vision as the control group Age: under 5 years of age for children who were blind and under 18 months for children with vision Setting: Israel	To assess the characteristic motor development pattern in children who were blind from Israel. Specific focus: Blind children, motor delay, and motor development.	The children who were blind were reported to be significantly delayed ($p < .001$) in all motor skills based on the standard milestone ages for children with vision. Children who were blind were significantly delayed in the pre-walking motor skills except for the skill of sitting from a supine position when compared to control participants and significantly delayed in all post-walking motor skills.

Table 28

Spatial Concepts and Balance Performance: Motor Learning in Blind and Visually Impaired Children (Pereira, 1990).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation B	Quantitative Multivariate analysis Pre and post test	67 children with visual impairments and 150 children with vision Age: 50 children with visual impairments were aged between 6 and 10 years old and 17 between 11 and 13 years old; children with vision were aged between 6 and 10 years old	To evaluate positional concepts and balance performance in children who were visually impaired, evaluate important conditioning factors, and determine the effect of two pedagogical approaches. Specific focus: Blindness, elementary education, instructional effectiveness, motor development, perceptual motor learning, psychomotor skills, skill development, spatial ability, teaching methods, and visual impairments.	Positional concept results were independent of visual acuity and field loss; however, balance performance is related to low visual acuity and a reduction in visual fields. Conditioning factors depended on descriptive positive reinforcement from the teacher, a variety of teaching cues, and the child's engagement and time on task. In addition, the pedagogical approach that focused on stronger cognitive appeal proved to be more effective in improving motor development.

Table 29

Effect of Vestibular Dysfunction on the Development of Gross Motor Function in Children with Profound Hearing Loss (Inoue et al., 2013).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation C	Quantitative Mann-Whitney *U* Test Kruskal-Wallis Test	89 children with profound sensorineural hearing loss Age: 20 to 97 months Gender: 44 females, 45 males Setting: Japan, Tokyo: tertiary referral center	To examine the effect of vestibular dysfunction on the development of gross motor function in children with profound hearing loss. Specific focus: Vestibular evoked myogenic potential, caloric test, rotational test, and gross motor development.	Based on the results, it was indicated that the age of independent walking was significantly delayed in the group of participants with combined superior and inferior dysfunction ($p < .01$) and in the group of participants with inferior dysfunction ($p < .05$) when compared to children with normal vestibular function. The age of independent walking was significantly delayed (> 18 months) in the combined superior and inferior dysfunction group and in the inferior dysfunction group compared with the normal group. Therefore, children with profound hearing loss usually have vestibular dysfunction, which impacts the child's gross motor development.

Table 30

The Influence of a Vestibular Dysfunction on the Motor Development of Hearing-Impaired Children (De Kegel, Maes, Baetens, Dhooge, & Van Waelvelde, 2012).

Strength Level & Recommendation Level	**Research Method**	**Population**	**Purpose**	**Summary of Results**
Level 1 Recommendation C	Quantitative *M ABC-2** Mann-Whitney *U* Test ANCOVA Bivariate and Multivariate Regression	41 children with no hearing loss and 48 children with hearing loss Age: 3 to 12 years old Gender: children with no hearing loss: 22 females and 29 males; children with hearing loss: 23 females and 25 males Setting: Belgium	To identify the predictive ability of vestibular function along with other factors on motor performance of children with hearing loss. Specific focus: Balance performance, vestibular function testing, and hearing impaired children.	Children in the study who had an absence of the VEMP** response performed significantly worse on static balance (p = .003) and had a larger postural instability (i.e., one leg stance with eyes closed, p = .013) and bipedal stance on a cushion with eyes closed (p = .008) than children with hearing loss that had a present VEMP response. The three most important predictor variables on motor performance by bivariate regression analyses are the vestibular-ocular reflex gain value of the rotatory chair test at 0.01 and 0.05 Hz frequency, and the VEMP asymmetry ratio. Therefore, children with hearing loss are at risk for balance difficulties in which VEMP testing can provide prediction of their balance performance.

Note. ***** = Movement Assessment Battery for Children- 2nd Edition, ****** = Vestibular Evoked Myogenic Potential test

Table 31

Balance Sensory Organization in Children with Profound Hearing Loss and Cochlear Implants (Suarez, Angeli, Suarez, Rosales, Carrera, & Alonso, 2007).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation C	Quantitative Kruskal-Wallis Test Wilcoxon Rank Sum	36 children with sensorineural hearing loss and 22 children with no hearing loss Age: 8 to 11 years old	To examine how deaf children use sensory information for postural control, with normal or abnormal vestibular responses. Specific focus: Postural control, cochlear implant, and vestibular plasticity.	The sway velocity and center of pressure distribution were higher in Condition 2 compared to Condition 1 in all participants. However, those with a hypoactive vestibular system (n = 8) had significantly higher value in sway velocity and center of pressure distribution in Condition 2 than those with a profound sensorineural hearing loss with normal vestibular function and those with no hearing loss. In addition, there was no significant difference in postural control with the cochlear implant turned on or off in Condition 2. Therefore, children with hearing loss primarily use visual and somatosensory information for postural control.

Table 32

Evidence of Progressive Delay of Motor Development in Children with Sensorineural Hearing Loss and Concurrent Vestibular Dysfunction (Rine, Cornwall, Can, LoCascio, O'Hare, Robinson, & Rice, 2000).

Strength Level & Recommendation Level	**Research Method**	**Population**	**Purpose**	**Summary of Results**
Level 2 Recommendation B	Quantitative ANOVA *T*-test Repeated Measures ANOVA Pearson Correlation	39 children with sensorineural hearing loss Age: 26 to 83 months Gender: 15 females, 24 males	To examine motor proficiency of children with sensorineural hearing loss. Specific focus: N/A	Regardless of age, children with sensorineural hearing loss scored significantly lower ($p \leq .01$) on the Gross Motor Scale and on the Locomotor category. There was no correlation found between vestibular function and z scores on the *Peabody Developmental Motor Scales* or skill categories. However, vestibular function did identify as a significant predictor of children with a delay in motor and/or balance development.

Table 33

Age Related Balance Changes in Hearing-Impaired Children (Siegel, Marchetti, & Tecklin, 1991).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation B	Quantitative *Z*-Test ANOVA *T*-Test	28 children with sensorineural hearing loss (> 65dB) Age: 14 to 10 years old Setting: School for the Deaf	To compare the scores on a standardized balance test of three age groups of children who are deaf with those of a sample of children with hearing. Specific focus: Child development, equilibrium, hearing disorders, posture, tests and measurements, and vestibular system.	The mean balance score for all three groups was significantly ($p < .05$) lower compared with normative value. ANOVA with Scheffè follow up displayed a significant ($p < .01$) difference between groups 1 and 2 and between groups 1 and 3 on mean balance scores. A *t*-test revealed no significant difference based on gender. However, age showed an effect on balance scores with older groups having significantly higher scores than the younger groups.

Note. dB = decibel

Table 34

Motor Development of Children who are Deaf (Dummer, Haubenstricker, & Stewart, 1996).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation B	Quantitative *TGMD** ANCOVA Partial Correlation	201 children with hearing loss (>55dB) Age: 4 to 18 years old Gender: 91 females, 110 males Setting: School for the Deaf	To examine the motor development of children who were deaf. Specific focus: N/A	Based on the results of the ANCOVA, age (in months), which was the covariate, was significant in performance of both the locomotor skills, F(l, 192) = 155.95, $p < .05$, and the object-control skills, F(1, 190) = 334.96, $p < .05$. Therefore, the mean scores on the *TGMD* increased as chronological age increased. Another ANCOVA resulted in age accounting for the most variance on the throw for distance, F(1, 127) = 747.04, $p < .05$, kick for distance, F(1, 156) = 274.54, $p < .05$, jump for distance, F(l, 183) = 414.33, $p < .05$, and 15-yard dash, F(1, 190) = 415.85, $p < .05$. Based on the norms of the *TGMD*, it was reported that children who are deaf demonstrated delays of 1 to 3 years in the acquisition of object-control skills. However, only the locomotor skills of skipping and leaping were performed at a mastery level at later ages than those without hearing loss when compared to the norms of the test.

Note. * = *Test of Gross Motor Development,* dB = decibel

Table 35

Balance and Gait Evaluation: Comparative Study Between Deaf and Hearing Students (Melo, Silva, Tassitano, Macky, & Silva, 2012).

Strength Level & Recommendation Level	**Research Method**	**Population**	**Purpose**	**Summary of Results**
Level 3 Recommendation B	Quantitative *Tinetti scale* *"Up and Go" test* *T*-Test Chi-Square Test	88 children (44 with sensorineural hearing loss) Age: 7 to 17 years old Gender: 44 females, 44 males Setting: Brazil schools	To evaluate the body balance and gait of deaf and hearing students and to compare data between groups, while considering gender and age. Specific focus: Child, postural balance, gait, hearing loss, sensorineural, and deafness.	In the *Tinetti scale* results for balance, the children who were deaf had no significant difference ($p = .47$) in performance compared to the children with hearing. However, in the *Tinetti's* gait characteristics the two groups significantly ($p < .001$) differed, with the children with hearing loss performed significantly lowered than the children with hearing, even when separated by gender. In the *"Up and Go" test*, only the 7 to 10 year old group of children with hearing loss performed significantly ($p < .001$) worse than the children with hearing. When compared by gender, both male groups did not differ but there was a significantly ($p = .027$) difference with female children with hearing performing better than female children with hearing loss. Therefore, children with hearing loss displayed difference in gait and had an increased risk for falling.

Table 36

Cochlear Implant Outcomes in Children with Motor Developmental Delay (Amirsalari et al., 2012).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation B	Quantitative *T*-Test Chi-square Test Wilcoxon and Mann Whitney *U* Test	262 children with sensorineural hearing loss (28 with motor developmental delay) Age: 1 to 9 years old Gender: 133 female, 129 male Setting: Iran	To examine the effects of motor developmental delay on the outcome of cochlear implants. Specific focus: Motor developmental delay, cochlear implant, and sensorineural hearing loss.	The results of the speech intelligibility ratings and the auditory perception scales were not significantly different between the group with normal motor development and the group with delayed motor development. The mean auditory perception scale score after surgery for children with delayed motor development was 5.03 and was 5.77 for children with normal motor development. The mean speech intelligibility ratting score after surgery for children with delayed motor development was 2.53, and was 2.66 children with normal motor development. Therefore, children with or without motor developmental delay can benefit from cochlear implantation.

Table 37

Motor Developmental of Deaf Children with and without Cochlear Implants (Gheysen, Loots, & Waelvelde, 2008).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation B	Quantitative *M-ABC* *KTK* *T*-Test Kruskal-Wallis Test	36 children with hearing loss (20 with cochlear implants) 43 children with hearing as control Age: 4 to 12 years old Gender: Children with hearing loss: 21 female, 15 male; children with hearing: 28 female, 15 male Setting: Belgium mainstreamed education	To examine the impact a cochlear implant has on motor development of children who were deaf. Specific focus: N/A	Motor performances of children with hearing were significantly ($p < .008$) better for the total score of the *M-ABC** and all scales of the one-leg standing test. Children with hearing had better manual dexterity, ball skills, and dynamic and static balance on the scales of the *M-ABC*. Children who were deaf displayed significantly ($p < .008$) lower balancing scores when standing on one leg, with eyes open and closed, compared to children with hearing. Only one significant ($p = .033$) difference was reported between the group of children with and without cochlear implants, which was walking backward on beams in the *KTK*** test. Therefore, cochlear implantation had no significant impact on motor development.

*Note. * = Movement Assessment Battery for Children (Dutch Version), ** = Körperkoordinationstest für Kinder*

Table 38

Comparing Motor Development of Deaf Children of Deaf Parents and Deaf Children of Hearing Parents (Lieberman, Volding, & Winnick, 2004).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation B	Quantitative *TGMD* *T*-Test Factorial ANOVA	29 children (14 with Deaf parents) Age: 4 to 9 years old Gender: 11 females, 18 males Setting: New York Deaf schools	To compare the motor development of Deaf children of Deaf parents with that of Deaf children of hearing parents. Specific focus: N/A	There was no significant ($p > .05$) difference between the two groups on the *TGMD* locomotor and object control subtest. A factorial ANOVA displayed a significant main effect ($p < .05$) on both the locomotor and object control subtests and no significant main effect of hearing status of parents. Therefore, the hearing status of their parents does not affect motor development in children who are deaf.

Table 39

The Effect of Interventional Proprioceptive Training on Static Balance and Gait in Deaf Children (Majlesi, Farahpour, Azadian, and Amini, 2014).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 2 Recommendation A	Quantitative *T*-Test Repeated Measures ANOVA	20 children (10 with hearing loss) Age: 8 to 14 years Gender: all male Setting: Iran	To examine the effect of a 12 session exercise balance program based on proprioception training on balance and gait in deaf children compared with children with hearing. Specific focus: Deaf children, postural sway, proprioceptive intervention, gait, and balance.	There were no significant differences between the two groups in age, height, weight, or body mass index prior to the intervention. After the intervention, children who were deaf had a significantly (p = .028) decrease in postural sway. The postural sway in all participants were significantly (p < .001) greater in anterior-posterior than medial-lateral. In addition, after the intervention, both groups average sway while standing on the foam block significantly (p = .02) decreased. Gait velocity was not significantly affected after the intervention in the group with hearing loss. Therefore, exercise programs for children with hearing loss can result in enhanced somatosensory ability and improved balance.

Table 40

Improvement of Motor Development and Postural Control Following Intervention in Children with Sensorineural Hearing loss and Vestibular Impairment (Rine et al., 2004).

Strength Level & Recommendation Level	**Research Method**	**Population**	**Purpose**	**Summary of Results**
Level 2 Recommendation A	Quantitative *PDMS* *SCT-3* *T*-Test	21 children with bilateral vestibular dysfunction Age: 3 to 8.5 years Gender: 12 female, 9 male Setting: Miami, School Program for the Deaf	To examine the effect of exercise intervention on motor development and postural control in children with sensorineural hearing loss and vestibular dysfunction. Specific focus: Vestibular, rehabilitation, children, substitutions, developmental delay, and sensory integration.	In the post-intervention testing, those in the exercise intervention group had significant ($p < .05$) improvements in the *PDMS** raw scores and intervention quotient compared to pre-intervention. The post-intervention *PDMS* quotients of the exercise intervention group were significantly ($p = .005$) higher than those of the placebo group. In postural control test, the post-intervention exercise group, not the placebo group, had significant ($p = .03$) improvements in the *SCT-3*** scores but no significant difference in the vision and somatosensory ratio scores. Therefore, exercise interventions focused on sensory integration for children with hearing loss and vestibular dysfunction can result in the arrest of motor developmental delays.

Note. * = *Peabody Developmental Motor Scales,* ** = Sensory Conditions Testing

Table 41

Physical Education and Children with CHARGE Syndrome: Research to Practice (Lieberman, Haibach, & Schedlin, 2012).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation A	Quantitative Descriptive Information Questionnaire	26 parents of a child with CHARGE* syndrome Age: 6 to 19 years old with CHARGE syndrome Setting: International CHARGE Syndrome Conference	To determine the status (i.e., physical education setting, communication, modification, successful and difficult units) of physical education provided to children with CHARGE syndrome. Specific focus: CHARGE syndrome, physical education, adapted evaluation, child, and disabled.	Physical education settings ranged from inclusive (42%) to segregated (23%) and a combination of both (19%). In addition 69% (n = 26) had physical education on their IEP. Communication used during physical education class was primarily American Sign Language (17%). Equipment modification was used most frequent (n =6). Children were most successful in individual motor skills then units and had difficulty with fundamental motor skills. Therefore, physical education placement, communication, and modification should be individualized to each student with CHARGE syndrome.

Note. * = coloboma of the eye, heart defects, atresia of the choanae, retardation of growth and/or development, genital and/or urinary abnormalities, and ear abnormalities and deafness.

Table 42

A Functional Classification System for the Deaf-Blind (Dunlap, 1985).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation C	Quantitative Correlation	251 individuals with deafblindness Age: infant through 21 years old Gender: 48% females, 52% males Setting: six southeastern states	To investigate a classification of individuals with deafblindness by activity skills rather than vision or hearing loss. Specific focus: N/A	A cluster solution of three groups was presented. Four variables were retained out of 11 in the stepwise process: Gross motor, language, leisure, and socialization. Difference in Groups 1 and 2 were physical activities and between Group 2 and 3 were cognitive activities. Therefore, gross motor activity skills may predict the cognitive abilities of individuals who are deafblind.

Table 43

A National Transition Follow-up Study of Youth Identified as Deafblind: Parent perspectives (Petroff, 1999).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 1 Recommendation B	Qualitative Survey	102 parents of individuals with deafblindness Setting: United States	To survey the characteristics and experiences of youth with deafblindness who are recently left school. Specific focus: N/A	Of the youth who were deafblind, 36.6% used a wheelchair and depend on others to be mobile (highest percentage) with the next highest percentage of youth, 30.1 %, walked independently without assistance or supervision. It was reported that majority of youth who are deafblind were divided into two very different categories. The largest group of the investigation sample fell under the following category: People who did not communicate with language (i.e., signed or spoken) and did not walk independently and people who communicated effectively using spoken and/or sign language and walked independently. Therefore, walking independently is an important characteristic of youth with deafblindness.

Table 44

Characteristics and Development of children with CHARGE Association/Syndrome (Salem-Hartshorne & Jacob, 2004).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation B	Quantitative *ABES* Correlation	100 families with a child who had CHARGE syndrome Age: Children with CHARGE syndrome ranged from 5 to 15 years old Gender: 50 female, 50 male Setting: range of states, including Canada	To examine the developmental outcomes in children with CHARGE syndrome and determine the developmental range and specific factors that are associated with development. Specific focus: N/A	The participant's earlier age of walking had the strongest positive relationship (r = .39; p < .01) to the *ABES* score. In addition, the degree of deafblindness (r = .33; p < .01) and medical involvement (r = .26; p < .01) were correlated with the age at walking. *ABES** score was negatively correlated to late walking age, degree of hearing loss, degree of deafblindness, and medical involvement. Sensory impairment and medical involvement were moderately correlated with age of walking. Therefore, age of walking had the strongest relationship to the *ABES* score of children with CHARGE syndrome.

*Note. * = Adaptive Behavior Evaluation Scale*

Table 45

Executive Function in CHARGE Syndrome (Hartshorne, Nicholas, Grialou, & Russ, 2007).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 2 Recommendation B	Quantitative *BRIEF* *T*-Test Correlation Regression	98 parents of individuals with CHARGE syndrome Age: Individuals with CHARGE syndrome: 5 to 18 years old Gender: 92.9% of the parents were mothers, 59.2% of the children were male	To examine the executive functions in individuals with CHARGE syndrome and how they may relate to behavior. Specific focus: N/A	All participants (n = 98) reported their child with CHARGE syndrome having a delay in motor milestones. The average age of walking was 3.08 years and the average age of crawling was 1.64 years. Age of walking was significantly ($p < .05$) associated with all three indexes on the *BRIEF**. Scores on the *BRIEF* that are 65 or higher are considered potentially clinically significant. Correlation between age of walking, age of crawling, and walking ability with the three indexes (i.e., Behavioral Regulation Index, Metacognition Index, and Global Executive Composite) on the *BRIEF* was performed. As age of walking is more delayed (i.e., age increases), scores on the *BRIEF* indexes increase and are more clinically significant. Therefore, age of walking was significantly ($p < .05$) associated with all three indexes on the *BRIEF*.

*Note. * = Behavior Rating Inventory of Executive Function*

Table 46

Balance and Self-efficacy of Balance in Children with CHARGE Syndrome (Haibach & Lieberman, 2013).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation A	Quantitative *ABC* ANOVA Correlation	22 children with CHARGE syndrome Control: 31 children Age: 6 to 12 years old Gender: 9 female, 13 male Setting: CHARGE Syndrome Foundation Conference	To examine the balance and self-efficacy of balance in children with CHARGE syndrome. Specific focus: CHARGE syndrome child, disabled balance, postural evaluation, and self-efficacy evaluation.	The individuals with CHARGE syndrome scored significantly ($p < .001$) lower on all of the balance tasks. The balance scale indicated that nine of the participants with CHARGE syndrome were at a low risk of falling with an additional eight at a medium fall risk, and four at a high fall risk. None of the children without CHARGE syndrome were at a fall risk. None of the children without CHARGE syndrome were at a fall risk. In addition, the participants with CHARGE syndrome scored significantly ($p < .001$) lower on the *ABC** scale. On a scale of 0 to 10 for balance confidence with a 10 being completely confident, 19 of the children with CHARGE syndrome indicated a balance confidence score of 0. Scores on the *ABC* were moderately ($r = .56$) correlated with the balance scale scores. Also, the balance scores were not significantly ($p = .465$) correlated by gender or age. Therefore, children with CHARGE syndrome are at risk for falling and lack balance confidence.

*Note. * = Activities-Specific Balance Confidence Scale*

Table 47

Vestibular Abnormalities in CHARGE Association (Murofushi et al., 1997).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation B	Quantitative Descriptive	5 individuals with CHARGE syndrome Age: 3 to 23 years old Gender: 4 female, 1 male	To examine vestibular abnormalities in individuals with CHARGE syndrome. Specific focus: Balance, CHARGE association, cochlea, hearing, inner ear anomaly, semicircular canal, vestibule, and vestibulo-ocular reflex.	Of the five, all had vestibular abnormalities (i.e., absence of semicircular canals) and only three obtained independently walking the other two were unable at approximately 6 years old. The three who were able to walk independently performed the milestone at 18 months, 3 years, and 5 years. However, all three demonstrated a walking gait with a wide base and stiff legged and had difficulties walking and balancing on uneven surfaces. Therefore, vestibular abnormalities may be the cause of abnormal walking gaits in individuals with CHARGE syndrome.

Table 48

Vestibular Areflexia as a Cause of Delayed Motor Skill Development in Children with CHARGE Association (Admiraal & Huygen, 1997).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation A	Quantitative Descriptive	6 individuals with CHARGE syndrome Age: 3 to 26 years old Gender: 5 female, 1 male Setting: Institute for the Deaf	To examine vestibular abnormalities in individuals with CHARGE syndrome. Specific focus: Childhood deafness, vestibular areflexia, optokenetic nystagmus, and pendular nystagmus.	Of the six individuals with CHARGE syndrome, ages 3 to 26 years old, all had vestibular areflexia and had some degree of bilateral hearing loss. Only one individual had horizontal pendular nystagmus. The age range of independent walking was between 2 to 4 years of age; however, one individual's age was not reported. Therefore, the investigators reported that intact saccular function is important for individuals with CHARGE syndrome to perform upright stance and independent walking.

Table 49

Vestibular Anomalies in CHARGE Syndrome: Investigations on and Consequences for Postural Development (Abadie et al., 2000).

Strength Level & Recommendation Level	Research Method	Population	Purpose	Summary of Results
Level 3 Recommendation A	Quantitative Mann-Whitney *U* Test Qualitative Interviews	17 children with CHARGE syndrome Age: 3 to 9 years old Gender: 6 female, 11 male Setting: France	To examine vestibular abnormalities in individuals with CHARGE syndrome. Specific focus: CHARGE syndrome, inner ear, vestibular function, semicircular canals, and otolith.	The participants' developmental milestone mean ± SD and range were: stable sitting 14 ± 3.3 months (8 to 18 months), crawling on the back 15.7 ± 7.8 months, standing with support 19.8 ± 4.9 months (12 to 27 months), standing without support 23.9 ± 6.1 months (17 to 33 months), and walking indoors 29.8 ± 8.7 months (18 to 52 months). Ten of the 17 participants had bilateral complete absence of all three semicircular canals and vestibular function abnormities. Of the 10 with an absence of semicircular canals, none were able to walk before 18 months of age. Therefore, vestibular investigations related to children with CHARGE syndrome are valuable for assessments and adaptations for intervention programs.

APPENDIX B

Institutional Review Board Approval

TWU
TEXAS WOMAN'S UNIVERSITY
DENTON DALLAS HOUSTON

Institutional Review Board
Office of Research and Sponsored Programs
P.O. Box 425619, Denton, TX 76204-5619
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DATE: July 20, 2015

TO: Ms. Elizabeth Foster
Communication Sciences & Disorders

FROM: Institutional Review Board - Denton

Re: Approval for Impact of Rate Limiters and Affordances on the Developmental Walking Milestone in Individuals with CHARGE Syndrome (Protocol #: 18216)

The above referenced study has been reviewed and approved by the Denton Institutional Review Board (IRB) on 7/14/2015 using an expedited review procedure. This approval is valid for one year and expires on 7/13/2016. The IRB will send an email notification 45 days prior to the expiration date with instructions to extend or close the study. It is your responsibility to request an extension for the study if it is not yet complete, to close the protocol file when the study is complete, and to make certain that the study is not conducted beyond the expiration date.

If applicable, agency approval letters must be submitted to the IRB upon receipt prior to any data collection at that agency. A copy of the approved consent form with the IRB approval stamp is enclosed. Please use the consent form with the most recent approval date stamp when obtaining consent from your participants. A copy of the signed consent forms must be submitted with the request to close the study file at the completion of the study.

Any modifications to this study must be submitted for review to the IRB using the Modification Request Form. Additionally, the IRB must be notified immediately of any adverse events or unanticipated problems. All forms are located on the IRB website. If you have any questions, please contact the TWU IRB.

cc. Dr. Erika S. Armstrong, Communication Sciences & Disorders
Dr. Lisa Silliman-French, Communication Sciences & Disorders
Graduate School

APPENDIX C

CHARGE Syndrome Foundation Letter of Support

CHARGE Syndrome Foundation, Inc.

August 23, 2015

Beth Foster ABD, CAPE
2220 Plum Ct
Little Elm, TX 75068

Dear Beth,

We are very excited to hear about the research study you are currently doing for your dissertation. The CHARGE Syndrome Foundation has given you permission to recruit participants for your study, "Impact of Rate Limiters and Affordances on the Developmental Milestone of Walking in Individuals with CHARGE syndrome." This permission extends to any recruitment at the 12th International CHARGE Syndrome Conference in Schaumburg, IL and through other Foundation venues, including but not limited to emailing individuals, our Facebook page and newsletter.

We look forward to seeing the results of your study. Good luck with everything.

Best wishes,

David Wolfe
President

318 Half Day Rd. #305 | Buffalo Grove, IL 60089
Phone: 800-442-7604
Email: info@chargesyndrome.org
Website: www.chargesyndrome.org

APPENDIX D

Data Use Transfer Agreement

DATA USE AGREEMENT

This Data Use Agreement ("Agreement") is made and entered into as of this 20th day of August, 2015 (the "Effective Date") by and between Kinesiology Department of Texas Woman's University with an address at 304 Administration Dr. Denton, TX 76204 ("Data Recipient"), and Saint Louis University, a Missouri benevolent corporation with an address at 221 North Grand Boulevard, St. Louis, MO 63103 ("Covered Entity").

WITNESSETH:

WHEREAS, Covered Entity may Disclose or make available to Data Recipient, and Data Recipient may Use, Disclose, receive, transmit, maintain or create from, certain information in conjunction with research; and

WHEREAS, Covered Entity and Data Recipient are committed to compliance with the Health Insurance Portability and Accountability Act of 1996 ("HIPAA") and regulations promulgated thereunder; and

WHEREAS, the purpose of this Agreement is to satisfy the obligations of Covered Entity under HIPAA and to ensure the integrity and confidentiality of certain information Disclosed or made available to Data Recipient and certain information that Data Recipient Uses, Discloses, receives, transmits, maintains or creates, from Covered Entity.

WHEREAS, Data Recipient agrees to limit its use of the Limited Data Set and protect the Limited Data Set in accordance with the terms of this Agreement and the HIPAA Privacy and Security Rules.

THEREFORE, in consideration of the mutual agreements, terms and conditions herein contained, Covered Entity and Data Recipient agree as follows:

A. DEFINITIONS

Terms used but not otherwise defined in this Agreement, including without limitation "Use" and "Disclose", shall have the same meaning as those terms in the Privacy Rule.

1. Individual shall have the same meaning as the term "individual" in 45 CFR Sect. 164.501 of the Privacy Rule and shall include a person who qualifies as a personal representative in accordance with 45 CFR Sect. 164.502(g) of the Privacy Rule.

2. Limited Data Set shall have the same meaning as the term "limited data set" in 45 CFR 164.514(e) of the Privacy Rule.

3. Privacy Rule shall mean the Standards for Privacy of Individually Identifiable Information at 45 CFR Part 160 and Part 164, Subparts A and E, as amended from time to time.

4. Protected Health Information or PHI shall have the same meaning as the term "protected health information" in 45 CFR Sect. 164.501 of the Privacy Rule, to the extent such information is created or received by Data Recipient from Covered Entity.

5. Required by Law shall have the same meaning as the term "required by law" in 45 CFR Sect. 164.501 of the Privacy Rule.

B. SCOPE AND PURPOSE

1. This Agreement sets forth the terms and conditions pursuant to which Covered Entity will Disclose certain PHI to the Data Recipient.

2. Except as otherwise specified herein, Data Recipient may make all Uses and Disclosures of the Limited Data Set necessary to conduct the research described herein: The purpose of this investigation is to examine the impact of individual characteristics (i.e., hearing loss, vision loss) on attainment of the independent walking developmental milestone in individuals with CHARGE syndrome. It is also to examine other characteristics that may have a relationship with the age at which individuals with CHARGE syndrome being independently walking ("Research Project").

3. For purposes of this Agreement, Recipient's primary representative for receipt, Use and Disclosure of the Data for the Purpose shall be: Elizabeth Foster

C. OBLIGATIONS AND ACTIVITIES OF DATA RECIPIENT

1. Data Recipient agrees to not Use or Disclose the Limited Data Set for any purpose other than the Research Project or as Required by Law.

2. Data Recipient agrees to use appropriate safeguards to prevent Use or Disclosure of the Limited Data Set other than as provided for by this Agreement.

3. Data Recipient agrees to promptly report to the Covered Entity any Use or Disclosure of the Limited Data Set not provided for by this Agreement of which it becomes aware.

4. Data Recipient agrees to ensure that any agent to whom it provides the Limited Data Set agrees to the same restrictions and conditions that apply through this Agreement to the Data Recipient with respect to such information.

5. Data Recipient agrees not to identify the information contained in the Limited Data Set or contact the individual.

6. Data Recipient agrees to share the results of the Research Project ("Results") with Covered Entity. Covered Entity's use of the Results shall be limited to its internal, non-commercial, research purposes only.

D. TERM AND TERMINATION

1. The Term of this Agreement shall be from the Effective Date until August 20th, 2016.

2. Upon Covered Entity's knowledge of a material breach by Data Recipient, Covered Entity shall have the right to immediately terminate this Agreement.

3. (a) Except as provided in paragraph (b) of this subsection, upon termination of this Agreement or upon request of Covered Entity, whichever occurs first, Data Recipient shall return or destroy the Limited Data Set received from Covered Entity.

(b) In the event that Data Recipient determines that returning or destroying the Limited Data Set is not feasible, Data Recipient shall provide to Covered Entity notification of the conditions that make return or destruction infeasible. Upon mutual agreement of the parties that return or destruction of the Limited Data Set is infeasible, Data Recipient shall extend the protections of this Agreement to such Limited Data Set and limit further uses and disclosures of such Protected Health Information to those purposes that make the return or destruction infeasible, for so long as Data Recipient maintains such Limited Data Set.

E. DISCLAIMER

THE COVERED ENTITY MAKES NO REPRESENTATIONS AND EXTENDS NO WARRANTIES OF ANY KIND WITH RESPECT TO THE DATA TO BE TRANSFERRED UNDER THIS AGREEMENT. ALL DATA IS PROVIDED BY THE COVERED ENTITY "AS IS." THE COVERED ENTITY EXPRESSLY DISCLAIMS ALL WARRANTIES, EXPRESS OR IMPLIED, INCLUDING THE WARRANTIES OF MERCHANTABILITY, FITNESS FOR A PARTICULAR PURPOSE, OR THAT THE USE OF THE MATERIALS WILL NOT INFRINGE ANY PATENT, COPYRIGHT, TRADEMARK, OR OTHER PROPRIETARY RIGHTS.

F. OWNERSHIP

The Covered Entity shall retain ownership of all data transferred by it to the Data Recipient under this Agreement. Nothing in this Agreement shall be construed to confer or grant any commercial rights or license to any patentable intellectual property that may

arise from use of the data or to any patentable intellectual property owned or created by the Covered Entity.

The Data Recipient shall retain ownership of the Results. Nothing in this Agreement shall be construed to confer or grant any commercial rights or license to any patentable intellectual property that may arise from use of the data or to any patentable intellectual property owned or created by the Data Recipient.

G. MISCELLANEOUS

1. A reference in this Agreement to a section in the Privacy Rule means the section as amended or as renumbered.

2. The parties agree to take such action as is necessary to amend this Agreement from time to time as is necessary for Covered Entity to comply with the requirements of the Privacy Rule and HIPAA.

3. The respective rights and obligations of Data Recipient under Section C of this Agreement shall survive termination of this Agreement.

4. Any ambiguity in this Agreement shall be resolved to permit Covered Entity to comply with the Privacy Rule.

5. There are no intended third party beneficiaries to this Agreement. Without in any way limiting the foregoing, it is the parties' specific intent that nothing contained in this Agreement gives rise to any right or cause of action, contractual or otherwise, in or on behalf of the individuals whose PHI is Used or Disclosed pursuant to this Agreement.

6. No provision of this Agreement may be waived except by an agreement in writing signed by the waiving party. A waiver of any term or provision shall not be construed as a waiver of any other term or provision.

7. The persons signing below have the right and authority to execute this Agreement and no further approvals are necessary to create a binding agreement.

8. In the event of any conflict between the terms and conditions stated within this Agreement and those contained within any other agreement or understanding between the parties, written, oral or implied, the terms of this Agreement shall govern. Without limiting the foregoing, no provision of any other agreement or understanding between the parties limiting the liability of Data Recipient to Covered Entity shall apply to the breach of any covenant in this Agreement by Data Recipient.

9. This Agreement shall be construed in accordance with and governed by the laws of the State of Missouri.

IN WITNESS WHEREOF, the parties have executed this Agreement effective upon the Effective Date set forth above.

COVERED ENTITY:

Saint Louis University

Stephanie Kimzey
Associate Director
Office of Technology Management

DATA RECIPIENT:

Name: Elizabeth Foster

Title: Graduate Assistant/PI

APPENDIX E

CHARGE Syndrome Clinical Database Section Questions

Section 2

Q3.10 Is ${q://QID365/ChoiceTextEntryValue/1} a male or female?

- ❍ Male (1)
- ❍ Female (2)

Q345 At how many weeks was ${q://QID365/ChoiceTextEntryValue/1} born and DOB?

_____ Weeks (1) _____ Date of Birth

Q366 Please enter ${q://QID365/ChoiceTextEntryValue/1}'s birth weight in either pounds and ounces or grams (e.g., 6 pounds 2 ounces OR 2778 grams). Valid ranges are 1-13 pounds or 400-5000 grams. If unknown, please leave blank.

Pounds (1-13) (1)
Ounces (2)
Grams (400-5000) (3)

Q368 Please enter ${q://QID365/ChoiceTextEntryValue/1}'s birth length in either inches or centimeters (e.g., 15 inches OR 38 centimeters). Valid ranges are 12-25 inches or 30-65 centimeters. If unknown, please leave blank.

Inches (12-25) (1)
Centimeters (30-65) (3)

Q371 Please enter ${q://QID365/ChoiceTextEntryValue/1}'s birth head circumference in either inches or centimeters (e.g., 15 inches OR 38 centimeters). Valid ranges are 10-17 inches or 25-45 centimeters. If unknown, please leave blank.

Inches (10-17) (1)
Centimeters (24-45) (3)

Section 3

Q359 Has ${q://QID360/ChoiceTextEntryValue/1} had a CT or MRI of the inner ear?

Answer If Has your child had a CT or MRI of the inner ear? CT - Yes Is Selected Or Has your child had a CT or MRI of the inner ear? MRI - Yes Is Selected

	Yes (1)	No (2)
CT (1)	❑	❑
MRI (2)	❑	❑

Q4.35 Inner ears - please check all that apply:

	Right ear			Left ear		
	Yes (1)	No (2)	Unsure (3)	Yes (1)	No (2)	Unsure (3)
Small/absent semicircular canals, confirmed by CT or MRI (1)	❍	❍	❍	❍	❍	❍
Cochlear malformation (e.g., Mondini defect or other), confirmed by CT or MRI (2)	❍	❍	❍	❍	❍	❍

Answer If Has your child had a CT or MRI of the inner ear? CT - Yes Is Selected Or Has your child had a CT or MRI of the inner ear? MRI - Yes Is Selected

Q4.36 Inner ears - auditory nerve on MRI

	Right ear				Left ear			
	Normal (1)	Small (2)	Absent (3)	Unsure (4)	Normal (1)	Small (2)	Absent (3)	Unsure (4)
Auditory nerve on MRI (1)	❍	❍	❍	❍	❍	❍	❍	❍

Q4.37 Does ${q://QID360/ChoiceTextEntryValue/1} have balance/vestibular (inner ear balance) problems?

- ❍ Yes (1)
- ❍ No (2)
- ❍ Unsure (3)

Q4.38 Did/does ${q://QID360/ChoiceTextEntryValue/1} use a 5-point crawl (head on ground)?

- ❍ Yes (1)
- ❍ No (2)
- ❍ Unsure (3)
- ❍ Not crawling yet (4)

Q4.39 Did/does ${q://QID360/ChoiceTextEntryValue/1} crawl or scoot on his/her back?

- ❍ Yes (1)
- ❍ No (2)
- ❍ Unsure (3)
- ❍ Not crawling/scooting yet (4)

Section 6

Q3 Approximately how many months total did ${q://QID359/ChoiceTextEntryValue/1} spend in the hospital during the first three years of life? Please enter a number.

Q4 Approximately how many surgeries has ${q://QID359/ChoiceTextEntryValue/1} had?

Q5 Indicate which of the following surgeries ${q://QID359/ChoiceTextEntryValue/1} has had. Please choose all that apply.

Section 7

Q7.2 Does ${q://QID356/ChoiceTextEntryValue/1} have loss of visual field (often due to coloboma)?

	Left eye			Right eye		
	Yes (1)	No (2)	Unsure (3)	Yes (1)	No (2)	Unsure (3)
Visual field loss - upper (1)	❍	❍	❍	❍	❍	❍
Visual field loss - central/macula (2)	❍	❍	❍	❍	❍	❍
Nystagmus (3)	❍	❍	❍	❍	❍	❍

Q7.3 What is ${q://QID356/ChoiceTextEntryValue/1}'s visual acuity as measured by the ophthalmologist (e.g., 20/40, 6/12, or unknown)?

Left eye (1)
Right eye (2)

Q7.5 What is ${q://QID356/ChoiceTextEntryValue/1}'s estimated level of vision?

	Normal (1)	Mild loss (2)	Moderate loss (finger counting) (3)	Light/dark shadows only (5)	Completely blind (6)	Unknown (7)
Left eye (1)	❍	❍	❍	❍	❍	❍
Right eye (2)	❍	❍	❍	❍	❍	❍

Q7.6 Does ${q://QID356/ChoiceTextEntryValue/1} wear glasses?

- ❍ Yes (1)
- ❍ No (2)

Answer If Does your child wear glasses? Yes Is Selected

Q7.7 ${q://QID356/ChoiceTextEntryValue/1}'s glasses are to correct which of the following? Please check all that apply.

- ❑ Farsightedness (i.e., needs glasses to read/see up close) (1)
- ❑ Nearsightedness (i.e., needs glasses to see far away) (2)
- ❑ Astigmatism (3)
- ❑ Sensitivity to bright light (wears tinted lenses) (4)
- ❑ Diploplia (wears prism lenses) (5)

Q357 Is ${q://QID356/ChoiceTextEntryValue/1} thought to have cortical vision loss/impairment?

- ❍ Yes (1)
- ❍ No (2)
- ❍ Unsure (3)

Q358 Does ${q://QID356/ChoiceTextEntryValue/1} have other visual loss/impairment?

- ❍ Yes (1)
- ❍ No (2)

Section 8

Answer If Is there other visual loss / impairment? Yes Is Selected

Q359 What other visual loss/impairment does ${q://QID356/ChoiceTextEntryValue/1} have?

Q41 If you have recent audiograms, please scan and upload them below. Note: Please wait until all files have finished uploading before going to the next page. If you have more than five files to upload, you may complete and submit this section multiple times with new information.

Q361 Audiogram 1

Q8.2 Does ${q://QID357/ChoiceTextEntryValue/1} have hearing impairment?

	Yes (1)	No (2)	Unsure (3)
Left ear (1)	❍	❍	❍
Right ear (2)	❍	❍	❍

Answer If Does your child have hearing impairment? Right ear - Yes Is Selected

Q8.3 What is ${q://QID357/ChoiceTextEntryValue/1}'s left ear impairment? Please choose all that apply.

- ❑ Conductive (1)
- ❑ Sensorineural (2)
- ❑ Unsure (3)

Answer If Does your child have hearing impairment? Left ear - Yes Is Selected

Q8.4 What is ${q://QID357/ChoiceTextEntryValue/1}'s right ear impairment? Please choose all that apply.

- ❑ Conductive (1)
- ❑ Sensorineural (2)
- ❑ Unsure (3)

Q8.5 What is ${q://QID357/ChoiceTextEntryValue/1}'s hearing level?

	Normal hearing (0-15 db) (1)	Slight hearing loss (15-25 dB) (2)	Mild (26-30 dB) (3)	Moderate (30-50 dB) (4)	Moderately severe (50-70 dB) (5)	Severe (71-90 dB) (6)	Profound (91+ dB) (7)	Unknown (8)
Left ear (1)	❍	❍	❍	❍	❍	❍	❍	❍
Right ear (2)	❍	❍	❍	❍	❍	❍	❍	❍

Q360 What is ${q://QID357/ChoiceTextEntryValue/1}'s speech reception threshold (in decibels)?

_____ Speech reception threshold - unaided (1)
_____ Speech reception threshold - aided (2)

Q8.6 Does ${q://QID357/ChoiceTextEntryValue/1} have any of the following? Please choose all that apply.

- ❑ Behind the ear hearing aid - left ear (1)
- ❑ Behind the ear hearing aid - right ear (2)
- ❑ Bone-anchored hearing aid (BAHA) (3)
- ❑ Cochlear implant - left ear (4)
- ❑ Cochlear implant - right ear (5)

Section 9

Q9.2 Please report whether ${q://QID369/ChoiceTextEntryValue/1} has reached each milestone

Answer If Please report whether your child has reached each milesto... Raise head - Yes Is Selected

Q9.3 Age in months at which ${q://QID369/ChoiceTextEntryValue/1} achieved milestone: Raise head (leave at zero if unsure)

_____ Age in months (leave at zero if unsure) (1)

Answer If Please report whether your child has reached each milesto... Sit with support - Yes Is Selected

Q9.4 Age in months at which ${q://QID369/ChoiceTextEntryValue/1} achieved milestone: Sit with support (leave at zero if unsure)

_____ Age in months (leave at zero if unsure) (1)

Answer If Please report whether your child has reached each milesto... Sit alone - Yes Is Selected

Q9.5 Age in months at which ${q://QID369/ChoiceTextEntryValue/1} achieved milestone: Sit alone (leave at zero if unsure)

_____ Age in months (leave at zero if unsure) (1)

Answer If Please report whether your child has reached each milesto... Crawl on tummy - Yes Is Selected

Q9.6 Age in months at which ${q://QID369/ChoiceTextEntryValue/1} achieved milestone: Crawl on tummy (leave at zero if unsure)

_____ Age in months (leave at zero if unsure) (1)

Answer If Please report whether your child has reached each milesto... Crawl on back - Yes Is Selected

Q9.7 Age in months at which ${q://QID369/ChoiceTextEntryValue/1} achieved milestone: Crawl on back (leave at zero if unsure)

_____ Age in months (leave at zero if unsure) (1)

Answer If Please report whether your child has reached each milesto... Pull to stand - Yes Is Selected

Q9.9 Age in months at which ${q://QID369/ChoiceTextEntryValue/1} achieved milestone: Pull to stand (leave at zero if unsure)

_____ Age in months (leave at zero if unsure) (1)

Answer If Please report whether your child has reached each milesto... Stand holding on - Yes Is Selected

Q9.10 Age in months at which ${q://QID369/ChoiceTextEntryValue/1} achieved milestone: Stand holding on (leave at zero if unsure)

_____ Age in months (leave at zero if unsure) (1)

Answer If Please report whether your child has reached each milesto... Cruise holding on - Yes Is Selected

Q9.11 Age in months at which ${q://QID369/ChoiceTextEntryValue/1} achieved milestone: Cruise holding on (leave at zero if unsure)

_____ Age in months (leave at zero if unsure) (1)

Answer If Please report whether your child has reached each milesto... Stand alone - Yes Is Selected

Q9.12 Age in months at which ${q://QID369/ChoiceTextEntryValue/1} achieved milestone: Stand alone (leave at zero if unsure)

_____ Age in months (leave at zero if unsure) (1)

Answer If Please report whether your child has reached each milesto... Walk alone - Yes Is Selected

Q9.13 Age in months at which ${q://QID369/ChoiceTextEntryValue/1} achieved milestone: Walk alone (leave at zero if unsure)

_____ Age in months (leave at zero if unsure) (1)

Answer If Please report whether your child has reached each milesto... Run - Yes Is Selected

Q9.14 Age in months at which ${q://QID369/ChoiceTextEntryValue/1} achieved milestone: Run (leave at zero if unsure)

_____ Age in months (leave at zero if unsure) (1)

Answer If Please report whether your child has reached each milesto... Jump on two feet - Yes Is Selected

Q9.15 Age in months at which ${q://QID369/ChoiceTextEntryValue/1} achieved milestone: Jump on two feet (leave at zero if unsure)

_____ Age in months (leave at zero if unsure) (1)

Answer If Please report whether your child has reached each milesto... Climb stairs with alternating feet - Yes Is Selected

Q9.16 Age in months at which ${q://QID369/ChoiceTextEntryValue/1} achieved milestone: Climb stairs with alternating feet (leave at zero if unsure)

_____ Age in months (leave at zero if unsure) (1)

Q9.22 What is ${q://QID369/ChoiceTextEntryValue/1}'s current level of proficiency with expressive language? Please choose all that apply.

- ❑ Gestures and pointing (4)
- ❑ Single words (1)
- ❑ Two word phrases (2)
- ❑ Sentences (3)
- ❑ Can carry on a conversation (5)

Q368 Please enter ${q://QID369/ChoiceTextEntryValue/1}'s most recent height in either inches or centimeters (e.g., 15 inches or 38 centimeters). Valid ranges are 15-80 inches or 38-200 centimeters.

Inches (15-80) (1)
Centimeters (38-200) (3)
Approximate date of measurement (mm/dd/yyyy) (4)

Q366 Please enter ${q://QID369/ChoiceTextEntryValue/1}'s most recent weight in either pounds and ounces or kilograms (e.g., 46 pounds 2 ounces or 78 kilograms). Valid ranges are 5-200 pounds or 1-90 kilograms.

Pounds (5-200) (1)
Ounces (2)
Kilograms (1-90) (3)
Approximate date of measurement (mm/dd/yyyy) (4)

APPENDIX F

Interview Protocol

Interview Protocol

Questions

1. Tell me the story of your child achieving independent walking?

Probing questions:

A. How did your child progress through developmental motor milestones (i.e., creeping, 5-point crawl, standing)?

2. What things were most helpful or contributed to your child walking?

Probing questions:

A. Were there any individual factors (i.e., motivation), pertaining directly to characteristics of your child?

B. Were there any environmental factors (i.e., indoors or outdoors)?

C. What about the task itself (i.e., independent walking)?

3. What were the roadblocks or difficulties you encountered that caused a delay in your child walking?

Probing questions:

A. Were there any individual factors (e.g., vision, hearing, and muscle weakness), pertaining to characteristics about your child?

B. Were there any environmental factors (e.g., house set-up, neighborhood, other people, sensory issues, or other services)?

C. What about the task of walking (e.g., legs were not strong enough, and balance issues)?

4. What else, if anything, would you like me to know about your child achieving independent walking, that we have not yet discussed?

Background Information

Are you the primary caregiver or parent of an individual (child) with CHARGE syndrome?

Parent's Age: _____ Current Child's Age: _____

At what age was your child diagnosed with CHARGE syndrome?_____

At what age did your child achieve developmental walking milestone? _____

What is your child's with CHARGE syndrome vision acuity (e.g., 20/400)?

_____

What is your child's with CHARGE syndrome hearing loss (e.g., 90 decibels)?

_____

How many days prior to walking was your child with CHARGE syndrome hospitalized?

_____

How many surgeries did your child with CHARGE syndrome have prior to walking?

_____

APPENDIX G

List of Referral Resources

List of Referral Resources

American Psychological Association (APA) Locator Service: http://locator.apa.org/
Toll-Free Referral Number: 1-800-964-2000

Mental Health of America Referrals: http://www.nmha.org/go/searchMHA

American Counseling Association Referrals
http://www.counseling.org/Resources/CounselorDirectory/TP/Home/CT2.aspx

National Family Association for Deaf Blind (NFADB): this organization supports individuals who are deafblind and their families. http://www.nfadb.org/

CHARGE Syndrome Foundation: http://www.chargesyndrome.org/
Parent to Parent: If you are interested in finding someone in your area please contact the foundation at 800-442-7604 or by sending an email to info@chargesyndrome.org for a list of families.

CHARGE Syndrome Listserv: An online support list for individuals with CHARGE, parents, family members, caregivers, doctors, therapists, or anyone with an interest in or is working with a person who has CHARGE Syndrome. http://groups.yahoo.com/group/CHARGE/join

CHARGE Syndrome Foundation Facebook Page:
http://www.facebook.com/groups/chargesyndrome

The CHARGE Family Support Group UK: http://www.chargesyndrome.org.uk/

Texas CHARGE Group: Texas families. http://texaschargers.org/

CHARGE Syndrome Association of Australasia Ltd: supports families in Australia and New Zealand. http://www.chargesyndrome.org.au/

Association CHARGE: This is a French support group. http://www.associationcharge.fr/

Dutch CHARGE Syndrome Network: http://www.chargesyndroom.nl/

Belgian CHARGE Syndrome Website: http://www.chargesyndroom.be/index.php

CHARGE Syndrome German: (Germany, Austria, Switzerland, etc.). http://www.charge-syndrom.de/

Asociación Españoña Síndrome de Charge: Family support group based in Spain
http://www.sindromecharge.es/

APPENDIX H

Participant Recruitment

RESEARCH STUDY

Texas Woman's University, Denton

Department of Kinesiology

The purpose of this study is to examine the impact of variables on the age of independent walking developmental milestone in individuals with CHARGE syndrome by collecting data from the CHARGE Syndrome Clinical Database Project (CSCDP) and interviewing parents (or primary caregivers) on your perspectives and experiences pertaining to your child achieving independent walking.

Who is Eligible?

- ❖ Parent or primary caregiver of individuals with CHARGE syndrome who have acquired independent walking

What will you be asked to do?

- ❖ Complete the CSCDP (Including sections covering: vision, hearing, and milestones).
- ❖ Participate in a 15 to 25 minute telephone or video interview to discuss possible influences on the (usually delayed) age of walking in your child.

Participation is completely voluntary.

There is a potential risk of loss of confidentiality in all, email, downloading, and internet transactions.

If you are interested in participating or have any questions, please contact:

Beth Foster at Email: efoster@twu.edu or 267-241-3728

Research Study: Walking Phone (267)-241-3728 Email: efoster@twu.edu	Research Study: Walking Phone (267)-241-3728 Email: efoster@twu.edu	Research Study: Walking Phone (267)-241-3728 Email: efoster@twu.edu	Research Study: Walking Phone (267)-241-3728 Email: efoster@twu.edu	Research Study: Walking Phone (267)-241-3728 Email: efoster@twu.edu	Research Study: Walking Phone (267)-241-3728 Email: efoster@twu.edu	Research Study: Walking Phone (267)-241-3728 Email: efoster@twu.edu

Participant Recruitment Script for Investigator Initiated Person Contact

Excuse me, Mrs. Smith?

Do you have a minute? My name is Beth Foster. I am a Doctoral student at Texas Woman's University. I am working on a research study about variables that may have an impact on the age of walking in individuals with CHARGE syndrome. Can your child or individual with CHARGE syndrome walk independently? Would you be interested in learning more details about the research study?

- If the individual says "no, not interested" then the conversation with end with Thank you for your time.
- If the individual says "yes" then the consent to participate form with be explained.

"If you agree to participate, there are two phases to the study. The total time commitment for this investigation is four hours. Phase I of the investigation will take approximately 2 to 3 hours to complete all sections of the CSCDP online survey. Phase II of the investigation will take approximately 15 to 25 minutes for the webcam interview and an additional 20 minutes to read over the interview transcript. In Phase I, we ask you to participate in the CHARGE Syndrome Clinical Database Project (CSCDP). I will use data you enter to examine possible variables that may impact the age of walking. In Phase II, I will conduct a 15 to 25 minute phone or video interview with you. I will ask you about your perceptions and experience pertaining to your child achieving independent walking. It is entirely up to you whether to participate or not and you may withdraw at any time and may skip questions you would prefer not to answer. Do you have any questions for me? Are you interested in participating in this study?"

Participant Recruitment Email

Dear Parent or Primary Caregiver of an individual with CHARGE syndrome:

My name is Beth Foster. I am a Certified Adapted Physical Educator and Doctoral student at Texas Woman's University (TWU) and a member of the CHARGE Syndrome Foundation. I am interested in determining your experiences and perspective pertaining to your child achieving independent walking. This study has been approved by the TWU IRB (protocol # 18216).

There are two phases to the investigation. In Phase I, you will be asked to participate (or review and update information already entered) in the CHARGE Syndrome Clinical Database Project (CSCDP) through the CHARGE Syndrome Foundation. This will provide baseline information, which will be used to identify variables that may have an effect or correlate with delayed independent walking developmental milestone in individuals with CHARGE syndrome. In Phase II of the investigation, you may be selected to participate in a telephone or video interview regarding your experience and perspectives of your child achieving independent walking. The interview will be conducted by myself and will be audio recorded.

Please review the attached consent to participate document; participants must agree to this consent.

If you have any questions, please feel free to contact Beth at efoster@twu.edu

If you agree to participate in this study, reply to this email, stating you are interested.

Thank you for your time and attention,

Sincerely,

Beth Foster, MS, CAPE

APPENDIX I

Consent to Participate in Research Form

TEXAS WOMAN'S UNIVERSITY
CONSENT TO PARTICIPATE IN RESEARCH

Title: Impact of Rate Limiters and Affordances on the Developmental Walking Milestone in Individuals with CHARGE Syndrome

Primary Investigator: Elizabeth A. Foster, MS, CAPE…….EFoster@twu.edu
940-898-2606
Researcher Advisor: Lisa Silliman-French, PhD……...LSillimanFrench@twu.edu
940-898-2594

Explanation and Purpose of the Research

You are being asked to participate in a research study, conducted by Elizabeth A. Foster, CAPE at Texas Woman's University (TWU). The purpose of this investigation is to examine the impact or correlation of individual characteristics with the age of independent walking developmental milestone in individuals with CHARGE syndrome and to determine parent or primary caregiver's perspectives on factors that caused the delay and those that contributed based on his or her child, environment, and the task of independent walking developmental milestone. You have been asked to participate in this study because you are a parent or primary caregiver of a child or individual with CHARGE syndrome.

Description of Procedures

In order to be a participant in this study, you must be at least 18 years of age or older and be a primary caregiver of an individual with CHARGE syndrome who has acquired independent walking.

In the first phase of this investigation, you will be asked to participate (or review and update information already entered) in the CHARGE Syndrome Clinical Database Project (CSCDP). The information will be used to examine variables that may have an effect or correlate to the delay in achieving independent walking developmental milestone. In the second phase of this investigation, you may or may not be selected for participation in an interview. If you are not selected to participate in this phase of the study, you will be thanked for your time and no further involvement will be required. However, if you would like a copy of the study summary, you may request it at any time. This summary will be delivered via email upon study completion. If you are selected for phase II, you will be contacted through email or telephone for an interview.

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Initials

If you are chosen for the second phase of this investigation, you will be asked to spend between approximately 15 minutes to 25 minutes in an audio recorded telephone or video interview with the primary investigator. The investigator will ask questions about your experiences with your child walking. You and the interviewer will choose a code name for use during the recorded interview before the start of the interview. In addition, you will be asked to use code names for all people you discuss during the interview to preserve privacy. The interview will be audio recorded and then transcribed to ensure accuracy when examining your responses. You will be sent an email of the transcription to read, evaluate, and email any changes if you would like to make to the transcription.

Potential Risks

One risk in this investigation is loss of confidentiality. Confidentiality will be protected to the extent that is allowed by law. There is a potential risk of loss of confidentiality in all, email, downloading, and internet transactions. The researcher will be the only individual who will have access to the information within the CHARGE Syndrome Clinical Database Project (CSCDP) with will be de-identifiable. The CSCDP questionnaire is accessible through Qualtrics. Qualtrics provides data security for Protected Health Information. Qualtrics meets the rigorous privacy standards imposed on health care records by the Health Insurance Portability and Accountability Act. All Qualtrics accounts are hidden behind passwords and all data are protected with real-time data replication. All email correspondence with identifying information will be stored in a password-protected database that will be deleted upon the study's completion. During the interview, should any names be inadvertently used in the interview, the researcher will change the names in the written transcripts. Materials related to the study, including any identifiable information (i.e., name, email, and telephone contact information) will be kept separate and stored in a locked cabinet and electronically will be stored in a password protected database that will be deleted after the study is complete. All interview audiotapes will be stored in a secure and locked cabinet. Interview recordings and transcripts will only be read or listened to by the primary researcher and research committee. Tapes will be destroyed or erased at the conclusion of the transcription process. Unidentified transcripts of interviews will be shredded within five years of the study's completion. You can choose a location for the phone or video interview to occur (i.e., home, office, work); the interviewer will conduct the webcam interview from a private location in a TWU academic office or personal home office.

Another risk in this study is the possibility of coercion. Participation is completely voluntary and potential participants will have a sufficient opportunity to consider whether to participate. Participants can leave the study at anytime in which their information will not be used in the study.

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Initials

A consent to participate form will be signed prior to participation in the study. Participants can ask questions pertaining to the study at anytime: prior, during, or after the investigation occurs.

Another risk in this study is the possibility of psychological or emotional harm. The researcher will ask you questions about your experiences with your child walking. The researcher will also ask you to offer your perceptions and experiences that contributed to your child walking and difficulties that delayed your child in walking. You may stop the interview at any time should you experience any psychological or emotional discomfort. You may choose which questions you wish to answer and decline to answer any questions that may cause you discomfort. The researchers will provide the participant with a list of resources when he or she signs the consent to participate form, in the event he or she experiences any feelings of discomfort.

Another possible risk in this study is fatigue. If you become tired or upset, you may take breaks as needed. You may also stop answering questions at any time and reschedule or end the interview.

Since the study questions will center on personal experiences and perceptions, there is a risk of the invasion of privacy. However, participation is voluntary and you may stop the interview at
any time, skip any questions that cause you discomfort, and leave the study at any time.

Data will be unidentified and stored in a password-protected file. Identifiable data (i.e., name, email, and phone contact information) will be separated and stored in its own password protected file and locked cabinet. As noted, the researcher will provide you through email with a list of resources in the event you experience any feelings of discomfort.

There is a risk of loss of time. Participation in the study is voluntary. Interviews are expected to last between 15 to 25 minutes. You may choose to leave the study at any time for any reason.

The researchers will try to prevent any problem that could happen because of this research. You should let the researchers know at once if there is a problem and they will help you. However, TWU does not provide medical services or financial assistance for any injuries that might happen because you are taking part in this research.

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Initials

Participation and Benefits

Your involvement in this investigation is completely voluntary and you may withdraw from the study at any time. Your involvement in this study may or may not be of direct benefit to you or your child with CHARGE syndrome. Participation could potentially affect future development of services or interventions pertaining to walking for individuals with CHARGE syndrome.

Questions Regarding the Study

You will be provided with a copy of this consent form. If you have any questions about the research study you should ask the researcher; her phone number is at the top of this form. If you have questions about your rights as a participant in this research or the way this study has been conducted, you may contact the Texas Woman's University Office of Research and Sponsored Programs at 940-898-3378 or via e-mail at IRB@twu.edu.

By signing below, you acknowledge that you have read this information and are giving your informed consent to participate in this study.

_____ _____

Signature of Participant Date

*If you would like to know the general results of this study tell us where you want them sent:

Email: _____

APPENDIX J

Motor Developmental Milestone Figures

5-Point Crawl

Figure 1. Developmental milestones in achieving normal walking gait. Note correct terminology for *crawl* (i.e., on belly) versus *creep* (i.e., on hands and knees). Ages given are *average* time of appearance. From *Adapted physical activity, recreation, and sport: Crossdisciplinary and lifespan*, 6th ed., by C. Sherrill. Copyright ©2004 by McGraw-Hill Education, Publishers (New York, NY). Used with permission. In addition, the figure to the right is the 5-point crawl which is common in children with CHARGE syndrome.

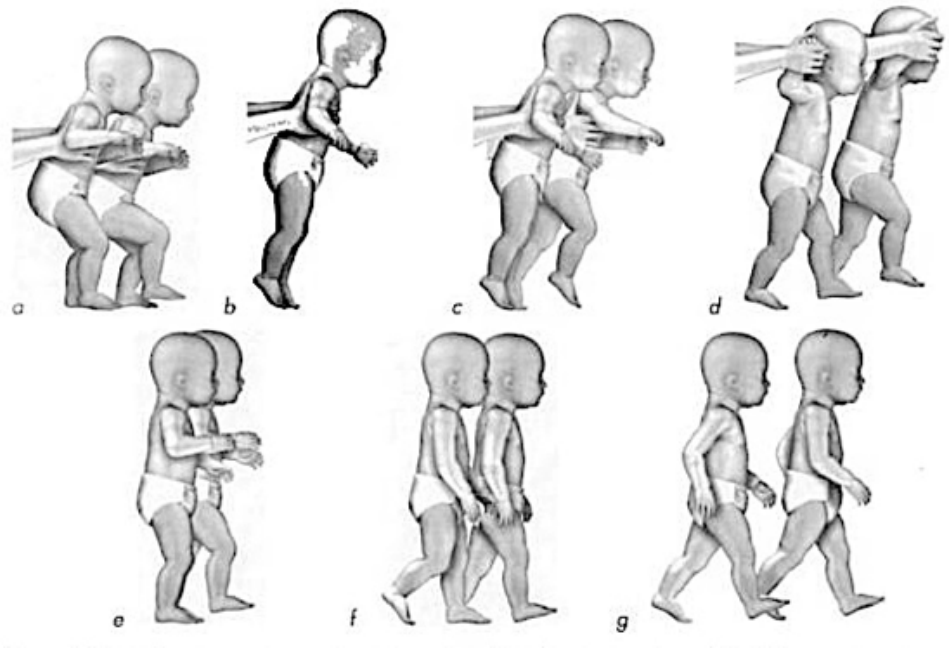

Figure 2. McGraw's seven stages of erect locomotion (i.e., walking). (a) Reflex stepping phase, (b) inhibitory or static phase, (c) transitions phase, includes stomping of foot or stand bouncing and the reappearance of stepping movements, (d) deliberate stepping phase with erect posture (e) independent stepping phase marking the start of independent walking, (f) heel-toe Progressions, and (g) adult pattern of walking with synchronous swinging of the arms with the opposite lower extremity (McGraw, 1940, 1945).