

Parenting Capacities of Primary Caregivers of Children with Disorders of Sexual Development

A Thesis Presented

by

Reagan D. Reed

to

The Department of Psychology

in partial fulfillment of the requirements for a Departmental Honors Award

April 28, 2016

Oklahoma State University



Table of Contents

Abstract.....	3
Introduction.....	4
Beyond the Medical Implications.....	5
Objective.....	5
Defining Parenting Capacity.....	6
Parenting capacities of caregivers of children with a chronic illness.....	7
<i>Parental overprotection</i>	7
<i>Perceived child vulnerability</i>	8
<i>Parenting Stress</i>	8
Early experiences and medical decision making among parents of children with DSD.....	9
Parenting capacities of caregivers of children with DSD.....	11
<i>Parenting stress</i>	11
<i>Parental overprotection</i>	11
<i>Perceived child vulnerability</i>	12
Psychological outcomes among parents of children diagnosed with DSD.....	12
Future Directions.....	13
Conclusion.....	14
References.....	15

Abstract

Atypical development of chromosomal, gonadal or anatomic sex (a disorder known as DSD) is announced at or shortly after birth and can put great stress on caregivers who are responsible for medical decision making, aftercare, and promoting future adjustment for children with this disorder. Consensus statements have recognized the stress associated with DSD for patients and their parents, however, much remains to be learned concerning parental characteristics and psychological outcomes among primary caregivers of children with DSD. Existing data and results of parenting studies of caregivers of children with chronic illness are presented as well as the limited findings within the DSD population. Psychological outcomes among parents of children diagnosed with DSD are discussed and future directions are offered.

Introduction

Disorders of sex development (DSD) are defined as “congenital conditions in which development of chromosomal, gonadal, or anatomic sex is atypical” (Hughes, Houk, Ahmed, & Lee, 2006). Disorders of this type were previously often referred to as “intersex conditions,” “hermaphroditism,” or “pseudohermaphroditism (male or female),” but this nomenclature has since been changed, in large part because of the negative response of individuals with ambiguous genitalia who found these terms to be derogatory. It is important to use terminology that is sensitive to the concerns of patients, and consequently, a Consensus document was published to develop a more appropriate term which would encompass many varieties of DSD, but exclude conditions which only cause precocious or delayed puberty (Hughes, 2008; Hughes et al., 2006). To date, much of what is known about DSD has stemmed from research illuminating genetic and genital variations within the population of individuals with DSD and from retrospective medical management and outcome studies, and they remain a largely understudied phenomenon.

The terminology agreed upon by professionals in the Consensus document is extremely important for the doctors and professionals involved in the medical management of individuals and families with DSD, as it provides a universal language allowing them clear communication. Optimal care for children and adults with DSD requires an experienced multidisciplinary team made up of pediatric subspecialists in endocrinology, surgery, and urology, psychologists, gynecologists, geneticists, neonatologists, and social workers, which will vary based on location and financial ability, but is important for intercommunication among medical providers (Hughes, 2008; Hughes et al., 2006; Hughes, Nihoul-Fékété, Thomas, & Cohen-Kettenis, 2007). It has been proposed, and widely accepted, that psychological care should be an integral part of the medical management regimen for DSD patients because of the sensitivity of the subject and the vulnerability that both children and adults face for psychological and social harm (Hughes et al., 2007). It is also important that parents of these patients are included in the medical management process so that they are educated about sexual development and are emotionally able to work with surgical doctors to reach a decision on the best way to proceed.

Beyond the Medical Implications

Children born with DSD pose numerous challenges for parents both emotionally and financially. Because the population of individuals diagnosed with this disorder is understudied and misunderstood, many parents are unaware and unable to understand that biological sex does not always conform to either “male” or “female.” In a society that commonly views biological sex as binary, parents can become overwhelmed with uncertainty after a DSD diagnosis is made. Parents are uncertain about their child’s future gender identity, find it difficult to understand complex medical information and terminology, and are fearful of the medical and psychological implications their decisions may have. In addition, they must learn to cope with continuous medical care, and must face potentially huge financial burdens and strains on the family (Sanders, Carter, & Goodacre, 2011; Sanders, Carter, & Goodacre, 2012).-

Atypical sex development and gender behavior carry a stigma that many parents feel burdened with concealing, and when children become aware of their atypical genitalia, parents must decide what to reveal to their children and how to do so (Alpern, Gardner, Kogan, Sandberg, & Quittner, 2016). Thus far, much of the DSD research that parents have access to has looked at the medical and surgical outcomes among DSD patients, the psychosocial and psychological well-being of children with DSD or gender identity disorders, ethical challenges in the medical decision making process, and health-related quality of life among children with DSD and their parent. Despite these extensive studies over the previous topics, there has been limited research reviewing the caregiver’s psychological well-being and parenting capacities after the DSD diagnosis and medical management decisions have been made.

Objective

The objective of this paper is to review the existing literature on parenting capacities among individuals who are the primary caregivers for children diagnosed with DSD. Research in the field of DSD is hindered by a lack of research on parenting capacities and their impact on both the family system and the psychosocial well-being of the affected caregivers. Existing literature is presented and possible future directions for research in this understudied population are discussed.

Defining Parenting Capacity

This review addresses three specific parenting variables, have been referred to globally as “parenting capacities.” According to NIH, parenting capacities are defined as parenting practices or behaviors (e.g., lack of appropriate parental monitoring, supervision, and communication, high family conflict and disorganization, parental stress and depression, lack of parent-child bonding and negative discipline methods) that can contribute to a variety of negative outcomes in children. A number of parenting capacities have been studied and found to be consistently associated with poor child and parent adjustment outcomes in the context of chronic illness. They include parental overprotection, perceived child vulnerability, and parenting stress (Mullins et al., 2004). The concept of parental overprotection, coined by Levy (1931), has been described as excessive protective behavior by a parental figure considering the developmental stage of the child, which leads to prolonged infantilization and discouragement of independence and social maturity (Thomasgard & Metz, 1997). The concept of perceived child vulnerability is based on the observation that parents, oftentimes of children with chronic illness, have an increased negative perception of their child’s vulnerability to illness or injury which then leads to a significant discrepancy between the child’s actual health and the parental belief that their child continues to be at risk for serious illness or premature death (Thomasgard & Metz, 1997). Because perceived child vulnerability oftentimes leads to prolonged infantilization and parental overprotection, the terms have often been used interchangeably. However, the two concepts vary from each other in that perceived child vulnerability is just a flawed perception or belief, while parental overprotection an actual behavior or act (Thomasgard & Metz, 1997). Lastly, the concept of parenting stress includes the level of stress present in the parent-child relationship which can be caused by parental distress, difficult child characteristics and dysfunctional or unsatisfactory parent-child interactions (Abidin, 1990). These three parenting capacities are important to review together because it is believed that these variables have the ability to influence one another and, consequently, further compound the cycle of poor child and parental adjustment to chronic illness. In the next section, the research on these constructs in chronic illness populations other than DSDs will first be reviewed.

Parenting capacities of caregivers of children with a chronic illness

As previously mentioned, it is important to examine the constructs of parental overprotection, perceived child vulnerability, and parenting stress in the population of parents with children with a chronic illness in light of the consistent finding that parental adjustment variables are seen as influencing child-adjustment, and vice versa. Parenting capacities may be exhibited in different ways given differences in illness implications (i.e., severity of illness, prognosis, intensity of treatment, and predictability of side effects), knowledge of these differences may be helpful in treatment of parenting capacities across parents of children with various chronic illnesses (Hullmann et al., 2010)

Parental overprotection has been linked to poor behavioral adjustment, risky sexual behavior and inadequate regimen adherence in youth outcomes, and thus it is extremely important for research on parental monitoring and overprotection to be conducted in samples of caregivers of children who are chronically ill (Ellis, Templin, Naar-King, & Frey, 2008). Additionally, high levels of parental overprotection, or lack of promoting autonomy of the child, restricts the child's freedom and interfere with their development of social skills (Power, Dahlquist, Thompson, & Warren, 2003). Parental overprotection is oftentimes demonstrated by excessive physical or social contact, infantilization, and excessive concern- for the child's well-being, and is likely to develop in families with chronically ill children because these children require intensive medical management and place considerable physical, psychological, and social demands on families involved (Holmbeck et al., 2002; Thomasgard & Metz, 1997).

In a study that looked at parental overprotection in families with a chronic illness (spina bifida), mothers were found to be more likely to be over protective than fathers (Holmbeck et al., 2002). This may occur because mothers of children with a chronic illness may be more responsible for the multiple caretaking demands that are placed on the family (Floyd & Zmich, 1991). Because caring for a chronically ill child involves strict adherence to medical management programs, and parents want to protect their children from adverse medical outcomes, parents may engage in excessive levels of parental protection because they perceive their children to be more vulnerable to illness or injury than other children. (Thomasgard & Metz, 1997).

Perceived child vulnerability is used to describe the anxiety that parents of chronically ill children have about their child's health that stems from their concern that their child does not look as healthy as they should or the belief that he or she usually catches illness going around more easily than other children (Hullmann et al., 2010). It has been suggested that these anxious cognitions about children's illness often leads to the "vulnerable child syndrome," which is a maladaptive pattern of parent-child interactions and child behavior problems (Leslie & Boyce, 1996). This parental anxiety causes parents to respond to their child's chronic illness with excessive concern which has the potential to communicate to their child that they are indeed more vulnerable than other children (Mullins et al., 2007).

In a study of children with chronic asthma, researchers found that parents who perceive their children as being vulnerable are more likely to keep them home from school and take them to a physician's office for acute asthma care (Spurrier et al., 2000). Additional research on perceived vulnerability indicated that increased levels of perceived vulnerability was also associated with negative academic and psychosocial outcomes for children (Hullmann et al., 2010). Consistent with these findings, children whose parents perceive them as being more vulnerable also reported more generalized social distress and distress in social situations (Spurrier et al., 2000).

Parenting stress is conceptualized as the level of stress in the parent-child relationship and has been linked directly, through its relationship with poor parenting, to maladjustment in both children and their parents (Friedman, Holmbeck, Jandasek, Zukerman, & Abad, 2004; Hullmann et al., 2010). Results of a meta-analytic review of caregivers of children with chronic illness found that parents of chronically ill children endorsed greater general parenting stress than caregivers of healthy children. The same study found that caregivers of chronically ill children are also more likely to experience illness-related parenting stress (i.e. frequent clinic appointments, demanding treatment regimens) (Cousino & Hazen, 2013). Additionally, it has been found that there are significantly positive associations between general parenting stress and posttraumatic stress disorder symptoms in caregivers of children with cancer (Kazak & Barakat, 1997).

Parenting stress has also been found to be related to negative outcomes related to the child's disease (Barakat et al., 2007; Patterson, Goetz, Budd, & Warwick, 1993). More

specifically, Barakat et al. (2007) found that caregivers of children with sickle cell disease who had elevated levels of disease-related parenting stress were associated with children with greater disease severity and were found to have utilized health care more frequently. In a study with similar findings, Patterson et al. (1993) found that parents of children with cystic fibrosis who had elevated levels of parenting stress were associated with children who exhibited poorer adherence, lung functioning, and overall growth than children with cystic fibrosis whose parents showed less stress.

In summary, it is apparent that parental adjustment variables such as parental overprotection, perceived child vulnerability, and parenting stress are all interrelated and can result in negative outcomes in children with various chronic illnesses. Parents who perceive their chronically ill child as being more vulnerable are likely to exhibit excessive levels of parental protection and are also more likely to report experiencing parenting stress, more specifically, illness-related parenting stress than parents of healthy children. Research on these three maladaptive parenting capacities has been conducted on families with several different chronic illnesses, but few have been conducted in the context of children with DSD.

Early experiences and medical decision making among parents of children with DSD

Disorders of sex development have the potential to exert substantial amounts of strain on the families of children with this condition. Duguid et al. (2006) observed that over 60% of parents in their study experienced difficulties exchanging information about their child's condition with relatives and friends and 68% reported being concerned about the stigma or ridicule associated with the condition (Duguid et al., 2007; Hughes, Houk, Ahmed, & Lee, 2006). In another study that aimed to identify significant aspects of the early parental experience of a DSD diagnosis, researchers found that many of the parents interviewed opted out of sharing information about their child's condition with close family and friends because they feared it would lead to stigmatization, believed the right to disclose information should be preserved for their child, and felt they would be unable to adequately explain the condition. Such beliefs resulted in increased parental distress and avoidance of situations in which their children's genitals might be seen (Crissman et al., 2011).

Another major contribution to the negative early experiences of parents is the initial lack of education and the shock that accompanies their child's diagnosis of a condition characterized

by external and internal genitals, gonadal structures, sex chromosomes and/or hormone levels that are not typical for only one sex (Crissman et al., 2011; Zeiler & Wickstrom, 2009). This initial state of uncertainty is reported as being extremely difficult for parents because the child is taken from the parents very soon after birth so that professionals can begin medically testing and treating the child with hormones in order to biologically define the child's sex. The testing can take several months to complete, which makes it difficult for some parents to connect with their baby because of the lack of a definite sex identification (Zeiler & Wickstrom, 2009).

Interestingly, Crissman et al. (2011) found that several of the parents in their study felt certain about knowing their child's sex and gender of rearing prior to the completion of medical testing, despite a discordance across karyotype, gonads, or genital anatomy, simply based on their personal intuitions and the appearance of the external genitalia. After testing is completed, some medical professionals state that a decision regarding the child's sex is required as soon as possible and recommend early surgical intervention for medical, functional, and cosmetic reasons as well as its potential to relieve parental distress and improve parent-child attachment (Hughes et al., 2006).

Parents are considered to be responsible for understanding their child's best interest and making the difficult decision of whether or not to consent to genital reconstructive surgery (Wiesemann, Ude-Koeller, Sinnecker, & Thyen, 2009). Although the long-term outcome and possible risk of genital surgeries in childhood is unknown, poor decision-making by parents during the child's childhood can lead to distress in all individuals involved later in their life (Karkazis, Tamar-Mattis, & Alexander, 2010; Wiesemann et al., 2009). Accordingly, the educational process and discussions about sexual differentiation should thoroughly provide an overview of sexual differentiation as it applies to their child and should discuss possible medical/surgical treatments and their limitations and reported outcomes (Dayner, Lee, & Houk, 2004). Parents tend to disfavor postponing genital surgery until later in life despite the acknowledged risk to sexual functioning that results from such procedures which could be indirect evidence supporting the DSD consensus statement (2006) that notes that cosmetic surgeries performed in the first year are thought to relieve parental distress (Fedele et al., 2010).

Parenting capacities of caregivers of children with DSD

In the extant chronic illness literature previously reviewed, researchers found that parents of children with chronic illnesses report significant levels of parenting stress and exhibit high levels of parental overprotection and perceived child vulnerability. While there are many published reports of these parenting capacities and their characteristics within the population of children with chronic illness, there have been few published reports of parenting capacities of caregivers of children with genital anomalies. Notably, the research that has been conducted with this population to date has reported that caregivers of children with DSD experience similar levels of parenting stress, overprotection and perceived child vulnerability as caregivers of children with Type 1 diabetes mellitus (Kirk et al., 2011).

Parenting stress is a common parenting capacity reviewed in the extant DSD literature. Notably, it has been found that caregivers report increased parenting-related stress if they are rearing a child in whom they chose not to assent to genitoplasty. More specifically, Fedele et al. (2010) found that female caregivers report feeling more parenting-related stress if their child's genitalia remained ambiguous, which they speculated to be caused by the female caregiver's likelihood to take primary responsibility for their child's early physical needs. Studies have also looked at several parental factors that are thought to lead to parental stress. One such study found that two of the major concerns that increased parental stress levels were concerns related to surgery and anesthetics, not necessarily concerns about sexual function and fertility (Duguid et al., 2007). In a study that examined the differential effect of developmental stage on parental adjustment, researchers found that parenting stress was significantly higher in parents of adolescents rather than those of school-aged children, which is likely related to findings that parents of adolescents experience higher rates of personal stress and parenting stress related to their interactions with their child (Hullmann, Fedele, Wolfe-Christensen, Mullins, & Wisniewski, 2011). Finally, both parenting stress and parental overprotection have been observed to be experienced at increased levels by caregivers of younger children with DSD than caregivers of older children (Hullmann et al., 2011).

Parental overprotection is exhibited by many caregivers of children with ambiguous genitalia who feel that they are responsible for protecting their child from possible harm or embarrassment. This heightened sense of responsibility felt by parents of children with DSD is

confirmed in interviews with parents who faced the dilemma of sharing information about the condition with their child too early or not sharing information early enough and the resulting fear that their child would be unaware of their differences which would impact their self-esteem and confidence in their child's future intimate relationships (Sanders, Carter, & Goodacre, 2012). Furthermore, results found by Hullmann et al. (2011) found that caregivers of infants and toddlers reported significantly more overprotective behaviors than caregivers of children in the other age groups. These findings can be explained by the developmental theory, which suggests that parents expect their children to become more independent and autonomous as they mature and thus they do not have to remain as protective (Williams, Holmbeck, & Greenley, 2002). Several studies have found that parents of children with DSD perceive them as being vulnerable, which may explain why parents of these children exhibit increased levels of parental overprotection (Kirk et al., 2011; Sanders et al., 2012).

Levels of *perceived child vulnerability* was found to be equivalent among female caregivers of children with DSD and those of children with T1DM. However, differences between these two chronic illnesses exist in that caregivers of boys with DSD reported increased levels of perceived child vulnerability than those of girls (Kirk et al., 2011). These researchers speculated that this could be attributed to the fact that there is greater agreement within the medical community about how to treat girls with DSD as compared to boys, or, that female caregivers experience concern about their son's social competency and dissatisfaction with genital appearance because of the poorer cosmetic outcome for males (Crawford, Warne, Grover, Southwell, & Hutson, 2009; Gupta, Bhardwaj, Sharma, Ammini, & Gupta, 2010; Kirk et al., 2011). Not only do excessive levels of perceived vulnerability communicate to the child that they are more vulnerable than other "healthy" children, but parents who are experiencing this excessive concern are also more likely to exhibit externalizing behaviors and experience anxiety (Kazak & Barakat, 1997; Mullins et al., 2007).

Psychological outcomes among parents of children diagnosed with DSD

As previously noted, caregivers of children with DSD and other chronic illnesses are at risk for exhibiting maladaptive parenting capacities (i.e., increased levels of parental overprotection, perceived child vulnerability and parenting stress) which may, in turn, lead to increased levels of psychological distress. In a study that aimed to identify the relationship

between maladaptive parenting capacities and psychological distress (i.e., anxious and depressive symptoms) in caregivers of children with DSD, findings indicated that increased levels of parenting stress were significantly related to higher levels of anxiety and depressive symptoms in caregivers, while increased levels of perceived child vulnerability were solely related to higher levels of anxiety (Wolfe-Christensen et al., 2014). Another study that evaluated the psychological outcomes of caregivers of children with DSD, specifically examined the degree of external genital malformation and subsequent distress among caregivers. These researchers found that severe under-masculinization of the external genitals was associated with caregiver depression among caregivers of children who were reared male (Wolfe-Christensen et al., 2012). The same study also found that caregiver depression and anxiety were not related to the over-masculinization of the external genitals of children reared female. Notably, it has also been found that male caregivers and female caregivers of children with DSD appear to be differentially affected by medical decisions regarding genitoplasty; male caregivers find the age at which their child should undergo surgery to be the most psychologically distressing issue, while female caregivers are much more distressed about deciding whether or not their child should undergo surgery at all (Fedele et al., 2010). Taken together, these findings indicate that maladaptive parenting capacities in the context of condition-specific characteristics, such as under or over-masculinization, and medical-decision making have the potential to increase the levels of psychological distress experienced by both male and female caregivers.

Future Directions

It is necessary that family based interventions are utilized in order to reduce maladaptive parenting capacities and psychological distress; these interventions can potentially improve the functioning of caregivers of children with chronic illness and DSD specifically (Wolfe-Christensen et al., 2014). However, in order to implement a model of family-centered care and intervention, it is essential that professionals are able to acknowledge and understand how a child's disorder or illness affects the entire family (Case-Smith, 2004). It has been speculated that caregivers experience much of their psychological distress due to fears that others in the community will observe their child's ambiguous genitalia and question their gender or that their child's atypical genital anatomy will lead to atypical sexual behavior. Therefore, it would be

beneficial for future research to examine the socio-cultural circumstances that influence parental preferences and decision making for gender assignment (Fedele et al., 2010).

Cultural and social considerations are thought by some researchers to take precedence over medical factors in the decisional context because parents are motivated to “normalize” their child’s appearance and communication of identity to meet perceived social and cultural expectations (Daniel, Kent, Binney, & Pagdin, 2005; Nelson, Caress, Glenny, & Kirk, 2012). Parents oftentimes face an intense urge to “normalize” their children because “people don’t accept that everybody’s different” and they believe it is the only way to protect them from the emotional and social pain they might potentially face in the future (Gough, Weyman, Alderson, Butler, & Stoner, 2008; Sanders, Carter, & Goodacre, 2011). Cultural considerations are extremely understudied but necessary for a comprehensive understanding of parental experiences. All cultures vary in their views of men and women, which is why research should be done to examine the impact it has on parental decision making in light of the unpredictability of gender identity, sexual orientation, or surgical outcomes in children with DSD.

Conclusion

New research in the field of DSD is currently emerging, and it appears that research in the area of parenting capacity variables and parental outcomes remains an important set of issues to address. The existing data indicates that primary caregivers of children with chronic illness, particularly DSD, exhibit increased levels of maladaptive parenting characteristics. Perhaps caregivers adopt overprotective parenting characteristics because they perceive their child as being vulnerable to judgement from others who might observe their ambiguous genitalia or they fear their child’s condition will lead to atypical sexual behavior. Future qualitative studies are required to better understand why some caregivers adopt these parenting capacities and how interventions may improve functioning for both the parents and the child.

References

- Alpern, A. N., Gardner, M., Kogan, B., Sandberg, D. E., & Quittner, A. L. (2016). Development of Health-Related Quality of Life Instruments for Young Children With Disorders of Sex Development (DSD) and Their Parents. *Journal of Pediatric Psychology*. doi:10.1093/jpepsy/jsw022
- Abidin, R. (1990). Parenting Stress Index/Short Form manual. Los Angeles, CA: Western Psychological Services.
- Barakat, L. P., Patterson, C. A., Weinberger, B. S., Simon, K., Gonzalez, E. R., & Dampier, C. (2007). A Prospective Study of the Role of Coping and Family Functioning in Health Outcomes for Adolescents With Sickle Cell Disease. *Journal of Pediatric Hematology/Oncology*, 29(11), 752-760. doi:10.1097/MPH.0b013e318157fdac
- Case-Smith, J. (2004). Parenting a Child With a Chronic Medical Condition. *American Journal of Occupational Therapy*, 58(5), 551-560. Retrieved from <http://argo.library.okstate.edu/login?url=http://search.proquest.com/docview/620528062?accountid=4117>
- Cousino, M. K., & Hazen, R. A. (2013). Parenting stress among caregivers of children with chronic illness: A systematic review. *Journal of Pediatric Psychology*, 38(8), 809-828. doi:http://dx.doi.org/10.1093/jpepsy/jst049
- Crawford, J. M., Warne, G., Grover, S., Southwell, B. R., & Hutson, J. M. (2009). Results from a pediatric surgical centre justify early intervention in disorders of sex development. *Journal of Pediatric Surgery*, 44(2), 413-416. doi:http://dx.doi.org/10.1016/j.jpedsurg.2008.10.101
- Crissman, H. P., Warner, L., Gardner, M., Carr, M., Schast, A., Quittner, A. L., Sandberg, D. E. (2011). Children with disorders of sex development: A qualitative study of early parental experience. *International Journal of Pediatric Endocrinology*, 2011(1), 10. doi:10.1186/1687-9856-2011-10
- Daniel, E., Kent, G., Binney, V., & Pagdin, J. (2005). Trying to do my best as a mother: Decision-making in families of children undergoing elective surgical treatment for short

- stature. *British Journal of Health Psychology*, *10*(1), 101-114.
doi:10.1348/135910704X14609
- Dayner, J. E., Lee, P. A., & Houk, C. P. (2004). Medical treatment of intersex: parental perspectives. *Journal of Urology*, *172*(4 Pt 2), 1762-1765; discussion 1765. Retrieved from <http://www.ncbi.nlm.nih.gov/pubmed/15371809>
- Duguid, A., Morrison, S., Robertson, A., Chalmers, J., Youngson, G., Ahmed, S. F., & Scottish Genital Anomaly, N. (2007). The psychological impact of genital anomalies on the parents of affected children. *Acta Paediatrica*, *96*(3), 348-352. doi:10.1111/j.1651-2227.2006.00112.x
- Ellis, D. A., Templin, T. N., Naar-King, S., & Frey, M. A. (2008). Toward conceptual clarity in a critical parenting construct: Parental monitoring in youth with chronic illness. *Journal of Pediatric Psychology*, *33*(8), 799-808. doi:http://dx.doi.org/10.1093/jpepsy/jsn044
- Fedele, D. A., Kirk, K., Wolfe-Christensen, C., Phillips, T. M., Mazur, T., Mullins, L. L., . . . Wisniewski, A. B. (2010). Primary caregivers of children affected by disorders of sex development: mental health and caregiver characteristics in the context of genital ambiguity and genitoplasty. *International Journal of Pediatric Endocrinology*. 2010, 690674. doi:10.1155/2010/690674
- Floyd, F. J., & Zmich, D. E. (1991). Marriage and the Parenting Partnership: Perceptions and Interactions of Parents with Mentally Retarded and Typically Developing Children. *Child Development*, *62*(6), 1434-1448. doi:10.2307/1130817
- Friedman, D., Holmbeck, G. N., Jandasek, B., Zukerman, J., & Abad, M. (2004). Parent Functioning in Families of Preadolescents With Spina Bifida: Longitudinal Implications for Child Adjustment. *Journal of Family Psychology*, *18*(4), 609-619. doi:10.1037/0893-3200.18.4.609
- Gough, B., Weyman, N., Alderson, J., Butler, G., & Stoner, M. (2008). 'They did not have a word': The parental quest to locate a 'true sex' for their intersex children. *Psychology & Health*, *23*(4), 493-507. doi:10.1080/14768320601176170

- Gupta, D., Bhardwaj, M., Sharma, S., Ammini, A. C., & Gupta, D. K. (2010). Long-term psychosocial adjustments, satisfaction related to gender and the family equations in disorders of sexual differentiation with male sex assignment. *Pediatric Surgery International, 26*(10), 955-958. doi:10.1007/s00383-010-2661-y
- Holmbeck, G. N., Johnson, S. Z., Wills, K. E., McKernon, W., Rose, B., Erklin, S., & Kemper, T. (2002). Observed and perceived parental overprotection in relation to psychosocial adjustment in preadolescents with a physical disability: The mediational role of behavioral autonomy. *Journal of Consulting and Clinical Psychology, 70*(1), 96-110. doi:10.1037/0022-006X.70.1.96
- Hughes, I. A. (2008). Disorders of sex development: a new definition and classification. *Best Practice & Research Clinical Endocrinology & Metabolism, 22*(1), 119-134. doi:http://dx.doi.org/10.1016/j.beem.2007.11.001
- Hughes, I. A., Houk, C., Ahmed, S. F., & Lee, P. A. (2006). Consensus statement on management of intersex disorders. *Journal of Pediatric Urology, 2*(3), 148-162. doi:http://dx.doi.org/10.1016/j.jpuro.2006.03.004
- Hughes, I. A., Nihoul-Fékété, C., Thomas, B., & Cohen-Kettenis, P. T. (2007). Consequences of the ESPE/LWPES guidelines for diagnosis and treatment of disorders of sex development. *Best Practice & Research Clinical Endocrinology & Metabolism, 21*(3), 351-365. doi:http://dx.doi.org/10.1016/j.beem.2007.06.003
- Hullmann, S. E., Fedele, D. A., Wolfe-Christensen, C., Mullins, L. L., & Wisniewski, A. B. (2011). Differences in adjustment by child developmental stage among caregivers of children with disorders of sex development. *International Journal of Pediatric Endocrinology, 2011*(1), 16. doi:10.1186/1687-9856-2011-16
- Hullmann, S. E., Wolfe-Christensen, C., Ryan, J. L., Fedele, D. A., Rambo, P. L., Chaney, J. M., & Mullins, L. L. (2010). Parental overprotection, perceived child vulnerability, and parenting stress: A cross-illness comparison. *Journal of Clinical Psychology in Medical Settings, 17*(4), 357-365. doi:http://dx.doi.org/10.1007/s10880-010-9213-4

- Karkazis, K., Tamar-Mattis, A., & Kon Alexander, A. (2010). Genital Surgery for Disorders of Sex Development: Implementing a Shared Decision-Making Approach. *Journal of Pediatric Endocrinology and Metabolism*, 23, 789.
- Kazak, A. E., & Barakat, L. P. (1997). Brief Report: Parenting Stress and Quality of Life During Treatment for Childhood Leukemia Predicts Child and Parent Adjustment After Treatment Ends. *Journal of Pediatric Psychology*, 22(5), 749-758.
doi:10.1093/jpepsy/22.5.749
- Kirk, K. D., Fedele, D. A., Wolfe-Christensen, C., Phillips, T. M., Mazur, T., Mullins, L. L., . . . Wisniewski, A. B. (2011). Parenting characteristics of female caregivers of children affected by chronic endocrine conditions: a comparison between disorders of sex development and type 1 diabetes mellitus. *Journal of Pediatric Nursing*, 26(6), e29-36.
doi:10.1016/j.pedn.2010.10.005
- Leslie, L. K., & Boyce, W. T. (1996). Consultation with the specialist. The vulnerable child. *Pediatric Review*, 17(9), 323-326.
- Levy, D. (1931). Maternal overprotection and rejection. *Archives of Neurology and Psychiatry*, 25, 886-889.
- Mullins, L. L., Fuemmeler, B. F., Hoff, A., Chaney, J. M., Van Pelt, J., & Ewing, C. A. (2004). The Relationship of Parental Overprotection and Perceived Child Vulnerability to Depressive Symptomatology in Children With Type 1 Diabetes Mellitus: The Moderating Influence of Parenting Stress. *Children's Health Care*, 33(1), 21-34.
Retrieved from <http://search.ebscohost.com/login.aspx?direct=true&db=aph&AN=12537090&site=ehost-live>
- Mullins, L. L., Wolfe-Christensen, C., Pai, A. L. H., Carpentier, M. Y., Gillaspay, S., Cheek, J., & Page, M. (2007). The relationship of parental overprotection, perceived child vulnerability, and parenting stress to uncertainty in youth with chronic illness. *Journal of Pediatric Psychology*, 32(8), 973-982. doi:<http://dx.doi.org/10.1093/jpepsy/jsm044>
- Nelson, P. A., Caress, A.-L., Glenny, A.-M., & Kirk, S. A. (2012). 'Doing the "Right" Thing': How parents experience and manage decision-making for children's 'Normalising'

- surgeries. *Social Science & Medicine*, 74(5), 796-804.
doi:<http://dx.doi.org/10.1016/j.socscimed.2011.11.024>
- Patterson, J. M., Goetz, D., Budd, J., & Warwick, W. J. (1993). Family Correlates of a 10-Year Pulmonary Health Trend in Cystic Fibrosis. *Pediatrics*, 91(2), 383-389. Retrieved from <http://pediatrics.aappublications.org/content/pediatrics/91/2/383.full.pdf>
- Power, T. G., Dahlquist, L. M., Thompson, S. M., & Warren, R. (2003). Interactions Between Children With Juvenile Rheumatoid Arthritis and Their Mothers. *Journal of Pediatric Psychology*, 28(3), 213-221. doi:10.1093/jpepsy/jsg009
- Sanders, C., Carter, B., & Goodacre, L. (2011). Searching for harmony: Parents' narratives about their child's genital ambiguity and reconstructive genital surgeries in childhood. *Journal of Advanced Nursing*, 67(10), 2220-2230. doi:<http://dx.doi.org/10.1111/j.1365-2648.2011.05617.x>
- Sanders, C., Carter, B., & Goodacre, L. (2012). Parents need to protect: influences, risks and tensions for parents of prepubertal children born with ambiguous genitalia. *Journal of Clinical Nursing*, 21(21-22), 3315-3323. doi:10.1111/j.1365-2702.2012.04109.x
- Spurrier, N. J., Sawyer, M. G., Staugas, R., Martin, A. J., Kennedy, D., & Streiner, D. L. (2000). Association between parental perception of children's vulnerability to illness and management of children's asthma. *Pediatric Pulmonology*, 29(2), 88-93.
doi:10.1002/(SICI)1099-0496(200002)29:2<88::AID-PPUL2>3.0.CO;2-D
- Thomasgard, M., & Metz, W. P. (1997). Parental overprotection and its relation to perceived child vulnerability. *American Journal of Orthopsychiatry*, 67(2), 330-335.
doi:10.1037/h0080237
- Wiesemann, C., Ude-Koeller, S., Sinnecker, G. H. G., & Thyen, U. (2009). Ethical principles and recommendations for the medical management of differences of sex development (DSD)/intersex in children and adolescents. *European Journal of Pediatrics*, 169(6), 671-679. doi:10.1007/s00431-009-1086-x

- Williams, P. G., Holmbeck, G. N., & Greenley, R. N. (2002). Adolescent health psychology. *Journal of Consulting and Clinical Psychology, 70*(3), 828-842. doi:10.1037/0022-006X.70.3.828
- Wolfe-Christensen, C., Fedele, D. A., Kirk, K., Mullins, L. L., Lakshmanan, Y., & Wisniewski, A. B. (2014). Caregivers of children with a disorder of sex development: associations between parenting capacities and psychological distress. *Journal of Pediatric Urology, 10*(3), 538-543. doi:10.1016/j.jpuro.2013.11.016
- Wolfe-Christensen, C., Fedele, D. A., Kirk, K., Phillips, T. M., Mazur, T., Mullins, L. L., Wisniewski, A. B. (2012). Degree of external genital malformation at birth in children with a disorder of sex development and subsequent caregiver distress. *Journal of Urology 188*(4 Suppl), 1596-1600. doi:10.1016/j.juro.2012.02.040
- Zeiler, K., & Wickstrom, A. (2009). Why do 'we' perform surgery on newborn intersexed children?: The phenomenology of the parental experience of having a child with intersex anatomies. *Feminist Theory, 10*(3), 359-377.
doi:<http://dx.doi.org/10.1177/1464700109343258>