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Examining attitudes towards sexuality in CHARGE syndrome

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By

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The current study aimed to examine attitudes of sexuality of individuals with CHARGE syndrome by exploring potential barriers that may exist due to differences in the attitudes towards sexuality in parents of individuals with CHARGE syndrome and those attitudes of adults with CHARGE syndrome. Participants included 24 adults with CHARGE syndrome and 31 parents of individuals with CHARGE syndrome. The *Attitudes of Sexuality-Individuals with Intellectual Disabilities* (ASQ-ID) was completed by all participants and was adapted to address this specific population for the current study. Using a multivariate analysis of variance (MANOVA), four themes of sexuality from the ASQ-ID (i.e., sexual rights, parenting, non-reproductive sexual behavior, and self-control) were examined by comparing differences of mean scores across parents of individuals with CHARGE syndrome and adults with CHARGE syndrome. Further, standard linear regressions were used to determine if age was a predictor of attitudes of sexuality within this population. Results indicated that while there was not a statistically significant difference between parents' overall scores and adults with CHARGE syndrome's overall score, statistically significant differences were found in two of the subscales. Specifically, adults with CHARGE syndrome were found to have more

positive views in the area of parenting, as compared to parents of individuals with CHARGE syndrome. Likewise, parents of individuals with CHARGE syndrome were found to have more positive views in the area of self-control, opposed to adults with CHARGE syndrome. Further, age was not found to be a predictor of attitudes of sexuality within this population. The results and implications of the findings are discussed, as well as limitations and future directions.

DEDICATION

I would like to first dedicate this dissertation my partner, my rock, William. You never cease to amaze me. Thank you for sticking by my side throughout this roller coaster of a ride. Thank you for celebrating with me in highs, and for your comfort and unwavering belief in me in the lows. There is no way I could have made it through this journey without you. I love you and appreciate you so much.

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CHAPTER I

INTRODUCTION

In 1975, the World Health Association (WHO) laid the framework for a comprehensive approach to sexual health by including social, emotional, and intellectual aspects within their definition of sexual health in addition to the typical prevention of sexually transmitted diseases (STDs; now commonly known as sexually transmitted infections [STIs]) and HIV approach (WHO, 1975). Moreover, in the past three decades, the development of sexuality and sexual health has evolved to encompass themes of physical, emotional, mental, and social well-being, including sexual rights in sexual health (WHO, 2015). Specifically, WHO has taken an initiative to bring light to the universal concern of discrimination and inequality individuals experience in regard to sexual health. Discrimination within sexual health embodies a plethora of situations that anyone may face at a given time (e.g., individuals perceived to have socially unacceptable sexual practices, teenage pregnancy, homosexuality, individuals who are HIV-positive, lack of sexual health education, etc.; WHO, 2015). However, for individuals with disabilities, the risk for inequality and discrimination within sexuality and sexual health is severely heighten, as this population is often considered vulnerable due to physical or cognitive impairments. Further, research suggests that more so than others, individuals with disabilities face a general lack of acceptance within society, but particularly with regard to expression of their sexuality and access to sexual health education and information (Barnard-Brak, Schmidt, Chesnut, Wei, & Richman, 2014).

Specifically, within the United States alone, efforts to address the sexual health crisis has included investigating the most appropriate methods of providing sexuality education to schools and communities (Chappell, 2018; Elia & Tokunaga, 2015). Empirical evidence has supported the notion that comprehensive sexuality education should be provided to all students, incorporating developmentally appropriate aspects from a young age (e.g., safety) and continuing throughout their education so that individuals know their rights and are able to make well-informed decisions about their health (Barnard-Brak et al., 2014).

Unfortunately for individuals with disabilities, these educational experiences are often not afforded to them (Fader Wilkenfeld & Ballan, 2012; Howard-Barr et al., 2005), and if they are able to receive some exposure to sexuality education in school, it often fails to address issues that align to their specific disability. Thus, the responsibility of sexual health education and development of sexual identity for individuals with disabilities lies largely on their parents (Swango-Wilson, 2008a). Although research suggests parents want to be involved in this process, they often view this task as challenging and feel as if they do not possess the necessary skills to adequately address comprehensive sexuality education (Dupras & Dionne, 2012; Swango-Wilson, 2008a). Additionally, parents of children with disabilities are often reluctant to acknowledge their child's potential desire or ability to engage in sexual relationships (Brown & Pirtle, 2008).

Having a lack of sexuality education and awareness often perpetuates negative sexual perceptions in individuals with disabilities, as these individuals begin to internalize the negative social perception and stigmas (Addlakha, 2007; Ballan, 2001; Brown & Pirtle, 2008). Attitudes and perceptions of sexuality have been explored in individuals with various disabilities, including intellectual disabilities (ID; Bernert & Ogletree, 2013; Medina-Rico, Lopez-Ramos, &

Quinonez, 2017) and physical disabilities (Addlakha, 2007; Krupa, MacNeill, & Ma, 2010) in hopes of improving their overall wellbeing and eliminate deprivation of human rights. However, the literature lacks exploration of perceptions of sexuality from individuals with medically complex developmental disabilities (Fader Wilkenfeld & Ballan, 2011). Additionally, while comparisons of perceptions of sexuality have been made between caregivers and individuals with disabilities, to date, a study has yet to investigate perceptions of sexuality from individuals with a disability and parents of individuals with a disability within a specific a population (Brown & Pirtle, 2008).

The present study aimed to join efforts in the current initiative to provide sexual rights to all by exploring the attitudes towards sexuality in CHARGE syndrome, a multifaceted genetic condition, where individuals present with a variety of both physical and cognitive disabilities and it is the leading cause of deafblindness (U.S. Department of Education, National Center on Deaf-Blindness, 2017). CHARGE syndrome is an acronym named to represent the collection of characteristics commonly exhibited by individuals with this genetic condition (i.e., coloboma, heart defects, atresia choanae, retarded growth and development, genital hypoplasia, and ear anomalies and deafness; Blake, Salem-Hartshorne, Daoud, & Gradstein, 2005). Often these individuals will present with facial and genital abnormalities, as well as severe medical complications. Likewise, individuals with CHARGE syndrome exhibit a wide range of cognitive functioning, which can contribute to the many barriers in sexuality this population faces. This study extended the literature by exploring attitudes of sexuality in individuals who present with deafblindness and other complex medical concerns.

In addition to addressing sexuality in individuals with complex medical concerns, amongst physical and cognitive disabilities, Brown and Pirtle (2008) indicated that future

research should investigate beliefs held by the individual with a disability and how they view their own sexual development and sex education, as well as examining if those beliefs differ from their parents or caregivers. This exploratory study sought to extend the literature by comparing attitudes of sexuality from adults with CHARGE syndrome to parents of individuals with CHARGE syndrome. Understanding perceptions of sexuality from both perspectives of individuals with a disability and adults involved with individuals with disabilities are the first steps in creating a positive change agent in education to prevent further discrimination and improving quality of life for all.

Research Questions

Research Question #1: Are there differences in attitudes of sexuality between caregivers of individuals with CHARGE syndrome and individuals with CHARGE syndrome?

Hypothesis: Individuals with CHARGE are likely to have higher attitudes of sexuality than caregivers of individuals with CHARGE syndrome.

Research Question #2: Does age of individuals with CHARGE syndrome predict attitudes of sexuality for individuals with CHARGE syndrome?

Hypothesis: Age will likely predict attitudes of sexuality.

Research Question #3: Does age of parents predict attitudes towards sexuality for individuals with CHARGE?

Hypothesis: The older the parent or caregiver, the more conservative they are likely to be (Cuskelly & Bryde, 2004; Murray & Minnes, 1994).

Research Question #4: Are there differences between individuals with CHARGE syndrome who have had exposure to sex education or not with regard to attitudes towards sexuality?

Hypothesis: Individuals with CHARGE will have more positive attitudes of sexuality having received sexuality education (Ballan, 2012; Barnar-Brak et al., 2014; Medina-Rico, Lopez-Ramos, & Quinonez, 2017).

CHAPTER II

LITERATURE REVIEW

While the topic of sexuality is still often considered taboo, sexual health is critical to the physical and emotional health of all individuals, couples, and communities. Sexual health has developed beyond physical wellbeing, to mirror more comprehensive theories of human development by embodying various aspects of development, such as physical, emotional, mental, and social wellbeing (WHO, 2015). This idea of sexual health now incorporates issues of negative perceptions, discrimination, and inequality of individuals in regard to sexual health, including the inability to access sexuality education and resources. Unfortunately, individuals with disabilities are often faced with multiple barriers that contribute to overall poor sexual health resulting in negative self-perceptions of sexuality (East & Orchard, 2014; Fader Wilkenfeld & Ballan, 2011). Examining these perceptions or attitudes of sexuality provides insight to overcome these barriers, such as knowledge gaps, misperceptions, and desired resources within a specific population. While this area of research has been explored in individuals with either cognitive or physical disabilities (Addlakha, 2007; Calam, 2012; East & Orchard, 2014; Medina-Rico et al., 2017; Suter, McCracken, Bernert, & Ogletree, 2013), attitudes of sexuality have yet to be explored in individuals with medically complex or multiple disabilities, one such population being CHARGE syndrome. CHARGE syndrome is a genetic condition characterized by the presentation of multiple anomalies and is the leading cause of congenital deafblindness (U.S. Department of Education, National Center on Deafblindness,

2017). While this condition generates many obstacles that can hinder an individual's sexual health and obtain sexuality education, to date, the literature is absent of any investigations of sexuality and individuals with CHARGE syndrome. Exploring attitudes of sexuality within this population will provide the researchers with a better understanding of sexual health needs of this population (e.g., lack of resources, lack of sufficient knowledge, misinformation, etc.) and why that might be. The capacity to understand the present situation requires an overview of human sexuality and its theoretical implications, as well as examination of the history of sexuality and disability, barriers and social injustices individuals with disabilities face regarding their sexual health, and the resulting attitudes of sexuality of individuals with disabilities. Further, the literature review will provide a deeper understanding of CHARGE syndrome and rationale for exploring attitudes of sexuality within this population.

Human Sexuality

Human sexuality is an ever-evolving aspect of humanity that encompasses sex, gender identities and roles, sexual orientation, eroticism, pleasure, intimacy, and reproduction throughout the lifespan (WHO, 2002). Sexuality is an important aspect of physical, psychological, and social health, as well as overall life satisfaction (Esmail, Darry, Walter, & Knupp, 2010). Expression of sexuality is a basic human right that should be afford to all human beings. Sexuality can be experienced and expressed through various facets, such as thoughts, fantasies, desires, beliefs, attitudes, values, behaviors, practices, roles, and relationships (WHO, 2002). By defining how we interact with others, what relationships we develop, and how we love and show affection (Swango-Wilson, 2008b), sexuality allows us to fully build relationships with others (Medina-Rico et al., 2017).

The development of one's sexuality is influenced by a plethora of internal and external factors (e.g., biological, psychological, social, economic, political, cultural, ethical, legal, historical, religious, and spiritual; WHO, 2002). A desire to understand this phenomenon has sparked international concern, as the topic of sexuality has fueled declarations and studies both here and abroad. Initially fueled by significant health disparities related to sexuality (e.g., HIV/AIDs epidemic, STIs, unwanted pregnancies, unsafe abortions, infertility, sexual violence, and sexual dysfunction), researchers have discovered an even larger international concern involving discrimination and inequality related to sexuality and sexual health: a violation of human rights (WHO, 2006; 2015).

WHO has taken an initiative to bring light to the universal concern of discrimination and inequality individuals experience in regard to sexual health. This has resulted in the development the physical wellbeing of sexual health, to mirror more comprehensive theories of human development by embodying various aspects of development, such as physical, emotional, mental, and social wellbeing (WHO, 2015). Sexual health now incorporates issues of negative perceptions, discrimination, and inequality of individuals in regard to sexual health, including the inability to access sexuality education and resources. Discrimination within sexual health embodies a plethora of situations that anyone may face at a given time (e.g., individuals perceived to have socially unacceptable sexual practices, teenage pregnancy, homosexuality, individuals who are HIV-positive, lack of sexual health education, etc.; WHO, 2015). While great strides have been made, there is still a greater need for continued exploration and understanding of sexuality. The sexual rights of all people must be respected, protected, and fulfilled in order to fully grasp and maintain sexual health (WHO, 2002).

Theoretical implications

Theoretical frameworks are used in the field and psychology and sociology as a means to gathering greater insight on human behavior. Several theoretical frameworks have made efforts to explain the development of sexual attitudes and behaviors in humans, such as Freud's theory of psychosexual development, social learning theory (Bruess & Schroeder, 2014), and Bronfenbrenner's (1977) ecological systems theory (Jones, Meneses da Silva, Soloski, 2011). These frameworks further serve as models for developing strategies and instruction to influence the behavior of individuals when applied to sexuality education programs (e.g., increase knowledge on sexual health, motivation to use contraceptives, self-esteem, etc.; Brindis, Sattley, & Mamo, 2005).

Ecological systems theory. The comprehensive understanding of sexual health presents with similar theoretical framework as the Ecological Systems Theory (Bronfenbrenner, 1977). Consequently, as Bronfenbrenner (1977) was developing this model of human development, the World Health Organization began discussing expanding sexual health to be more inclusive of all aspects of human development (WHO, 1975). The Ecological Systems Theory incorporates a comprehensive working system of cognitive, emotional, biological, and behavioral aspects of human development. The theory conceptualizes human development within an interplay of systems known as the microsystem, mesosystem, ecosystem, macrosystem, and chronosystem (Bronfenbrenner, 1977). The microsystem describes the environment that the individual is directly operating in. In regard to sexuality, this section would incorporate an individual's sexual desires, values, personal scripts. Further, this system incorporates an individual's family and family values of sexuality, partner and partner's values and expectations, peers, and well as an individual's biological make up (Jones et al., 2011). A mesosystem includes the interactions that

take place between the microsystems of one's life (Bronfenbrenner, 1977). In terms of sexuality and sexual health, this is viewed as the social and intimate aspects of sexuality. This could also include stressors that affect your sexual desire (Jones et al., 2011).

According to Bronfenbrenner (1977) the exosystem includes institutions that are not located in an individual's immediate environment but influence an individual's daily life. Such institutions can include school, healthcare, community, and media, all of which involve aspects of sexuality in which you receive education, be confronted with sexual issues, or receive healthcare treatment (Jones et al., 2011). Further, the macrosystem are beliefs and messages from much larger systems operating in society, such as influence of culture and societal norms (Bronfenbrenner, 1977). Sexuality could be determined or influenced by gender norms, culture/ethnicity norms, and religion (Jones et al., 2011). Lastly, Bronfenbrenner (1977) describes the chronosystem as developmental changes throughout the life span, including puberty, menopause, and aging. This system is also unique in that it can impact one's desire or psychological process surrounding sex (e.g., miscarriage) or impacts and individuals experience with sexuality (e.g., abuse; Jones et al., 2011). While theoretical implications of the EST provide some basis of conceptualizing the interworking of human development into sexuality, it does however present with some limitations. Specifically, sexuality of individuals with disabilities might be missed in this view due to lack of educational and healthcare opportunities regarding sexuality (exosystem), negative societal misperceptions held regarding their sexuality (macrosystem), and potential abnormal sexual development (chronosystem).

Social learning theory/Social cognitive theory. Social learning theory is considered best practice as the framework for sexuality education programs (Future of Sex Education [FoSE], 2012), as theories that take into account both human behaviors and social influence have the

ability to target more areas for intervention, thus producing more meaningful change (Brindis et al., 2005). Social learning theory is found to be the core of many sexuality education programs, specifically those that are implemented in the United States (Kirby et al., 2007; Bruess & Schroeder, 2014). Originally based on the work of Rotter (1954) and more recently Bandura (i.e., social cognitive theory; 1991), many elements of social learning theory were taken from behaviorism and assumed that behavior is goal directed and individuals model behavior on that of others due to expectations of rewards. However, social learning theory diverges from operant learning by believing that behaviors can be chosen or increase in frequency and intensity without direct reinforcement (e.g., perceived reinforcement). Simply speaking, according to social learning theory, reinforcers in society have the ability to shape behavior and attitudes.

Social learning theory has been applied to various aspects of promoting sexual health across topics of sexual development, adolescent sexuality and contraceptive use, and health-related sexual behavior (Eisen & Zellman, 1990; Hogben & Byrne, 1998). One way in which this framework has been beneficial to acquiring sexual health knowledge, is through modeling. Social learning theory believes the act of learning occurs through the process of modeling, as humans engage in the behaviors that they have previously seen (Bruess & Schroeder, 2014). This aspect is particularly important with regard to sexuality education, as sexual decision making, and behaviors of individuals can be influenced by other's appropriate and inappropriate display of sexual behaviors. However, some disabilities, such as visual or hearing impairments, might not allow individuals to learn from models within their environment due to the inability to see or hear these models at all. Similarly, as a result of these impairments, individuals may not see the full model of sexual behaviors and misinterpret these acts resulting in a lack of knowledge and understanding of sexuality. Sexuality education programs rooted in social

learning theory have used modeling (with and without modifications) and reinforcement of specific behavior skills to teach the use of contraception and how to appropriately communicate with your partner about contraception (Hogben & Byrne, 1998).

Additionally, this theory acknowledges the impact of perceptions on attitudes and behavior. It allows program developers to take in account the social influence and the importance of the beliefs of young people in order to promote positive sexual behaviors (Bruess & Schroeder, 2014). For example, if an adolescent's group of friends tell him that they engage in sexual behavior, and the individual has not had sex yet, regardless if the friends were telling the truth, that fact that the individual believes his friends have sex will play a key role in his decision to engage in sexual activity (Bruess & Schroeder, 2014).

Further, this theory supports the assumption that sexual behaviors can be taught (Hogben & Byrne, 1998). When applied to pregnancy prevention, social learning theory postulates that a female will take birth control to avoid pregnancy and will continue to engage in taking birth control through reinforcement of obtaining her goal of not being pregnant and avoiding the negative social outcomes. She is also more likely to engage in conversations with her partner on using contraception if her teacher models the appropriate ways to handle that conversation (Kirby, Barth, Leland, & Fetro, 1991).

Nonetheless, the benefits of considering multiple theoretical frameworks for sexuality development allows for issues to be considered with multiple influences (e.g., individual, societal), information to be disseminated in different approaches (e.g., preventative, reactive), and interventions to be designed for various purposes (e.g., individual, target, universal). Thus, making sexuality education more robust and increasing the potential to address the needs of more individuals. However, while beneficial to providing an understanding of sexuality, theoretical

frameworks are just the structure upon where effective content, strategies, and interventions are built.

Individuals with Disabilities

Disability can be defined as a physical or mental impairment that impacts one or more major areas of daily functioning (e.g., school, work, relationships; Szydlowski, 2016). This definition can be applied to individuals who have record of impairment or are perceived by others to have an impairment. Disabilities vary greatly in severity and can include conditions such as visual, hearing, or motor impairment, cerebral palsy, genetic conditions (e.g., Down syndrome), developmental disorders, intellectual disabilities, mental or emotional health problem, or individuals with learning disabilities (Szydlowski, 2016). Despite how rare disabilities appear, the U.S. Bureau in 2010 reported that 2.8 million youth under the age of 15 in the United States had some form of physical, intellectual, or emotional disability (Brault, 2012), with many of these disabilities going unnoticed by others. However, due to series of historical civil rights movements in the United States, students with disabilities are afforded by law the same educational opportunities as their non-disabled peers.

Disabilities in Education

Educational opportunities began to change for students with disabilities beginning in the early 20th Century. This movement began in retaliation to the exclusion of individuals with disability from schools on the grounds of the *Brown v. Board of Education* (1954) landmark case ruling, in regard to equal educational opportunities for all people, as advocates for students with disabilities claimed that students should have equal rights to an education and equal treatment despite having a disability. Parents of individuals with disabilities joined forces with advocacy

groups and a movement for appropriate education for students with disabilities brought attention to this cause all over the nation. From here, a series of case action lawsuits (e.g., *Pennsylvania Association for Retarded Citizens* [PARC; 1972], *Mills v. Board of Education* [1972]) provided free public education for individuals with disabilities, ages 6-21, that exceeded solely academic experiences (e.g., including functional life skill instruction; Yell et al., 1998).

The federal government also made striking efforts to amend education for students with disabilities with the Elementary and Secondary Education Act of 1965, 20 U.S.C. 6301 et seq., which provided funding for public schools for education of students in specific categories including disabilities, as well as the Title VI amendment which added funding for programs specific to individuals with disabilities. In 1970, Title VI became the Education for the Handicapped Act (EHA), which is known as the fundamental framework of future special education legislation (Yell et al., 1998).

Three years later Section 504 of the Rehabilitation Act of 1973 was passed as the first real efforts to protect individuals with disabilities from discrimination. Following several amendments of previous laws, a collaboration of state and federal law was formed as the Education for All Handicapped Children Act of 1975, that mandated various education rights of students with disabilities (i.e., nondiscriminatory testing, least restrictive environment, procedural due process, free and appropriate education), and delineated appropriate educational governing bodies and funding sources (Yell et al., 1998). In 1990, this law took the form of the Individuals with Disabilities Education Act (IDEA), providing further clarification to special education categories, early intervention, and transition services (Yell et al., 1998). IDEA continues to serve as the federal legislation for special education services for students with

disabilities, with the most recent revisions in 2017 and includes access to social-emotional, adaptive skills, and behavioral services- which may include sexuality education.

Sexuality and Disability

While state and federal governments have made great strides to provide equal education, individuals with disabilities are still often not afforded the same educational opportunities, particularly with regard to sexuality education. The importance of sexuality is not limited to the typically developing individual, as the topic of sexuality is even more important for individuals with disabilities, as they are at greater risk of sexual abuse and sexual violence (Azzopardi-Lane & Callus, 2015; Medina-Rico et al., 2017). Researchers have found children with disabilities are nearly four times as likely as their nondisabled peers to be sexually abused (Skarbek et al., 2009; Sullivan & Knutson, 2000). They are afforded less access to sexual information and are less likely to know about rights and laws regarding sexual consent and abuse (Lynden, 2007), while often presenting with a lack of skills to make sensible decisions regarding their safety and well-being, including consenting to sexual acts (Swango-Wilson, 2008b).

Sexuality Education

Historically, in the United States, sexuality education has been limited and biased by not offering an educational experience fostering sexual health for all students (Barnard-Brak et al., 2014; Elia & Tokunaga, 2015). Many sex education curriculums are inadequate for individuals with disabilities. Thus, students with disabilities are often excluded from sexuality education discussions and do not receive any sexual health education at all (Boehning, 2006), particularly as their disabilities become more profound (Barnard- Brak et al., 2014). Furthermore, when sexuality issues are discussed, they typically contain content from a biological perspective, rather

than a social perspective (Ballan, 2012). Sexuality education should be comprehensive for all students (Chappell, 2018; Elia & Tokunaga, 2015), including aspects of anatomy, development, intimacy, relationships, safety, and protection (Ballan, 2012; Parchomiuk, 2013).

Often, these curricula need to be specifically tailor to address the disability of students and approached using a multidisciplinary team that includes an educator, nurse, social worker and personal care attendant (Fader Wilkenfeld & Ballan, 2011) to insure all information is accessible. When considering students with deafblindness, an intervenor and interpreter will need to be included. Further, students with disabilities are less likely to benefit from indirect curriculum methods (Boehning, 2006) and may require one-on-one instruction to acquire skills (Dukes & McGuire, 2009). Boehning (2006) developed best practices for sexuality education for individuals with disabilities. These included best practices for both content skills (e.g., effectively and politely refusing an offer, maintaining self-control, conversation initiation, and safety) and instructional supports (e.g., 3-dimension models, dolls, drawings, pictures, and diagrams explaining genitalia and its functions, video tapes; Boehning, 2006) for providing quality sexuality education. Moreover, Retznik (2017) suggested the emphasis of development of positive self-perception and self-imagine as a critical component of sexuality education for individuals with physical disabilities.

Lack of sexuality education has been linked to increased risk for negative effects, such as sexual abuse, human immunodeficiency virus (HIV), sexually transmitted infections (STIs), and engagement in risky sexual behavior. Individuals with disabilities have less access to sexual health information and are less likely to know about rights, laws, sexual consent, and abuse (Aderemi, 2014; Ballan, 2012; Lynden, 2007; Medina-Rico, et al., 2017). Curtiss and Ebata (2016) found that a general barrier to providing individuals with disabilities sexuality education

is the lack of knowledge and training by caregivers and teachers. From their research, Curtiss and Ebata (2016) determined there was a need for more workshops and trainings to help instructors and practitioners feel more prepared in this area, as skilled educators can be instrumental in the achievement of skills to make safe, healthy, and informed decisions regarding issues of sexuality for individuals with disabilities (Fader Wilkenfeld & Ballan, 2011; Howard-Barr et al., 2005).

In addition to a lack of knowledge and training, Eisenburg and colleagues (2013) examined further barriers to teaching sexual health in the classroom from the perspective of 368 middle school and high school teachers. They found that reported barriers included: lack of time, lack of financial resources, lack of curriculum, concern about parents' responses, concerns about students' responses, concerns about responses from administration, and school of district policy (Eisenberg et al., 2013). Additional barriers impeding the right to sexuality education are rooted in negative societal attitudes and perceptions of individuals with disability and sexuality (Krupa et al., 2010).

Attitudes Towards Sexuality and Disability

Societies often hold particular views about how and when sex should happen, between whom, and at what stage of one's life. For individuals with disabilities, these views are often quite negative, as societal perceptions about sexuality and individuals are fueled by myths and a lack of knowledge worldwide (Addlakha, 2007; Di Giulo, 2003; Fitzgerald & Withers, 2013). In efforts to further investigate attitudes of sexuality, Cuskelly and Bryde (2004) developed the *Attitudes towards Sexuality Questionnaire* (ASQ) to examine attitudes of sexual expression of individuals with disabilities. This instrument contained items relating to eight aspects of sexuality (i.e., sexual feelings, sex education, masturbation, personal relationships, sexual

intercourse, sterilization, marriage, and parenthood (Cuskelly & Bryde, 2004). After comparing responses from parents, staff involved with individuals with disabilities, and a community sample, Cuskelly and Bryde (2004) found that while generally attitudes were positive, the older a respondent was, the less positive they were. Further, parents of individuals with ID and staff involved with individuals with ID were less positive about parenthood in comparison to the other seven categories. This instrument was further developed by Cuskelly and Gilmore (2007) to reflect some of the stereotypical views of sexuality and establish normative data. Findings from this study resulted in a modified version of this instrument, the *Attitudes to Sexuality Questionnaire* (Individuals with an Intellectual Disability; ASQ-ID), which was identified by a factor analysis to address four themes of sexuality: sexual rights, parenting, non-reproductive sexual behavior, and self-control. Using this particular scale, many researchers have explored attitudes of sexuality of individuals with ID from staff involved with individuals with ID (Gilmore & Chambers, 2010; Meaney-Tavares & Gavidia-Payne 2012; Pebdani, 2016; Winarni, Hardian, Suharta, & Ediati, 2018), parents of individuals with ID (Tamas, Jovanovic, Rajic, Ignjatovic, & Prkosovacki, 2019; Winarni et al., 2018), and community samples (Di Marco, Licciardello, Mauceri, & La Guidara, 2013; Tamas et al., 2019; Winarni et al., 2018).

Further, individuals with disabilities are often perceived as asexual or hypersexual (Ballan, 2001), as men, particularly, are thought to have limited self-control over sexual urges (Gilmore & Chambers, 2010; Pebdani, 2016). Many people, including families and educators, do not acknowledge that individuals with disabilities desire intimacy, relationships, or are sexual beings (East & Orchard, 2014). Individuals with disabilities are not thought require the same sexual rights and freedoms as the typically developing population (Gilmore & Chambers, 2010;

Pebdani, 2016). These misperceptions attribute to lack of sexuality educational opportunities afforded to individuals with disabilities.

Educator opinions regarding sexuality and disability. Teachers often perceive students with disabilities as sexually innocent (Chappell, 2018) and having an inability to have intimate relationships (Aderemi, 2014). While some educators viewed sexuality as a basic human right, and while they felt confident in teaching individuals with disabilities sexuality education, they expressed concerns regarding individuals with disabilities' ability to consent (Fader Wilkenfeld & Ballan, 2011). Contrary, studies have found that educators and medical professionals, alike, express concern with having the necessary skill set to communicate relevant information on the topic of sexuality effectively (Aderemi, 2014; Fader Wilkenfeld & Ballan, 2011).

When investigating the sexuality education opinions of teachers of students with ID, Barnard-Brak and colleagues (2014) found that 54% of teachers reported that their students would benefit from sexuality education based on previous courses completed, although these results were higher for individuals with a milder intellectual impairment.

Parents and caregiver perceptions of sexuality and disability. Caregivers play an important role in the development sexual identity. They are in the position to provide experiences needed for appropriate social development, such as exploration of sexuality and awareness of self-protection skills (Swango-Wilson, 2008b). While parents want to participate in the sexuality education of their children with disabilities, they believe that the task is a difficult challenge to accomplish effectively (Dupras & Dionne, 2012; Swango-Wilson, 2008a) and are unprepared to deal with sexual issues. Parents of children with disabilities are often reluctant to acknowledge their child's potential to desire to engage in sexual relationships, although disabled children will develop in many areas in the same manner as their typically

developing peers (Brown & Pirtle, 2008). Further, caregivers are fearful that their child will suffer assault or be ridiculed for engaging in sexual behaviors (East & Orchard, 2014), thus parents tend to control their child's daily activities, reduce social activities and only address sexual topics in a reactive approach to correcting inappropriate behavior (Dupras & Dionne, 2012).

Sexually inaccurate or negative societal perceptions of individuals with disabilities perpetuate the lack of education provided to students, as some feel information about sexual health is not needed or not applicable to this population. These stigmas leave individuals susceptible to misinformation or increased vulnerability and extend to barriers that hinder resources and opportunities (Koller, 2000). Further, individuals with disabilities often report taking on the negative self-perception projected by society, which can lead to increased emotional and social concerns (Bernert & Ogletree, 2013).

Sexuality of Individuals with Disabilities

Individuals with disabilities often face difficulties and frustrations when trying to develop their own kind of sexual identity (Di Giulo, 2003; East & Orchard, 2014), as they internalize negative social assumptions (Di Giulo, 2003), feel socially rejected, and acquire negative attitudes about sex (Addlakha, 2007). Their experiences reflect many tensions between what they are denied access to because of their disability, including sexual education information and instruction and limited sexual expression by caregivers (Azzopardi-Lane & Callus, 2015; Bernert & Ogletree, 2013).

Individuals with cognitive disabilities (Azzopardi-Lane & Callus, 2015; Bernert & Ogletree, 2013; Medina-Rico et al., 2017) and physical disabilities (Addlakha, 2007; East & Orchard, 2014; Suter et al., 2012), alike, report facing challenges and difficulties in sexuality

development and an overarching desire for intimacy. Bernert and Ogletree (2013) found that individuals with ID largely held negative perceptions of sex, specifically in regard to fear in the first act, fear of experiencing negative consequences, and perceived or experienced lack of pleasure. Individuals with ID also report feeling constrained due to lack of privacy and limited finances, as they often depend on others for support (Azzopardi-Lane & Callus, 2015).

In addition to similar negative perceptions of sexuality those with cognitive disabilities face, individuals with physical disabilities often face negative perceptions and experiences unique to their physical handicap. For example, individuals with hearing impairments often lack incidental learning opportunities in sexuality (Suter et al., 2012), while individuals with vision impairments report difficulty navigating menstruation (Addlakha, 2007) and obtaining visual cues, such as reading body language (Krupa et al., 2010). Individuals with mobility issues or limited use of limbs, such as those with Cerebral Palsy (CP), often report frustration with the lack of information regarding the impact of their physical restraints on their sexual health (Krupa et al., 2010).

Negative sexual perceptions held by individuals with disabilities are often attributed to a lack of education and awareness in this topic. Krupa and colleagues (2010) found that many individuals report that the lack of sexual health education negatively impacted their self-esteem, self-worth, and identity, as well as put them at greater risk for unpleasant or dangerous sexual experiences. Therefore, understanding perceptions of sexuality from both perspectives of individuals with a disability and adults involved with individuals with disabilities are the first steps in creating a positive change agent in education to prevent further discrimination and improving quality of life for all.

Unfortunately, individuals with disabilities often find themselves in situations where no one wants to address their sexuality (Brown & Pirtle, 2008). Most report that sexuality was not addressed growing up (East & Orchard, 2014). In fact, individuals with disabilities largely feel as if this topic was avoided altogether in their childhood or ill-received by healthcare professionals when questions were presented (Krupa et al., 2010). For those fortunate enough to get some exposure to sexuality education, mothers and friends were reported to be the most frequent sources of this information, followed by teachers and school nurses (East & Orchard, 2014).

In support of the positive effects of sex education for individuals with disabilities, several studies have indicated that sex education and discussions of sexuality successfully increase the knowledge of the individual with ID regarding sexual activity, contraception, reproduction, and personal hygiene (Ballan, 2001; Brown & Pirtle, 2008). Findings from a longitudinal study by McCaffree and Matlack (2001) found that participants believed their comprehensive sexuality education course had a long-term positive impact on their lives in the areas of self-exploration, self-efficacy related to sexual health, and perceived knowledge and comfort when dealing with sexual issues. Similarly, Schaafsma, Kok, and Stoffelen, (2015) found that sexual knowledge and attitudes of individuals with ID were improved through sex education. Furthermore, studies have shown that sex education can positively influence important determinants, such as social or behavioral skills (Miltenberger et al. 1999; Egemo-Helm et al. 2007; Hayashi et al. 2011) and decision-making capabilities (Duke & McGuire, 2009; Khemka et al. 2005; Schwartz & Robertson, 2018).

Like individuals without disabilities, individuals with disabilities need to feel as if they have the right to openly discuss topics that are important to them in their lives or situations that

they do not understand in order to acquire the appropriate skills to effectively navigate through society (Azzopardi-Lane & Callus, 2015). Research shows that knowledge is power with regard to reducing risks of sexual abuse. The ability to prevent and/or report sexual abuse is contingent upon an individual's skills to recognize, name, and understand body parts (Kenny & Wurtele, 2008), know and understand consent (Landford, 2016), and communicate with a caring adult regarding perceived or real danger (Future of Sex Education, 2018). In fact, convicted child sex offenders reported in one study that they were better able to take advantage of children with inadequate information about sex, including inadequate understanding of body parts (Elliot, Browne, & Kilcoyne, 1995). While there is limited research directly linking sex education to prevention of sexual abuse, a review of twenty-four studies reported that child's self-protection skills and knowledge can be increased by participation in sexuality education programs (Walsh, Zwi, Woolfenden, & Shlonsky, 2015). Likewise, Borges, Banyard, and Moynihan (2008) determined that even very brief educational programs can produce knowledge of sexuality and understanding of consent.

There is a continued need to explore and disseminate knowledge in the field of sexuality and disability. Societal attitudes and perceptions are driven by education and knowledge. If there is no exposure to sexuality and disability, it follows suit that society would have a narrow understanding of these issues (Esmail et al., 2010), which inherently perpetuates marginalization, stigmas, and barriers to attaining sexual health for individuals with disabilities.

CHARGE Syndrome

CHARGE syndrome is a genetic condition where multiple anomalies are present at birth. The name of this condition is an acronym derived from common abnormalities used to characterize this syndrome: Coloboma of the eyes, Heart defects, Atresia of the choanae,

Retardation of growth, Genital hypoplasia, Ear anomalies and/or deafness (Pagon et al., 1981). CHARGE syndrome is a rare condition affecting 1 in 10,000 births. There are no known demographic disparities, as it affects both males and females equally and is found in various races and ethnicities across the world (National Organization for Rare Disorder, 2019). Currently, there are two methods of diagnosis for CHARGE syndrome: a criterion checklist (Blake et al., 2005) and genetic testing for a mutation of the CHD7 gene. Diagnostic criterion involves a host of physical disabilities including facial nerve palsy, short stature, ear abnormalities, genital underdevelopment, mobility issues, and the most common, coloboma of the eye/vision impairments and hearing impairments (Blake et al., 1998). As a result of these physical and medical complications, many individuals with CHARGE undergo a dozen or more procedures and surgeries throughout their life that limit opportunities for social engagement, academics, and practicing of adaptive living skills.

Some of these anomalies (e.g., genital underdevelopment, growth retardation) can be attribute to an irregular production of growth hormones and sex hormones (Kirk, 2011). Further, problems related to sex hormones complicate typical development particularly around puberty, as very few males enter puberty spontaneously. Delayed or defective production of sex hormones from the gonads (i.e., testicles in boys and ovaries in girls), are treated through hormone therapy in gradual dosages of estrogen for women and testosterone for men. In addition, genital abnormalities can include undescended testicles and small penis in males and smaller clitoris and labia minora in females (Kirk, 2011). Moreover, while little data exist on fertility rates within the population, both men and women with CHARGE syndrome have been known to naturally produce offspring (Kirk, 2011).

Further, CHARGE syndrome is identified as the leading cause for deafblindness, as approximately 80%-90% of individuals meet qualifications for deafblindness even when hearing or vision impairments are mild (Hartshorne, Hefner, Davenport & Thelin, 2011; U.S. Department of Education, National Center on Deafblindness, 2017). This often results in individuals having difficulty with taking in information, as their disabilities make it difficult to absorb information from the environment and interact the world around them. Likewise, individuals with CHARGE syndrome exhibit a wide range of cognitive and adaptive functioning, with some individuals living independently and others requiring substantial daily care to support their needs (Salem-Hartshorne & Jacob, 2004).

All of these factors (i.e., hearing/vision impairments, medical complications, range of cognitive and adaptive functioning, physical abnormalities, etc.) contribute to the many barriers in sexuality this population faces. The extent to which sexuality of individuals with CHARGE syndrome are affected by these barriers; however, is not known. While the topic of sexuality has been extensively studied in similar populations, to date, there has been no exploration of sexuality within the CHARGE population.

The Current Study

While previous studies have sought out to explore perceptions of individuals with intellectual/developmental disabilities and physical disabilities distinctly, Fader Wilkenfeld and Ballan (2011) suggested that future research should investigate the knowledge, attitudes, and beliefs of sexuality and sexuality education for individuals with medically complex disabilities, as these individuals may possess unique attitudes and perceptions due to the intricacy of their presentation. In addition, Sellwood, Raghavendra, Jewell (2017) reported a lack of research regarding the sexuality of individuals with congenital physical and communication disabilities

(e.g., deaf, blind, deafblind), and the need for further exploration concerning aspects intimacy within this population.

Developing a better understanding of sexuality needs for this population by exploring attitudes of sexuality is particularly important, as individuals with deafblindness are considered extremely vulnerable (Hutton, 2000; Simcock, 2016). Moss and Blaha (2001) indicate significant contributing factors to their vulnerability are the inability to disclose abuse due to communication challenges and access to limited information regarding sexuality and sexual abuse. For congenital deafblind individuals touch is considered a learning tool (Moss & Blaha, 2001); individuals learn that it is acceptable to be touched (Kiekopf, 2007). Lack of information and dependence on others could have individuals tolerating abusive behaviors or perceiving them as sensory experiences (Moss & Blaha, 2001; Kiekopf, 2007). Thus, identifying and rectifying distorted knowledge and information gaps regarding sexual health is critical to enhancing safety and improving quality of life for individuals who are deafblind (Todd, 2012).

One of the most significant obstacles these individuals encounter to their sexual health is the inability to gather information that sighted, hearing peers learn incidentally (Davis, 1971; Getch, 2001; Miller, 1999; Neff, 1978; Todd, 2012). While their peers are naturally exposed to information about sexuality, these learning opportunities are often inaccessible to those who are deafblind; this may leave them unaware of even the most basic concepts, such as their own gender (Davis, 1971; Miller, 1999; Neff, 1978; Todd, 2012). Sexuality education can be instrumental to further inclusion in society, as inappropriate sexual behaviors are judged harshly, regardless of a person's disability status (Moss & Blaha, 2001). As sexuality education explicitly instructs individuals on how to initiate and maintain friendships and other important relationships, it could, conceivably, have an impact on their feelings of vulnerability by

providing them with increased social contact (Todd, 2012). In short, teaching sexuality to the deafblind population, regardless of level of functioning, is tantamount to acknowledging that these individuals have social/sexual rights and responsibilities (Miller, 1999).

One study to date has explored parental and staff attitudes towards instruction in human sexuality for individuals with sensory impairments (i.e., deaf, blind, deafblind; Love, 1983). This study found that both parents and staff strongly recognize the need for education of sexuality for this population (Love, 1983). In order to minimize the risk of child sexual abuse, Belote (2012) with the California Deafblind Service suggests that instruction of issues of sexuality should commence at an early age, including respect for privacy, anatomy, and terminology (i.e., both medical and slang terms), and these skills should be assessed for generality. While individuals with deafblindness report feeling vulnerable in various situations, they do not appear to describe themselves as being in a constant state of vulnerability (Simcock, 2016). They desire intimacy and social relationships (Stratton, 2011). Unfortunately, the literature largely looks at the negative outcomes associated with the vulnerability of this population, and there is limited exploration of positive aspects of sexuality including intimacy or the experience of individuals who are deafblind (Simcock, 2016). Thus, the current study extends the literature by exploring the attitudes of sexuality of individuals with CHARGE syndrome, a multifaceted genetic condition and the leading cause of deafblindness, where individuals often present with comorbid physical and developmental or intellectual disabilities, most often accompanied by deafblindness.

In addition to addressing sexuality in individuals with both cognitive and physical disabilities, Brown and Pirtle (2008) indicated that future research should investigate beliefs held by the individual with a disability and how they view their own sexual development and sex

education, as well as examining if those beliefs differ from their parents or caregivers. With this in mind, the purpose of the current study was to examine various aspects of sexuality in CHARGE syndrome by exploring potential barriers that may exist due to differences in the attitudes towards sexuality in parents of individuals with CHARGE syndrome and those attitudes of adults with CHARGE syndrome. No previous study has investigated sexual health, sexual education, or sexuality in CHARGE syndrome.

Furthermore, the current study aimed to identify concerns and discrepancies regarding perceptions of themes surrounding a comprehensive sexuality education for individuals with CHARGE syndrome from both adults involved with individuals with CHARGE syndrome and individuals with CHARGE syndrome themselves. Additionally, the information obtained from individuals with CHARGE syndrome receiving a sexuality education was reviewed along with perceptions of sexuality from various points of view to identify how sexuality education should be delivered to this population. This study was an initial step to understand barriers preventing the delivery of sexuality education to this underserved population, including identifying and rectifying distorted knowledge about sexuality. This study also provides a foundation for dissemination of sexual health information for both individuals with CHARGE syndrome and parents of individuals with CHARGE syndrome by determining areas of discrepancy. Attitudes on sexuality are important to consider since they can provide valuable clues in the conceptualization of educational programs in order to have an effect on achieving a better quality of life for individuals with disabilities (Tamas et al., 2019).

This study examined the following research questions:

Research Question #1: Are there differences in attitudes of sexuality between caregivers of individuals with CHARGE syndrome and individuals with CHARGE syndrome?

Research Question #2: Does age of individuals with CHARGE syndrome predict attitudes of sexuality for individuals with CHARGE syndrome?

Research Question #3: Does age of parents predict attitudes towards sexuality for individuals with CHARGE?

Research Question #4: Are there differences between individuals with CHARGE syndrome who have had exposure to sex education or not with regard to attitudes towards sexuality?

CHAPTER III

METHODOLOGY

Sampling

Due to the exploratory nature of this study within a low incidence population, minimum sample size for the current study was determined in comparison to sample sizes of previously reported studies in the literature within the CHARGE syndrome population. Similar to Hudson, Macdonald, and Blake (2015), Wulffaert and colleagues (2009), and Haibach and Lieberman (2013), a minimum of 40 participants (i.e., 20 parents of individuals with CHARGE and 20 adults with CHARGE syndrome) 18 years old and above were recruited to participate in the procedures.

Participants and Setting

Participants were recruited from the international CHARGE Syndrome Foundation research web page, as well as various CHARGE syndrome social media outlets (i.e., Facebook CHARGE syndrome page, CHARGE Syndrome twitter account), and email listservs. Following IRB approval (Appendix E), direct links to the survey were posted on various social media platforms for parents or legal guardians of individuals with CHARGE syndrome, and adults with CHARGE syndrome (i.e., ages 18 and above) that were interested in participating in the study. Further, for individuals that indicated they were interested in participating the study at the International CHARGE Syndrome Conference, they were sent a direct link to the survey via email. All participants had either a previous diagnosis of CHARGE syndrome, genetically or

clinically diagnosed, or be the parent of an individual with CHARGE syndrome. This study did not require parents of individuals with CHARGE syndrome and adults with CHARGE syndrome to be directly linked.

In all, 67 parents of individuals with CHARGE syndrome, and 52 adults with CHARGE syndrome were initially recruited. However, only 31 parents of individuals with CHARGE syndrome, and 24 adults with CHARGE syndrome completed the entirety of the survey and were therefore included in the study. Demographic information regarding parent participants and adults with CHARGE syndrome participants is included in Table 1.

Table 1

Demographic Information of Participants (N = 55)

Characteristics	Parent of individual with CHARGE syndrome sample (n = 31)	Adult with CHARGE syndrome sample (n = 24)
Sex		
Female	25	15
Male	5	9
Nationality		
Australia	0	1
Canada	0	5
Germany	1	0
India	1	0
New Zealand	1	1
United Kingdom	1	1
United States of America	27	16

Table 1 (continued)

Characteristics	Parent of individual with CHARGE syndrome sample (n = 31)	Adult with CHARGE syndrome sample (n = 24)
Religion		
Christianity	19	12
Buddhism	1	0
Hinduism	1	0
Islam	0	1
Judaism	1	1
Nonreligious	9	4
Neo-Paganism	0	1
Primal-indigenous	0	1
Unitarian-Universalism	0	2
Age	M = 47.27 (31-66)	M = 27 (18-40)
Marital Status		
Married	25	0
Divorced	6	0
Single/Never married	0	24

Table 1 (continued)

Characteristics	Parent of individual with CHARGE syndrome sample (n = 31)	Adult with CHARGE syndrome sample (n = 24)
High school graduate/equivalent	7	7
Some college but no degree	3	6
Associate degree	3	3
Bachelor's degree	8	6
Master's degree	5	1
Doctoral degree	2	0
Professional degree	3	0
Do you have children?		
Yes	31	0
No	0	24
How many children do you have?	M = 2.52 (1-7)	N/A
How old is your child with CHARGE syndrome?	M = 19.69 (4 months - 36 years)	N/A

Table 1 (continued)

Characteristics	Parent of individual with CHARGE syndrome sample (n = 31)	Adult with CHARGE syndrome sample (n = 24)
Gender of child with CHARGE syndrome		
Male	14	N/A
Female	17	N/A
Did you require the use of an intervener to complete this survey?		
Yes	N/A	1
No	N/A	23

Instruments

Demographic Questionnaire

In order to gain a better understanding of participants with CHARGE syndrome and participants who are parents or legal guardians of a child with CHARGE syndrome, demographic questionnaires were created. These questionnaires included: the *Individual with CHARGE Syndrome Demographic Questionnaire,* and the *Parent Demographic Questionnaire*. Due to complexity of CHARGE syndrome, demographic information, as well as the major and minor phenotypic features of CHARGE syndrome were collected. Demographic information from the *Individual with CHARGE Syndrome Demographic Questionnaire* included the following: (a) person completing the questionnaire (i.e., parent of individual with CHARGE syndrome or adult

with CHARGE syndrome), (b) date of birth, (c) age, (d) race, (e) gender, (f) country of origin and region, (g) marital status (h) number of children, (i) level of education, (j) age of CHARGE syndrome diagnosis, (k) major/minor characteristics of CHARGE syndrome, (l) additional clinical diagnosis, (m) special education services, (n) living situation (e.g., if they have a caregiver or live alone), and (o) if they received sexuality education. Demographic information from the *Parent Demographic Questionnaire* included the following: (a) identifying information on person completing the questionnaire (i.e., date of birth, age, race, gender, country of origin and region, marital status, number of children, and level of education), (b) identifying information of their child with CHARGE Syndrome (i.e., same as listed above in the Individual with CHARGE Syndrome Demographic Questionnaire, and (c) if their child has received sexuality education to their knowledge.

Attitudes towards Sexuality Questionnaire - Intellectual Disabilities (ASQ-ID)

The initial ASQ (Cuskelly and Bryde, 2004) was developed to compare the attitudes of parents of an adult with an ID, support staff working with an adult with an ID, and a community sample. It contained 33 items grouped into eight subthemes (i.e., subscales) including: sex education, masturbation, relationships, sexual feelings, sexual intercourse, sterilization, parenthood, and marriage. While this questionnaire was found to have suitable test–retest reliability of $r = 0.91$, as well adequate internal consistency substantiated by a Cronbach's alpha score of 0.90, researchers suggested the need for further development of this scale by examining a factor structure (Cuskelly & Bryde, 2004). Thus, the revised ASQ-ID (Cuskelly & Gilmore, 2007) was developed providing a factor structure for the items on the scale and gender specified questions, as recommended by Cuskelly and Bryde (2004).

The ASQ-ID was used in the current study to measure the attitudes of parents of individuals with CHARGE syndrome, as well as attitudes of sexuality in adults with CHARGE syndrome due to its ability to measure attitudes towards various aspects of sexuality regarding adults with an intellectual disability. This questionnaire has a total of 28 items (i.e., specific to one gender; 56 total items for combined genders), rated on a 6-point Likert scale weighted by ratings of *strongly agree* to *strongly disagree.* Higher total scores are associated with more liberal or positive attitudes towards the sexuality of adults with IDs, with total scores ranging from 28 (lowest possible score; 56 for combined gendered items) – 168 (highest possible score; 336 for combined gendered items; Cuskelly & Gilmore, 2007). A factor analysis conducted by Cuskelly and Gilmore (2007) grouped items into four factors producing the subscales: "sexual rights," "parenting," "non-reproductive sexual behavior," and "self-control." Each subscale produced excellent to adequate internal consistency, as determined by using Cronbach's alpha. The "sexual rights" (alpha = .93) subscale contains 13 items with a subscale score ranging from 13-78. The "parenting" subscale (alpha = .88) contains 7 items with a subscale score ranging from 7-42, and the "non-reproductive sexual behavior" subscale (alpha = .84) consist of 5 items with a subscale score that ranges from 5-30. Finally, the "self-control" subscale (alpha = .67) includes 3 items and produces a score between 3-18. A correlational analysis revealed that all four subscales significantly correlated at $p < .001$ (Pearson's r range from .43 to .66). For the purposes of this study, items from this measure was slightly modified to target the specific population of this study. Modifications to items included replacing "individuals with intellectual disabilities" to "individuals with CHARGE syndrome." While modifications could innately affect the reliability of this measurement, this instrument was selected due to the wide range of cognitive functioning, including ID, that presents in individuals with CHARGE syndrome (Smith

& Blake, 2010). Further, there is empirical support of the utility of this instrument in exploratory studies investigating the attitudes of sexuality using similar sample sizes (Winarni, Hardian, Suharta, & Ediati, 2018). This instrument was combined within the respective demographic questionnaire and made available on the internet through Qualtrics.com.

Procedural Overview

After IRB approval (see Appendix E), a recruitment letter containing the link to the survey was posted on the CHARGE Syndrome Foundation research web page, as well as various CHARGE syndrome social media outlets and sent through listserv e-mails to individuals who have previously indicated an interest in participating in CHARGE sexuality research. Through the recruitment letter, participants were provided with information regarding their implied consent and confidentiality along with the link to the Qualtrics survey. Recruitment flyers were also presented at the 2019 International CHARGE Syndrome Conference. Data were collected using the Qualtrics survey system. Participants were able to take the survey at any location they wish; however, due to the sensitive nature of this topic, a cautionary statement was provided prior to the survey encouraging participants to complete the survey in a private location. After obtaining implied consent, as indicated by continuing to partake in the survey, participants were asked to complete a demographic form (embedded in the questionnaire) and various questions regarding their perceptions of themes regarding sexuality through the ASQ-ID.

Data Analysis

Data was analyzed based on the previously determined research questions. Analyses were based upon those conducted in earlier investigations of this instrument on individuals with intellectual disabilities (Cuskelly & Bryde, 2004; Cuskelly & Gilmore, 2007; Tamas et al., 2019; Winarni et al., 2018). Individual subscale scores and a total score of the ASQ-ID were calculated from all subscales, and all data was entered into SPSS and double coded. Any discrepancies were visually analyzed until 100% agreement was reached. Total ASQ-ID scores and subscale scores were calculated for all participant responses to female questions, male questions, and both female and male scores combined (see Table 1).

Further, to ensure psychometric quality of adapted measures, reliability was assessed through Cronbach's alpha and correlational analysis. Similar to Cuskelly and Gilmore (2007), each subscale produced excellent to adequate internal consistency, as determined by using Cronbach's alpha: "sexual rights" subscale, alpha=.88; "parenting" subscale alpha=.93; "non-reproductive sexual behavior" subscale, alpha=.90; and "self-control" subscale, alpha=.79. A correlational analysis revealed that all four gender combined subscales significantly correlated (Pearson's *r*s range from .30 to .62, *p*s < .05). While there were inherent limitations in the analysis and interpretation of quantitative data obtained from a small-scale exploratory and attitudinal research study, these are expected as this study aims to target critical issues of a sensitive topic in a low incidence disability population.

Comparison of Means

The first question for analysis was to determine if there are differences in total attitude scores between adults with CHARGE syndrome and parents of a child with CHARGE syndrome. An independent samples *t*-test was conducted to determine if there are differences between the two groups' (i.e., independent variable: individuals with CHARGE syndrome, parents of an individual with CHARGE syndrome) total attitude score (i.e., dependent variable). Assumptions for independent samples *t*-test analysis were checked and include the following: presence of outliers by using a visual analysis of box plots, normal distributions of data by using the Sharpiro-Wilk test for normality, and homogeneity of variance by using Levene's test for equality of variances.

Comparison of Group Differences

Further analyses of research Question 1 was conducted with regard to differences among subscale scores of the instrument. A one-way multivariate analysis of variance (MANOVA) was conducted to determine if a difference exists on attitudes of sexuality (i.e., subscales as dependent variables) due to having exposure to sexuality education (i.e., independent variable). Assumptions for independent samples t-test analyses were checked and include the following: presence of outliers by using a visual analysis of box plots, normal distributions of data by using the Sharpiro-Wilk test for normality, and homogeneity of variance by using Levene's test for equality of variances. Assumptions for a one-way MANOVA analyses were tested and include the following: testing for univariate and multivariate outliers using boxplots and Mahalanobis distance, normality using the Shapiro-Wilk test of normality, multicollinearity using Pearson correlation coefficients, linearity using scatterplot matrices, sample size adequacy, and equality

of variance-covariance matrices and homogeneity of variances using Box's test of equality of variance-covariance matrices.

Analysis of Predictive Descriptors

The second (i.e., age of individual with CHARGE) and third research questions (i.e., age of parent) were analyzed to determine if age of an individual better predicts attitudes of sexuality. A simple linear regression was used to examine Question 2 in addition to Question 3, as the dependent variable (i.e., total attitudes score) can be predicted by the independent variable (i.e., age of in an individual with CHARGE syndrome [research question 2] and age of parent of an individual with CHARGE syndrome [research question 3]). Assumptions for simple linear regression analyses were checked and include the following: a linear relationship between the dependent and independent variables, independence of observations, no significant multivariate outliers, homoscedasticity, and normal distribution of residuals.

CHAPTER IV

RESULTS

In the present study, four primary questions were asked: 1) Are there differences in attitudes of sexuality between caregivers of individuals with CHARGE syndrome and individuals with CHARGE syndrome? 2) Does age of individuals with CHARGE syndrome predict attitudes of sexuality for individuals with CHARGE syndrome? 3) Does age of parents predict attitudes towards sexuality for individuals with CHARGE? 4) Are there differences between individuals with CHARGE syndrome who have had exposure to sex education or not with regard to attitudes towards sexuality? To answer these questions independent sample t-test, MANOVA, and linear regressions were conducted.

Preliminary Analysis

First, to determine if female and male questions should be analyzed separately as gender specific items or analyzed as combined totals for all participants, a correlational analysis was conducted. The correlational analysis revealed that each gender specific subscale (e.g., male parenting subscale items compared to female parenting subscale items) significantly and substantially correlated at $p < .05$ for all four subscales: "sexual rights" $p = .90$, "parenting" $p = .90$, "nonreproductive sexual rights" $p = .84$, and "self-control" $p = .71$. Thus, combined totals for each of the four subscales, opposed to gender specific subscales, were used for the following analyses.

Research Question 1: Independent Sample T-Test and MANOVA

To answer the question if there were differences in attitudes of sexuality between caregivers of individuals with CHARGE syndrome and individuals with CHARGE syndrome, an independent samples *t*-test was initially conducted to compare overall attitude of sexuality scores. A one-way MANOVA was then used as a follow up analysis to take a closer look at differences within the four subscales of attitudes of sexuality.

Independent Samples T-test

To compare overall differences in attitudes of sexuality between caregivers of individuals with CHARGE syndrome and individuals with CHARGE syndrome an independent samples t-test was conducted. When checking assumptions, one outlier was identified through visual inspection of a boxplot and removed, and attitudes of sexuality were normally distributed for both groups, as assessed by Shapiro-Wilk's test ($p > .05$). The assumption of homogeneity of variances was violated, as assessed by Levene's test for equality of variances ($p = .031$), thus a Welch *t*-test was used to determine if there were differences in attitudes of sexuality between parents of individuals with CHARGE syndrome and adults with CHARGE syndrome. Although parents of individuals with CHARGE syndrome's mean attitudes of sexuality score was slightly lower ($M = 281.40$, $SD = 33.29$) than adults with CHARGE syndrome's mean attitudes of sexuality score ($M = 290.91$, $SD = 20.07$), it was not found to be a statistically significant difference, $M = -9.51$, 95%, CI [-24.25, 5.21], $t(48.68) = -1.298$, $p = .200$. Both parents and adults with CHARGE syndrome were found to have relatively positive attitudes of sexuality (range 56-336).

MANOVA

To further analyze group differences among attitudes of sexuality subscales (i.e., parenting, nonreproductive sexual behavior, sexual rights) and between caregivers of individuals with CHARGE syndrome and adults with CHARGE syndrome, a one-way multivariate analysis of variance was run. In the analysis, the independent variable was participant with Level 1 = parents of individuals with CHARGE syndrome and Level 2 = adults with CHARGE syndrome, while dependent variables included total combined gender scores on the four subscales: (a) parenting, (b) self-control, (c) nonreproductive sexual behavior, and (d) sexual rights. Preliminary assumption checking revealed eight univariate outliers, which were identified through visual inspection of box plots and were removed, leaving a total of 27 parents of individuals with CHARGE syndrome and 20 adults with CHARGE syndrome to be included in the analysis. When looking at the assumption of normality, skewness and kurtosis all scores fell into an acceptable of range (-1-1). There was no perfect multicollinearity, as assessed by Pearson correlation (Table 2) and no multivariate outliers in the data, as assessed by Mahalanobis distance. Homogeneity of variance-covariances matrices was found, as assessed by Box's test of equality of covariance matrices (p = .237). The correlations of the dependent variables of sexual rights, parenting, non-reproductive sexual rights, and self-control can be found in Table 2.

Table 2

Pearson Correlation of Dependent Variables

	Sexual Rights	Parenting	Nonreproductive sexual rights	Self-control
Sexual Rights	1.00	.621	.554	.560
Parenting	.621	1.00	.303	.296
Nonreproductive sexual rights	.554	.303	1.00	.323
Self-control	.560	.296	.323	1.00

The results of the MANOVA showed there was statistically significant difference between the two groups of participants on the four ASQ-ID subscales, $F(4, 42) = 10.203$, $p <$ 001; Wilks' $\Lambda = .507$; partial $\eta^2 = .493$. The univariate results showed there was no statistically significant difference based on participant group on the subscale scores of "sexual rights" ($p =$ .909), and "nonreproductive sexual behavior" ($p = .423$), compared to parents of individuals with CHARGE syndrome or adults with CHARGE syndrome. However, a statistically significant difference was found in regard to "parenting" and "self-control." Specifically, in regard to "parenting" $F(1,45) = 8.215$, p = .006, and an effect size of $\eta^2 = .154$, and "self-control" $F(1, 45) = 8.196$, $p = .006$, and an effect size of $\eta^2 = .154$, using a Bonferroni adjusted α level of .0125. The effect sizes of both "parenting" and "self-control" are large, suggesting a great difference between attitudes of sexuality with parents and individuals themselves with CHARGE syndrome in regard to parenting and self-control. Specifically, adults with CHARGE syndrome reported more positive views ($M = 79.35$, $SD = 4.42$) for attitudes of parenting for individuals with

CHARGE syndrome, than did parents of individuals with CHARGE syndrome (M = 72.48, SD = 10). Moreover, with regard to attitudes of self-control for individuals with CHARGE syndrome, parents were found to hold slightly more positive views (M = 29.17, SD = 3.81) than adults with CHARGE syndrome (M = 27.45, SD = 3.37). These significant differences should be noted when addressing difference attitudes of sexuality in CHARGE syndrome with parents and individuals themselves with this condition.

Research Question 2: Linear Regression

To determine the effect of age on attitudes of sexuality for individuals with CHARGE syndrome a linear regression was conducted. The independent variable for this analysis was age and the dependent variable was attitudes of sexuality total score. When checking assumptions, to assess linearity a scatterplot of individuals with CHARGE syndrome's ASQ-ID scores against their age with superimposed regression line was plotted. Visual inspection of these two plots indicated a linear relationship between the variables. There was homoscedasticity and normality of the residuals, and no outliers were identified. Age was not found to significantly predict attitudes of sexuality for individuals with CHARGE syndrome, $F(1, 18) = 1.423$, $p = .248$, $R^2 = .073$, adjusted $R^2 = .022$. Age was found to only account for 7% of the variance in attitudes of sexuality for individuals with CHARGE syndrome.

Research Question 3: Linear Regression

Further, to determine if age was a predictor of attitudes of sexuality for parents of individuals with CHARGE syndrome an additional linear regression was conducted. In this analysis, the explanatory variable was age and the outcome variable was the attitude of sexuality score. To assess linearity a scatterplot of parents of individuals with CHARGE syndrome's

ASQ-ID scores against their age with superimposed regression line was plotted. Visual inspection of these two plots indicated a linear relationship between the variables. There was homoscedasticity and normality of the residuals, and no outliers were identified. Age was not found to significantly predict attitudes of sexuality for individuals with CHARGE syndrome, $F(1, 28) = .18$, $p = .674$ $R^2 = .006$, adjusted $R^2 = -.029$. Age was found to account for less than 1% of the variance in attitudes of sexuality for parents of individuals with CHARGE syndrome.

Research Question 4

Finally, the researchers hoped to examine differences between individuals with CHARGE syndrome who have had exposure to sex education to those who had not with regard to attitudes towards sexuality. Due to the lack of homogeneity of variance (i.e., 95% [N = 23] of the sample reported having received some form of sexuality education), this question was not able to be analyzed. However, when taking a closer look at who provided individuals with CHARGE syndrome sexuality education, 58% reported receiving sexuality education from their parents (n = 14), 95% reported receiving sexuality education from an educator (e.g., teacher, coach, principal, school nurse; n = 23), 12% reported receiving sexuality education from a doctor (n = 3), and 29% reported receiving information from a friend (n = 7).

Summary

In summary, there was not a statically significant difference between parent's total attitude of sexuality scores and individuals with CHARGE syndrome's total attitude of sexuality scores, suggesting that parents of individuals with CHARGE syndrome and adults with CHARGE syndrome hold similar views of sexuality. However, when taking a closer look at the four attitudes of sexuality subscales (i.e., "sexual rights," "parenting," "non-reproductive sexual

behavior," and "self-control"), a statistically significant difference was found between parents and individuals with CHARGE syndrome in regard to the parenting and self-control subscales. This suggests that while, overall, parents and adults with this condition share similar and positive attitudes of sexuality for this population, there are noted differences with regard to specific areas of sexuality. Specifically, adults with CHARGE syndrome appeared to have more positive views of parenting in CHARGE syndrome than parents of individuals with CHARGE syndrome. Simply stated, individuals with CHARGE express positive views and abilities to parent children; while parents of individuals with CHARGE express less open views to their child's ability to parent. On the other hand, parents seemed to have slightly more positive views of self-control, than did individuals with CHARGE, themselves. This suggests that parents believe their child can engage in appropriate sexual self-control; while individuals with CHARGE themselves view less self-control for sexual urges. Age, however, was not found to be a predictor of attitudes of sexuality for parents of individuals with CHARGE syndrome nor adults with CHARGE syndrome. Further, most individuals with CHARGE syndrome who participated in the study reported having received some level of sexuality education, thus exploring the impact of receiving sexuality education on attitudes of sexuality was not able to be conducted.

CHAPTER V

DISCUSSION

The purpose of the following study was to examine attitudes of sexuality of individuals with CHARGE syndrome by exploring potential barriers that may exist due to differences in the attitudes towards sexuality in parents of individuals with CHARGE syndrome and those attitudes of adults with CHARGE syndrome. Examining these perceptions or attitudes of sexuality provides insight to overcome these barriers, such as knowledge gaps, misperceptions, and desired resources within a specific population. As the topic of sexuality continues to be taboo for many cultures, much of the sexuality and disability literature is in its infancy. Current literature in this area has focused largely on individuals with intellectual disabilities or physical disabilities (Addlakha, 2007; Calam, 2012; East & Orchard, 2014; Medina-Rico et al., 2017; Suter et al., 2013), and has in turn, generally failed to explore attitudes of sexuality in populations with medically complex or multiple disabilities (Fader Wilkenfeld & Ballan, 2011) and congenital physical and communication disabilities (e.g., deaf, blind, deafblind; Sellwood, Raghavendra, & Jewell, 2017). Signature characteristics of CHARGE syndrome, a multifaceted genetic condition, include a range of cognitive, social, and physical disabilities, which are often medically complex in nature. The current study served as an exploratory investigation, as it is the first to investigate the topics of sexual health, sexual education, and sexuality in CHARGE syndrome.

The findings of the current study examined a number of areas specifically related to attitudes of sexuality in CHARGE syndrome from parents of individuals with CHARGE syndrome and adults with CHARGE syndrome. The first research question sought out to determine if there were differences in attitudes of sexuality between caregivers of individuals with CHARGE syndrome and individuals with CHARGE syndrome. The researcher hypothesized that individuals with CHARGE were likely to have higher attitudes of sexuality than caregivers. In order to evaluate the data, an independent samples *t*-test was conducted to determine if there were differences between the two groups total attitude score on the ASQ-ID. Then, to further explore differences across themes of sexuality captured by the ASQ-ID (i.e., sexual rights, nonreproductive sexual behavior, self-control, and parenting), a MANOVA was conducted. The second and third research questions aimed to examine if age could better predict attitudes of sexuality for adults with CHARGE syndrome (i.e., research Question 2) and parents (i.e., research Question 3). While the literature provides mixed findings with regard to age as predictor of attitudes of sexuality, the researcher hypothesized that the older the parent or caregiver was, the more conservative they are likely to be (Cuskelly & Bryde, 2004; Murray & Minnes, 1994). Conversely, the researcher hypothesized that the older an individual with CHARGE syndrome is, the more positive their attitudes of sexuality would be. To evaluate these questions, two separate standard linear regressions were conducted. The final research question sought out to examine differences between individuals with CHARGE syndrome who have had exposure to sex education or not with regard to attitudes towards sexuality. The researchers hypothesized that individuals with CHARGE who report having had some exposure to sexuality education will have more positive attitudes of sexuality (Ballan, 2012; Barnar-Brak et al., 2014; Medina-Rico, M., Lopez-Ramos, & Quinonez, 2017). Due to the lack of

homogeneity of variance (i.e., 95% [N=23] of the sample reported having received some form of sexuality education), this question was not able to be analyzed.

While, overall, the results did not support the present study's hypotheses, some important findings were discovered in this investigation. Particular to Hypothesis 1, while individuals with CHARGE syndromes' total attitude of sexuality mean score was slightly higher than parents' mean score, the differences were not found to be statistically significant. This suggests that parents of individuals with CHARGE syndrome and adults with CHARGE syndrome hold similar positive, global views of sexuality. However, when looking closer at individual themes of sexuality (i.e., sexual rights, parenting, non-reproductive sexual behavior, and self-control), some statistically significant differences were found between parents and individuals with CHARGE syndrome.

The scores obtained by parents differed significantly on the subscales of *parenting* and *self-control* from the scores obtained by adults with CHARGE syndrome. Specifically, adults with CHARGE syndrome appeared to have more positive views of parenting in CHARGE syndrome than parents of individuals with CHARGE syndrome. The parenting subscale measures attitudes towards individuals with CHARGE syndrome becoming parents and their ability to rear children. The current study found that parents of individuals with CHARGE syndrome were less confident that individuals with CHARGE syndrome have the abilities to care for a child, while adults with CHARGE syndrome felt more positively about their abilities to raise children and their rights to parenthood. Using this particular scale, several studies reported similar findings pertaining to more conservative views of parenting for individuals with intellectual disabilities. For example, parents of individuals with intellectual disabilities scored lowest in the area of parenting in the following studies: Cuskelly and Bryde (2004), Cuskelly and

Gilmore (2007); Tamas et al., (2019). Similarly, direct care staff, who are analogous to parents in residential facilities, have also been found to be less supportive of parenting in individuals with intellectual disabilities (Cuskelly & Bryde, 2004; Gilmore & Chambers, 2010) in comparison to the other themes of sexuality. These findings could be due to a lack of independence displayed by individuals with disabilities and concern for their capacity to provide the level of care and support offspring need. This area of parenting appears to be more complex than other themes of sexuality and has implications that extend beyond the rights of an individual.

Although attitudes towards individuals with disabilities parenting are often found less positive and more intricate than other areas of sexuality, these perceptions can change through trainings and education. Meaney-Tavares and Gavidia-Payne (2012) and Pebdani (2016) found that following a sexuality training, care staff had significantly more positive views of parenting for individuals with intellectual disabilities. Providing resources and information on how to support parents with disabilities and their families in the community, lead to more positive attitudes of sexuality and better opportunities for these individuals. Further, it should be noted with regard to the current study, that while adults with CHARGE syndrome expressed positive views on parenting, none of the participants with CHARGE syndrome reported having any children. Though a characteristic of this condition involves abnormalities of the genitalia and reproductive system, both men and women with CHARGE syndrome have been known to naturally produce offspring (Kirk, 2011). There is much room for continued exploration in the area of parenting within this population from both parents and individuals with CHARGE syndrome.

Another statistically significant difference found between adults with CHARGE syndrome and parents of individuals with CHARGE syndrome among themes of sexuality, was found in the area of self-control. The self-control subscale was composed of questions related to sexual desires and feelings. Parents of individuals with CHARGE appeared to have more positive views of self-control of their child with CHARGE than adults with CHARGE syndrome reported. These findings are similar to those of Tamas and colleagues (2019) where parents of individuals with intellectual disabilities were found to obtain higher scores in the area of self-control when rating their child with an ID when compared to professionals. However, it is interesting that in the current study individuals with CHARGE syndrome held less-positive views in this area of sexuality than did those without the condition (i.e., parents). These findings suggest that parents of individuals with CHARGE syndrome felt that individuals with CHARGE did not have stronger sexual feelings than other individuals, did not need to use medication to inhibit their sexual drive, and were not more easily stimulated than others, while adults with CHARGE syndrome felt less control over these areas of sexuality. There are several factors that could contribute to these findings. First, given their birth anomalies which impact growth and sexual hormones, individuals with CHARGE syndrome could potentially be experiencing increased sex drives or unprovoked penile erections as a result of these treatments to initiate and sustain growth and puberty. With the increased drive, and associated communication and social deficits related to CHARGE syndrome, individuals may be more likely to think about or engage in inappropriate sexual behaviors. When thinking of automatically maintained behaviors (i.e., behaviors that are driven by internal stimuli), individuals often rely on behavioral and/or medical treatment to address those concerns. If parents are more incline to think that their children are able to control these inappropriate sexual behaviors, they may be less likely to reach out to other

professionals regarding these issues. Moreover, less positive views of self-control in individuals with CHARGE syndrome could also be attributed to feelings associated with statements that have been made to them over time by parents or professionals regarding their inability or need to control sexual behaviors, opposed to personal experiences with self-control. These views could also be attributed to a lack of understanding from individuals with CHARGE that all individuals experience sexual thoughts and desires, even if that is not something that is commonly discussed. Continued exploration in the area of self-control in sexuality for individuals within this population is warranted, as well as education and resources for caregivers, as well as for individuals with CHARGE.

Likewise, Hypothesis 2 and Hypothesis 3 were also not supported by the findings of this current study, as age was not found to be a predictor of attitudes of sexuality for parents of individuals with CHARGE syndrome nor adults with CHARGE syndrome. While age has been noted as an important factor in affecting attitudes towards sexuality (Le Gall, Mullet, & Shafighi, 2002), results of this study supported findings from Winarni et al., (2018) and Tamas et al., (2019) where age did not appear to predict attitudes of sexuality. Cuskelly and Gimore (2007) found that individuals over the age of 60 present with less accepting attitudes of sexuality. It is plausible that our results did not find age to be a predictor, because two participants in this study were found to be over the age of 60.

Implications

There are many implications that can be taken from the results of the study. First, it was important to understand how parents of individuals with CHARGE syndrome feel about sexuality, as well as individuals with CHARGE syndrome themselves. Past research has examined the importance of exploring attitudes of sexuality, as it provides insight to overcome

barriers, such as knowledge gaps, misperceptions, and desired resources within a specific population. These stigmas leave individuals with disabilities susceptible to misinformation or increased vulnerability and extend to barriers that hinder resources and opportunities (Koller, 2000). The results suggest that attitudes of sexuality are generally positive for this population. Further, barriers such as gender biases and lack of sexuality education did not seem to be present in this population, as attitudes for males and females with CHARGE syndrome correlated and most individuals with CHARGE syndrome that participated in this study reported receiving some level of sexual education. Perhaps due to this education, our sample of participants felt more comfortable to participate in this study, which may impact the ability to generalize the results to individuals not exposed to sexuality education. However, findings do suggest there is still room for growth and education on specific domains of sexuality. Therefore, continued research of sexuality is still warranted in this population as many questions are left unanswered. Nevertheless, this study serves as the first known attempt to explore areas sexuality in CHARGE syndrome and laid a foundation for initial understanding of areas of growth within in this subject.

These findings can also serve as a tool for professionals to provide better sexual health education, resources, and services to individuals with CHARGE syndrome and their families. We now understand that individuals with CHARGE syndrome feel positively about their abilities to raise children, while their caregivers strongly disagree. Professionals should feel equipped to provide education on family planning and alternatives to family planning for individuals with CHARGE syndrome, as well as community resources that provide assistance for parents with disabilities. This also suggests a need for additional research regarding specific concerns parents of individuals with CHARGE have related to their child engaging in child rearing. Further, we learned that individuals with CHARGE syndrome do not feel very strongly that they have self-

control over their sexual urges, while parents feel that individuals with CHARGE syndrome do not experience sexual urges at a heighten rate and are able to exhibit self-control. Given this finding, professionals should provide better education on biological norms of sexual urges to individuals with CHARGE syndrome for those with and without hormone treatment, as well as information on when one could be experiencing something that is atypical and should require a consult with a medical professional. This prompts the need for all individuals with CHARGE to participate in sexuality education as part of the general education curriculum and adapted to meet their needs. Additionally, such individualized education could be incorporated into their adaptive or health behaviors goals within special education plans.

Limitations

As a small-scale exploratory and attitudinal research study, there were inherent limitations in the analysis and interpretation of quantitative data obtained. This was expected as this study aimed to target critical issues of a sensitive topic in a low incidence disability population. Thus, one of the most predominant limitations of the study was the smaller sample size. Further, the sample largely consisted of female participants across both groups (parent participants= 81% female; individual with CHARGE participants= 63% female) and therefore may not generalize to fathers of individuals with CHARGE or males with CHARGE syndrome. Additionally, it is likely that participants who agreed to take part in this study investigating a sensitive topic felt more comfortable discussing sexuality, and perhaps, held more accepting attitudes. Relatedly, this sample may have had more exposure to sexuality education, prompting greater comfort with participation.

Furthermore, this study only examined attitudes of sexuality of individuals with CHARGE syndrome and did not include a comparison sample. This would have been helpful in

determining where participants attitudes of sexuality lie overall, compared to their attitudes of sexuality of individuals with CHARGE syndrome. In addition, this study only sought out to explore parent's views and adults with CHARGE syndrome's views of sexuality. Consideration should also be given to exploring views of other individuals involved with this population (e.g., medical professionals, educators, care staff), as their attitudes have the potential to influence opportunities and care for individuals with CHARGE syndrome.

Finally, attitude research innately produces limitations, particularly when measuring attitudes related to disability and sensitive topics, such as sexuality. Due to the tendency for participants to report politically or socially correct responses, the extent to which these attitudes translate to actual behaviors is unknown (Gilmore, 2010).

Future Research

Given the results of the study and the number of avenues still unexplored with regard to sexuality in CHARGE syndrome, there are a multitude of directions for future research in this area. While this study primarily focused on examining attitudes of sexuality in CHARGE syndrome, future research should consider including a comparison sample. This will allow researchers to determine how participants view attitudes of sexuality for individuals CHARGE syndrome related to their attitudes of sexuality of the general population. Having the opportunity to compare participant's perceptions of sexuality of CHARGE syndrome to the non-disabled population could provide a greater insight of marginalization and discriminations related to sexuality within this population.

Another avenue of future research could look further into the discrepancies of attitudes of sexuality between parents of individuals with CHARGE syndrome and adults with CHARGE syndrome. This study found that age was not a predictor of attitudes of sexuality for this

population, so identifying what factors (e.g., culture, severity of disabilities, education level, etc.) contribute to differences in attitudes of sexuality could aid addressing these discrepancies within this population. Specifically, determining what factors led to differences in attitudes of parenting and sexual control between the two groups could provide more insight for education and resources regarding these sensitive issues. Overall, this investigation found that parents and adults with CHARGE syndrome generally accepted that individuals with CHARGE syndrome are sexual beings and are largely comfortable with these individuals expressing their sexuality. It is possible that attitudes may differ when considering level of disability. Future research should examine if severity of complications related to CHARGE syndrome impact attitudes of sexuality.

Just as several studies have sought out to explore attitudes of sexuality in individuals with various disabilities (Addlakha, 2007; Calam, 2012; East & Orchard, 2014; Medina-Rico et al., 2017; Suter et al., 2013), the current study aimed to examine attitudes of sexuality in a novel population, CHARGE syndrome. This study served as an initial exploration of sexuality in CHARGE syndrome. While the target participants were parents of individuals with CHARGE syndrome and adults with CHARGE syndrome, results of this study suggested that gathering more information on attitudes of sexuality for individuals with CHARGE syndrome from other individuals involved with this population (e.g., medical professional, educators, paraprofessionals) is also warranted. This study found that outside of family, adults with CHARGE syndrome largely reported receiving sexuality education from medical professionals and educators. Thus, understanding attitudes of sexuality for individuals with CHARGE syndrome from other individuals involved with this population is necessary to examine potential biases and discrepancies imbedded in the sexuality education that is provided.

While the purpose of this study was to provide further insight to attitudes of sexuality in CHARGE syndrome, findings may have resulted in the formation of additional questions related to sexuality within this population. An area of interest related to examining attitudes and perspectives of sexuality is to inform treatment, education, and resources. Meaney-Tavares and Gavidia-Payne (2012) and Pebdani's (2016) findings support that sexuality education and trainings lead to more positive views of sexuality for individuals with disabilities. Since results of the current study determined there were differences among participants in areas of sexuality, future research should consider providing training and education on various topics of sexuality, specifically self-control and parenting, to determine if exposure to sexuality education produces a change in attitudes of sexuality for individuals with CHARGE syndrome. Moreover, future research should explore the extent to which attitudes of sexuality translate to actual behaviors. One way to analyze this area could be to look at attitudes of sexuality in matched samples of individuals with CHARGE syndrome and their parents. Improving attitudes of sexuality for a specific population is a critical step in decreasing marginalization and discrimination. However, it is when those positive perceptions translate to behavior, change will be imminent.

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APPENDIX A

EXAMPLE RECRUITMENT MATERIALS

Examining the attitudes towards sexuality in CHARGE syndrome

<u>*EXAMPLE* of Recruitment Materials Page</u>

Participants will be recruited through a variety of means including the following sources:

1. CHARGE Syndrome Facebook Page
2. MSU Bulldog CHARGE Lab Twitter
3. Yahoo! Listserve CHARGE Syndrome
4. Email recruitment letter
5. Recruitment flyers or posters at national/international CHARGE Syndrome Conferences

Recruitment Source: Social Media

Participants will be recruited from the following social media outlets:

1. CHARGE Syndrome Facebook Pages
2. MSU Bulldog CHARGE Lab Twitter
3. Yahoo! Listserve CHARGE Syndrome

Recruitment Materials

The Mississippi State Bulldog CHARGE Syndrome Research Lab is currently conducting a research study exploring the attitudes towards sexuality in CHARGE syndrome. Specifically, this study aims examine the attitudes towards sexuality from adults with CHARGE syndrome, as well as parents or legal guardian of an individual with CHARGE syndrome. While attitudes of sexuality have been examined in various populations of individuals with disabilities, there is little to no research involving those with low incidence conditions.

SO WHAT DO WE NEED FROM YOU?

We request that interested individuals click on the link below to complete an online survey (15-20 minutes).

WHO CAN PARTICIPATE?

1. Parents of individuals with a diagnosis of CHARGE (clinical or genetic diagnosis)
2. Individuals 21 years of age and older with diagnosis of CHARGE (clinical or genetic diagnosis)

Please contact Dr. Daniel Gadke or Dr. Kasee Stratton, Assistant Professors and Licensed Psychologists, if you are interested in participating or would like to inquire about any further information: dgadke@colled.msstate.edu or kstratton@colled.msstate.edu

Recruitment Source: Mail Letter

1. Email recruitment letter

2. Main recruitment letter

Recruitment Letter

DATE

Dear Parent/Caregiver:

We hope this [letter/email] finds you and your family well. Over the years, I have been presented with multiple questions and concerns regarding the topic of sexuality from both individuals with CHARGE syndrome and parents of individuals of CHARGE syndrome. As a result of these concerns, we are requesting your participation of a study, **Examining the attitudes towards sexuality in individuals with CHARGE syndrome.**

SO WHAT DO WE NEED FROM YOU?

We request that interested individuals go to the link below to complete an online survey (15-20 minutes).

WHO CAN PARTICIPATE?

1. Parents of individuals with a diagnosis of CHARGE (clinical or genetic diagnosis)
2. Individuals 21 years of age and older with diagnosis of CHARGE (clinical or genetic diagnosis)

Please contact Dr. Daniel Gadke or Dr. Kasee Stratton, Assistant Professors and Licensed Psychologists, if you are interested in participating or would like to inquire about any further information: dgadke@colled.msstate.edu or kstratton@colled.msstate.edu

All the best to you and your family,

Emily S. Mathis, M.S., BCBA

Doctoral Candidate

Mississippi State University

eas216@msstate.edu

Recruitment Source: Conference

1. Conference presentation/display (see attachment "Conference Recruitment Flyer")

BULLDOG CHARGE SYNDROME RESEARCH LAB

SEEKING PARTICIPANTS

Examining the Attitudes Towards Sexuality in CHARGE Syndrome

RESEARCH STUDY

Who we need?

- Individuals ages 18 and above with CHARGE syndrome
- Parents of individuals with CHARGE Syndrome

What can you do?

- Click on the link below to complete a questionnaire (approximately 20 minutes) or email kstratton@colled.msstate.edu to get the link sent to you
- Your name will go into a drawing to win a $50 Visa Card following completion of the questionnaire

Link:

This Research Investigation is being conducted by Dr. Kasee Stratton, Assistant Professor, at Mississippi State University. For more information please contact: kstratton@colled.msstate.edu

APPENDIX B

DEMOGRAPHICS QUESTIONNAIRE- PARENT

Parents of Individuals with CHARGE Syndrome Demographics Questionnaire

(to be entered in Qualtrics)

---

1. Are you the child's? *(Please Circle Below)*

   MOTHER FATHER GUARDIAN OTHER *(please specify)*

The following questions about the participant completing the survey:

2. Name: _____
3. Email: _____
4. What country do you currently live? _____
5. Date of Birth *(Month/Day/Year)*:_____/_____/_____
6. Gender:
   a. Male
   b. Female
7. Religion:
   a. List several common religions? Or have option to write in?
8. Highest Level of Education:
   a. List*
9. Level of Income
   a. List*
10. Marital Status
    a. Single
    b. Married
    c. Divorced
    d. Widowed/widower
11. How many children do you have? _____

The following questions about your child with CHARGE Syndrome.

12. Child's Name: _____
13. Child's Date of Birth *(Month/Day/Year)*:_____/_____/_____

14. Child's Gender *(Please Circle)* MALE FEMALE

15. At what age was your child diagnosed as having CHARGE? _____ months old OR _____ years old

16. Who made the diagnosis of CHARGE? (e.g. geneticist, ENT, pediatrician)

_____

GENE TESTING:

17. Has your child been tested for the *CHD7* gene mutation? _____ YES _____ NO

a. *If yes:* Did you child test positive or negative for the mutation?

_____ Positive _____Negative

b. When was your child tested? (Month/Year) _____/_____

CHARGE CHARACTERISTICS: *(Please check all that apply)*

	Check all that apply	**Characteristic**	**Description**
	Example: X	Child has CHARGE Syndrome	
18.		Coloboma of the eye	Coloboma of the iris, retina, choroid, macula or disc (not the eyelid); microphthalmos (small eye) or anophthalmos (missing eye): CAUSES VISION LOSS
19.		Choanal atresia or stenosis	The choanae are the passages that go from the back of the nose to the throat. They can be narrow (stenosis) or blocked (atresia). It can be unilateral (one-sided) or bilateral (both sides), bony or membranous.
20.		Anosmia (missing or decreased sense of smell)	Cranial Nerve I- missing or decreased sense of smell
21.		Swallowing problems	Cranial Nerve(s) IX/X - Swallowing difficulties, aspiration
	Check all that apply	**Characteristic**	**Description**
21.		Facial Palsy	Cranial Nerve VII - Facial palsy (one side or both)
22.		CHARGE outer ear	Short, wide ear with little/no lobe, "snipped off" helix (outer fold), inner fold which is discontinuous with tragus, triangular concha, floppy often stick out

23.		CHARGE middle ear	Malformed bones of the middle ear (ossicles): CAUSES CONDUCTIVE HEARING LOSS
24.		CHARGE inner ear	Malformed cochlea (Mondini defect); small or absent semicircular canals: CAUSE HEARING LOSS AND BALANCE PROBLEMS
25.		Sensorineural Hearing Loss	"Nerve loss"
26.		Vestibular Problems	Balance problems
27.		Frequent Middle Ear Infections	
28.		Heart Defects	Can be any type, but many are complex, such as tetralogy of Fallot
29.		Cleft lip +/- cleft palate	Cleft lip with or without cleft palate, cleft palate, submucous cleft palate
30.		TE (Tracheosophageal) fistula	Espphageal atreaisa, Trancheo-espphageal fistula (TEF), H-shaped TEF; connection between wind pipe and esophagus)
31.		Kidney Abnormalities	Small kidney, missing kidney, misplaced kidney, reflux
32.		Genital Abnormalities (Hypoplasia)	*Male*: small penis, undescended testes *Female*: small labia, small or missing uterus *Both*: lack of puberty without hormone intervention
33.		Growth deficiency	Growth hormone deficiency Other short stature
34.		Typical CHARGE Face	Square face w/ broad prominent forehead, arched eyebrows, large eyes, prominent nasal bridge with square root, thick nostrils, prominent nasal columella (between the nostrils), flat midface, small mouth, occasional small chin, larger chin with age. Facial asymmetry even without facial palsy
35.		Abdominal Defects	Umbilical hernia, omphalocele
36.		Palm crease	Hockey-stick palmar crease
37.		Spine Anomalies	Scoliosis, kyphosis, hemivertibrae
38.		Obsessive-Compulsive Behavior or Perseverative Behavior	Perseverative behavior in younger individuals, obsessive compulsive behavior (OCD) in older individuals
39.		Other	Please describe:
40.		Other	Please describe:

41. Please indicate any diagnoses given to your child for her/his behavior (such as Autism, ADHD, Intellectual Disability, etc):

42. Please indicate what educational diagnosis appears on your child's Individualized Education Plan (IEP) (e.g. Deafblind, Hearing impairment, Vision Impairment, Multiple Disabilities)?

---

43. What medications and herbal supplements is your child taking on a regular basis?

_____

_____

44. To the best of your knowledge, how well does your child see? (with glasses or contact lenses, if used)

(Circle number of ONE choice in each column)

LEFT	RIGHT	
1	1	NORMAL VISION
2	2	SOME TROUBLE SEEING
3	3	MODERATE DIFFICULTY
4	4	MUCH DIFFICULTY
5	5	TOTALLY BLIND

45. To the best of your knowledge, how well does your child hear? (with hearing aids or other hearing devices, if used)

(Circle number of ONE choice in each column)

LEFT	RIGHT	
1	1	NORMAL HEARING
2	2	SOME TROUBLE
3	3	MODERATE DIFFICULTY
4	4	MUCH DIFFICULTY
5	5	TOTALLY DEAF

46. Does your child have problems with sleep? *(Please Circle)* YES NO

47. How many surgeries has your child had? _____ Surgeries

48. To the best of your knowledge, has your child received sexuality education?
YES NO

If yes, who provided that education?

a) Parent
b) School Educator (e.g., teacher, coach, principal, school nurse)
c) Doctor
d) Friend
e) Mentor

APPENDIX C

DEMOGRAPHICS QUESTIONNAIRE – INDIVIDUALS WITH CHARGE SYNDROME

Individuals with CHARGE Syndrome Demographics Questionnaire

(to be entered in Qualtrics)

---

1. Name: _____
2. Date of Birth *(Month/Day/Year)*:_____/_____/_____
3. Email: _____
4. Do you have an intervener or anyone assisting you with completing this survey? YES NO
5. What country do you currently live? _____
6. Gender:
    a. Male
    b. Female
7. Religion:
    a. List several common religions? Or have option to write in?
8. Highest Level of Education:
    a. List*
9. Marital Status
    a. Single/Never married
    b. Married
    c. Divorced
    d. Widowed/widower
10. Do you have any children? _____

The following questions about your experiences with CHARGE Syndrome.

11. At what age were you diagnosed as having CHARGE? _____ months old OR _____ years old
12. Who made the diagnosis of CHARGE? (e.g. geneticist, ENT, pediatrician)

GENE TESTING:

13. Have you been tested for the *CHD7* gene mutation? _____ YES _____ NO

a. *If yes:* Did you test positive or negative for the mutation?

_____ Positive _____Negative

b. When were you tested? (Month/Year) _____/_____

CHARGE CHARACTERISTICS: *(Please check all that apply)*

	Check all that apply	**Characteristic**	**Description**
	Example: X	I have CHARGE Syndrome	
14.		Coloboma of the eye	Coloboma of the iris, retina, choroid, macula or disc (not the eyelid); microphthalmos (small eye) or anophthalmos (missing eye): CAUSES VISION LOSS
15.		Choanal atresia or stenosis	The choanae are the passages that go from the back of the nose to the throat. They can be narrow (stenosis) or blocked (atresia). It can be unilateral (one-sided) or bilateral (both sides), bony or membranous.
16.		Anosmia (missing or decreased sense of smell)	Cranial Nerve I- missing or decreased sense of smell
17.		Swallowing problems	Cranial Nerve(s) IX/X - Swallowing difficulties, aspiration
	Check all that apply	**Characteristic**	**Description**
18.		Facial Palsy	Cranial Nerve VII - Facial palsy (one side or both)
19.		CHARGE outer ear	Short, wide ear with little/no lobe, "snipped off" helix (outer fold), inner fold which is discontinuous with tragus, triangular concha, floppy often stick out
20.		CHARGE middle ear	Malformed bones of the middle ear (ossicles): CAUSES CONDUCTIVE HEARING LOSS
21.		CHARGE inner ear	Malformed cochlea (Mondini defect); small or absent semicircular canals: CAUSE HEARING LOSS AND BALANCE PROBLEMS
22.		Sensorineural Hearing Loss	"Nerve loss"
23.		Vestibular Problems	Balance problems
24.		Frequent Middle Ear Infections	

25.		Heart Defects	Can be any type, but many are complex, such as tetralogy of Fallot
26.		Cleft lip +/- cleft palate	Cleft lip with or without cleft palate, cleft palate, submucous cleft palate
27.		TE (Tracheosophageal) fistula	Espphageal atreaisa, Trancheo-espphageal fistula (TEF), H-shaped TEF; connection between wind pipe and esophagus)
28.		Kidney Abnormalities	Small kidney, missing kidney, misplaced kidney, reflux
29.		Genital Abnormalities (Hypoplasia)	*Male*: small penis, undescended testes *Female*: small labia, small or missing uterus *Both*: lack of puberty without hormone intervention
30.		Growth deficiency	Growth hormone deficiency Other short stature
31.		Typical CHARGE Face	Square face w/ broad prominent forehead, arched eyebrows, large eyes, prominent nasal bridge with square root, thick nostrils, prominent nasal columella (between the nostrils), flat midface, small mouth, occasional small chin, larger chin with age. Facial asymmetry even without facial palsy
32.		Abdominal Defects	Umbilical hernia, omphalocele
33.		Palm crease	Hockey-stick palmar crease
34.		Spine Anomalies	Scoliosis, kyphosis, hemivertibrae
35.		Obsessive-Compulsive Behavior or Perseverative Behavior	Perseverative behavior in younger individuals, obsessive compulsive behavior (OCD) in older individuals
36.		Other	Please describe:
37.		Other	Please describe:

38. Please indicate any other social/emotional/behavioral diagnoses you have been given (such as Autism, ADHD, Intellectual disability, anxiety, depression, etc):

_____

_____

39. Please indicate what educational diagnosis appeared on your school Individualized Education Plan (IEP) (e.g. Deafblind, Hearing impairment, Vision Impairment, Multiple Disabilities)?

_____

40. What medications and herbal supplements do you take on a regular basis?

_____

41. How well do you see? (with glasses or contact lenses, if used)

(Circle number of ONE choice in each column)

LEFT	RIGHT	
1	1	NORMAL VISION
2	2	SOME TROUBLE SEEING
3	3	MODERATE DIFFICULTY
4	4	MUCH DIFFICULTY
5	5	TOTALLY BLIND

42. How well do you hear? (with hearing aids or other hearing devices, if used)

(Circle number of ONE choice in each column)

LEFT	RIGHT	
1	1	NORMAL HEARING
2	2	SOME TROUBLE
3	3	MODERATE DIFFICULTY
4	4	MUCH DIFFICULTY
5	5	TOTALLY DEAF

43. How many surgeries have you child had? _____ Surgeries

44. Have you ever received education on sexuality? YES NO

If yes, who provided that education?

a) Parent
b) School Educator (e.g., teacher, coach, principal, school nurse)
c) Doctor
d) Friend
e) Mentor

APPENDIX D

ATTITUDES TO SEXUALITY QUESTIONNAIRE (ASQ-ID)

ATTITUDES TO SEXUALITY QUESTIONNAIRE

Cuskelly, Bryde & Gilmore

Published in Cuskelly & Gilmore (2007)

Permission granted from Dr. Cuskelly in July 2018

Questions in red were omitted from total scores based on findings from Cuskelly & Gilmore (2007)

In this section of the questionnaire, we are asking your views only about **female sexuality.** We realize that your answers might be different if we asked about males but please think only about **females** here without making any comparisons.

1. With the right support, women with CHARGE Syndrome can rear well-adjusted children.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

2. Provided no unwanted children are born and no-one is harmed, consenting adult women with CHARGE Syndrome should be allowed to live in a heterosexual relationship.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

3. Consenting women with CHARGE Syndrome should be allowed to live in a homosexual relationship if they so desire.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

4. It is best to wait for the girl or woman with CHARGE Syndrome to raise questions about sexuality before discussing the topic with her.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

5. Women with CHARGE Syndrome have less interest in sex than do other women.

Strongly	Disagree	Mildly	Mildly	Agree	Strongly

Disagree Disagree Agree Agree

6. If women with CHARGE Syndrome marry, they should be forbidden by law to have children.

Strongly Disagree | Disagree | Mildly Disagree | Mildly Agree | Agree | Strongly Agree

7. Women with CHARGE Syndrome should be allowed to engage in non-sexual romantic relationships.

Strongly Disagree | Disagree | Mildly Disagree | Mildly Agree | Agree | Strongly Agree

8. Medication should be used as a means of inhibiting sexual desire in women with CHARGE Syndrome.

Strongly Disagree | Disagree | Mildly Disagree | Mildly Agree | Agree | Strongly Agree

9. Masturbation should be discouraged for women with CHARGE Syndrome.

Strongly Disagree | Disagree | Mildly Disagree | Mildly Agree | Agree | Strongly Agree

10. Discussions on sexual intercourse promote promiscuity in women with CHARGE Syndrome.

Strongly Disagree | Disagree | Mildly Disagree | Mildly Agree | Agree | Strongly Agree

11. Women with CHARGE Syndrome should only be permitted to marry if either they or their partners have been sterilised.

Strongly Disagree | Disagree | Mildly Disagree | Mildly Agree | Agree | Strongly Agree

12. Masturbation in private for women with CHARGE Syndrome is an acceptable form of sexual expression.

Strongly Disagree | Disagree | Mildly Disagree | Mildly Agree | Agree | Strongly Agree

13. Women with CHARGE Syndrome typically have fewer sexual interests than other women.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

14. Generally women with CHARGE Syndrome are able to make distinctions between sexual thoughts and sexual actions.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

15. Women with CHARGE Syndrome are unable to develop and maintain an emotionally intimate relationship with a partner.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

16. Sex education for women with CHARGE Syndrome has a valuable role in safeguarding them from sexual exploitation.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

17. In general, sexual behaviour is a major problem area in management and caring for women with CHARGE Syndrome.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

18. Sexual intercourse should be permitted between consenting adults with CHARGE Syndrome.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

19. Group homes or hostels for adults with CHARGE Syndrome should be either all male or all female, not mixed.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

20. Care staff and parents should discourage women with CHARGE Syndrome from having children.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

21. It is best not to discuss issues of sexuality with girls with CHARGE Syndrome until they reach puberty.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

22. Women with CHARGE Syndrome have the right to marry.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

23. It is a good idea to ensure privacy at home for women with CHARGE Syndrome who wish to masturbate.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

24. Whenever possible, women with CHARGE Syndrome should be involved in the decision about their being sterilized.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

25. Sexual intercourse should be discouraged for women with CHARGE Syndrome.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

26. Advice on contraception should be fully available to women with CHARGE Syndrome whose level of development makes sexual activity possible.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

27. Women with CHARGE Syndrome are more easily stimulated sexually than people without CHARGE Syndrome.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

28. Marriage between adults with CHARGE Syndrome does not present society with too many problems.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

29. Sterilisation is a desirable practice for women with CHARGE Syndrome.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

30. Sex education for women with CHARGE Syndrome should be compulsory.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

31. Masturbation should be taught to women with CHARGE Syndrome as an acceptable form of sexual expression in sex education courses.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

32. Marriage should not be encouraged as a future option for women with CHARGE Syndrome.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

33. Women with CHARGE Syndrome should be permitted to have children within marriage.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

34. Women with CHARGE Syndrome have stronger sexual feelings than other women.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

In this section of the questionnaire, we are asking your views only about **male sexuality.** We realize that your answers might be different if we asked about males but please think only about **males** here without making any comparisons.

1. With the right support, men with CHARGE syndrome can rear well-adjusted children.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

2. Provided no unwanted children are born and no-one is harmed, consenting adult men with CHARGE syndrome should be allowed to live in a heterosexual relationship.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

3. Consenting men with CHARGE syndrome should be allowed to live in a homosexual relationship if they so desire.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

4. It is best to wait for the girl or woman with CHARGE syndrome to raise questions about sexuality before discussing the topic with her.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

5. Men with CHARGE syndrome have less interest in sex than do other men.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

6. If men with CHARGE syndrome marry, they should be forbidden by law to have children.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

7. Men with CHARGE syndrome should be allowed to engage in non-sexual romantic relationships.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

8. Medication should be used as a means of inhibiting sexual desire in men with CHARGE syndrome.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

9. Masturbation should be discouraged for men with CHARGE syndrome.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

10. Discussions on sexual intercourse promote promiscuity in men with CHARGE syndrome.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

11. Men with CHARGE syndrome should only be permitted to marry if either they or their partners have been sterilised.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

12. Masturbation in private for men with CHARGE syndrome is an acceptable form of sexual expression.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

13. Men with CHARGE syndrome typically have fewer sexual interests than other men.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

14. Generally men with CHARGE syndrome are able to make distinctions between sexual thoughts and sexual actions.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

15. Men with CHARGE syndrome are unable to develop and maintain an emotionally intimate relationship with a partner.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

16. Sex education for men with CHARGE syndrome has a valuable role in safeguarding them from sexual exploitation.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

17. In general, sexual behaviour is a major problem area in management and caring for men with CHARGE syndrome.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

18. Sexual intercourse should be permitted between consenting adults with CHARGE syndrome.

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19. Group homes or hostels for adults with CHARGE syndrome should be either all male or all female, not mixed.

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26. Advice on contraception should be fully available to men with CHARGE syndrome whose level of development makes sexual activity possible.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

27. Men with CHARGE syndrome are more easily stimulated sexually than people without CHARGE syndrome.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

28. Marriage between adults with CHARGE syndrome does not present society with too many problems.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

29. Sterilisation is a desirable practice for men with CHARGE syndrome.

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Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

32. Marriage should not be encouraged as a future option for men with CHARGE syndrome.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

33. Men with CHARGE syndrome should be permitted to have children within marriage.

Strongly	Disagree	Mildly	Mildly	Agree	Strongly

Disagree		Disagree	Agree		Agree

34. Men with CHARGE syndrome have stronger sexual feelings than other men.

Strongly Disagree	Disagree	Mildly Disagree	Mildly Agree	Agree	Strongly Agree

APPENDIX E

IRB APPROVAL

MISSISSIPPI STATE UNIVERSITY

Office of Research Compliance

Institutional Review Board for the Protection of Human Subjects in Research
P.O. Box 6223
53 Morgan Avenue
Mississippi State, MS 39762
P. 662.325.3294

www.orc.msstate.edu

NOTICE OF DETERMINATION FROM THE HUMAN RESEARCH PROTECTION PROGRAM

DATE:	July 12, 2019
TO:	Daniel Gadke, Phd, Counseling Ed Psyc & Foundations, Jasmine Sorrell;Kasee Stratton-Gadke;Tawny McCleon;Tianlan Wei Emily Mathis, MS, Counsel Ed Psych & Foundation, Jasmine Sorrell, BS, Counsel Ed Psych & Foundation, Kasee Stratton-Gadke, PhD, Counseling Ed Psyc & Foundations, Tawny McCleon, Phd, Counseling Ed Psyc & Foundations, Tianlan Wei, PhD, Counseling Ed Psyc & Foundations
PROTOCOL TITLE:	Examining Attitudes Towards Sexuality in CHARGE Syndrome
PROTOCOL NUMBER:	IRB-19-209
APPROVAL PERIOD:	Approval Date: July 12, 2019 Expiration Date: July 11, 2024

Under an expedited review procedure, the research project identified above was approved on July 12, 2019 by the Mississippi State University Institutional Review Board (MSU IRB). The application qualified for expedited review under CFR 46.110, Category 7.

This memorandum is your record of the IRB approval of this study. Please maintain it with your study records.

Please note that the MSU HRPP accreditation for our human subjects protection program requires an approval stamp for consent forms. The approval stamp will assist in ensuring the HRPP approved version of the consent form is used in the actual conduct of research. **If applicable, you must use the stamped consent form for obtaining consent from participants.**

The MSU IRB approval for this project will expire on July 11, 2024. If you expect your project to continue beyond this date, you must submit an application for renewal of this HRPP approval. HRPP approval must be maintained for the entire term of your project.

If, during the course of your project, you intend to make changes to this study, you must obtain approval from the HRPP prior to implementing any changes. Upon becoming aware of an unanticipated problem that suggests participants or others are at greater risk of harm than was previously known or recognized, a problem report must be submitted to the HRPP as soon as possible, but always within 10 days. Serious problems must be reported verbally within one business day, in addition to the submission of the written Problem Report.

You are required to maintain complete records pertaining to the use of humans as participants in your research. This includes all information or materials conveyed to and received from participants as well as signed consent forms, data, analyses, and results. These records must be maintained for at least three years following project completion or termination, and they are subject to inspection and review by the HRPP and other authorized agencies.

Please notify this office when your project is complete. Upon notification, we will close our files pertaining to your project. Reactivation of the HRPP approval will require a new HRPP application.

If you have any questions relating to the protection of human research participants, please contact the HRPP by phone at 662.325.5220 or email irb@research.msstate.edu. We wish you the very best of luck in your research and look forward to working with you again.

Approval Period:	July 12, 2019 through July 11, 2024
Review Type:	EXPEDITED
IRB Number:	IORG0000467