Article

Parent Perceptions of Movement Skills in Children with and without CHARGE Syndrome

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Abstract: CHARGE Syndrome (CS) is a low incidence disorder with unique characteristics including deafblindness, growth deficiencies, heart defects, and atresia choanae. The purpose of this study was to examine parents' perceptions of motor competence in their child with CS as well as the value and support they provide on motor competence compared to parents of children without disabilities. Parents of 33 children with CS (*M*age = 6.8 years SD=2.6) and parents of 38 peers without disabilities (*M*age = 7.0 years SD=2.3) completed a demographic and a 32-item parents' perception questionnaire regarding their confidence in their child's motor competence. From the results there were significantly lower motor competence scores in parents' ratings of children with CS; however, they participated with their child in movement skills and tracked their child's movement skills more often than parents of children without disabilities. No group differences were found in the belief that their child can change their movement skills, or in the importance of motor competence. These findings will assist in the understanding of the impact of parenting a child with CS upon their perception of their child's motor competence and the level of support they provide their child. This information indicates the important role that parents of children with severe disabilities have in their child's motor development.

Keywords: severe disability; youth; motor development; deafblindness; parental support

Introduction

CHARGE syndrome (CS) is a low incidence disorder affecting between 1 per 10,000 to 15,000 live births (Deuce, Howard, Rose, & Fuggle, 2012), but is the leading genetic cause of deafblindness (National Center on Deaf-Blindness, 2018). CS is a complex genetic disorder causing numerous birth and sensory deficits, such as hearing, vision, balance, and smell. CHARGE is an acronym created by Pagon and colleagues (1981) that signify the physical and health issues that were first linked to the syndrome: Colobama of the eye, Heart malformations, Atresia of the choanae, Restrictions of growth and development, Genitourinary abnormalities, and Ear malformations. Since this time, CHARGE diagnosis is typically based upon genetic testing for the presence of a CHD7 gene mutation (Janssen et al., 2012). The presence of this gene mutation is often combined with major features of CS such as colobomas, choanal atresia, malformed cochlea, ear malformations, cranial nerve anomalies, and middle ear bone malformations. Minor features of CS include heart defects, kidney abnormalities, restrictions in growth, cleft lip and palate, and genital abnormalities (Blake et al., 1998).

Each child with CS is quite unique as the characteristics and severity of each characteristic presented vary widely (Blake & Prasad, 2006). Although there are wide variations of CS characteristics and experiences, most children with CS have some level of hearing and vision loss, with colobomas present in 70-90% of individuals with CS (CHARGE Syndrome, 2019). Most individuals with CS also have either damaged or missing vestibular organs, which in conjunction with the vision and hearing losses, enhance challenges to balance and movement (Möller, 2011). These sensory losses contribute to delays in motor development and communication challenges (Hartshorne, Hefner, & Davenport, 2005).

Children with CS are typically faced with numerous other challenges to their motor development including executive function deficits, low muscle tone, prolonged hospitalizations (Hartshorne, Nicholas, Grialou, & Russ, 2007; Lieberman, Haibach, & Schedlin, 2012), lack of experienced teachers in teaching children with deafblindness (Lane, Lieberman, Beach, Perreault, & Columna, 2020; Lieberman et al., 2012; Perkins, Columna, Lieberman, & Bailey, 2013), and reduced opportunities to participate in physical activities and recreation in comparison to their peers without disabilities (Imel, Hartshorne, Slavin, & Kanouse, 2020; Lieberman, Kirk, & Haegele, 2018; Lieberman et al., 2012). Movement delays as indicated by later onsets of motor milestones are apparent in children with CS at very young ages. Perhaps the most striking delay is the onset of independent walking, which has been found to be delayed 25 months later than infants without disabilities (Dammeyer, 2012). Age of walking has been negatively associated with numerous motor competence measures including balance, locomotor, and ball skills (Perreault et al., 2020), as well as other critical developmental measures such as sleep (Harshorne, Heussler, Dailor, Williams, Papadopoulos, & Brandt, 2009), language (Dammeyer, 2012), medications (Wachtel, Hartshorne, & Dailor, 2007), and ability to adapt to changes (Salem-Hartshorne & Jacob, 2005). Children with CS struggle with maintaining static balance as well as dynamic balance (Haibach & Lieberman, 2013; Haibach-Beach, Perreault, Lieberman, & Foster, 2020), which is likely due to their sensory deficits in vision, hearing, and the vestibular system (Möller, 2011).

Children with CS also have significantly lower motor competence than their peers without disabilities (Haibach-Beach, Perreault, Lieberman, & Foster, 2019; Perreault Haibach-Beach, Lieberman, & Foster, 2021). For the present study, the term motor competence is used as a global term to describe the measurement of movement skills, such as fundamental motor skills (e.g., ball and locomotor skills; Barnett, Stodden, Hulteen, & Sacko, 2020; Robinson, Stodden, Barnett, Lopes, Logan, Rodrigues & D'Hondt et al., 2015). In particular, children with CS have the biggest delays in skills which require sufficient balance and coordination to be able to perform including the hop, underhand throw, and skip (Perreault, et al., 2021). Although children with CS demonstrate lower levels of motor competence when compared to their peers without disabilities, other external factors may influence their motor development.

Starting at birth, parents are critical in a child's development (Harter, 1987; Reed, 1991) and tend to play an important role in promoting children's motor competence (Reed, 1991). Parents create environments for their children to participate and practice motor tasks (Silva, Flôres, Corrêa, Cordovil, & Copetti, 2017), and support their children's environment by providing toys and equipment to develop motor competence (Cools, De Martelaer, Samaey, & Andries, 2011; Barnett, Hinkley, Okely, & Salmon, 2013). Parents of children with disabilities may be even more essential to their child's motor development than children without disabilities. For example, parents of youth with visual impairments provide elevated levels of support to their children; they encourage their children to be active, provide transportation, and financial assistance (Linsenbigler, Petersen, & Lieberman, 2018). However, parents may act as a barrier by overprotecting their children from getting hurt (McHugh & Pyfer, 1999; Stuart, Lieberman, & Hand, 2006). Knowing that parents are play a critical role to their children's motor development, the overprotecting barrier may impact their child's motor competence in a negative way.

The tripartite model of efficacy beliefs (Lent & Lopez, 2002) helps explain the potential influence of parents' perceptions on a child's motor competence. This model extends Bandura's social cognitive theory to other key factors that are believed to enhance behaviors, such as effort and performance (Jackson & Beauchamp, 2010). One of these key factors is the influence from relationships and interactions with significant people, such as parents. This source of efficacy is important because it can reflect how much time the child will persist in various activities. If the 'other' does not believe in the child, the child may withdraw from the activity (Lent & Lopez, 2002). This is especially important given that development of fundamental motor skills are considered foundational to acquiring more complex movements (Clark & Metcalfe, 2002). In addition, motor competence in childhood is directly related to active engagement in physical activity (Lubans, Morgan, Cliff, Barnett & Okely 2010; Robinson et al., 2015). Children that tend to possess low proficiency in their motor competence, opt for less physically active lifestyles in order to avoid movement challenges (Wrotniak, Epstein, Dorn, Jones, & Kondilis, 2006). When a child has low motor competence, the likelihood of limited physical activity engagement and associated health problems, such as obesity and diabetes, tend to be high (Robinson et al., 2015). It has been shown that physical inactivity during childhood will result in physical inactivity in adolescence and into adulthood (Telama, 2009). Therefore, movement skill development experiences are critical for lifelong physically active lifestyles.

Although there is some research on motor competence in children with CS, there has been no research conducted on the parents' perceptions of children with CS with regards to motor competence. As asserted by the tripartite model of efficacy beliefs, parental perceptions, in particular, the value parents place upon motor competence and physical activity plays an important role in children's desire to be physically active (Lent & Lopez, 2002). It is possible that parents of children with CS may deprioritize motor competence (Bowling, Blaine, Kaur, & Davison,2019) due to the many additional communication, medical, and behavioral challenges children with CS experience (Hartshorne et al., 2021). Therefore, the purpose of this study was to examine parents' perceptions of motor competence in their child with CS as well as the value and support they provide upon motor competence compared to parents of children without disabilities. These findings will assist in the understanding of how parents of children with CS prioritize motor competence. In addition, this information can assist in the development of a more effective.

Materials and Methods

Participants

Parents of 33 children (11 boys, 22 girls) with a mean age of 6.75 year (SD = 2.57 years) with CS who attended the 2019 International CS Conference in Dallas, TX participated in this study. The control group was recruited from the western New York local area and consisted of parents of 38 children without disabilities of similar age (M = 6.97 years, SD = 2.27) and gender distribution (19 males, 19 females). All parents included in the study were mothers, with the exception of three fathers who were all parents of children without disabilities. Participants completed a demographics survey about their child's age, sex, and CHARGE characteristics (if applicable). Based upon parental report, 26 of the children with

CS had severe to profound hearing loss and the other 7 participants had mild to moderate hearing loss in one or both ears. Fifteen participants were missing their semicircular canals, 10 had partially formed semicircular canals, 3 had fully formed semicircular canals, and 6 did not report. All participants had vision loss which varied from B1, blindness to low vision B4 per the IBSA/USABA visual classification scales (IBSA, 2021), with 14% indicating blindness, B1, in one eye. Most (61%) of the participants experienced growth deficiencies and 81% indicated their child had heart defects. In addition, approximately half of the parents indicated their child had atresia choanae. The participants averaged 12.1 extended hospitalization stays. The participants age of independent walking was mean age of 38.5 months (SD = 17.5).

Measure

Parents' perceptions were measured by the Child Movement Skills Research: Parent Questionnaire (Clancy, Jackson, Dimmock, & Thornton, 2017; Jackson, Whipp, Chua, Pengelley, & Beauchamp, 2012). This 32-item questionnaire focuses on how well parents think their child can carry out different movement skills, such as kicking, striking, throwing, running, galloping, etc. The first set of questions (12 items) measures how confident parents are in their child's ability to perform 12 different movement skills compared to his or her same age peers (e.g., *Right now, compared to his/her peers of the same age, how confident are you in your child's ability to do the following things consistently well…*). Parents respond using a 5-point Likert scale, with 1 having no confidence at all and 5 having complete confidence. The next set of questions (13 items) measures how important parents think these movement skills are, and whether or not they think it is possible for (a) their child, and (b) people in general, to do much about how good they are at these movement skills (e.g., *How much do you agree with the following statements?).* Parents rate how much they agreed with the statements on a 7-point Likert scale, with 1 being strongly disagree and 7 being strongly agree. The following set of questions (three items) measures how often the parents provided support (encouragement, participation, transport) to their child's movement skills (e.g., *How often do you do the following?*). Parents respond using a 5-point Likert scale, with 1 being never or rarely and 5 being daily. The final set of questions (four items) measures how often a particular method (seeking opportunities, planning activities, setting goals, tracking practice) was used to encourage or support their child to participate in movement skills (e.g., *How often do you use the following tactics?).* Parents respond using a 5-point Likert scale, with 1 being never and 5 being always. There was excellent internal consistency, from reliability analyses, for each set of items (α = .83 - .96) except items measuring if parents feel that people in general can do much to improve their motor competence (α = .039); thus, these items were removed from the analysis.

Procedure

The Institutional Review Board at the SUNY Brockport approved all procedures. Informed consent was obtained from each participant. Participants completed a demographics questionnaire about their child's age, sex, CS characteristics followed by the parents' perceptions questionnaire. This study is part of a larger study examining motor competence in youth with CS that included a subset of this sample (n=6) who participated in a pilot intervention.

Data Analysis

Parent's ratings of his or her child's motor competence for the locomotor skills (i.e., hop, gallop, run, jump, leap, and slide) were averaged into one overall locomotor rating score. This process was repeated for the ball skills ratings (i.e., catch, throw, strike, kick, roll, and dribble) to produce an overall ball skills rating score and for all skills ratings to produce a total skill rating score. Next, the six items associated with whether or not parents think it is possible for their child to do much about their movement skills were averaged to create a single score. All other items were kept as individual scores. Alpha was set at .05 for all analyses. Mann-Whitney U tests were conducted to determine differences in scores on the parents' perceptions questionnaire between the parents of children with and without CS.

Results

There were significant group differences in parents' ratings of their children's motor competence across all skills (Table 1). Specifically, the parents of the children with CS rated their child's motor competence for locomotor (U = 71, *p* < .001, *r* = .76), ball skills (U = 108.5, *p* < .001, *r* = .71), and total skills (U = 76.6, *p* < .001, *r* = .75) lower than the parents of children without disabilities. There were no group differences in parents' belief that their child can change his or her motor competence. There were also no group differences in how important parents view their child's motor competence. When it came to support, the parents of children with CS participated more often with their child in movement skills than the parents of children without disabilities (U = 225, *p* < .001, *r* = .56). The parents of children with CS also tracked their child's practice of movement skills more often than the parents of children without disabilities (U = 440.5, *p* = .04, *r* = .24). There were no group differences in how often parents provided other types of support.

Table 1. Comparison of perceptions between parents of children with and without CHARGE syndrome.

	CHARGE (*N* = 33)		Controls (*N* = 38)		*z*	*p*	*r*
Variable	Median	Mrank	Median	Mrank			
Locomotor Rate	2.50	19.15	4.67	50.63	6.44	<.001	.76
Ball Skill Rate	2.67	20.29	4.50	49.64	6.00	<.001	.71
Total Skill Rate	2.58	19.32	4.58	50.49	6.35	<.001	.75
Child Change	4.00	31.94	4.00	39.53	1.63	.103	.19
Skill Importance	6.00	35.59	6.00	36.36	0.16	.872	.02
Encourage	5.00	36.36	5.00	34.78	0.37	.711	.04
Participate	4.00	47.47	3.00	25.42	4.76	<.001	.56
Transport	3.50	35.48	3.50	35.51	0.01	.995	.001
Seek Options	4.00	36.98	4.00	34.43	0.60	.550	.07
Plan Activities	4.00	36.81	4.00	33.43	0.73	.465	.09
Set Goals	3.00	39.11	3.00	32.46	1.41	.159	.17
Track Practice	3.00	40.73	2.50	31.09	2.03	.043	.24

Discussion

This study examined differences in parental perceptions of motor competence between children with and without CS and the value and support parents of children with CS provide upon motor competence compared to parents of children without disabilities. Overall, parents of children with CS had lower perceptions of their child's motor competence in comparison with parents of children without disabilities. This may likely be due to the issue of multiple surgeries and prolonged hospitalizations not often found in children without disabilities. Previous research has found that children with CS are delayed in motor competence in comparison to their peers without disabilities (Haibach-Beach et al., 2019; Perreault et al., 2021), which often concerned parents. It has been well documented that children with CS already experience delayed walking (Foster et al., 2020; Haibach-Beach et al, 2020), balance deficits (Haibach et al., 2013; Haibach-Beach et al., 2020), and vestibular challenges (Möller, 2011), which can contribute to these delays in motor development. As such, children with CS have significantly lower locomotor, ball skills, and total skill scores compared to their peers without disabilities. The consistent lower performance in motor competence in children with CS may be the basis for the lower expectations for their children.

A key component in the tripartite model of efficacy beliefs (Lent & Lopez, 2002) is other efficacy and the extent to which, important others in an individual's life play on certain tasks such as motor competence. Parents may be an important source of other efficacy in a child's life, with or without disabilities. If parents do not believe in the child's movement skills and show their support, the child may lose interest in the skill or activity altogether (Kef & Dekovic, 2004). This current study found that the parents of children with CS spent more time engaging in movement skills with their child than parents of children without disabilities. This finding may likely be due to the fact that they know their children will perform behind their same-age peers without the additional help and support in this area. It was clear that these parents valued motor competence as they were shown to track their child's performance on a more regular basis than parents of children without CS.

In a recent study by Lane and colleagues (2020), it was found that parents believed that motor competence acquired through physical education would help their children develop life skills such as socialization, independence, balance, stamina, and mobility. Gaining appropriate foundational skills would also open up typical opportunities for their children such as sports teams, recreation, and fitness. Parents of children with CS have also indicated that they prefer additional support such as a paraeducator to be with their child in physical education (Lieberman et al., 2012). This 1:1 nature when teaching physical activity is more beneficial for children with multiple and severe disabilities than small groups or whole class instruction (Grenier & Lieberman, 2018). This value in a more 1:1 setting with more attention for their child is evident in the findings of this study provide support that more attention should be placed upon individualized work at home with tracking progress. Although this is the first study of its kind for children with severe disabilities, it is believed that the tripartite model lends itself well to this approach.

Future research should explore the child's perception of parental support. Children's perception of parental support may act as a moderator on the parental influences on the child's physical activity outcomes (Davison et al., 2013). In addition future research should explore the beliefs of adapted physical educators and paraeducators in addition to parents as teachers also play a vital role in motor competence.

According to the current research by Lane and colleagues (2020), it was also found that increased communication between the parents and the physical education teacher is beneficial for the child. This increased focus on improving motor competence at home can transfer to the physical education environment and benefit the teacher and the child with CS if communication can be improved. Naturally, communication can be improved by these parents once they know the skill level of their child. The focus on tracking the skill level found in this study can ensure they have data to communicate with the teachers.

The development of fundamental movement skills is considered the building blocks to more complex motor skills (Clark & Metcalfe, 2002) which are critical components that influence physical activity levels and health indices throughout a person's life (Lubans et al, 2010; Robinson et al., 2015). The focus on motor competence shown in this study will improve the likelihood that individuals with CS will be independent and also have more choices related to physical activity and sport in the future. Studies by Lieberman and colleagues (2002; 2003; 2018) found that children and adults who were deafblind often did not engage in their preferred recreation or physical activities leading to a lack of self-determination. This increased focus on motor competence can likely decrease the gap found between children with and without CS and increase the likelihood that they will have opportunities to participate in their preferred physical activities.

Limitations

The current study includes some limitations. Given the low incidence and geographical distribution of CS in the general population, a random sample was not a viable option. Thus, the children with CS were recruited from a biannual international conference on CS – a convenience sample. The conference brings in a large number of professionals and families from across the United States and other countries. Thus, the sample may not be representative of the entire CS population and possibly subject to sampling error. Additionally, we only included children with CS who can ambulate independently in the study; thus, the findings cannot be generalized to children with CS who do not meet these criteria. Given the limited research in this area, future studies should continue to assess motor competence in children with CS to have a more complete understanding of motor competence in this population. It is particularly important that researchers examine the psychometrics of parental perceptions of motor competence with actual motor competence in youth with severe disabilities. The authors emphasize that research has shown weak associations between parental reports and actual motor competence (Zysset et al., 2018); however, the purpose of this study was not only to examine parents' perceptions of their child's motor competence but also how they value motor competence. Parents of children with disabilities deprioritize motor competence (Bowling et al., 2019) yet parental influence is an important influence upon their child's motor competence (Lent & Lopez, 2002). Since preliminary evidence suggests children with CS have a large deficit in motor competence compared to their typically developing peers, understanding the role of the parents can help inform home interventions designed to improve motor competence in various contexts.

Perspectives

Parents play an important socializing influence upon physical activity levels in all children. Parents of children with disabilities have been found to rate fitness and skill improvement as well as friendship and teamwork as top reasons for participating in physical activity and are more likely to encourage physical activity when there are opportunities for their child to perform in self-contained or inclusive environments which encourage friendship and teamwork (Yao, Shapiro, & Liao, 2016). Young adults with visual impairments have also indicated the importance of support from parents in physical activity enjoyment (Haegele, 2019). Parents of children with CS tend to have lower expectations of their child's motor competence in comparison to their peers without disabilities; however, these parents understood the importance of developing motor competence as illustrated by their increased support of working with their children and tracking their motor competence. It is imperative that researchers and practitioners continue to provide parents of children with CS the tools to assist their children to reach their potential (Haibach-Beach et al., 2019; Perreault et al., 2020). Each child with CS has unique physiological characteristics and it is critical to provide additional support and opportunities to be physically active in order for these children to be able to acquire motor competence. In addition, parents should seek to modify the equipment and provide a variety of instructions and verbal cues to assist their child (Foster et al., 2019; Lieberman & Haibach, 2016). Future research should examine the associations of parental perceptions upon motor competence in children with CS. Motor competence and a physically active lifestyle are important to increase a healthy lifestyle and lead to self-determined adults.

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References

Barnett, L., Hinkley, T., Okely, A. D., & Salmon, J. (2013). Child, family and environmental correlates of children's motor skill proficiency. *Journal of Science and Medicine in Sport*, *16*(4), 332-336. https://doi.org/10.1016/j.jsams.2012.01.011

Barnett, L. M., Stodden, D. F., Hulteen, R. M., & Sacko, R. S. (2020). Motor competence assessment. In T. Brusseau, S. Fairclough, & D. Lubans. *The Routledge Handbook of Youth Physical Activity*. (pp. 384-408). New York, NY: Routledge.

Blake, K.D., Davenport, S.L., Hall, B.D., Hefner, M.A., Pagon, R.A., Williams, M.S., … Graham, J.M. (1998). CHARGE association: An update and review for the primary pediatrician. *Clinical Pediatrics, 37*, 159-173. https://doi.org/10.1177/000992289803700302

Blake, K.D., & Prasad, C. (2006). CHARGE syndrome. *Orphanet Journal of Rare Diseases,1*, 34. https://doi.org/10.1186/1750-1172-1-34

Bowling, A., Blaine, R. E., Kaur, R., & Davison, K. K. (2019). Shaping healthy habits in children with neurodevelopmental and mental health disorders: Parent perceptions of barriers, facilitators and promising strategies. *International Journal of Behavioral Nutrition and Physical Activity*, *16*(1), 52. https://doi.org/10.1186/s12966-019-0813-6

CHARGE Syndrome (2019). Retrieved from https://rarediseases.org/rare-diseases/charge- syndrome

Clancy, C., Jackson, B., Dimmock, J. A., & Thornton, A. L. (2017). Parents and the Pygmalion Effect: Exploring relations between parent efficacy beliefs and children's fundamental movement skills. *Journal of Sport & Exercise Psychology*, *39*, S56-S56.

Clark, J. E., & Metcalfe, J. S. (2002). Mountain of motor development: A metaphor. In J. E. Clark & J. Humphrey (Eds.), *Motor development: Research and reviews* (Vol. 2, pp. 163–190). Reston, VA: NASPE Publications.

Cools, W., De Martelaer, K., Samaey, C., & Andries, C. (2011). Fundamental movement skill performance of preschool children in relation to family context. *Journal of Sports Sciences*, *29*(7), 649-660. https://doi.org/10.1080/02640414.2010.551540

Dammeyer, J. (2012). Development and characteristics of children with Usher syndrome and CHARGE syndrome. *International Journal of Pediatric Otorhinolaryngology, 76,* 1292-1296. https://doi.org/10.1016/j.ijporl.2012.05.021

Davison, K.K., Mâsse, L.C., Timperio, A., Frenn, M.D., Saunders, J., Mendoza, J.A., Gobbi, E., Hanson, P., & Trost, S.G. (2013). Physical activity parenting measurement and research: Challenges, explanations, and solutions. *Childhood Obesity*, 9 (Suppl. 1), S103-S109. https://doi.org/10.1089/chi.2013.0037

Deuce, G., Howard, S., Rose, S., & Fuggle, C. (2012). A study of CHARGE Syndrome in the UK. *The British Journal of Visual Impairment, 30*(2), 91-100. https://doi.org/10.1177/0264619612443883

Foster, E., Silliman-French, L., & Grenier, M. (2020). Parents' perceptions of constraints impacting the development of walking in children with CHARGE syndrome. *Research and Practice for Persons with Severe Disabilities, 45*(3), 196-211. https://doi.org/10.1177/1540796920927656

Foster, E., Lieberman, L., Perreault, M., & Haibach-Beach, P. (2019). Constraints model for improving motor skills in children with CHARGE syndrome. *Palaestra, 33*(2), 39-44. https://doi.org/10.1177/0145482X20939469

Grenier, M., & Lieberman, L.J. (Eds) (2018). *Physical education for children with moderate to severe disabilities*. Champaign, IL: Human Kinetics.

Haegele, J. (2018). Youth leisure-time physical activity from the perspectives of young adults with visual impairments. *European Journal of Adapted Physical Activity, 11*(2), 1-13. https://doi.org/10.5507/euj.2018.010

Haibach-Beach, P., Perreault, M., Lieberman, L., & Foster, E. (2020). Independent Walking and Balance in Children with CHARGE Syndrome. *British Journal of Visual Impairment*. 00(0), 1-10. https://doi.org/10.1177/0264619620946068

Haibach-Beach, P., Perreault, M, Lieberman, L., & Foster, E. (2019). Gross motor skill performance in children with CHARGE Syndrome: Research to Practice. *Research in Developmental Disabilities*, 91. https://doi.org/10.1016/j.ridd.2019.05.002

Haibach, P., & Lieberman, L.J. (2013). Balance and the self-efficacy of balance in children with CHARGE Syndrome. *The Journal of Visual Impairment and Blindness, 107,* 297-309. https://doi,org/10.1177/0145482X1310700406

Harter, S. (1987). The determinants and mediational role of global self-worth in children. In: N. Eisenberg, (Ed.), *Contemporary Topics in Developmental Psychology* (pp. 219-242). New York, NY: John Wiley.

Hartshorne, T.S., Hefner, M.A., & Davenport, S.L.H. (2005). Behavior in CHARGE syndrome: Introduction to the special topic. *American Journal of Medical Genetics, 133A*, 228-231. https://doi.org/10.1002/ajmg.a.30541

Hartshorne, T.S., Heussler, H.S., Dailor, A.N., Williams, G.L. Papadopoulos, D., & Brandt, K.K. (2009). Sleep disturbances in CHARGE syndrome: types and relationships with behavior and caregiver well-being. *Developmental Medicine & Child Neurolology, 51*, 143–150. https://doi.org/10.1111/j.1469-8749.2008.03146.x

Hartshorne, T. S., Nicholas, J., Grialou, T. L., & Russ, J. M. (2007). Executive function in CHARGE syndrome. *Child Neuropsychology*, *13*(4), 333-344. https://doi.org/10.1080/09297040600850944

Imel, G., Hartshorne, T.S., Slavin, L.J., & Kanouse, S.K. (2020). Participation in and barriers to recreation in CHARGE Syndrome. *Palaestra, 34*, 38-43.

IBSA. International Blind Sports Association. (2021, March8). *Classification*. Retrieved from https://www.ibsasport.org/classification/

Jackson, B., & Beauchamp, M. R. (2010). Self-efficacy as a metaperception within coach–athlete and athlete–athlete relationships. *Psychology of Sport and Exercise*, 11(3), 188-196. https://doi.org/10.1016/j.psychsport.2009.12.005

Jackson, B., Whipp, P. R., Chua, K. P., Pengelley, R., & Beauchamp, M. R. (2012). Assessment of tripartite efficacy beliefs within school-based physical education: Instrument development and reliability and validity evidence. *Psychology of Sport and Exercise*, *13*(2), 108-117. https://doi.org/10.1016/j.psychsport.2011.10.007

Janssen, N., Bergman, J.E.H., Swertz, M.A., Tranebjaerg, L., Lodahl, M., Schoots, J., Hofstra, R.M.W., van Ravenswaaij-Arts, C.M.A., & Hoefsloot, L.H. (2012). Mutation update on the CHD7 gene involved in CHARGE syndrome. *Human Mutation, 33*, 1149-1160. https://doi.org/10.1002/humu.22086

Kef, S., & Deković, M. (2004). The role of parental and peer support in adolescents well-being: a comparison of adolescents with and without a visual impairment. *Journal of Adolescence*, *27*(4), 453-466. https://doi.org/10.1016/j.adolescence.2003.12.005

Lane, K., Lieberman, L.J., Haibach-Beach, P., Perreault, M., & Columna, L. (2020). Parental perspectives on physical education services for children with CHARGE

Syndrome. *Special Education Journal*, *55*(2), 90-100. https://doi.org/10.1177/0022466920942769

Lent, R. W., & Lopez, F. G. (2002). Cognitive ties that bind: A tripartite view of efficacy beliefs in growth-promoting relationships. *Journal of Social and Clinical Psychology*, *21*(3), 256-286. https://doi.org/10.1521/jscp.21.3.256.22535

Lieberman, L. J., Haibach, P., & Schedlin, H. (2012). Physical education and children with CHARGE Syndrome: Research to practice. *Journal of Visual Impairment and Blindness, 106*(2) 106-119. https://doi.org/10.1177/0145482X1210600205

Lieberman, L. J. & Haibach, P. S. (2016). *Gross Motor Development Curriculum: For Children With Visual Impairment*. Louisville, KY: American Printing House for the Blind.

Lieberman, L. J., & Kirk, T.N., & Haegele, J.A (2018). Physical education and transition planning experiences relating to recreation among adults who are deafblind: A recall analysis. *Journal of Visual Impairment and Blindness*, *112*, 73-86. https://doi.org/10.1177/0145482X1811200107

Lieberman, L. J., & MacVicar, J. (2003). Play and Recreation Habits of Youth who are Deaf-blind. *Journal of Visual Impairment and Blindness*, 97(12), 755-768. https://doi.org/10.1177/0145482X0710100706

Lieberman, L. J., & Stuart, M.E. (2002). Self-determined recreation and leisure choices of individuals with deaf-blindness. *Journal of Visual Impairment and Blindness,* 96(10), 724-735. https://doi.org/10/1177/0145482X0209601004

Lieberman, L. J., Stuart, M. E., Hand, K., & Robinson, B. (2006). An investigation of the motivational effects of talking pedometers among children with visual impairments and deaf-blindness. *Journal of Visual Impairment & Blindness*, 100(12), 726-736. https://doi.org/10.1.1.837.4406

Linsenbigler, K., Petersen, S., & Lieberman, L. (2018). Barriers to physical activity for children with visual impairments: How far have we come and where do we still need to go? *Palaestra, 32*(1), 26-31.

Lubans, D.R., Morgan, P.J., Cliff, D.P., Barnett, L.M., & Okely, A.D. (2010). Fundamental movement skills in children and adolescents: Review of associated health benefits. *Sports Medicine, 40*(12), 1019-1035. https://doi.org/10.2165/11536850- 000000000-00000

McHugh, E., & Pyfer, J. (1999). The development of rocking in children who are blind. *Journal of Visual Impairment & Blindness, 93*(2), 82-95. https://doi.org/10.1177/0145482X9909300203

Möller, C. (2021). Overview of balance and the vestibular system. In T.S. Hartshorne, M.A. Hefner, K.D. Blake (Eds.), *CHARGE syndrome*. (pp. 65-76). San Diego, CA: Plural Publishing.

National Center on Deaf-Blindness (2018, October). The 2017 National Child Count of Children and Youth who are Deaf-Blind. Retrieved from https://nationaldb.org/reports/national-child-count-2017

Pagon, R.A., Graham Jr., J.M., & Zonana, J.Y.S.L. (1981). Coloboma, congenital heart disease, and choanal atresia with multiple anomalies: CHARGE association. *Journal of Pediatrics, 99*, 223-227. https://doi.org/10.1016/s0022-3476(81)80454-4

Perkins, K., Columna, L., Lieberman, L.J., & Bailey, J. (2013). Parental perceptions toward physical activity for their children with visual impairments and blindness. *Journal of Visual Impairments and Blindness, 107,* 131-142. https://doi.org/10.1177/0145482X130700206

Perreault, M., Haibach-Beach, P., Lieberman, L., & Foster, E. (2021). Motor competence in children with CHARGE syndrome. *Research and Practice for Persons with Severe Disabilities*. https://doi.org/10.1177/1540796921998011

Perreault, M., Haibach-Beach, P., Lieberman, L., & Foster, E. (2020). Relationship between motor skills, balance, and physical activity in children with CHARGE syndrome. *Journal of Visual Impairment & Blindness*, *114*(4), 315-324. https://doi.org/10.1177/0145482X20939469

Reed, E. S. (1991). Cognition as the cooperative appropriation of affordances. *Ecological Psychology*, *3*(2), 135-158. https://doi.org/10.1207/s15326969eco0302_5

Robinson, L. E., D. F. Stodden, L. M. Barnett, V. P. Lopes, S. W. Logan, L. P. Rodrigues & E. D'Hondt (2015). Motor competence and its effect on positive developmental trajectories of health. *Sports Medicine, 45*(9), 1273-1284. https://doi.org/10.1007/s40279-015-0351-6

Salem-Hartshorne, N., & Jacob, S. (2005). Adaptive behavior in children with CHARGE syndrome. *American Journal of Medicine Genetics A, 133A*, 262-267. https://doi.org/10.1002/ajmg.a.30546

Silva, S. D., Flôres, F. S., Corrêa, S. L., Cordovil, R., & Copetti, F. (2017). Mother's perception of children's motor development in southern Brazil. *Perceptual and Motor Skills*, *124*(1), 72-85. https://doi.org/10.1177/0031512516676203

Stuart, M.E., Lieberman, L., & Hand, K.E. (2006). Beliefs about physical activity among children who are visually impaired and their parents. *Journal of Visual Impairment and Blindness, 100*(4), 223-234. https://doi.org/10.1177/0145482X0610000405

Telama, R. (2009). Tracking of physical activity from childhood to adulthood: A review. *Obesity Facts, 3*, 187-196. https://doi.org/10.1159/000222244

Yao, W.A., Shapiro, D., & Liao, C. (2016). Parents motivation for participation in physical activity for children with impairments. *European Journal of Adapted Physical Activity, 9*(1), 15-26. https://doi.org/10.5507/euj.2016.002

Wachtel, L.E., Hartshorne, T.S., & Dailor, A.N. (2007). Psychiatric diagnoses and psychotropic medications in CHARGE syndrome: a pediatric survey. *Journal of Developmental Physical Disability, 19*, 471-483. https://doi.org/10.1007/s10882-007-9064-6

Zysset, A.E., Kakebeeke, T.H., Messerli-Bürgy, N., Meyer, A.H., Stülb, K., Leeger-Aschmann, ... Jenni, O.G. (2018). The validity of parental reports on motor skills performance level in preschool children: A comparison with a standardized motor test. *European Journal of Pediatrics, 177,* 715–722. https://doi.org/10.1007/s00431-017-3078-6