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FACTORS ASSOCIATED WITH PARENT DEPRESSIVE SYMPTOMS AND FAMILY QUALITY OF LIFE IN FAMILIES WITH AND WITHOUT ADOLESCENTS AND YOUNG ADULTS WITH SPINA BIFIDA

By

Monique Ridosh

A Thesis Submitted in

Partial Fulfillment of the

Requirements for the Degree of

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August 2014

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ABSTRACT
FACTORS ASSOCIATED WITH PARENT DEPRESSIVE SYMPTOMS AND FAMILY QUALITY OF LIFE IN FAMILIES WITH AND WITHOUT ADOLESCENTS AND YOUNG ADULTS WITH SPINA BIFIDA

by

Monique Ridosh

The University of Wisconsin-Milwaukee, 2014
Under the Supervision of Professor Kathleen Sawin

The purpose of this study was to explore which context and process factors contribute to parent depressive symptoms (PDS) and family quality of life (FQOL) in families with adolescents/young adults (AYA) with and without spina bifida (SB). Secondary analysis was conducted on data (N = 209) from a multi-site cross-sectional study of adaptation in AYA with SB. Measures included: Behavior Rating Inventory of Executive Function (Behavioral Regulation Index and Metacognition Index), FACES III (Cohesion subscale), Family APGAR, Family Inventory of Resources for Management (Family Mastery and Health subscale), a single-item measure of stress, Beck Depression Inventory (BDI-II) and The FQOL Scale. Descriptive statistics, hierarchical multiple regression and Sobel test for mediation were used for the analysis. Cronbach's alphas ranged from 0.80 - 0.97. Fifty-four percent of the parents had an AYA with SB, 86% parents were Caucasian, 19% experienced depressive symptoms and the average age of the AYA was 15.2 years. Income, family resources and parent stress but not presence of SB explained 38% of the variance of PDS. Presence of SB, family satisfaction, parent stress and PDS explained 49% of the variance of FQOL. PDS partially mediated the relationship of family resources and FQOL. Further exploratory analysis indicated that in parents of AYA with SB, family satisfaction and PDS explained 47% of the variance of FQOL. In the

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comparison group, family resources and parent stress explained 49% of the variance of FQOL. It is important for health care providers to screen parents for PDS, address effective use of family resources, and implement strategies to reduce stress. Attention to FQOL in families who have an AYA with SB is particularly important. Further research is needed to identify other factors that contribute to PDS and FQOL.

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I dedicate this work to my family, friends and colleagues.
To my son for reminding me always of the gift of being his mother. To my husband for his commitment to our family, enduring love and support. To my parents for forming me into the mother, wife, daughter, sister, friend and nurse that I am today.

I would like to thank my Major Professor, Dr. Kathleen Sawin for her years of commitment, guiding my learning to inquire and find meaning through family research. I also wish to acknowledge Dr. Gayle Roux for her mentorship and positioning me to grow my inner strength in the process of seeking new knowledge. I would like to acknowledge all committee members for their contributions of time, expertise, guidance and support to facilitate my learning and the dissemination of this work.

To my friends and colleagues, I thank you for your continuous encouragement, acceptance and support.

You have all contributed to my Family Quality of Life.

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Chapter 1

Caring for a child with a chronic health condition is a life changing experience for families. Chronic health conditions affect individuals and families in ways that alter their daily lives. While families endeavor to adapt some do better than others. Throughout their lives, adaptation is a dynamic state of being. For those families who poorly adapt, the health of the individual and family are at increased risk for complications and other conditions. While caregiving demands of a child with a chronic health condition (CHC) have been linked to physical and mental health of caregivers (Raina et al., 2005), needs of parents are typically unaddressed in our current health system and literature. Reimbursement mechanisms are primarily directed to care of individuals with disease diagnoses while beginning to allocate a portion of funds for health promotion and prevention of illness (The Patient Protection and Affordable Care Act, 2010). The experience of having a child with a chronic condition changes the way that parents perceive their life situation. What the family identifies as important may affect how they live their lives and how they maintain their health and the health of their child.

Children with CHC include children with special health care needs ". . . who have or are at increased risk for a chronic physical, developmental, behavioral, or emotional condition and who also require health and related services of a type or amount beyond that required by children generally" (McPherson et al., 1998, p. 138; Newacheck, Rising, & Kim, 2006; van der Lee, Mokkink, Grootenhuis, Heymans, & Offringa, 2007). In the US, approximately 10 million children live with a CHC (National Survey of Children's Health, 2007). As science advances in the care of CHCs, children live in more complex states of health under the care of their parents.

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Spina bifida (SB) is the complex CHC under study as an exemplar. Prevalence of SB in children and adolescents 0 – 19 years old in the US is estimated at 3.1 cases in 10,000, about 24,860 in 2002 (Shin et al., 2010). SB results from a neural tube malformation during early stages of fetal development. The secondary conditions of SB include physical mobility impairment, neuropsychological deficits, bladder and bowel dysfunction, and social competence difficulty. These secondary conditions affect the individual, family, and community across the life course. Survival of youth with SB has improved with advances in care (Davis et al., 2005) resulting in a higher incidence of adolescents transitioning to adulthood. Many young adults continue to receive care from childhood neurology clinic providers into their mid-twenties (Ridosh, Braun, Roux, Bellin, & Sawin, 2011). Caregivers experience increased burden while caring for the child, adolescent, and young adult with a chronic condition impacting their own physical and mental health (Grosse, Flores, Ouyang, Robbins, & Tilford, 2009; Raina et al., 2005; Valença, de Menezes, Calado, & de Aguiar Cavalcanti, 2012). Understanding factors that contribute to their family's quality of life may help to prevent burden of secondary conditions on the individual, family, and society.

Conceptual Framework

Two conceptual frameworks were used to develop a general conceptual orientation of factors relevant to families with a child with CHC. The two frameworks were the Transactional Stress and Coping Model (Thompson & Gustafson, 1996) and the Ecological Model of Secondary Conditions (Sawin, Buran, Brei, & Fastenau, 2003). The Transactional Stress and Coping Model refers to maternal meditational processes of stress, coping and family functioning and outcomes of maternal and child adjustment

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(Thompson & Gustafson, 1996). Family members and the family unit strive to adapt to the stress of living with chronic conditions (Thompson, Gil, Burbach, Keith, & Kinney, 1993). This model explains factors related to adaptation in families with children with sickle cell disease, insulin-dependent diabetes mellitus and in families with children with chronic conditions compared to those without condition (Hocking & Lochman, 2005; McClellan & Cohen, 2007). Assumptions of the model are that cognitive processes of stress appraisal and expectations of efficacy of locus of control, methods of coping, and supportive, conflicted or controlling family functioning patterns of the individual and family have an impact on adaptation more so than severity of illness or socioeconomic status (Thompson & Gustafson, 1996). The model delineates the outcomes of maternal adjustment and child adjustment as related.

The second model that influenced the general conceptual orientation was the Ecological Model of Secondary Conditions (Sawin et al., 2003). This model includes risk factors and protective processes associated with adaptation of adolescents with CHCs. Three risk or context factors: condition-specific (e.g. severity of condition), demographic (e.g. age, gender, socio-economic status), and neuropsychological (e.g. executive functioning) and three protective processes adolescent/young adult (AYA) resilience (e.g. future expectations), family resourcefulness (e.g. satisfaction) and perceived health-care adequacy (family centered care) explain relationships with adaptation outcomes (e.g. physical health, mental health, and quality of life outcomes) for adolescents.

Where these models intersect are in identifying context (demographic, condition) and processes (stress appraisal, coping, family functioning/satisfaction) related to outcomes, mental health and quality of life outcomes. Context was defined as the

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environment in which parental adaptation outcomes occur. Process was defined as the perceptions and activities that lead to parental adaptation outcomes. Outcomes were defined as the result of the process. In this study, parent depressive symptoms (PDS) and family quality of life (FQOL) were the adaptation outcomes of interest. The context factor categories derived from the models were demographic, condition, and child factors (parent perception of executive functioning). The processes were family functioning and stress as a parent factor. PDS were a proximal outcome and the distal outcome was FQOL. A theoretical framework of factors related to the outcomes was generated from two reviews of literature.

A secondary analysis was possible using an existing dataset of a study of secondary conditions and adaptation in AYA with SB. The Ecological Model of Secondary Conditions grounded the primary study. The integration of the two models provided the foundation for organization of concepts in the literature and generated hypotheses.

Purpose

The aim of this study is to explore which context and process factors contribute to PDS and FQOL in families with adolescents/young adults (AYA) with and without a chronic health condition (CHC), specifically spina bifida (SB). A measurement model was derived from the theoretical framework of factors related to outcomes and available data. See Figure 1 for measurement model. This study will advance science by (1) identification of factors related to PDS and FQOL from a large multi-site United States sample, (2) identification of a possible mediator of FQOL, (3) identification of factors

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related to outcomes by subsamples with SB and comparison, and (4) evaluation of an overall global measure of FQOL using a 3-item scale.

The Primary Study: Secondary Conditions and Adaptation in Spina Bifida

The primary study tested the Ecological Model of Secondary Conditions conceptual framework in adolescents and young adults (AYA) with SB. The model proposed that three risk factors (demographic characteristics, neurological severity, and neuropsychological deficits) and three protective processes (adolescent resilience, family resourcefulness and health care adequacy) were predictors of secondary conditions and adaptation outcomes in AYA (i.e., physical health, mental health, social competency, health-related quality of life, and academic achievement). Demographic characteristics included age, gender and socioeconomic status. Adolescent resilience variables included decision-making, responsibility, attitude, hope, coping, sexuality beliefs, communication efficacy, and future expectations. Family resourcefulness included cohesion, satisfaction, level of protection, mastery, and family activity. Perceived health-care adequacy included SB needs and family centered care. According to the model, neuropsychological (NP) deficits mediated the impact of neurological severity (level of lesion, hydrocephalus status, and neurological complications) on outcomes. The primary study sample from multiple sites included 112 parents of AYA with SB and 97 parents of AYA without SB. Teachers were asked to provide school and behavioral data. Data were collected by interviews of parents and AYA, neuropsychological (educational) testing, and mailed information from the adolescent's teacher. Experienced and trained health professionals conducted interviews via telephone.

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Background

The nursing discipline is concerned with the interaction of the concepts of person, health, environment and nursing (Fawcett, 1978). This interaction occurs at multiple levels and systems to include the individual, family, community, and population. Nursing is concerned by nature of its social contract with factors society values including physical, emotional, and spiritual health and well-being. As the body of knowledge in nursing evolves to meet changing societal needs, development of theory through research to guide practice is needed to promote health and well-being of families. Specifically, addressing the needs of families with children with CHC will advance the nursing disciplines' body of knowledge to fit the needs of society. Knowledge development to understand both PDS and FQOL and the factors contributing to them will add to family science.

Care of the family includes addressing the well-being of its members. Parents of children, specifically parents of adolescents with and without SB are the focus of the current study. In parents of adolescents generally, up to 40% struggle with lower self-esteem, lower life satisfaction, higher anxiety and depression (Steinberg, 2001). Since there was an abundance of literature in the general category of depressive symptoms, the review of literature was limited to families with children with SB. FQOL in families with children with SB was only evaluated in two studies therefore review of this body of literature was expanded to families with children with any CHC. The following will define and describe both outcome variables for the current study, PDS and parent perception of FQOL. The outcome variables will be further explained in chapters two and three manuscripts, which synthesize the literature on these two outcomes.

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Parent Depressive Symptoms

An estimated one in 10 adults in the US suffers from current depression, 9.1% in 2006 - 2008 (Centers for Disease Control and Prevention, 2010). It is estimated this burden extends to at least 15 million children who live with a depressed parent (Ertel, Rich-Edwards, & Koenen, 2011; National Research Council and Institute of Medicine, 2009). The impact of depression reaches beyond the individual to familial and societal concerns that are multigenerational and universal.

Depression is the presence and severity of different symptoms of depression to include sadness, loss of interest or pleasure, feelings of guilt or low self-worth, disturbed sleep or appetite, feelings of tiredness, and poor concentration (Marcus et al., 2012, p. 6). PDS are the specific symptoms that characterize depression in parents, number and severity of symptoms can be minimal, mild, moderate or severe. PDS are an important concept for parents affecting their worldview. Psychological distress, more broadly addressed a range of symptoms including anxiety, phobia, paranoid ideation and psychosis. The variety of measures for psychological distress in the literature made it difficult to determine severity and compare symptomatology across studies for synthesis. Although measuring psychological distress more broadly identifies range of symptoms, measuring PDS more specifically is a pragmatic indicator of mental health outcome clinically relevant to evaluate and treat. The current study will address PDS as an adaptation outcome.

Although research of adult clinical depression is abundant, a specific focus on 'parents' of children with complex chronic health conditions such as spina bifida was limited. The literature available did include a comprehensive review on the relationships

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between parenting, parent depressive symptoms and child health outcomes (National Research Council and Institute of Medicine, 2009). A review of earlier literature of the concept of PDS found that the broader concept of psychological distress was examined in families with children with SB prior to 2005. More recently, a focus on depressive symptoms was noted as a response to the shift in the way depression was diagnosed. Factors related to each of these concepts (psychological distress and PDS) are identified in the review of literature. Discussion of concepts and measures are found in chapter 2.

While studies were limited in the review of literature of PDS in parents of children with SB, up to 48% of parents experienced depressive symptoms. A review of literature of PDS identified 32-67% of psychological distress and PDS were explained by similar context factors (demographic factors, presence and severity of SB, and child factors) and process factors (family functioning and parent factors such as stress and coping) (Ridosh, Sawin, & Klein-Tasman, 2014). Furthermore, while these context variables were important they were not sufficient alone to explain depressive symptoms. The process variables (family functioning, parent stress and coping) contributed a greater amount of variance in PDS (Ridosh, Sawin, & Klein-Tasman, 2014). Concepts were identified in the literature review and described in chapter 2. In addition, there is some evidence that parents of children with SB have more PDS than those without a chronic health condition (Ridosh, Sawin, & Klein-Tasman, 2014).

Family Quality of Life

Research on FQOL is in early stages of theory development. FQOL is being studied in the disciplines of psychology, education, and nursing. There is a growing body of evidence in FQOL focusing on individuals with intellectual disability. Knowledge

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from the discipline of nursing can inform inquiry to establish valid and reliable measures of FQOL in the family experience of living with a member with a chronic health condition. The definition of FQOL for this study was generated from the review of the literature in families with children with a CHC.

The literature revealed two conceptualizations of FQOL: overall global FQOL and domain-specific FQOL. Domains included family relationships, family interaction, parenting, influence of values, health, careers, community, support from services, support from others, disability-related support, leisure, finances, physical material well-being, and emotional well-being (Ridosh, Sawin, & Schiffman, 2014). Further two types of overall FQOL were identified. One was the summary of specific domains and the other was a global "gestalt" of FQOL. Domain-specific conceptualizations of FQOL are useful to researchers across disciplines to understand the specific components of FQOL that "make up" FQOL. The overall global concept was found to be helpful in identifying a person's individualized evaluation of FQOL weighted by the factors important to the individual. For this study, FQOL is defined as an overall appraisal of the domains of life important to the family. The distal adaptation outcome for the current study was overall global FQOL.

Measurement of FQOL is emerging. It has been measured mostly in families with a member with intellectual disability, scarcely measured in the context of complex health conditions such as cancer and spina bifida (Mellon, 2002; Mellon & Northouse, 2001; Ridosh, Sawin, & Brei, 2013; Sawin, Brei, Buran, & Fastenau, 2002). Various measures of FQOL, including scales or subscales scores, various dimensions of satisfaction and/or importance and attainment scores, and overall global measures have been used across studies. Reporting of overall FQOL, whether by a sum of domain-specific scales or a

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global single item enabled synthesis of findings. Understanding of the various units of measurement provides different perspectives of FQOL conceptualization. Researchers have begun to study FQOL by gathering data from individuals and their family members.

Two domain specific instruments are used in the literature. An overall score is addressed by the sum of the domains. The first instrument, the Beach FQOL Scale was developed as a tool to assess family outcomes in families with children with developmental disabilities by measuring domains of family life (Hoffman, Marquis, Poston, Summers, & Turnbull, 2006). Domains include family interaction, parenting, emotional well-being, physical/material well-being and disability-related support (Hoffman et al., 2006). An international research initiative developed the second instrument, FQOL Survey-2006 (Werner et al., 2009). This instrument assesses family outcomes in families with a member with intellectual disability in domains of family life including health, financial well-being, family relationships, support from others, support from services, influence of values, careers, leisure and recreation, and community integration in the context of importance, opportunities, initiative, attainment, stability, and satisfaction (Brown et al., 2006). One group of researchers supported an overall FQOL-2006 latent construct, where each domain loaded onto the second order factor (Isaacs et al., 2012). The instrument also includes two single item global measures, one a measure of overall FQOL and the second a measure of satisfaction with FQOL.

Others have used a series of similar single items as a global measure of overall individual and FQOL. These investigators asked the parent to describe their adolescents' quality of life, their own quality of life and their FQOL (Sawin, Brei, Stevens, Neufeld, &

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Buran, 2006). For this study, an overall global measure of FQOL was proposed using these three questions in combination.

A review of the literature revealed that demographic variables (income, service adequacy, waiver status), severity of condition, and child factors (child behavior problems, future expectations, neuropsychological functioning) were related to FQOL (Ridosh, Sawin, & Schiffman, 2014). In the studies that addressed process variables, family functioning were most predictive of FQOL. Demographic, condition, child factors (context), family functioning and parent stress (processes) were consistently predictive of FQOL in families with children with a CHC (Ridosh, Sawin, & Schiffman, 2014). Although family-professional partnership (family functioning) mediated the relationship of service adequacy and FQOL in one study (Summers et al., 2007), demographic, child and parent factors also accounted for portions of variance in FQOL. The literature review presented in chapter 3 will describe the difference between these conceptualizations and their measure and identify factors related to FQOL.

Research Question and Hypotheses

The following is the proposed research question: What are the context and process factors related to PDS and FQOL in families who have adolescents with and without SB? The research hypotheses include the following:

H_0 1. The context factors (demographic [child age, income, parent gender, race], presence of SB, child [parent perception of executive function]), process factors (family functioning [cohesion, satisfaction, resources], parent stress), delineated in the measurement model are related to the proximal outcome (PDS);

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H_0 2. The context factors (demographic [child age, income, parent gender, race, ethnicity], presence of SB, child [parent perception of executive function]), process factors (family functioning [cohesion, satisfaction, resources], parent stress), and proximal outcome (PDS) delineated in the measurement model are related to the distal outcome of FQOL;

H_0 3. Depressive symptoms mediate the relationship of context and process factors to FQOL.

If the context variable presence of SB is significant in the multiple regression analysis, exploratory analysis will be conducted to determine which context and process factors contribute to PDS and FQOL in families who have adolescents with and without SB.

Current Study: Secondary analysis

The current study used secondary analysis to explain parent outcomes, PDS and FQOL in parents of AYA with and without SB. Variables included in the measurement model were limited to those available in the database in both groups, with and without SB and had empirical support.

The design of the study was a descriptive, correlational secondary analysis. Preliminary analysis used correlations to determine which context and process factors were related to PDS and FQOL and supported selection of factors that were included in hierarchical multiple regression in the total sample of parents ($N = 209$) with AYA 12 – 21 years old. Regression analysis tested the relationship between possible independent variables (child age, income, parent gender, race, ethnicity, SB presence, parent perception of executive function, family functioning and parent stress) and dependent variables (PDS and FQOL). Relationship of independent variables with PDS and then

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PDS with FQOL had to be significant to test for mediation. The selection of the number of variables entered in the equation depended on correlations, power analysis, and conceptual fit based on the theoretical framework guiding the study. The Sobel test was used to determine mediation. The Sobel test is used when there is one mediator, one independent variable and one outcome variable to estimate the direct effect on the outcome that is mediated by the independent variable (Dudley & Benuzillo, 2004). Additional exploratory analyses were done when a significant difference was noted between parents with AYA with and without SB to explore which context and process factors contributed to FQOL in these two groups. Two different regression analyses were conducted to determine if there were different patterns of factors related to outcomes. Methods of the current study are further described in Chapter 4. The following describes the conceptual definitions of the context, process, and outcomes proposed for current study based on available data from primary study.

Conceptual Definitions

Context

Demographic. Demographic data will include child age, income, parent gender, race, and ethnicity as variables.

- Child age, the length of time that a person has lived in number of years, serves as an indicator of developmental stage;
- Income, combined family income serves as a proxy for socioeconomic status and access to resources;
- Parent gender, the state of being male or female who may have different gender-based perspectives;

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- Race, category of group of people who self-identify as part of group based on place of origin. Categories include Black, Caucasian, American Indian, Other (specify), racial group may share genetic and/or health risk factors;
- Ethnicity, a group of people sharing the same culture regardless of race categorized as Hispanic or not Hispanic, this group of persons may share health beliefs and behaviors. A two question format was used for race and ethnicity reporting (Race and Ethnic Standards for Federal Statistics and Administrative Reporting, 1997)

Condition factor. Presence of SB was a variable to identify AYA with and without SB. AYA, either had diagnosis of SB, a complex CHC or had no major medical conditions.

Child factor. Parent perception of executive functioning (EF) will be an indicator of a component of child neuropsychological functioning. Executive function is "a collection of related yet distinct abilities that provide for intentional, goal-directed, problem-solving action" (Gioia & Isquith, 2004, p. 138). Specifically, the indicator will reflect inhibition, mental flexibility, and emotional control necessary for effective functioning.

Process

Family functioning. Family functioning is defined as the attributes of a family system that characterize how they operate or behave (McCubbin & McCubbin, 1987). Family cohesion, satisfaction and resources were considered central family functioning concepts for this study.

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- Family Cohesion. Family cohesion is an indicator of emotional bonding and the degree of individual autonomy among family members (Olson, 1986);
- Family satisfaction. Family satisfaction is an indicator of family functioning by measuring the individual's satisfaction with family adaptation, partnership, growth, affection, and resolve (Austin & Huberty, 1989);
- Family Resources. Resources are an indicator of mastery over family events, family support resources, family esteem, and communication (McCubbin, Comeau, & Harkins, 1981);

Parent factor. Stress is an overall appraisal process in which perception of demands exceed resources in the relationship between person and environment. Stress can be acute, intermittent, or chronic and can contribute in the short term to a state of balance yet when prolonged can be damaging physiologically in the long term (Lazarus & Folkman, 1984; McEwen, 1998).

Adaptation Outcomes

For this study, adaptation outcomes are defined as the proximal outcome of PDS and distal outcome of FQOL.

Parent depressive symptoms. PDS are the symptoms of depression present in the last 2 weeks and severity of different symptoms of depression. "Depression is a common mental disorder, characterized by sadness, loss of interest or pleasure, feelings of guilt or low self-worth, disturbed sleep or appetite, feelings of tiredness, and poor concentration" (Marcus et al., 2012, p. 6).

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Family quality of life (FQOL). The definition of FQOL for this study was an overall appraisal of the domains of life important to the family.

Orientation to the Dissertation

The following chapters in the dissertation, *Factors Associated with Parent Depressive Symptoms and Family Quality of Life in Families with and without Adolescents and Young Adults with Spina Bifida* will outline literature related to PDS and FQOL, findings of a descriptive correlational study exploring factors related to PDS and FQOL, and discuss implications for practice, research and policy. Three manuscripts are included as part of the final dissertation.

Chapter two includes the first manuscript, *Depressive Symptoms in Parents of Children with Spina Bifida: A review of the literature* synthesizes findings of factors related to PDS. This review is limited to studies that include parents of children with spina bifida (SB). Prevalence of PDS and specific context and process factors known to explain variance in PDS are identified.

Chapter three includes the second manuscript, *Family Quality of Life in Families of Children with a Chronic Health Condition: A review of the literature* addresses FQOL and includes factors related to FQOL in families with children more broadly. Specific context and process factors known to explain variance of FQOL in parents of children with CHC are identified.

Both of the manuscripts review findings and are organized by context, process, and outcome. PDS are considered a proximal outcome in the proposed study and FQOL is a distal outcome. Following a review and critique of the literature a theoretical framework of FQOL is proposed.

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Chapter 4 includes the third manuscript, *Factors Associated with Parent Depressive Symptoms and Family Quality of Life in Families with and without Adolescents and Young Adults with Spina Bifida,* a data-based article of results of the study. This manuscript includes procedures specific to secondary analysis in evaluation of missing values and findings from regression and mediation analyses. A discussion of the findings as well as implications for practice and research is included.

Chapter 5 synthesizes implications for theory, practice, research, and policy. Practice implications highlight levels of prevention and recommendations for early detection, screening and treatment of parents at risk for depression in primary care. Future research trajectory to build the science of FQOL is suggested to include use and testing of new measure of FQOL in addition to identification of other related factors not yet studied. Policy recommendations are based on current affordable care legislation, US Preventive Services Task Force guidelines and leveraging existing resources.

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Chapter 2

Depressive Symptoms in Parents of Children with Spina Bifida: A review of the literature

Abstract

Purpose. The purpose of this review was to synthesize the literature on depressive symptoms in parents of children with spina bifida.

Design and Methods. A search was conducted using databases (CINAHL, MEDLINE, and PsycINFO). Fifteen studies were identified that met inclusion and exclusion criteria.

Findings. This review identified both: (a) a high prevalence of parental depressive symptoms (PDS); and (b) specific factors, demographic, condition, and child factors, family functioning and parent factors explained 32-67% of parent depressive symptoms (PDS).

Conclusions. Although context factors were important, they alone were not sufficient to explain PDS. Process factors contributed more variance in PDS. This body of literature was limited by a lower level of evidence, small number of studies, and overall internal and external validity issues.

Clinical Relevance. Although a portion of variance remains unexplained, findings warrant implementation of parent depression screening in families with children with spina bifida. This review identified factors related to PDS and highlighted gaps in the literature to guide future research of families with children with chronic conditions.

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Parents of children with a chronic health condition (CHC) face substantial challenges in managing their child's condition, dealing with everyday life, and promoting the health of all family members. These challenges can put parents at higher risk for negative physical and mental health outcomes. For example, parents of children with asthma are at risk for depressive symptoms because of associated poverty, child behavior problems, poor emotional support, and poor family functioning (Tu, Perreault, Seguin, & Gauvin, 2011). In parents of children with epilepsy, family income, child behavior problems and family satisfaction likewise predict parental depressive symptoms (Shore, Austin, Huster, & Dunn, 2002).

Parents of children with spina bifida (SB) may be particularly at risk for depression due to increased care demands. These parents, caring for a child with this neurological condition, which has multisystem involvement, have "a long complicated journey" (Sawin & Thompson, 2009, p. 284). Families experience limitation in social interactions and stigma linked to child bowel and bladder continence, neuropsychological deficits, and physical mobility impairments.

Mental health outcomes have been a concern of investigators studying families with SB for over 20 years. In the earlier literature (before 2005), the focus was on a broad concept, psychological distress (PDISS). More recently, the literature has transitioned to address parental depressive symptoms (PDS), a more pragmatic concept for screening, evaluation and treatment. The change in diagnostic criteria in the DSM IV for depression led to the emergence of more specific measures of PDS. The new criteria were published in 1996. However, they were not adopted by many clinicians and researchers until the early 2000s. Although a meta-analysis that summarized the prevalence of PDISS and the

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factors related to them was published in 2005 (Vermaes et al., 2005), no other review has been conducted since the shift to PDS. It is important to determine if the prevalence of negative mental health outcomes and the factors associated with these outcomes have changed since this conceptual shift. Additionally, a synthesis of the early and later literature would help us understand why some parents of children with SB adapt well to these challenges and others experience depressive symptoms.

Background

Depression, a global public health issue, is a leading cause of disability affecting an estimated 350 million worldwide (Marcus et al., 2012). Preventing depression is an initiative of the World Health Organization. This initiative addresses vulnerabilities and risk factors for mental health and well-being across the life course (Marcus et al., 2012). Depression affects the individual, family and society—its impact is multigenerational. One in 10 mothers in the United States (US) suffer from depression and mothers with depression were more likely to be unemployed or earn low income, less educated, single and less than 35 years old (Ertel, Rich-Edwards & Koenen, 2011). About half of these women received services for depression. Blacks experienced more adversity and Whites had more comorbid conditions (Ertel et al., 2011). Highlighted in a review of depression in parents, parenting, and children were the multigenerational challenges of parent-child relationships, parent adversity and comorbidities of substance abuse or trauma, and physical and mental health treatment of families (parent and child) (National Research Council and Institute of Medicine, 2009). The review by the National Research Council and Institute of Medicine (2009) recommended a need to identify depressed parents and

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support a national prevention strategy of parent depression and adverse outcomes of children.

While depressive symptoms have been explored in parents generally, they have not been well addressed in parents with a child with a chronic health condition (CHC) who face complex demands in their everyday lives. Parents of children with CHC may have distinct risk and protective factors remaining undefined. The negative impact of symptoms of depression in parents are associated with childhood health outcomes such as delays in growth (Surkan, Kennedy, Hurley, & Black, 2011), child neuropsychological and behavior functioning problems (Ashman, Dawson, & Panagiotides, 2008), and psychopathology (Weissman et al., 2006). Presence of parent depressive symptoms and lack of treatment is associated with increased prevalence of child behavior problems and greater risk of depressive symptoms in children creating a cycle of poor health outcomes for the entire family. It is estimated that this burden extends to at least 15 million children who live with a depressed parent (Ertel et al., 2011; National Research Council and Institute of Medicine, 2009). When PDS is present in families of children with a CHC, the impact can be seen in the health of the parent, child and family. Although the presence of the CHC itself may not be directly related with physical and mental health outcomes, other factors are associated with adaptation of the family when a CHC is present. SB, a complex CHC with multiple comorbidities, which typically require a high level of parental care and involvement, is a suitable exemplar.

Prevalence of SB in children and adolescents 0 – 19 years old in the US is estimated at 3.1 cases in 10,000 (Shin et al., 2010). SB results from a neural tube malformation during early stages of fetal development. Parenting a child with SB

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includes challenges of child's learning difficulties due to impairments in working memory, numeral literacy, verbal communication and problem solving abilities. Significant impact on independence and social integration in society is evident for the individual, family, and community across the life course. Parents, as primary caregivers, experience increased burden while caring for the child, adolescent, and young adult with a chronic condition impacting their own physical and mental health (Grosse, Flores, Ouyang, Robbins, & Tilford, 2009; Raina et al., 2005; Valença, de Menezes, Calado, & de Aguiar Cavalcanti, 2012).

Recent reviews of psychosocial outcomes in parents of children with SB focused on family functioning and social adjustment (Holmbeck & Devine, 2010; Holmbeck, Greenley, Coakley, Greco, & Hagstrom, 2006). The presence of depressive symptoms in these parents, initially conceptualized as psychological distress and more recently specifically by parent depressive symptoms, is not well understood in this population. The purpose of this review is to synthesize the literature on depressive symptoms in parents of children with SB, specifically addressing the questions (a) what is the prevalence of parent depressive symptoms conceptualized as either psychological distress (PDISS) or parental depressive symptoms (PDS), and (b) what are the factors related to PDS?

Two theoretical models influenced the overall conceptual approach (e.g., concept, process and outcome) that guided this review of PDS, the Ecological Model of Secondary Conditions and Adaptation in SB (Sawin, Buran, Brei, & Fastenau, 2003) and the Transactional Stress and Coping Model (Thompson & Gustafson, 1996). Context is defined as the environment in which parental adaptation outcomes occur. Context factors

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are specific to the child, condition and demographic characteristics such as gender and SES. Process is defined as the perceptions and activities that lead to parental adaptation outcomes. Process factors include those variables specific to family process such as stress appraisal, coping and family functioning. Adaptation outcomes are defined as the result of the process and include mental health outcomes, specifically depressive symptoms.

The Ecological Model of Secondary Conditions and Adaptation in SB includes context factors (risk), protective processes and adaptation outcomes in adolescents with SB. The basic structure of the model delineating relationships between the context and process factors to adaptation outcomes such as physical, mental, and quality of life outcomes in adolescent/young adults (AYA) with SB is also useful in understanding parent outcomes. In the Transactional Stress and Coping Model, managing stress, coping and family functioning are maternal mediational processes. This model delineated the factors related to two outcomes, maternal and child adjustment (Thompson & Gustafson, 1996). Thus, this review was organized by the general conceptual categories of context, process, and outcomes, specifically.

Design and Methods

Primary research studies were located in the following steps. First an initial search was conducted in CINAHL, MEDLINE, and PsycINFO databases using combination of keywords “parent*”, “depress*”, and “spina*”. Inclusion criteria were studies published after 1990, English language, peer reviewed articles, and pertaining to parent depression outcome and spina bifida. Search terms “myelo”, “distress”, and measures (BDI, CES-D, and SCL-90-R) did not yield any additional articles. The initial search yielded 27 records. Review of a recent unpublished study and a manual search of references, yielded another

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15 studies. Abstracts of 42 articles were reviewed and 15 articles met the inclusion criteria. Excluded from the sample were articles addressing child outcomes, intellectual disability, spinal cord injury, and CHC other than SB. Review articles addressing related concepts (family functioning, psychosocial adjustment of the child) were omitted since their focus was on family functioning of the child (Holmbeck & Devine, 2010; Holmbeck, Greenley, Coakley, Greco, & Hagstrom, 2006). See Figure 2 for search strategy. The search timeframe was broad to capture the early conceptualization of depressive symptoms as "psychological distress" and the more recent definitive conceptualization of PDS congruent with diagnostic criteria.

This review synthesized findings from 14 primary research studies and one meta-analysis. Seven of 15 studies in the meta-analysis were included in the current review as primary studies. The results of the meta-analysis are reported separately. The meta-analysis addressed psychological adjustment, specifically PDISS. All studies before 2005 with the exception of King, King, Rosenbaum, and Goffin (1999) (examined both PDISS and PDS) used the conceptualization of parental psychological distress (PDISS) and were considered "early" while all following the meta-analysis (Vermaes et al., 2005) specified later findings related to parental depressive symptoms (PDS). Table 2 summarizes prevalence of parental depressive symptoms (measured by PDS and PDISS) in spina bifida and factors related to PDS. Figure 3 summarizes the concepts identified and the number of studies that address each concept.

Results and Discussion

Early (PDISS) and later (PDS) findings in the review are presented by prevalence, factors related to depressive symptoms and a critique of literature addressing design,

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concepts, and instruments. Lastly, gaps in the literature are discussed. An evidence table (see Table 1) summarizes the studies on depressive symptoms in parents of children with SB delineating study authors, year, levels of evidence, study questions, concepts measured, significant findings and strengths/limitation of each study reviewed.

The studies synthesized prior to 2005 in the meta-analysis addressing psychological distress in parents of children with SB had limitations acknowledged by the authors (Vermaes et al., 2005). Inclusion of some of the studies used for the meta-analysis in the current review provides a means to synthesize data with studies conducted more recently to delineate prevalence and factors related to PDS. Comparison of meta-findings (effect sizes) with individual study findings was not possible. Identification of factors with significant relationships contributed to a comprehensive understanding of factors related to PDS.

Meta-analysis of Early Studies

The aim of the meta-analysis by Vermaes, Janssens, Bosman, and Gerris (2005) was to identify if parents of children with SB have more psychological distress than controls, if mothers and fathers differ in their levels of psychological distress, and to delineate which factors correlated with variations in psychological adjustment. Vermaes et al. (2005) provided some evidence of factors related to PDISS in meta-analysis and synthesized literature on parents of children with SB. Mothers of children with SB had .73 standard deviations higher PDISS than comparison group (a medium to large effect size). The data reported in this meta-analysis regarding factors other than parent gender were associations based on one to three studies with similar factors, therefore limited. Effect size *r* was reported and interpreted magnitude as small ($r = 0.1$), medium ($r = 0.3$)

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and large (r = 0.5) effects. Socio-economic variables (race, socioeconomic status (SES), parent education level and employment) combined had a small effect (effect size r = -0.13) on PDISS. This finding illustrates while demographic variables were important, impact was small and limited in specificity to identify risk population not allowing for differentiation of disparate groups. Relevant findings from the meta-analysis were that parents of children with SB, specifically mothers more likely experienced greater PDISS. Family income, SES and condition severity factors had a small effect while child behavior and emotional problems had a moderate to large effects on PDISS. Stress, coping, parenting satisfaction/competence, marital adjustment and positive family environment had moderate to large relationships with PDISS. Quantity of social support and satisfaction with social support had a moderate relationship with PDISS.

Analysis of Primary Studies: Prevalence Depressive Symptoms

Studies addressed depressive symptoms however, no clinical evaluation or confirmation and diagnosis of depression were reported. Criterion for "caseness" of depressive symptoms was only reported in four studies using T-score greater than 63 on Global Severity Index of the SCL-90-R tool (Friedman, Holmbeck, Jandasek, Zukerman, & Abad, 2004; Holmbeck et al., 1997; Kronenberger & Thompson, 1992a, 1992b). Only two other studies reported criteria for clinically relevant depressive symptoms, BDI greater than 10 (Valença et al., 2012) and GCS greater than 30 (Brei, Woodrome, Fastenau, Sawin, & Buran, 2013). More than half of the studies found PDS ranged from 14 - 48 % (see Table 1). The early studies measuring PDISS and the later studies measuring PDS reported similar prevalence rates of depressive symptoms (from 19-44% and 19-48% respectively). Only one of the studies (Hobdell, 2004) found an overall

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prevalence rate of distress or PDS less than 19% and four studies (Brei et al., 2013; Kronenberger & Thompson, 1992a, 1992b; Valença et al., 2012) 44% or higher. A pattern of lower rates of depressive symptoms 14 – 25% was noted in the few studies examining parents of children less than 9 years old. Most studies had a wide age range (2 months – 18 years) and generally did not report relationship of age of the child to PDS.

Analysis of Primary Studies: Factors Associated with Depressive Symptoms

Context Factors. Context factors associated with depressive symptoms included demographic, condition, and child factors (see Table 2).

Demographic Factors. Several studies identified a significant relationship between gender of parent (Holmbeck et al., 1997; Ulus et al. 2012), SES or race and extent of depressive symptoms (Kronenberger & Thompson, 1992a, 1992b; Barakat & Linney, 1992; Barakat & Linney, 1995; Valença et al., 2012). A study exploring differences between mothers and fathers with and without children with a CHC found fathers experienced more psychological symptoms than mothers and a rate of 25.6% in fathers of a child with SB and 16.3% in fathers of a child without SB (Holmbeck et al., 1997). In the same study, the rate of psychological symptoms for mother of a child with SB was 19.2% compared to 11.1% in mothers of a child without SB. (Holmbeck et al., 1997). In contrast, Ulus et al. (2012) found that mothers of a child with SB experienced significantly greater PDS than fathers. In addition, the factors related to PDS differed with stress and coping related to PDS for fathers and family functioning for mothers.

A few early studies that included race in a block of demographic variables (race, child age, child gender, family SES) found mothers' race was related to PDISS

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(Kronenberger & Thompson, 1992a, 1992b). Race was the only significant demographic variable reported to predict 17 – 22% of the variance in PDISS.

SES alone was rarely related to outcomes but there was some evidence that SES in families with SB was lower than comparison groups (Barakat & Linney, 1992; Barakat & Linney, 1995). Select early and later studies in the US and Brazil found indicators of SES related to PDS (Barakat & Linney, 1995; Valença et al., 2012). The number of family members was a significant predictor of parental distress in one early study (Barakat & Linney, 1995).

Child age was a factor related to PDS in one study in analysis by Grosse, Flores, Ouyang, Robbins, and Tilford (2009) with parents of children with SB ages 0 – 6 years old. Parents reported "feeling blue more than a little of the time", but not in parents of children 7 – 17 years of age (Grosse et al., 2009, p. 577). No other studies included child age as a factor in analysis. About half of the studies included samples of children across all ages groups up to 18 years of age (Grosse et al., 2009; Kronenberger & Thompson, 1992a, 1992b; Lemanek, Jones, & Lieberman, 2000; Ulus et al., 2012; Valença et al., 2012). Only one study specifically focused on AYA, which reported the highest prevalence of PDS (Brei et al., 2013).

Presence of SB. There was some support for the impact of SB on parental outcomes in the small number of studies using SB and comparison samples. One found no impact (Barakat & Linney, 1995) while Holmbeck and colleagues found the presence of SB related to PDISS for fathers (Holmbeck et al., 1997, Friedman et al., 2004) and another found 32% of mothers reported PDS in contrast to 12% of comparison mothers (Grosse et al., 2009). In studies of only families with SB, there was some support for the

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relationship of the severity of SB to outcomes. SB severity was related to PDS in three studies (Grosse et al., 2009; Ok & Kurzrock, 2011; Valença et al., 2012) but not in a fourth (Ulus et al., 2012). However, condition severity was inconsistently defined across studies, which limited the ability to clearly understand the impact of aspects of severity on depressive symptoms. Measures of condition severity found to be related to outcome included number of shunt operations, lesion level, functional disability, mobility, bladder and bowel continence, sensation and bowel movements, number of accidents, abdominal pain from constipation, and laxative use. One study used a composite score of condition severity to include number of shunts and bladder and bowel continence (Brei et al., 2013). Another study chose multiple indicators of severity to include sensation and bowel movements, number of accidents, abdominal pain from constipation, and laxative use (Ok & Kurzrock, 2011).

Child factors. Child behavior problems (BP) were related to PDISS in three studies across all age groups (Friedman et al., 2004; King et al. 1999; Lemanek et al., 2000). Indicators of BP included Conduct Disorder, Hyperactivity Disorder, Emotional Disorder, and Somatization (King et al., 1999), and child internalizing and externalizing problems (Friedman et al., 2004; Lemanek et al., 2000). King et al. (1999) found child BP were the most significant predictor of parent depressive symptoms (largest path coefficient among variables tested).

In the broader CHC literature, child behavior problems were generally measured with the Child Behavior Checklist. Parents of children with other CHC such as asthma (McQuaid, Kopel, & Nassau, 2001), congenital heart disease (Landolt, Ystrom, Stene-Larsen, Holmstrom, & Vollrath, 2013), and sickle cell disease (Thompson, Gil, Burbach,

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Keith, & Kinney, 1993) report more behavior problems in comparison to children without conditions. In one large US epidemiological study, child behavior or emotional problems were related to both maternal and paternal depressive symptoms. (Weitzman, Rosenthal, & Liu 2011). Since much of the literature reports cross-sectional data, it is difficult to evaluate whether unidirectional or bi-directional relationships exist between child behavior problems and parent depressive symptoms. However, two longitudinal studies show child behavior problems at earlier time point predict later maternal depressive symptoms (Friedman et al., 2004; Landolt et al., 2013), suggesting causal relationship.

Finally, receptive language, mental processing speed, oculomotor skills, executive functioning, and fine motor skills were components of neuropsychological functioning, which were negatively associated with PDS in adolescents and young adults (AYA) with SB (Brei et al., 2013). Parents with AYA with SB experienced the highest prevalence of PDS, 48% (Brei et al., 2013).

In summary, presence and severity of SB, parent gender, SES, and child age were related to PDISS or PDS in a limited number of studies. Child behavior problems had the largest relationship with PDS. A specific child factor, child neuropsychological functioning had a moderate relationship with PDS in the study with the highest prevalence of PDS. See Figure 3 for framework including context factors that emerged from findings of this review.

Process Factors. Process factors expected to be associated with depressive symptoms included family functioning and parent factors. Each study reviewed found at least one process factor related to PDS (see Table 2).

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Family functioning. Family functioning is defined as family system attributes that characterize how the family operates or behaves (McCubbin & McCubbin, 1987). When operationalized as the process of family cohesion, social support, and support satisfaction, family functioning was found to be negatively related to parental distress and PDS (Barakat & Linney, 1992; Brei et al., 2013; King et al., 1999; Kronenberger & Thompson, 1992a, Ulus, et al., 2012). Studies found that lower levels of satisfaction with support were related to higher PDISS (Barakat & Linney, 1992; King et al., 1999; Kronenberger & Thompson, 1992a). Similar findings reported in one earlier and one later (44% PDISS; 48% PDS) study found controlling family environment, marital quality/support (Kronenberger & Thompson, 1992a) and family protective factors (family cohesion, satisfaction, mastery and esteem) (Brei et al., 2013) were predictors of depressive symptoms. Satisfaction with support was important across all child age groups most notably in Barakat and Linney's (1992) study, social support and support satisfaction explained 42% of the variance in the outcome. Most recently, Ulus et al. (2012) found family functioning, mother's role and father's problem solving and general functioning related to PDS.

Parent factors. Chronic sorrow, negative coping, higher stress and lack of parental competence were related to PDS and varied according to child age. The only parent factor relevant in families with infants and young children was chronic sorrow (Hobdell, 2004). Other parent factors begin to relate to PDS in the school age years when managing ongoing stress puts demands on parent coping. Use of negative coping strategies was related to PDISS in parents with school age children (Barakat & Linney, 1995). Avoidant coping, more specifically behavioral disengagement, less ability to adapt

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to change and venting of emotions in addition to parenting satisfaction was related to PDISS in one study (Holmbeck et al., 1997). Parents who vented their emotions to friends were more at risk for depressive symptoms (Holmbeck et al., 1997; Kronenberger & Thompson, 1992a).

Parent perceived stress of everyday life in families of children with CHC is more than stress about aspects of the child's condition. Stress is an overall appraisal process in which perception of demands exceed resources in the relationship between person and environment. Stress can be acute, intermittent, or chronic and can contribute in the short term to a state of balance yet when prolonged can be damaging physiologically in the long term (Lazarus & Folkman, 1984; McEwen, 1998). Parent stress, number of leisure days reported, anxiety levels, and caregiver burden, in these studies were indicators of stress (Grosse et al., 2009; Kronenberger & Thompson, 1992a; Valença et al., 2012). Holmbeck et al. (1997) also found stress from role restriction and social isolation related to PDS. Stress was alleviated in one intervention study testing a surgical procedure that improved bowel continence. Parents were more likely to leave their home and socialize after this procedure and this process related to PDS (Ok & Kurzrock, 2011). Parent perception of competence and parenting satisfaction were significantly related to depressive symptoms in two studies (Holmbeck et al., 1997; Lemanek et al., 2000). In a Brazilian sample, depressive symptoms were related to higher anxiety and caregiver burden (Valença et al., 2012). Generally, studies explored either family functioning or parent factors, but not together. A notable pattern was that either family functioning or parent factors were significant in each study reviewed. Perhaps exploring both within

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same study sample may enhance understanding of distinct contributions of relevant process factors.

In summary, every study had either a family functioning or parent factor related to PDS. Across five primary studies, relationship of family member cohesion, social support, and support satisfaction to PDS was supported. When context, child behavior or neuropsychological functioning was considered, family functioning had a moderate to large relationship with PDS.

Context and Process Factors. Multivariate analysis used in a few studies examined both context and process variable contribution to outcomes (Barakat & Linney, 1995; Brei et al., 2013; Kronenberger & Thompson, 1992a, 1992b). The process variables generally had an either similar (Kronenberger & Thompson, 1992a) or larger contribution to understanding of PDISS than context variables (Barakat & Linney, 1995; Brei et al., 2013). Controlling for race in both samples, process factors differed and family functioning variables (controlling family environment & marital quality/support) explained a greater amount of variance (total variance 50%) than stress (total variance 32%) (Kronenberger & Thompson 1992a, 1992b). Barakat and Linney (1995) found the most variance of PDISS explained by both context and process factors (67%) when specifically evaluating negative parent coping strategies. Although the context factors (SES, race, and child factors) explained 20% of the variance in PDS, adding the process variables problem focused, emotion-focused, and avoidant parent coping explained an additional 47% of the outcome (Barakat & Linney, 1995). Finally, a recent study found 57% of variance in PDS was explained by neuropsychological functioning (a child context factor) and family functioning process factors (family cohesion, satisfaction,

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mastery and esteem) (Brei et al., 2013). When multivariate analysis included context factors in analysis, process factors contributed more variance in PDS/PDISS. In addition, in families with school-aged children parents' negative coping strategies were related to PDS. There is not sufficient evidence to understand differences in family functioning and parent factors between age groups.

The results of this review were organized by categories to identify factors related to depressive symptoms. The overall pattern of context and process variables related to depressive symptoms were consistent whether the outcome evaluated was PDISS or PDS. However, the later literature began to explore factors important in clinical practice such as neuropsychological functioning (Brei et al., 2013), leisure and socialization (Grosse et al., 2009; Ok & Kurzrock, 2011).

The evidence presented was limited by methodological shortcomings in the studies reviewed. The following critique addresses the design, concepts, and instruments measuring depressive symptoms in studies examining relationships of context and process factors related to outcome variable of depressive symptoms. See Table 2 for context and process factors related to depressive symptoms summary and Table 1 for relevant findings and strengths and limitations.

Methodological Review

Design. The guidelines for appraisal of level of evidence by Melnyk and Fineout-Overholt (2011) were used in this review with level I as the highest and level VII as the lowest (see Table 1). All 15 studies were quantitative and about half of the studies (7) were single descriptive correlational studies at level of evidence VI. Five comparative descriptive (2-group: group with SB and comparison) design studies were conducted

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between 1992 and 2009 at level IV evidence. One was a longitudinal study with a 2-year lag between time points allowing for comparison of factors across time (Friedman et al., 2004). One quasi-experimental study in 2011, the only intervention study for impact of surgical procedure of bowel care management on quality of life, was at level III evidence. One study, the meta-analysis was at the highest level of evidence I. While the meta-analysis was a stronger design it was limited by the small number of studies utilized and lack of conceptual homogeneity among variables used to calculate effect sizes. The evidence in depressive symptoms body of literature is descriptive of factors associated with but not causal of PDS.

The studies completed in the 1990s primarily focused on psychological adjustment and process factors of social support (Barakat & Linney, 1992; King et al., 1999; Kronenberger & Thompson, 1992a), stress (Holmbeck et al., 1997; Kronenberger & Thompson, 1992b), and coping (Barakat & Linney, 1992). Inclusion of "family" was noted in studies in the late 1990s (Holmbeck et al., 1997; King et al., 1999). In the last decade, a shift to understand outcomes of adaptation is noted (Grosse et al., 2009; Lemanek, Jones, & Lieberman, 2000; Ok & Kurzrock, 2011; Valença et al., 2012). Most recently, a specific aim was to examine relationship of risk and protective factors and PDS (Brei et al., 2013).

Sample and location. The external validity of these studies is limited by small sample sizes and sampling methods. Total sample sizes ranged from 23 – 164 participants. Several studies had multiple reports using the same sample to address different research questions (Barakat & Linney, 1992; Barakat & Linney, 1995; Friedman et al., 2004; Holmbeck et al., 1997; Kronenberger & Thompson, 1992a, 1992b).

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Although it can be beneficial to use data from the same sample, it complicates synthesizing results for a literature review.

Adequate sampling method was evident in Friedman et al. (2004) and Holmbeck et al. (1997) studies using same sample. The similarity of SB and comparison groups in sample may be due to the recruitment method. Investigators contacted schools where participants with SB attended to recruit matched comparison families, thus increasing the likelihood of similar race, ethnicity, SES, and age. Recruitment strategies that did not result in matched samples included those from pediatric clinics, childcare centers, newspaper advertisements, custodial services of local university and referral from participants (Barakat & Linney 1992; Barakat & Linney 1995; Gross et al., 2009). Overall, this group of level IV comparison studies was weak and results relating to group differences should be interpreted with caution.

Convenience samples of families with SB were primarily from clinics in Midwest United States with the exception of studies in Canada (King et al., 1999), Brazil (Valença et al., 2012), and Turkey (Ulus et al., 2012). The Canadian and Turkish samples both found family functioning process factors as related to PDS. The study conducted in Brazil found relationships between context factors of condition severity and SES and process factor of caregiver burden and anxiety related to depressive symptoms (Valença et al., 2012). See Table 1 for sample characteristics.

This body of literature is mostly limited to data from one informant, mother's report. Although studies identified their participants as parents or families, the primary informant was the mother. Three studies specifically use mother and father pairs as groups to understand differences between gender of parents (Hobdell, 2004; Lemanek et

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al., 2000; Ulus et al., 2012), none of the studies addressed family as the unit of analysis. Data from a variety of sources would facilitate analysis between subjects such as cluster analysis to determine types of families with similar factors related to PDS. Child age or developmental stage variables may better explain parent outcomes in future studies.

Analysis. Primarily studies used bivariate analysis, a few used multivariate methods in this body of literature to explain PDISS (Barakat & Linney, 1992; Barakat & Linney, 1995; Kronenberger & Thompson, 1992a, 1992b) and two specifically addressed PDS (Brei et al., 2013; Grosse et al., 2009). Regression analyses offered greater ability to explain multiple independent variables and their portion of variance in the dependent variable rather than simply stating there is a bivariate relationship. Variance explained across studies ranged from 32% to 67%. Logistic regression was used by one study (Grosse et al., 2009) to explain relationship of variables by SB severity (level of lesion). Expanding multivariate analysis would be critical to understanding relationships that are more complex.

Concepts and Instruments. Variability in measures of PDS was evident across studies. There was inconsistency in the conceptual definitions of factors in the studies and the instruments used to measure factors (see Table 1). Although the majority of the early studies (before 2005) addressed broad and complex concept of PDISS (n = 9), later studies more specifically addressed PDS (n = 5). The most common instruments used to measure PDISS were the Symptom Checklist-90-revised (SCL-90-R) (6) and the Brief Symptom Inventory (BSI) (3), which is a short form of the SCL-90-R instrument. About half of the studies used global severity index (GSI) of the larger instrument as a measure of overall severity of PDISS. This approach provided a broad measure of PDISS that

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addressed a range of symptoms. Further, it was not possible to determine overall severity of psychological symptoms, specifically depressive symptoms since measures are incongruent.

After 2005, the majority of studies focused specifically on PDS and most used measures consistent with symptoms identified as part of diagnostic criteria. This was a positive development as PDS can be specifically measured as a clinically relevant indicator of mental health thus facilitating evaluation and further diagnosis and treatment. Five instruments, Beck Depression Inventory (BDI), Generalized Contentment Scale (GCS), Center for Epidemiologic Studies Depression Scale (CES-D), Fecal Incontinence and Constipation Quality of Life (FICQOL), and 2 items from the Short Form Health Survey (SF-36), measured depressive symptoms. The first three scales have published reliability and validity data that support their specific measure of PDS. The FICQOL and the 2 items for the SF36 both have specific items that address PDS although their reliability and validity have not been established. Less than half of the studies reviewed measured depressive symptoms. Other studies measured depressive symptoms as a component of overall psychological status. Although different measures were used for PDISS and PDS, the prevalence identified using the measures and the context and process factors related to them were similar. For example, demographic context factors, parent gender and socioeconomic status, which had small relationship with PDISS were also found related to PDS in studies after meta-analysis was conducted in 2005 (Ulus et al., 2012; Valença et al., 2012). Presence and severity of SB, operationalized as severity in more recent literature, were related to both PDISS and PDS. Process factors to include family and parent factors were similar before and after the Vermaes et al. (2005) meta-

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analysis. A few new parent factors were examined in recent studies to understand impact of leisure time and travel/socialization (Grosse et al., 2009; Ok & Kurzrock, 2011). Restricted leisure (one or no leisure days per month) experienced by families with children with SB (27%) versus comparison (4%) group related to PDS (Grosse et al. 2009), while surgical intervention for bowel management affected travel/socialization. PDS was significantly improved post-surgery as parents were less often prevented from the leaving the home (Ok & Kurzrock, 2011). A more expanded conceptualization of parent leisure and socialization are needed to better understand the protective influence of leisure activities.

Summary

In summary, this synthesis has addressed a relatively small number of studies conducted in families with children with SB in relation to depressive symptoms. The level of evidence is mostly between level III-VI with only one study at level I and one at III. The meta-analysis (Vermaes et al., 2005) provided a review of factors to further explore in future research specific to families with a child with SB. An understanding of the importance of both context and process factors in the study of depression outcomes is reinforced by the review findings. Similar findings were noted in early and later literature of factors related to both PDISS and PDS.

Strengths of this review were that studies did examine concepts related to parent (not child) outcomes contributing to the literature of parents of a child with SB, an understudied population. This allowed for review of factors related to PDS and select instruments to report valid and reliable measures of PDS. These descriptive studies were invaluable in identifying potential factors associated with depressive symptoms for

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further exploration. The weaknesses of the studies included poorly matched samples for those that had comparison groups, relatively small convenience samples, use of primarily the maternal caregiver as an informant, and inconsistent measurement, especially in the early studies.

This body of literature provides preliminary evidence (a) for a high prevalence of depressive symptoms in parents (up to 48%), and (b) identification of context (demographic, condition, and child factors) and process (family functioning and parent factors) factors which could potentially explain PDS. Although context factors were important, they were not sufficient alone to explain depressive symptoms. In the small number of studies identifying both context and process factors, process factors contributed a significant additional explanation of variance in PDS.

Gaps and Implications for Research and Practice

While early and later findings were similar, the use of PDS as the outcome measure did facilitate report of a more specific outcome to determine prevalence and more precise outcome measure for development of interventions. Addressing PDS as an outcome will be useful for targeted clinically focused interventions and clinical effectiveness research. The prevalence of PDS among families with children with SB warrants further study. A better understanding of context and process factors related to PDS is possible using multivariate analysis to determine contribution of factors such as condition severity, child neuropsychological functioning, and family functioning. Further, possible mediating role of family functioning process variables and PDS in parents of children with SB could be explored. Although a comprehensive understanding of the

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factors related to PDS remains limited, findings warrant implementation of parent depression screening in families with children with SB.

Further, the measurement of SB severity needs to be explored. This issue would be advanced by development of a measure of condition severity that allows for understanding of components of severity may help to tailor design of interventions based on aspects of condition. Process factors of family functioning, parental stress and coping are important modifiable factors that can become integral components of intervention research. A newly emerging concept of parent leisure activities can be further explored to understand aspects of the activities useful and protective for parents. Although we know some predictors of PDS, are demographic, condition, neuropsychological functioning, family functioning, parent stress and coping factors, better understanding of their mediating and moderating relationships can support development of intervention programs.

Early childhood development was understudied in this population and is a critical period for development of child neuropsychological functioning that needs further study. Understanding emerging neuropsychological deficits in children can help to identify problems early (Heffelfinger & Koop, 2009). Stress such as early childhood adversity and exposure to PDS, can have long-term implications for neuropsychological development and trajectory of chronic health conditions (Shonkoff, Boyce, & McEwen, 2009). Although there were studies that investigated a wide range of ages, the unique needs of parents of adolescents with SB also seem to be understudied. Combining all ages might overlook the unique challenges of each age group and the trajectory of parent depression across child's developmental stages. Longitudinal research is also critical to

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understanding factors pertinent for parents of children in specific age groups. Sample sizes of studies need to be increased through multi-site and interdisciplinary partnerships to advance statistical methods investigating causal factors. Better understanding of risk and protective factors across the life course will guide researchers and clinicians to improve outcomes for parents affecting the individual and family over time.

Review Limitations

The study samples in this review were mostly from clinic populations and were convenience samples. The majority of comparison samples were poorly matched, potentially contributing to significant group differences. This review found variability in reporting indices of condition severity that made it difficult to reach a conclusion of differences between levels of lesion. Since almost half of the studies had mixed samples in age ranging from infant to young adult, the conclusions by age must be interpreted with caution. The use of the term "parent" may have limited the ability to identify studies of caregivers more broadly although preliminary review showed the "caregiver" literature was related to adult dependents. Parents of adult children were omitted, however this synthesis allowed for targeted recommendations for parents of children. Although efforts were made to be inclusive of terms such as psychological adjustment and psychosocial distress, this review focused on PDISS and PDS may not be inclusive of all research on mental health of parents of children with SB. Measurement of PDS is limited to symptoms reported by parents in the last two weeks. The more specific focus on PDS in the recent literature may not capture other symptoms such as anxiety or symptoms of substance abuse. Alternative measures such as the PROMIS (Patient-Reported Outcomes Measurement Information System) Mental Health Summary or Anxiety Scale may be

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helpful (Hays, Bjorner, Revicki, Spritzer, & Cella, 2009). While the purpose of this review was to synthesize the literature on depressive symptoms in parents of children with SB, that limited ability to generalize findings to other chronic conditions.

Conclusion

This review adds to the literature a theoretically-based synthesis of findings related to PDS in families with children with SB. Factors related to PDS were identified and gaps highlighted to guide future research of families with children with SB and potentially other CHC. While a portion of variance remains unexplained, findings warrant implementation of parent depression screening in families with children with SB.

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Chapter 3

Family Quality of Life in Families of Children with a Chronic Health Condition: A review of the literature

Abstract

The purpose of this manuscript was to review the concept and measurement of Family Quality of Life (FQOL), delineate parents' report of family quality of life and synthesize the literature on factors related to FQOL in families of children with chronic health conditions. Twelve studies were identified from 2002 to 2013 in databases (CINAHL, MEDLINE, and PsycINFO) and references from retrieved articles. Parents reported high perceptions of overall FQOL and domains-specific FQOL. Domains included family relationships, family interaction, parenting, influence of values, health, careers, community, support from services, support from others, disability-related support, leisure, finances, physical material well-being, and emotional well-being. Factors related to FQOL were income, services, condition severity, and child factors (child behavior problems, future expectations, neuropsychological functioning) family functioning (family cohesion, family resources, family satisfaction, social support, support satisfaction) and parent factors (depressive symptoms, hope, leisure, stress). In this review, family functioning had the largest relationship with FQOL.

Note: Following Chapter 3 is a paragraph that describes the integration of factors related to PDS and FQOL from Chapters 2 and 3.

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Family quality of life (FQOL) is an important emerging concept in the study of families of children with a chronic health condition (CHC). In the United States (US), approximately 10 million children live with a CHC (National Survey of Children's Health, 2007), which is defined as having or being at risk for "a chronic physical, developmental, behavioral, or emotional condition" (McPherson et al., 1998; Newacheck, Rising, & Kim, 2006; van der Lee, Mokkink, Grootenhuis, Heymans, & Offringa, 2007). Parents with a child who has a CHC, experience increased caregiving demands that may influence their FQOL. The purpose of this manuscript was to (a) review the concept and measurement of FQOL, (b) describe parent perception of overall and domain-specific FQOL, and (c) synthesize the literature on factors related to FQOL in families of children with CHC.

Background

Definitions of FQOL and related concepts provide a background for this review of literature to better understand concept and measurement. The concept of quality of life has been defined as "an individual's perception of their position in life in the context of the culture and value systems in which they live, and in relation to their goals, expectations, and concerns" (World Health Organization, 1997, p. 1). A related concept, health-related quality of life is used to describe individual quality of life in context of a health condition. The lived experience of the individual can be multidimensional or an overall global perception of quality of life. Family, a group of individuals who identify themselves as part of the family experience FQOL. The concept of FQOL has emerged from the perspective of family with children. A small number of studies identified in the

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literature used the concept of "FQOL" in a variety of ways. Three authors conceptually defined FQOL. The earliest definition proposed by Poston (2003) is:

> Family quality of life can be defined as the conditions, consistent with the family's values where the family's needs are met (i.e., daily family life, emotional well-being, financial well-being, physical environment, health, parenting, advocacy); family members enjoy their life together as a family (i.e., family interaction); and family members have the opportunities to do things that are important to them (i.e., social well-being and productivity) (p. 346).

Brown et al. (2006) conceptualize FQOL as " . . . the degree to which family quality of life is enjoyable, meaningful, and supported by the types of resources that are important to family members, as well as the struggles faced by families (p. 3). Thereafter Zuna, Summers, Turnbull, Hu, and Xu (2010) defined FQOL as "a dynamic sense of well-being of the family, collectively and subjectively defined and informed by its members, in which individual and family-level needs interact" (p. 262).

Two of these definitions (Brown et al., 2006; Poston, 2003) suggest a multi-dimensional concept, an individual's perspective of components of family life while the third proposed by Zuna et al. (2010) is defined as a collective overall wellbeing. The two multi-dimensional definitions have similar domains including (a) family life that is meaningful or consistent with family values, (b) enjoyment, and (c) resources. A reflection of family struggle is a unique aspect of Brown et al.'s (2006) perspective. These definitions are limited by the a-priori delineation of specific domains that are important to the family. Zuna et al.'s (2010) approach is a collective conceptualization based on family needs that may or may not be reflective of the beliefs in a variety of

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families. Important aspects of family life may vary between members. A common limitation of these definitions is that none includes the family member's ability to differentially prioritize domains of FQOL important to them. These multiple conceptualizations of FQOL have led to several measures of the construct.

In the quality of life literature, there are two conceptualizations, overall QOL and domain-specific QOL. Overall QOL can be a summary of domains or it can be an overall global perception. Some researchers feel that this overall global perception of QOL that reflects the individual's emphasis on domains important to them may be useful as an outcome (Ferrans, 1996; Grady, Jaowiec, & White-Williams, 1999; Sawin, Brei, Buran, & Fastenau, 2002). Similarly, a global concept of parents' perception of FQOL can include the domains important to the family. The second conceptualization, domain-specific QOL can also apply to FQOL where specified domains that represent aspects of family life are delineated. It is not clear which of the conceptualizations of FQOL as overall concept or a concept with multiple domains (domain-specific) or a combination of the two can be useful in advancing family science.

It is important to differentiate FQOL, which focuses on a sense of well-being of the family, from a related concept family functioning. Family functioning is defined as the attributes of a family system that characterize how they operate or behave (McCubbin & McCubbin, 1987). It includes attributes such as family cohesiveness, satisfaction, mastery, hardiness, or resourcefulness. While empirically family functioning and FQOL are related (r = 0.34 - 0.60) (Ridosh, Sawin, & Brei, 2013; Sawin et al., 2002), they are not the same concept.

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The construct of family quality of life can be operationalized as a family outcome or result of the efforts of families to balance those interactions and relationships to stabilize the family and environment on a continuum, dynamic and salient to the family at the present moment.

Measurement. Four different measures of FQOL were reported in the literature of families with children. Two measures with specified domains were the Beach FQOL Scale (Hoffman, Marquis, Poston, Summers, & Turnbull, 2006) and FQOL-2006 Survey (Brown et al., 2006). A single and 3-item measure of FQOL did not specify domains (Ridosh et al., 2013; Sawin, Buran, Brei, & Fastenau, 2003). See Table 3 for summary of instruments of FQOL, their psychometric properties.

Conceptual model

Two theoretical models influenced the overall conceptual approach to the review of literature. The Transactional Stress and Coping Model identifies maternal processes (managing stress, coping and family functioning) related to outcomes of maternal and child adjustment (Thompson & Gustafson, 1996). The second model, the Ecological Model of Secondary Conditions (Sawin et al., 2003), includes contextual risks and protective processes associated with adaptation of adolescents with CHCs. Three contextual risk factors and three protective processes explain relationships with adaptation outcomes (e.g. physical, mental, and quality of life outcomes) for adolescents. Both of these models suggest a linear relationship whereby context (environment) followed by process leads to outcomes. This broad conceptual approach using the categories context, process and outcome guides the identification of factors related to FQOL in the literature. Context is defined as the environment in which parental

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adaptation outcomes occur (demographic, condition and child factors). Process is defined as the perceptions and activities that lead to parental adaptation outcomes. Outcome is defined as the result of the process and includes adaptation. Understanding both context and process factors together better explains factors related to outcomes. This review used the general orientation from both models (context, process and outcomes). Parent perception of FQOL is the adaptation outcome of interest.

Methods

This review was designed to synthesize the literature on the family outcome, FQOL and the relationships of context and process factors to FQOL. Primary research reports were located in the following steps. First, a search was conducted in CINAHL, MEDLINE, and PsycINFO databases using keyword "Family quality of life". Inclusion criteria were articles published from 2000 to 2013, published in English language, peer reviewed empirical research articles, and pertaining to FQOL as an outcome. Exclusion criteria were articles related to child outcomes, individual quality of life, caregiver burden, and families with adult children. Titles and abstracts of 36 articles were reviewed. A review of references and studies available to the researcher identified seven additional studies that met inclusion criteria for a sample of 43 records screened. After review of titles and abstracts 13 records were not eligible due to the exclusion criteria. Twenty-nine studies were reviewed and 17 were excluded since they did not meet inclusion criteria. The final sample included 12 primary research studies. See Figure 4 for a flow diagram of the search strategy.

The overall FQOL score and domain scores reported by the authors (means and SD) were used to describe prevalence. When more than one study reported an overall or

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domain score, a mean of all studies reporting that score was calculated for this analysis by the primary author (MMR) and was reported in the result tables. Domains were reported from highest to lowest frequency. If a study deviated from the pattern using a sample with specific characteristics, (e.g., from an international study or a study using a unique population) that deviation was noted.

The factors related to FQOL were identified by either correlation and/or regression analysis and reported by context (demographic, condition and child factors), process (family functioning and parent factors) variables. Magnitude of the relationship was reported when data were available. A summary of factors related to FQOL including the amount of variance explained in each study was reported in result tables.

This review analyzed 12 research studies. First FQOL was described, then factors related to FQOL synthesized. A critique of the quality of the literature was summarized. Finally, a theoretical framework was generated from findings of factors related to FQOL.

Results

Characteristics of the sample from the 12 studies used for this review are summarized in Table 5. Studies were published from 2002 to 2012, samples sizes ranged from 43-442 but were typically less than 200, and studies primarily represented families with a child from birth to 21 years of age. Two studies included children and dependent adults.

Parent perceptions of overall global and domain-specific FQOL were high (see Tables 6 and 7). Three of the five studies using the Beach FQOL Scale reported an overall global score, indicating overall satisfaction with FQOL ($\mu_{\text{sum overall}} = 3.80$, $\sigma = 0.67$; range 3.56 to 3.99; on a 5 point scale). Only two studies using the FQOL-2006

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Survey reported the FQOL global single item ($\mu_{\text{sum overall}} = 3.80$, one $\sigma = 0.91$; range 3.71 – 3.90; on a 5 point scale), which was in range of neither satisfied/dissatisfied to satisfied (Rillotta, Kirby, Shearer, & Nettelbeck, 2012; Werner et al., 2009). When analyzed by instrument the average of the scores were very similar (Davis & Gavidia-Payne, 2009; Eskow, Pineles, & Summers, 2011; Ridosh et al., 2013; Sawin et al., 2002; Summers et al., 2007; Werner et al., 2009). The single item and 3-item scales using a 100 point response pattern anchored on "excellent" were also high ($\mu = 72.5$ - 80.5; $\sigma = 15.62$ - 21.6) (Ridosh et al., 2013; Sawin et al., 2002).

Four of the studies using the Beach FQOL Scale report at least select domain scores (see Table 7) (Davis & Gavidia-Payne, 2009; Eskow et al., 2011; Jackson, Wegner, & Turnbull, 2010; Summers et al., 2007). The physical/material well-being domain ranked highest, followed by family interaction and parenting ($\mu_{\text{sum score}} > 4.00$). Disability related support was close to this criteria ($\mu_{\text{sum score}} = 3.92$). The lowest ranking domain was ($\mu_{\text{sum score}} = 3.30$) emotional support (see Table 7). Although there were only a few studies using this tool, the patterns were consistent across three studies, particularly with the emotional well-being scale, which was substantially below the other domains. Only Eskow, Pineles, and Summers (2011) reported lower scores (means < 4.0) on three domains (parenting, disability-related support and emotional well-being) and these scores were primarily in the registry sample. The registry sample consists of families on a waiting list for a US Medicaid Waiver Program that provides additional support such as home and community-based services to families with children (Eskow et al., 2011). The study investigating FQOL in families with children who were hearing impaired had domain subscales scores above the other studies. Most of their domain subscales were

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above 4.22 except for the emotional well-being domain that had a mean of 3.65 (Jackson et al., 2010).

Five studies using FQOL-2006 Survey (see Table 8) reported domain mean scores for both the Satisfaction and Attainment Dimensions (Ajuwon & Brown, 2012; Clark, Brown, & Karrapaya, 2012; Neikrug, Roth, & Judes, 2011; Rillotta et al., 2012; Werner et al., 2009). It is important to note that these studies were all international and reflected divergent cultures. Family relationships ranked highest in both satisfaction and attainment ($\mu_{\text{sum score}} > 4.0$) and satisfaction was consistently reported high ($\mu_{\text{sum score}} = 4.16$; $\mu_{\text{domain scores}} = 4.01 – 4.36$) (Ajuwon & Brown, 2012; Clark et al., 2012; Neikrug et al., 2011; Rillotta et al., 2012). The Canadian study had lowest domain mean score of 3.91 in family relationships (Werner, 2009). FQOL-2006 survey domains included influence of religious, spiritual, and cultural values, which were high in four studies ($\mu_{\text{sum score}} = 4.02$; $\mu_{\text{domain scores}} = 3.82 – 4.22$) (Ajuwon & Brown, 2012; Clark et al., 2012; Neikrug et al., 2011; Rillotta, et al., 2012). Attainment of "Health of the family" was also high across samples in five studies using FQOL-2006 survey ($\mu_{\text{sum score}} = 4.01$; $\mu_{\text{domain scores}} = 3.57 - 4.44$) but slightly lower in satisfaction ($\mu_{\text{sum score}} = 3.82$; $\mu_{\text{domain scores}} = 3.57 - 3.90$) (Ajuwon & Brown, 2012; Clark et al., 2012; Neikrug et al., 2011; Rillotta et al., 2012; Werner et al., 2009). However, in a recent psychometric evaluation of this survey, the Health domain was the least reliable ($\alpha = 0.53$) in sample across three countries (Isaacs et al., 2012) and therefore should be evaluated for a specific culture before broad use.

Moderate satisfaction ($\mu = 3.32$) and low attainment ($\mu = 2.86$) of community integration was described in a sample from Israel (Neikrug et al., 2011). Although differing by rank, the four lowest satisfaction FQOL ($\mu_{\text{sum scores}}$ less than 3.5) were from

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support from services, support from others, leisure and finance domains—also the lowest of attainment scores.

In summary, overall FQOL scores reflected relatively high perceptions of FQOL (3 out of 5 or 75 out of a 100). There is no way to determine how parents using the single item or 3-item global measures weighted potential domain components to determine their overall FQOL. The domain scores on the Beach FQOL tool and the FQOL-2006 Survey reflected substantial variance. The domains, family relationships and values were higher and support from services and support from others were lower using FQOL survey. In contrast, using the Beach tool, physical/material (health services/finances) ranked highest. Similarly, the Beach tool captured least satisfaction with social support (emotional and disability-related).

Factors Related to FQOL

Context. Six studies (Davis & Gavidia-Payne, 2009; Eskow, et al., 2011; Hu, Wang, & Fei., 2013; Ridosh et al., 2013; Sawin, et al., 2002; Werner et al., 2009) reported factors related to FQOL (see Table 9). Demographic factors related to FQOL were income and service. Together with severity of condition, income explained 1.6% of the variance in FQOL in a sample of low-income families from China (Hu et al., 2012). In the US, income was related to FQOL in two studies of families who had a child with a CHC. First, combined income of parents of an AYA with SB was moderately related to FQOL (Ridosh et al., 2013). Second, while controlling for income and age of the child, service through waiver status in families who had a child with autism participating in a US state program predicted FQOL (Eskow et al., 2011). Additionally, service adequacy in the US study evaluating mediating effect of professional partnership on FQOL was

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important (Summers et al., 2007). Only the study by Hu et al. (2012) explored severity of condition and found it a predictor of FQOL.

Three child factors, behavior problems, future expectations and neuropsychological functioning, were moderately to strongly correlated with FQOL (r = 0.33-0.61). In a sample with young children, intensity of child behavior problems measured by the Child Behavior Subscale of the Parent Hassles Scale was related to FQOL. Greater intensity of the childhood behavior problems was a predictor of lower FQOL, family income was no longer significant when child factor considered (Davis & Gavidia-Payne, 2009). In the samples with AYA with SB, future expectations, such as maintaining relationships, having a good job, and other accomplishments, were moderately to strongly related to single-item and 3-item scores (Ridosh et al., 2013; Sawin et al., 2002). Neuropsychological functioning, measured by the Behavior Rating Inventory of Executive Function was moderately related to FQOL. Families with AYA with higher executive functioning and adolescent future expectations had higher FQOL (Ridosh et al., 2013).

Process. Family functioning was related to FQOL in six of the studies reviewed using both overall global and domain-specific measures of FQOL. Five studies (Davis & Gavidia-Payne, 2009; Ridosh et al., 2013; Sawin, et al., 2002; Summers et al., 2007; Werner et al., 2009) reported process factors identified by correlations and/or regression analysis. In studies using correlations, family functioning was moderately to strongly correlated with FQOL (r = 0.45- 0.62) (Ridosh et al., 2013; Sawin et al., 2002; Werner et al., 2009). In a Canadian sample, family relationships (family satisfaction) were reported as moderately correlated with global FQOL item (r = 0.45) from FQOL-2006 survey

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(Werner et al., 2009). Family cohesion, family resources and family satisfaction were highly related to FQOL (r = 0.41 – 0.62) in studies of AYA with SB using overall single and 3-item FQOL measures (Ridosh et al., 2013; Sawin et al., 2002). In the earlier of these studies family satisfaction and parental hope explained 50% of the variance in FQOL (Sawin et al., 2002).

In studies with samples with young children, social support and support satisfaction were related to FQOL. Specifically support from family (R^2 = 0.17) and support satisfaction (professional support) (R^2 = 0.10) were significant (Davis & Gavidia-Payne, 2009; Summers et al., 2007). Support satisfaction (family-professional partnership) was a partial mediator of service adequacy and FQOL (Summers et al., 2007), the only mediation tested.

Parent factors related to FQOL were primarily found in studies using single item and 3-item measures of FQOL in families with SB (Ridosh et al., 2013; Sawin et al. 2002), except for leisure time in Canadian sample from FQOL-2006 survey (Werner et al., 2009). Parent factors (depressive symptoms, hope, leisure, stress) were strongly correlated to FQOL (r = 0.47 - 0.72) in three studies (Ridosh et al., 2013; Sawin et al. 2002; Werner et al., 2009). Stress of the condition and stress of everyday life had moderate relationship with FQOL in one study of AYA with spina bifida (r = 0.30 - 0.47) (Sawin et al., 2002).

In summary, process factors were related to FQOL across the majority of studies. Family functioning had the largest relationship with FQOL. In this review, findings suggest context (demographic, child) and process factors (family functioning, parent factors) were consistently related to FQOL in families with children with CHC.

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Methodological Critique

As a group the studies reviewed were limited by the level of evidence, issues of sample size and composition, lack of consistency of measurement and level of analysis.

Design and sample. Appraisal for level of the evidence was based on Melnyk and Fineout-Overholt (2011) hierarchy of evidence criteria. The highest level of evidence (I) is a meta-analysis and lowest (VII), a report from an expert or committee. The higher the level of evidence, the greater strength of the findings. In the current review, 11 studies were descriptive studies at level of evidence VI. One study (Eskow et al., 2011) used a two-group design categorized as level IV. The majority of studies were conducted by two research teams (Beach Center on Disability and Surrey Place Center International Family Quality of Life Project) in samples of families with children with intellectual disabilities. Another initiative has begun research of families with children with CHC, specifically spina bifida (SB). These descriptive studies are appropriate for preliminary development of a new concept, but studies with stronger designs will be needed to advance the understanding of FQOL. The quality of the descriptive studies is limited by the characteristics of the families in samples, the sample size, and level of analyses of many of the studies (see Table 5 sample characteristics).

Only two studies conducted since 2009 explored and described FQOL in the context of families with a child with an intellectual disability (Jackson et al., 2010; Ridosh et al., 2013). These two addressed FQOL in families with SB (Ridosh et al., 2013) and hearing impairment (Jackson et al., 2010). Although children with these diagnoses are not typically intellectually impaired, children in both groups can have substantial learning problems.

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Issues with the samples limited the quality of the studies reviewed. Studies generally reported data from maternal primary caregivers. Only half of the studies reviewed had adequate sample size and thus the results of the others must be seen as preliminary. Four studies had a sample size between 103-207 (Jackson et al., 2010; Neikrug et al., 2011; Rillotta et al., 2012; Summers et al., 2007) and two large studies had samples of 442-855 participants (Eskow et al., 2011; Hu et al., 2012). Probability sampling methods were used in two studies, (Clark et al., 2012; Hu et al., 2012)—one randomly selected sample of families receiving services in Malaysia (Clark et al., 2012) and the other used a stratified sample in urban and suburban communities and diverse age groups living in Beijing, China (Hu et al., 2012). Due to the small number of studies using rigorous sampling methods, comparison across studies was difficult. Only the Hu et al. (2012) study reported factors related to FQOL and their findings were generally consistent with two US studies (Eskow et al., 2011, Ridosh et al. 2012). Finally, low response rates (16 – 28%) (Davis & Gavidia-Payne, 2009; Eskow et al., 2011; Summers et al., 2007) with the exception of Chinese sample at 72% (Hu et al., 2012) limited the usefulness of results.

The international study of FQOL has both strengths and limitations. The breadth of settings potentially allows investigators to compare and contrast FQOL across various communities and cultures. Five studies were conducted in the US, three from the Midwest (Ridosh et al., 2013; Sawin, et al., 2002; Summers et al., 2007), one from the Northeast (Eskow et al., 2011), and one across 42 US states (Jackson et al., 2010). Seven studies were conducted outside of the US—Australia (Davis & Gavidia-Payne, 2009; Rillotta et al., 2012), China (Hu et al., 2012), Canada (Werner et al., 2009), Israel

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(Neikrug et al., 2011), Malaysia (Clark et al., 2012), and Nigeria (Ajuwon & Brown, 2012). The primary measure of FQOL in the US was the Beach and in other countries was the FQOL-2006 survey. However, these settings vary widely by culture, economy, health care systems and resources. Additional studies are needed to fully understand if FQOL is similar across countries and cultures. Given the limitations of the samples in this review the results need to be seen as preliminary.

Instruments and analyses. Although the reliability of the Beach FQOL Scale is good, the factors and subscales measure a family's perception of satisfaction on only the specific aspects included in the tool. The majority of the studies using the FQOL-2006 survey focused on describing the dimensions and domains and in only two instances reported the global FQOL item score (Rillotta et al., 2012; Werner et al., 2009) (see Table 8). In contrast, only one study using the Beach FQOL Scale limited their analysis to frequencies (Jackson et al., 2010). The Beach FQOL Scale inconsistently reported domain means and overall FQOL scores (see Tables 6 & 7). The most advanced analyses occurred in the study of factors related to FQOL where three used correlations (Ridosh et al., 2013; Sawin et al., 2002; Werner et al., 2009), four studies used regression analysis (Davis & Gavidia-Payne, 2009; Eskow et al., 2011; Hu et al., 2012; Sawin et al., 2002), and one a mediation analysis (Summers et al., 2007).

In summary, this body of literature is limited by design, samples and analysis procedures. Overall findings do represent some descriptive data of FQOL but generalizability is limited due to power, and response rates. Although many of these studies have limitations, the results can be useful in identifying potential factors related to FQOL for further study.

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Discussion

Synthesis of studies exploring FQOL in families of children was limited by early conceptual development of FQOL. The inconsistency in the few definitions led to a variety of measures restricting ability to make conclusions in our understanding factors influencing family outcomes. Although domain-specific definitions and instruments provide useful measures of FQOL, preliminary evidence suggests an overall measure is also valid and reliable. A definition and measure of overall FQOL, in addition to prescribed domains of life and their individual measurement would facilitate future study of FQOL as an outcome measure. From this review of the literature, a definition of global FQOL is proposed: FQOL is an overall appraisal of the domains of life that are important to the family.

FQOL is a weighted perception of the domains important to the reporter about the family as a whole, a sum of a family member's perspectives of the individual, the child, and their family's quality of life. The nature of FQOL is a dynamic one. A measure of appraisal is captured when parent report of FQOL allows for parents to ascribe their own weight to domains of life important to them and report their own score representing different domains of life at different times. The single item or three-item scales serves such a purpose and allows a parent to weigh their overall perception of FQOL on continuum from poor to excellent, A summary of psychometric properties of FQOL measurement can be found in Table 4.

Currently, measures of FQOL reflect various dimensions of FQOL, overall and domain-specific FQOL are inconsistent making comparison difficult. The current summative domain-specific measures determine the degree to which the domains are

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aspects of family quality of life. Both the Beach FQOL Scale and the FQOL-2006 are domain-specific, while offering the capacity for a total FQOL score, whether from the total number of items or the single global item.

Since family relationships were the most highly rated component of FQOL in international samples, understanding what contributes to strength of relationships is important. Three of the samples included children over 18 (although mean ages ranged from 7 to 25 years), families with young adult children may have built stronger family relationships over time contributing to internal family resources. International studies are important to understand FQOL across all cultural groups but cultural and health care resources must be considered across studies. Data on ethnicity within samples would add context of the demographic factors that remain unexplained as related to FQOL. These context factors will be important for knowledge translation to practice. Larger samples, not only multi-site but also ethnically diverse and from developing/developed countries will inform further development of the science.

Analyses of factors related to FQOL are limited by the few studies that report multivariate analyses. Research analyses of mediation and moderation, predictive models using hierarchical regression and structural equation modeling will strengthen the evidence. This research will inform both intervention and evaluation of families with children with CHC.

There is a dearth of contextual data related to child factors in the study of FQOL. International studies did not evaluate child factors except for one study in Australia. Studies mainly reported data from maternal main-caregivers, multiple informant data

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may help to better explain FQOL especially in developing countries where multiple versus primary caregivers include extended families, siblings or grandparents.

There is some evidence of family functioning factors being related to FQOL, but understanding specific aspects of family functioning and possible parent factors that may be more important than others in the context of variety of samples remains unclear. Family functioning factors most predictive of FQOL had multiple indicators. Differentiating family functioning factors (cohesion, resources, satisfaction, social support, support satisfaction) that are internal and external to the family will be important to develop predictor models of FQOL. Understanding parent factors such as depressive symptoms, hope, leisure, and stress and their unique or combined contribution to FQOL as mediators and moderators will better explain adaptation outcomes of families with CHC. Use of a theoretical framework for design of studies was only explicit in studies of families with SB; therefore a comprehensive framework is indicated for future research.

Proposed Theoretical Framework of Factors Related to FQOL

The results of this review and the conceptual model of both Ecological Model of Secondary Conditions (Sawin et al., 2003) and the Transactional Stress and Coping Model (Thompson & Gustafson, 1996), were used to generate a theoretical framework of the factors related to parent perception of FQOL (see Figure 5). Context factors are proposed as the environment in which the FQOL occurs. The context factors, income, service adequacy, waiver status, severity of condition, child behavior problems, child future expectations and neuropsychological functioning, are proposed to have direct and indirect relationships to FQOL. Process factors, perceptions and activities that lead to FQOL outcomes are family cohesion, family resources, family satisfaction, social

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support, support satisfaction, hope, leisure, stress and parent depressive symptoms. Process factors are proposed to have direct relationships with FQOL. Several assumptions are made regarding the proposed theoretical framework. First parents' perception of FQOL whether overall FQOL or domain-specific is a family outcome variable, which can be reported by an individual family member. Second, select process factors may mediate the relationship of context factors to outcomes. Identification of more empirical evidence to support factors and relationships identified, testing of other potential mediation relationships and consideration of additional context and process factors can contribute to understanding of FQOL in families with children with CHC.

Review Limitations. The small number of studies of "family quality of life" and parent outcomes limited this review. Only research studies that reported findings of FQOL using quantitative or mixed methods were included. While some qualitative data was available in studies using mixed methods, these data were scarcely available in the primary research reports. Since the state of the science is in its earliest stages of conceptual development, further investigation of the psychometric properties of existing instruments and further evaluation of qualitative findings would add to the conceptual clarity of FQOL.

Conclusion

This review described what is known about FQOL in families with children with CHCs to advance the science of FQOL. A review of parent report of FQOL, identification of factors related to FQOL, critique of the evidence, and gaps in the literature were described. This review resulted in a simplified definition of global FQOL and a theoretical framework summarizing relationships for future study.

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Synthesis of Chapters 2 and 3

The literature review conducted on parent depressive symptoms in parents of children with SB and the literature review of FQOL in families of children with a CHC identified similar context and process variables related to the adaptation outcomes, PDS and FQOL. Based on the Thompson and Gustafson (1996) and Sawin et al.'s (2003) models, the parental mental health outcome, PDS, was identified as a proximal outcome in the proposed model and FQOL a distal outcome. The proximal outcome, PDS, may mediate the relationship of context or process variables to FQOL (see Figure 7).

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Chapter 4

Factors associated with Parent Depressive Symptoms and Family Quality of Life in Families with and without Adolescents and Young Adults with Spina Bifida

Abstract

The aim of this study was to explore factors related to parent depressive symptoms (PDS) and family quality of life (FQOL) in families. This secondary analysis used data (N = 209) from a multi-site correlational study of adaptation in adolescents/young adults (AYA) with and without spina bifida (SB) to explore parent outcomes. Outcome measures included the Beck Depression Inventory (BDI) and The FQOL Scale. Thirty-eight percent of the variance of PDS was explained by income, family resources and parent stress, but presence of SB was not a significant predictor. Presence of SB, family satisfaction, parent stress and PDS explained 49% of the variance of FQOL. PDS partially mediate the relationship of family resources and FQOL. For parents in SB subsample, family satisfaction and PDS explained 47% of the variance in FQOL. While family resources and stress, not PDS explained 49% of the variance in FQOL in the comparison subsample. Addressing PDS and FQOL in health care encounters is essential.

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With advances in healthcare over the last several decades, children with multiple health conditions, which previously limited longevity are thriving and surviving into adulthood (Davis et al., 2005). One of these conditions, spina bifida, a congenital disability caused by a neural tube malformation in fetal development, impacts the lives of adolescents/young adults (AYA) and their families. Severity of SB varies widely as a result of multiple surgeries, limitations in physical mobility, difficulty with bladder and bowel management, and social competence difficulty. Parenting a child with SB involves attending to a child's learning difficulties due to impairments in working memory, numeral literacy, verbal communication and problem solving abilities. The care of these children is complex, unpredictable and may require heavy family involvement that often affects family and parental well-being. Addressing overall well-being of parents such as mental health and quality of life is a public health priority (Marcus et al., 2012; U.S. Department of Health and Human Services, 2010). Specifically, parent depressive symptoms (PDS) and family quality of life (FQOL) are important outcomes to understand the lived experience of parents of children with a chronic health condition (CHC). However, there is little in the literature about either PDS or FQOL and the factors related to them. The aim of this study was to explore which context and process factors have direct and/or indirect relationships with PDS and FQOL in families with AYA with and without a chronic health condition (CHC), specifically spina bifida (SB).

Background

Overall well-being is threatened when adults experience depressive symptoms. The health of the family is compromised when these adults are parents. Parental depressive symptoms affect function in daily life of relationships, parenting and work life

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(National Research Council and Institute of Medicine, 2009). Depressive symptoms include sadness, pessimism, loss of pleasure or interest, changes in sleep and appetite, feelings of worthlessness, concentration difficulty, agitation and irritability (American Psychiatric Association, 2013). PDS can affect a parent's ability to effectively manage the increased demands of family life.

Although adult mental health is addressed in the general population, literature on parent depression is limited and is focused mostly in mothers with infants in the post-partum period. Even in this population only 12% of mothers diagnosed with depression received treatment (Horowitz & Cousins, 2006). In a large Canadian population health study, parents of children with health conditions had greater odds of overall poor health and were twice as likely to also have a chronic condition or activity limitation of their own as comparison parents of children without CHC (Brehaut et al., 2009). The parents of young children with health problems were more than twice as likely to experience depressive symptoms as parents of children without health problems (Brehaut et al., 2009). Barriers exist to the identification and treatment of depressive symptoms in parents (National Research Council and Institute of Medicine, 2009). Parents of adolescents and specifically those with CHC are often overlooked.

Quality of life (QOL) is defined as "an individual's perception of their position in life in the context of the culture and value systems in which they live, and in relation to their goals, expectations, and concerns" (World Health Organization, 1997, p. 1) Two conceptualizations of quality of life (QOL) in the literature include overall QOL and domain-specific QOL. Broad domains of QOL include health and functioning, psychological/ spiritual, social and economic, and family (Ferrans, 1996). While in

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Ferran's (1996) work family is a domain of QOL, the parent's perception of their families is not an individual members' perception of their position in life rather it is the appraisal of the family. FQOL is a parallel emerging construct. Research in FQOL has been complicated by conceptual and methodological complexities (Ridosh, Sawin, & Schiffman, 2014). When domains specific to FQOL are proposed they include family relationships, family interaction, parenting, influence of values, health, careers, community, support from services, support from others, disability-related support, leisure, finances, physical/material well-being, and emotional well-being (Brown et al., 2006; Hoffman, Marquis, Poston, Summers, & Turnbull, 2006). However, domain specific approaches do not allow the family member to "weigh" which domain(s) are important to their family's quality of life. Thus, for this study, FQOL was defined as an overall appraisal of the domains of life that are important to the family (Ridosh et al., 2014).

Measurement of FQOL is developing. The FQOL literature to date primarily focused on families who have a child with an intellectual or developmental disability. There is no current literature on the concept of FQOL in parents of children without CHC and limited literature on families with adolescents or young adults. Current studies of FQOL focus on the satisfaction of specific domains of QOL and disability-related resources available to the family.

Factors related to PDS and FQOL

A recent review of depression in parents generally, focused on the relationship between depression, parenting practices and child physical and mental health outcomes, found stress and adversity contributed to depression (National Research Council and

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Institute of Medicine, 2009). Demographic variables were important contributors to PDS. Ten percent of mothers who were less than 35 years of age, had lower education, lower income, were unemployed and single were found to have PDS (Ertel, Rich-Edwards, & Koenen, 2011). Race has been inconsistently related to depression and may be a confounding variable differentially affecting specific group. For example, Black mothers had a higher rate of adversity and White mothers were more likely to have comorbidities (Ertel et al., 2011).

Recurrent depressive episodes were noted as a risk factor with worsening duration of each depressive episode and lowering of the threshold of response to stress. Other related factors to PDS in adults were categorized as biological factors (genetic, neurological, hormonal, immunological, and neuro-endocrinological responses related to stress appraisal), environmental (acute negative life events, chronic stress, childhood experience with adversity), personal vulnerabilities (cognitive thinking (negative), interpersonal relationships (marital and parenting problems), personality characteristics (neuroticism and ruminative), and comorbidities (anxiety, substance abuse, behavioral and personality disorders and medical illnesses) (National Research Council and Institute of Medicine, 2009).

Factors related to PDS and FQOL are important to consider in families with children with CHC. The few studies (Brei, Woodrome, Fastenau, Sawin, & Buran, 2013; Kronenberger & Thompson, 1992a, 1992b; Valença, de Menezes, Calado, & de Aguiar Cavalcanti, 2012) exploring PDS in parents of children with SB identified a PDS prevalence of 44% or higher. Factors related to PDS in parents were synthesized in a recent review (Ridosh, Sawin, & Klein-Tasman, 2014). Investigators studying factors

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related to PDS in parents found the amount of variance explained ranged from 32 to 67%. Demographic factors related to PDS included income, parent education, parent gender, race, SES, and child age. Child factors included SB presence and severity, child behavior problems, child emotional problems, receptive language, and parent perception of executive functioning. Family and parent factors included family-centered caregiving, family cohesion, family environment, family resources, family satisfaction marital quality/support, social support and support satisfaction (family functioning) and anxiety, caregiver burden, coping, parenting, presence of a partner, sorrow, and stress (parent factors) (Ridosh, Sawin, & Klein-Tasman, 2014).

The perception of FQOL reported in the literature was moderately high (greater than 3.5 on a 0 – 5 scale) (Davis & Gavidia-Payne, 2009; Eskow, Pineles, & Summers, 2011; Ridosh, Sawin, & Brei, 2013; Sawin, Brei, Buran, & Fastenau, 2002; Summers et al., 2007). Demographic and child factors related to FQOL were income, condition severity, and child factors (child behavior problems, future expectations and parent perception of executive functioning [EF]). Family and parent factors related to FQOL were family functioning (family cohesion, family resources, family satisfaction, social support and support satisfaction) and parent factors (depressive symptoms, hope, leisure, and parent stress) (Ridosh, Sawin, & Schiffman, 2014). While family functioning was consistently predictive of FQOL, measures of family functioning varied.

Factors related to both PDS and FQOL were identified from the primarily descriptive correlational literature that had conceptual and methodological limitations. Few studies examined factors in families of adolescents or had comparison groups of children without conditions and many used a range of measures lacking specificity of

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outcomes of interest, PDS and FQOL. Although it is known that the prevalence of depressive symptoms in parents of children with SB is high, little is known about factors contributing to PDS in these families (Ridosh, Sawin, & Klein-Tasman, 2014). No literature evaluates how PDS are related to parent perception of FQOL. The current study is grounded in a conceptual framework generated from the reviews of the literature on PDS and FQOL.

Conceptual framework

Two conceptual frameworks were used to develop a general conceptual orientation of factors related to adaptation in families with a child with CHC. The two frameworks were the Transactional Stress and Coping Model (Thompson & Gustafson, 1996) and the Ecological Model of Secondary Conditions (Sawin, Buran, Brei, & Fastenau, 2003). The Transactional Stress and Coping Model refers to maternal meditational processes of stress, coping and family functioning and outcomes of maternal and child adjustment (Thompson & Gustafson, 1996). The second model was the Ecological Model of Secondary Conditions (Sawin et al., 2003). This model includes risk factors and protective processes associated with adaptation of adolescents with CHCs, including condition, demographic, neuropsychological, AYA resilience, family resourcefulness, and perceived health-care adequacy explain relationships with adaptation outcomes (e.g. physical health, mental health, and quality of life outcomes) for adolescents.

The integrated conceptual model that guided this study delineates common factors related to both outcomes (see Figure 7). The variables were organized by three categories context, process, and outcomes—proximal (PDS) and distal (FQOL). Context is defined

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as the environment in which parental adaptation outcomes occur such as demographic, condition and child factors. Process is defined as the perceptions and activities that lead to parental adaptation outcomes (family functioning and stress). Adaptation outcomes are defined as the result of the process (PDS and FQOL).

Context factors similar across literature of both outcomes PDS and FQOL were demographic (income), the severity of a CHC, and child factors (child behavior problems, parent perception of executive functioning). Process factors included family functioning (family cohesion, family resources, family satisfaction, social support and support satisfaction) and parent stress. In this model, context and process variables have direct and/or indirect relationships with both the proximal outcome and the distal outcome. Further PDS are theorized to mediate context and/or process variables on FQOL. The proposed study will explore the relationships of these variables in parents of adolescents/young adults. Specifically the study will evaluate how PDS relates to FQOL and determine if and how PDS influences relationships of other variables to FQOL. Understanding relationships will contribute to a theoretical framework of FQOL.

Hypotheses

Three hypotheses were proposed. If differences by presence of SB are identified further exploratory analysis will be conducted by subsample.

H_0 1. The context factors (demographic [child age, income, parent gender, race, ethnicity], presence of SB, child [parent perception of executive function]), and process factors (family functioning [cohesion, satisfaction, resources], parent stress), are related to the proximal outcome (PDS);

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H_0 2. The context factors (demographic [child age, income, parent gender, race], presence of SB, child [parent perception of executive function]), process factors (family functioning [cohesion, satisfaction, resources], parent stress), and proximal outcome (PDS) are related to the distal outcome of FQOL;

H_0 3. Parent depressive symptoms mediate the relationship of context and process factors to FQOL.

Methods

This secondary analysis was conducted on data from a cross-sectional correlational study of a sample of 209 parents of AYA, 112 parents of AYA with SB and 97 with AYA without SB from a multi-site study of adaptation in AYA with SB (Sawin, Buran, Brei, & Fastenau, 2003). IRB approval was obtained for both the original AYA adaptation study and secondary analysis. In the current study, available data included measures of the context and process variables delineated in the measurement model: Factors Related to PDS and FQOL (see Figure 1).

Participants

The convenience sample of AYA and their parents was recruited for the primary study from four children's hospital spina bifida programs in the Midwest and the Eastern United States (US). Comparison families were recruited by referral from SB families in the study, advertisement in each hospital and referral from primary care providers. Eligibility criteria included English speaking, families with AYA 12 to 21 years of age and without diagnoses of moderate or severe intellectual disabilities. The participants with SB had no major medical condition (i.e. life threatening, progressive, or

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incapacitation disability) unrelated to SB and the comparison sample had no major medical conditions (see Table 10 for characteristics of the sample).

Measures

The variables included in this study were guided by the study's conceptual framework and data available from the primary study are delineated in the Measurement model (see Figure 1) and described below. See Table 11 for internal reliabilities of scale scores in this study.

Context. Demographic variables were reported by parents on the Demographic and Clinical Information Form. Family income was reported as a four category variable (less than $20,000, $20,000 to less than $35,000, $35,000 to less than $50,000, or $50,000 or over). Race was identified by interviewee as Black, Caucasian, American Indian, or asked to specify if other and ethnicity, Hispanic or non-Hispanic. Child age was calculated using birth date and date of interview.

Presence of SB. Groups were identified as either SB present or comparison group by the primary study staff at each site.

Child factor. Parent perception of executive functioning (EF) was measured by Behavior Rating Inventory of Executive Function (BRIEF) (Gioia, Isquith, Guy, & Kenworthy, 2000). The 86-item BRIEF uses a response pattern from 1 (*never*) to 3 (*often*). A T-score correcting for age and gender is generated for The Behavioral Regulation Index (BRI) and the Metacognition Index (MCI). The first reflects the child's ability to control behaviors, inhibit behavior, shift between activities/situations and control emotional responses. The latter measures the ability to initiate activities, plan, organize, and monitor performance. The BRIEF was created for parent report of children

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up to 18 years of age. At the time of the original data collection there was no version of this measure for parents to report executive function for those over age 18. There is support from two studies that AYA with SB lagged 4-5 years behind their peers on autonomy, independence, cognitive processes and initiative (Davis, Shurtleff, Walker, Seidel, & Duguay , 2006; Holmbeck et al., 2002). Thus, the T-scores for 18 year olds were used by the original AYA adaptation study for those older than 18 years of age. Preliminary reliability has been established in this population, good internal consistency ($\alpha = 0.80 - 0.98$) and test-retest reliability ($r = 0.83$), (Gioia et al., 2000; Mahone, Zabel, Levey, Verda, & Kinsman, 2002; Ridosh et al, 2013).

Process. Family functioning was measured by three instruments, The Cohesion subscale of the FACES III, The Family APGAR, The Family Mastery and Health subscale of the Family Inventory of Resources for Management (FIRM). The first a well-established 10-item subscale of family cohesion addresses the families' closeness and shared values using response pattern from 1 (*almost never*) to 5 (*almost always*) (Olson, 1986; Sawin & Harrigan, 1994). Reliabilities have been high ($\alpha= 0.80 - 0.84$) in previous studies of AYA with SB (Sawin, Brei, Buran, & Fastenau, 2002; Ridosh et al., 2013). The second is a 5-item measure of family satisfaction (Smilkstein, Ashworth, & Montano, 1982) revised and simplified by Austin and Huberty (1989) uses a response pattern from 1 (*never*) to 5 (*always*). The scale measures satisfaction with family adaptation, partnership, growth, affection, and resolve. It has established reliability ($\alpha=0.71-0.91$), test-retest reliability ($r = 0.83$) and validity in the literature in families with AYA with SB (Bellin, Bentley, & Sawin 2009; Bellin & Rice, 2009; Bellin et al., 2010; Ridosh et al., 2013; Sawin et al., 2002; Smilkstein et al., 1982). The third is a modified 18-item

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subscale of family resources that reflects personal family and social support resources (McCubbin, Comeau, & Harkens, 1981). The investigators of the original study of AYA adaptation omitted two items from this scale based on low item-to-total correlations in previous work (Sawin et al., 2002). Reliability (α= 0.87-0.92) has been strong in families with chronic illness generally and specifically SB (Halvorsen, 1991; Knecht, 1991; Ridosh et al., 2013; Sawin et al., 2002; Sawin, Buran, Brei, & Fastenau, 2003).

Parent factor. Stress of everyday life was measured by a single-item rating of the parent's stress of everyday life from 0 (no stress at all) to 100 (very great stress). There is support in the literature for single-item measurement of concepts such as stress of everyday life (Gilliss, 1983; Knapp & Brown, 1995; Youngblut & Casper, 1993). This item was found to be strongly related to parent and adolescent outcomes (Sawin et al., 2002).

Outcomes. The Proximal Outcome, PDS, was measured by the Beck Depression Inventory-II (BDI) (Beck, Steer, & Brown, 1996). This 21-item scale measured presence and severity of different symptoms of depression in the last 2 weeks using response pattern from 0 to 3. Minimal depressive symptoms are defined as a sum score of 0 – 13, mild as 14 – 19, moderate as 20 – 28, and severe symptoms as 29 – 63 (Beck et al., 1996). There is strong support for validity and reliability (α = 0.90) in adults (Beck, Steer, & Carbin, 1988; Brouwer, Meijer, & Zevalkink, 2013). Cronbach's alpha was high (α = 0.88) in parents of AYA with SB (Ridosh et al., 2013), validity and reliability in parents of AYA without SB is unknown.

The Distal Outcome, FQOL was measured with a 3-item scale (parent perception of their teen's QOL, their own QOL and their FQOL) reported on a scale from 0 (poor) to

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100 (excellent). Factor analysis in a small related-study supported this single factor scale, which had a reliability of 0.84 (Ridosh et al., 2013). The use of a 3-item FQOL scale is further supported by a factor analysis using principal component analysis with Varimax rotation using data from the original AYA adaptation sample. All 3 items loaded on one factor (factor loadings were 0.94 parent's perception of family quality of life, parent's own quality of life 0.90 and parent perception of teen's quality of life 0.86), the Scree plot supported one factor and there was a single eigenvalue greater than 1.

Data Analysis

Power analysis indicated that with a medium effect size, $p = .05$, power of .80 and 16 independent variables (8 in each block of the hierarchical regression) a minimum sample of 116 will be needed for this secondary analysis (Soper, 2013). Data on 218 cases were evaluated and nine cases omitted, as they were missing data for three or more variables of interest resulting in a sample of 209 parents who completed the comprehensive study interview used for this analysis. Descriptive statistics were used to describe the sample and missing values analysis (MVA) was used to examine the patterns of missing data. At the item level, up to 3.3% missing data were identified in the dataset by MVA using SPSS (Version 22.0). There was no pattern to the missing data when explored by groups. Little's MCAR test was not significant therefore missing data were 'missing completely at random' (MCAR) and likely ignorable (Penny & Atkinson, 2011; Rigby, 2009; Tabachnick & Fidell, 2013; Verchota & Ke, 2014). There was no pattern to the missing values (except for the BRIEF discussed below) therefore 'casewise deletion' was acceptable (Rigby, 2009). The BRIEF scoring protocol indicated that for each person up to two missing items per subscale could be replaced with a score of 1 to calculate the

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scale raw score. This was accomplished with the BRI. The Metacognition Index (MCI) had more missing data than could be corrected per protocol. However, most items missing pertaining to engagement in school were not missing at random as they could be traced to cases with young adults not in school. In addition the MCI scale had good reliability ($\alpha = .96$). Thus, an exception was made for the two cases and the same replacement protocol (score of 1) was used with 3-4 missing items per subscale.

Chi-square statistic and independent samples t-test were used to identify any significant differences in the demographic characteristics of the sample between SB and comparison group to evaluate whether there was support to use the total sample for the multivariate analysis. Chi-square statistic showed no significant difference between groups (SB vs comparison) in age of AYA, parent interviewed, race/ethnicity, or gender of AYA in study. There was a significant difference between groups in scores for family income, χ^2 (207) = 16.67, $p < .001$. Thus, the total sample was used for the correlation and regression analysis and income was included as a control variable in step 1 of the regression. Preliminary analysis to evaluate the relationship among the context and process variables and their relationship to the outcomes was conducted using Pearson correlation coefficient for continuous variables and Spearman Rho for those with categorical variables (see Table 12). The preliminary correlations and theoretical framework were used to select the variables for the hierarchical multiple regression (HMR). Preliminary correlations between variables were analyzed. Both context and process variables were evaluated. Variables with significant correlations with at least one of the outcomes were considered for retention in multivariate analyses.

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Excluded from the regression were parent gender and ethnicity, which had little to no variability and not correlated to PDS or FQOL. Race and income did have variability but only income was significantly related to outcomes (r = 0.28 – 0.32), race was not therefore excluded from regression analysis. Income, AYA age, presence of SB and parent perception of EF (BRI and MCI) were the context variables included. Stress of everyday life and the three measures of family functioning were each correlated with the outcomes at r = 0.43 - 0.63, therefore all four measures were retained as process variables for analysis. This resulted in eight factors retained for the PDS analysis and nine for the FQOL analysis. See Table 12 for total correlations.

HMR analysis was then conducted using the total sample to address the hypotheses. The two regression analyses tested factors related to PDS and FQOL. To address the first hypothesis a HMR with PDS as the dependent variable was conducted by entering the context variables in block 1 then process variables in block 2. To address the second hypothesis HMR was conducted with FQOL as the dependent variable, context and process variables were again entered in block 1 and 2, and PDS was entered in block 3. Finally, variables were evaluated to determine if a context and/or process variable was significant in block 2 but not the subsequent block 3 when PDS was entered.

The relationships were evaluated to determine if criteria for mediation were met (Von Eye, Mun, & Mair, 2009). To test for mediation, relationship must be significant for three paths between (a) context or process factor (independent variable) and PDS (mediator); (b) PDS (mediator) and FQOL (dependent variable); and (c) context or process factor (independent variable) and FQOL (dependent variable) (Dudley & Benuzillo, 2004). If these criteria are met and if in the final regression the addition of

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PDS reduces the size of the relationship of one or more process variables to FQOL, then a Sobel test will be calculated to determine if PDS mediates context/process variable on outcome (Sobel, 1982; Von Eye et al., 2009). To test mediation using Sobel, regression coefficients and standard errors are derived from path a from independent variable to the mediator and path b from mediator to dependent variable accounting for independent variable (Preacher & Leonardelli, 2006). A significant Sobel test confirms mediation relationship. Full or partial mediation is determined by evaluating significance of path mediator to dependent variable accounting for independent variable and the standardized coefficient beta weight of the regression with mediator is less than without the mediator. If the path with the mediator is significant, partial mediation is supported. If path with the mediator is not significant, full mediation is supported (Von Eye et al., 2009).

If presence of SB is significant in the hierarchical multiple regression, exploratory analyses using the two subsamples of parents with and without SB will be conducted. Analysis will include evaluation of correlations and hierarchical multiple regressions exploring the relationship of context and process variable to outcomes by subsample. If different patterns by subsample are found, differences in study variables will be evaluated by independent samples t-test to better understand the patterns. With a medium effect size, $p = .05$, power of .80 and 10 independent variables a sample of 96 will be needed for the exploratory analysis. Both subsamples were adequate for this analysis (SB subsample $n = 112$; comparison sample $n = 97$).

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Results

Preliminary Analysis

Frequencies. In the total sample, primarily female (94%) parents were well educated either attending or completed college/vocational training (23%) and married (74%). Race and ethnicity lacked diversity with 3% Hispanic and 86% Caucasian, 11% Black, 3% other races. Just over half of the parents interviewed had an AYA with SB. The AYA mean age for the total sample was 15.2 years (see Table 10 for characteristics of the sample).

Demographic characteristics by subsample were similar except for income. Female parents were interviewed (SB group 94%/comparison 93%). Race and ethnicity lacked diversity with 4% Hispanic and 90% Caucasian, 5% Black, 4% other races in SB group. Comparison group race and ethnicity was slightly more diverse 2% Hispanic and 80% Caucasian, 17% Black, 3% other races. The AYA mean age for the group with SB was 15.1 years (σ = 2.9) and comparison was 15.4 (σ =2.6). Combined family income was significantly lower for families with AYA with SB, 18% earned less than $20,000 and 50% earned greater than $50,000. In comparison group 4% earned less than $20,000 and 73% earned greater than $50,000 (See Table 15 for Independent t-test results).

In the total sample, parents perceived slightly greater difficulty with child EF than parents of children in general population, about half a standard deviation difference in t-score mean (μ_{BRI} = 54.12, σ = 10.83; μ_{MCI} = 56.88, σ = 11.89). Parents reported having highly cohesive families (μ = 40.28, σ= 5.64, range 25-50). Family satisfaction was high, parents reported being almost always satisfied with the way their immediate family was available when help is needed, talked things over and shared problems, and expressed

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affection (μ = 4.13, σ = 0.62). Family resources such as flexibility, emotional support, cooperation were perceived as usually available to the family in the last year (μ = 3.13, σ = 0.46). The mean parent stress score was moderate in the total sample but had a large variance (μ = 53.33, σ = 26.32). Although the mean score reflected minimal depressive symptoms, the range was large (μ = 7.98, σ = 7.75, range = 0 – 46). FQOL in the total sample was rated high, in the upper quartile (μ = 85.62, σ = 13.23).

More parents of AYA with SB, 22%, experienced depressive symptoms in contrast to 14% of parents of AYA without SB. Nine percent of parents with SB experienced mild depressive symptoms, 10% moderate and about 4% severe depressive symptoms. In the comparison group 10% of parents experienced mild depressive symptoms and 4% moderate symptoms, none reported severe symptoms. Although FQOL was significantly different by groups, both groups reported high FQOL (SB group μ = 82.47, σ = 14.77; comparison group, μ = 89.25, σ = 10.10). Parents with AYA with SB had slightly lower FQOL with greater variance. See Table 15 for independent samples t-tests. See Table 11 for descriptive statistics of continuous variables.

Correlations. Bivariate correlations with outcome variables are described (see Table 12) for the total sample. The context factor correlations with significant but small relationships to PDS included child age, income, and parent perception of EF (both BRI and MCI). Income, presence of SB and parent perception of EF (both BRI and MCI) were related to FQOL. All process factors had moderate to large correlations to outcomes. Factors were inversely related to outcomes in the expected direction. Parent depressive symptoms were highly related to FQOL (r = - 0.54).

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Factors related to PDS and FQOL

The first hypothesis, testing relationship of context and process variables on PDS, was supported (see Figure 6). In the HMR first block, the context variables explained 26% of PDS with AYA age, family income and parent perception of EF metacognition index (MCI) were significant predictors. Presence of SB was not significant. Process variables added 12% of the variance. In the final block income, family resources and parent stress explained 38% total variance of PDS (see Table 13), age and parent perception of EF MCI were no longer significant when the process variables were entered.

The second hypothesis, testing the relationship of context, process and proximal outcome PDS variables and distal outcome of FQOL was supported. In the second HMR, the context factors in the first block, income and parent perception of EF MCI explained 22% of the variance in FQOL. When the process variables were added in the second block the context variables (income and EF) were not significant, presence of SB became significant. Significant process variables in the second block were family satisfaction, family resources, and parent stress that explained an additional 22% of the variance in FQOL. In the final block, presence of SB, family satisfaction, parent stress and PDS were significant and PDS added 5% of the variance in FQOL. This model explained 49% total variance of FQOL (see Table 14). Family resources subscale was the only variable to change significance when PDS was added therefore it will be used in evaluation. As SB was a significant variable in the FQOL regression, the proposed exploratory analysis was conducted to determine if there were different patterns of context and process factors related to FQOL for parents with and without an AYA with SB.

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Mediation Analysis. The third hypothesis, testing the mediation relationship of a context and/or process variable and FQOL was partially supported. The assumptions were met to proceed to Sobel test. The following relationships were significant: family resources (independent variable) and PDS (mediator), PDS (mediator) and FQOL (dependent variable), and family resources (independent variable) and FQOL (dependent variable). The relationship of family resources (independent variable) and FQOL (dependent variable) accounting for PDS (mediator) was significant and the beta weight (β) of family resources was smaller than without the mediator ($\beta = .55$ to $\beta = .38$). Partial mediation was supported and significant using Sobel test ($z = 4.56$, $p < .001$).

Exploratory Analysis of Factors by Subsample

When examined by subsample (families with an AYA with SB and the comparison sample), there were differences in the clinical context variables and the process variables (see Table 15). Differences exist for income, parent perception of EF (both BRI and MCI), family resources, PDS, and FQOL variables. In the correlation analysis the context factor relationships to FQOL were small (Spearman Rho 0.21 – 0.34; Pearson $r = 0.22 – 0.37$). In both subsamples, AYA age, parent gender, race and ethnicity were not correlated to FQOL. In the subsample with SB but not the comparison sample income was correlated to FQOL; parents with lower income had lower FQOL. Greater difficulty with child EF was related to lower FQOL in both groups. In both subsamples, all process factors were related to FQOL with mostly moderate to large correlations to outcomes. Stress of everyday life and the three measures of family functioning were correlated with FQOL ($r = 0.34 – 0.55$). Income and parent perception of EF were the

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context variables retained for the regression by subsamples while all four process variables were retained (See Table 16 for correlations by subsample).

In the HMR of FQOL for parents of AYA with SB, 18% of the variance in FQOL was initially explained by income. However it did not remain significant when the process variables that explained another 21% of the variance were added. Family satisfaction was the only significant process variable. In the final step of the model PDS added 8% explanation of the variance. In this final model family satisfaction and PDS were significant explaining 47% total variance of FQOL. PDS did not meet the criteria for a mediating variable (see Tables 17 & 18 for model summary of factors related to FQOL by group).

In the HMR of FQOL for parents in the comparison subsample, the only significant context variable was parent perception of EF explaining 17% of variance in block 1. However, this did not remain significant when the process variables were entered in block 2 contributing 31% variance. The significant process variables were family resources and parent stress. In block 3, the addition of PDS did not add any significant explanatory power to the model, R^2 change (0.008) was not significant (p = 0.235). In addition, depression itself was not significant. Thus, the results of block 2 are salient and this model explained 49% of the total variance in FQOL. The only remaining significant variables in the model were family resources and parent stress.

Discussion

Compelling findings in this study were the prevalence of PDS and FQOL as well as the different patterns of factors related to each of the outcomes in the total sample. Depressive symptoms were noted in 19% of parents in the total sample. This finding is

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higher than known prevalence of adults generally (9.1%) (Centers for Disease Control, 2010). Twenty-two percent of parents of an AYA with SB experienced depressive symptoms in contrast to 14% of comparison parents. This is similar to previous studies of parents who had a child with (19.2 - 48%) and without SB (11 - 25%) (Brei, et al., 2013; Holmbeck et al., 1997; Kronenberger & Thompson, 1992a, 1992b; Valença et al., 2012) .The BDI score (greater than 13) is delineated in the literature as the cut score for presence of mild depressive symptoms, 20 – 28 moderate, and greater than 28 severe (Beck & Steer, 1996). Only one study of parents of children with SB used a lower cut score of 10, which indicates minimal depressive symptoms (Valença et al., 2012). Detecting mild depressive symptoms can trigger screening and treatment at a point in the trajectory that prevents increasing severity of symptoms.

Difference of prevalence of FQOL in the two subsamples is a new finding. Only one other study reported findings of FQOL by subsamples (those in a waiver program and those in a wait-list registry) but it did not report whether the difference was significant (Eskow et al., 2011).

Perhaps the most striking finding was the difference in relationships between the context variables and the proximal and distal outcomes in the total sample and subsamples. Although the amount of variance explained by the context variables in the total sample was similar (26% in PDS and 22% in FQOL) income and not SB explained PDS while the opposite was true for FQOL. The presence of SB did not contribute any explanatory power to PDS when considered together with other context factors. The critical factor was income, a measure of socioeconomic status (SES). In another study using a well matched sample of parents with and without a preadolescent who had SB but

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no family income differences between groups, the presence of SB did predict PDS in fathers (Holmbeck et al., 1997). With the exception of one study (Valença et al., 2012) none of the other studies that supported a relationship between presence of SB or severity of SB and PDS also included income in analysis. Using a different measure of PDS and a different measure of SES (mother's education) a study of adolescents with SB found no relationship between SES and PDS (Brei et al., 2013). The choice of how to measure SES appears important. The inclusion of income as a measure of SES needs to be considered in all future investigations of PDS in families with child who has a CHC, especially SB. It is possible that there is a complex relationship between income and SB in that lower family income in families in SB subsample may be associated with loss of wages for the parent caring for a child with CHC or varied based on severity of the child's condition, neither of which were evaluated in this study and should be considered in the future.

A different pattern existed for the relationship of context variables with FQOL in the total sample. In the final model the only context variable that remained significant was presence of SB. The experience of caring for a child with SB requires parent's available time to maintain health of the child. This increased time can limit work opportunities and leisure activities and impact FQOL. Specific developmental issues such as the delay in achieving typical autonomy skills, the impact of learning issues on self-management of SB, the challenges in transition to adulthood, and the lack of employment in the SB population all have implications on the intensity of parenting, the expected trajectory of family life and the family's fiscal health.

The relationship between the process variables and the two dependent variables in the total sample were more similar. For both stress and a component of family

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functioning were critical factors. When families, had adequate resources and lower stress they were better able to adapt. Even in families with lower income and greater difficulty with child executive functioning, the process factors were protective and families were better equipped to handle life's challenges. Further, family functioning was consistently supported in the literature as related to PDS (Barakat & Linney, 1992; Brei et al., 2013; King, King, Rosenbaum, & Goffin, 1999; Kronenberger & Thompson, 1992a; Ulus, et al., 2012) and FQOL (Davis & Gavidia-Payne, 2009; Ridosh et al., 2013; Sawin, et al., 2002; Summers et al., 2007; Werner et al., 2009).

PDS was found to partially mediate the effect of family resources on FQOL in the total sample only. This relationship did not exist in the subsample. In the total sample, when depressive symptoms were present the impact of family resources decreased. The only other partial mediator found in the literature of FQOL is the family-professional partnership influence of service adequacy on FQOL (Summers et al., 2007). These findings suggest both support from family and support from others such as professional relationships do affect FQOL. A comprehensive approach to intervention would focus on enhancing family strengths and resources simultaneously with screening for and treating PDS.

The exploratory analysis revealed a somewhat different picture although the variables significant in the total sample remained significant in one or the other of the subsamples. Although parents in the SB subsample had a small but significantly lower FQOL scores, the models for both subsamples explained a similar amount of variance. Income was the significant factor for the SB subsample in the first step while EF (MCI subscale) was significant in step one for the comparison sample. In both, when process

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factors were entered the context factors were no longer significant suggesting that the process factors may mediate relationship of context factors and outcome. While PDS had a direct effect on FQOL in the SB subsample analysis it did not for the comparison subsample. For the SB subsample the most important factors to address are family satisfaction and parent depressive symptoms. More PDS and lower family satisfaction predicted lower FQOL. PDS for parents in the SB subsample may be a negative lens that can affect their ability to use family resources. This finding is consistent with the relationship of family satisfaction to FQOL in the literature (Davis & Gavidia-Payne, 2009; Jackson, Wegner, & Turnbull, 2010; Ridosh et al., 2013; Sawin et al., 2002; Summer et al., 2007).

The underlying factors of better family resources and lower stress were protective for parents in comparison group. Family resources remained a significant factor in the comparison subsample analysis of FQOL. The family mastery and health subscale of the Family Inventory of Resources for Management specifically addresses immediate family resources (only parents and children, not extended family, relatives or friends), specifically family strengths such as family decision-making, responsibilities, cooperation, perception of health of the family, and spending time together. The only family functioning indicator not significant in any of the regression analysis was family cohesion. Perhaps the effect of family cohesion was reduced as a result of parents' relationship with their adolescent and young adults as they gain more independence. In a sample of younger children, the effect of family cohesion may be more important.

The process variables significant in this study support the crucial role of family functioning. The family's ability to manage the increased demands of daily life requires

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strong family resources and positive coping strategies. If the family has family strengths and resources (satisfaction, emotional support, cooperation, flexibility) with which to address their challenges FQOL is higher. Congruent with review of literature, when families had positive family functioning, hopeful, experienced family-professional partnership, support from family and support from others they experienced better FQOL (Davis & Gavidia-Payne, 2009; Sawin et al., 2002; Summers et al., 2007).

It is interesting to note that neuropsychological functioning initially appeared to be salient for PDS and FQOL in the total sample but only in the comparison group in exploratory analysis. However, it did not remain significant once family functioning process factors were considered. Metacognitive executive function includes capacity for memory, planning and organization, the higher the score using the BRIEF tool the greater difficulty with executive functioning (EF). Other studies where neuropsychological functioning and child behavior problems were related to outcomes found family functioning important (Brei et al., 2013; King et al., 1999). Bivariate relationships of the indictors of executive functioning with PDS were in the expected direction. It is possible that family functioning mediates the impact of parent perception of EF on PDS or FQOL. Testing this potential mediation should be considered in later studies.

This study provides preliminary evidence on the somewhat divergent patterns of factors related to study outcomes in families who have an AYA with and without SB. Factors related to PDS did not vary by presence of SB while factors related to FQOL did. These findings provide direction for nursing practice and future research.

The theoretical framework of factors related to proximal outcome (PDS) and distal outcome (FQOL) was partially supported for the total sample. Context factors

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delineated in the model had direct relationships with both proximal and distal outcomes in the total sample. Support was also evident for the direct relationship of the process factors with the proximal outcome and distal outcome. The analyses were suggestive of a potential mediation of context-outcome relationship by process variables that needs to be explicitly tested in the future. The direct relationships of the process variables with the distal outcome were supported in both subsamples. PDS was directly related to FQOL only in the SB subsample. The proposed mediation by the proximal outcome on the relationship between process and distal outcome was only partially supported and only in the total sample. In contrast to the finding in the total sample, PDS was not found to be a mediator in the SB or comparison subsamples. Only process factors had direct relationships with FQOL in the comparison subsample. Preliminary evidence supported the potential mediation of context variables by process variables.

Implications for Nursing Practice and Research

Implications for Nursing Practice

This study suggests that integrating depression screening is indicated for primary care of parents with particular attention to parents who have a child or adolescent with a complex health condition such as SB. Early detection and treatment of depressive symptoms are needed to promote health and wellbeing of families. Only 1.4% of adults report depression screening was a part of their own primary care visit in 2010 (National Ambulatory Medical Care Survey, 2010, p. 19). As important, health providers of children with chronic conditions can conduct parent depression screening and referral in specialty practices; nurses can champion this effort. When a parent is determined to be at risk, nurses can conduct family assessment and provide practical supportive interventions

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to build and sustain family resources. Nurses can partner with families to identify goals and develop a treatment plan ensuring service adequacy and family-centered care.

Depression screening should especially target families at risk for low income such as single parents, unemployed, or those enrolled in public insurance program. Support for targeting low income families is noted across studies of families with children with SB (Barakat & Linney, 1992, 1995; Valença et al., 2012) as well as families generally or without a CHC. Families at risk for lower income because of job loss, continuing care needs, and restrictions in opportunities for employment require special attention. Preserving the ability to earn income is an important aspect of family life that is overlooked as priorities shift in caring for a newly diagnosed CHC in a child. Often loss of access to affordable health care occurs in times of transition when a family is adjusting to having a child with increased care demands. These are vulnerable periods in family life that could be better resourced by anticipatory guidance from providers. Health providers can play an important role in linking families to resources at these times to prevent loss of benefits, resources and work. Economic self-sufficiency should be a goal of the family that health providers support. Development of interventions addressing economic self-sufficiency, stress reduction and the strengthening of family resources are important, but effectiveness of utilization of available resources depends on identification and treatment of PDS.

The findings of this study show increased family resources are related to lower PDS. Parents with depressive symptoms may not be able to recognize and effectively utilize support from family and support from others. Nurses need to be vigilant for PDS that may prevent parents from utilizing resources needed for child and family health.

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Family centered care is an expectation of contemporary practice. Understanding a parents' perspective of FQOL is central to fulfilling this expectation. Individualized interventions that address building family functioning, specifically family strengths, satisfaction and resources are foundational to this effort. However, there are many roadblocks to implementing a family focus in health care delivery today. Systems interventions such as medical or health care home may be helpful. Community-based home visitation programs and early intervention programs can also address issues important to the family.

The instrument used in this analysis can serve as a clinically relevant short summative outcome measure of FQOL. Assessing the overall appraisal of the domains of life that are important to the family can be a way for health providers to measure the effectiveness of their interventions. Using a standard outcome measure can also help providers refine dosage and timing of interventions.

Knowing factors related to FQOL by subsample helps providers tailor strategies to daily life of parents in a meaningful way. Reducing stress of daily life by identifying practical solutions to problems, enhancing communication within a family, promoting shared decision-making in the context of their lives to increase satisfaction is essential. In families with AYA with SB understanding risk factors for depression will help to focus interventions for those families. Screening and treatment of depression remains especially important in parents with AYA with SB. Research of families must continue to discover factors that explain quality of life, in the context of the family—strengthening FQOL for families with and without CHC.

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Implications for Future Research

While the study model explained a large portion of variance of PDS and FQOL in families with an AYA with and without SB, more than half of the variance remains unexplained. Expansion of both context and process variables examined would be crucial to future research. The study results suggested that process variables may mediate the impact of context variables on outcomes. Further research should explicitly test this hypothesis. SB as a CHC appears to have a major impact on FQOL. Confirming this relationship in other larger more diverse samples of families who have an AYA with SB is indicated. Further, determining if FQOL differs across CHC and developmental stages of the child needs to be explored. Results of additional analysis are foundational to the development and testing of individualized interventions for families with children and AYA. Specifically, further investigation of the role of parent perception of EF on PDS and FQOL should be explored.

Measurement of several variables can be strengthened. The variable of parent perception of EF could be strengthened by obtaining parent report of young adult EF. In addition, other measures of metacognition may be helpful to understand the impact of EF on outcomes. The variable "presence or absence of SB" is limited and inclusion of a measure of severity might be helpful in future investigation of FQOL for the SB sample. In addition, parent leisure and socialization measures are needed to better understand the protective influence of leisure activities on FQOL outcomes. Significance of leisure activity was limited to a count of number of days of leisure a month or as an indicator of socialization, how many days family left home after surgical procedure. Leisure and socialization may be two different concepts, leisure time with others rather than in

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isolation may help to build and sustain resources. As important, leisure time would promote self-care activities for the parent and relieve stress.

This study provides further evidence of the psychometrics for a 3-item measure of FQOL in families with an AYA who has a CHC and preliminary evidence in families without a CHC. In a few studies of families with children with SB, FQOL measured with single item and 3-item scale was found to be high (Ridosh et al., 2013; Sawin et al., 2002). Factor analysis provided support for validity of the measurement of FQOL and Cronbach's alpha (0.88) supported its reliability in this sample. Future studies addressing both stability and construct validity are needed. Both confirmatory factor analysis and use of this measure in families with children who have other CHC could address the latter.

Future research should consider family resources' direct and indirect effect on FQOL. Mediation may be carried by patterns (Von Eye, Mun, & Mair, 2009) such as adolescent beliefs. In the literature there is support for AYA beliefs affecting parent perception of FQOL. Future expectations of AYA were strongly related to FQOL in a study that also found parent depressive symptoms strongly related to FQOL in parents with AYA with SB (Ridosh et al, 2013). These variables were also related to FQOL outcomes in a small study of young adults with SB (Sawin, Whitmore, & Ridosh, 2013). Other variables such as parent and AYA perspectives of future expectations and beliefs such as attitude, self-efficacy, perceived health competence, and perceived severity of SB may be explored in multivariate analysis to more fully explain PDS and FQOL.

Lastly, a recommendation for future research is to consider identification of barriers to depression screening for parents in a variety of settings. Prevention and risk reduction strategies must be piloted to better inform policy makers of return on

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investment of interventions. For now, enhancing family functioning, management of stress and PDS, and improvement of family's ability to generate and utilize family resources is the priority.

Limitations

Secondary analysis limitations need to be considered. Measures are limited by data collected and restricted to sampling method and size determined by original study investigators. An *a-priori* sample size calculation determined power would be adequate for a specific number of predictors desired for testing in the total sample. The calculation determined a sample size of 200 would be adequate for desired power (Soper, 2013).

The data available for cross-sectional analysis was collected for primary study of AYA adaptation that limited ability to evaluate all possible parent factors identified in the literature review. The cross-sectional data did not allow for evaluation of causal relationships. Order of entry of variables in blocks, categories of variables in hierarchical regression determined by theoretical framework guiding the study were limited by the assumptions of a linear relationship between context, process, and outcome. Reciprocal relationships, although potentially present, were not hypothesized in this study. Future studies can be designed to examine causal relationships over time. This would be important in testing FQOL as an outcome measure for family-centered interventions.

The original AYA adaptation multi-site study used a convenience sampling method. This sample represented families whose AYA with or without SB had no intellectual disabilities. Thus, it may not be appropriate to apply these findings to families of AYA with intellectual disabilities. In addition, the results of this study may only be applied to parents of AYA. The original AYA adaptation study was limited by its

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recruitment of the comparison sample. Although the characteristics of the sample were only significantly different by income, other differences not measured may also be different and affect the findings. Attention to detailed family match exemplified in Holmbeck et al.'s (1997) work should be considered for future studies.

These data were limited by self-report from one informant, mothers of AYA. Although a single reporter does not capture two-parent family perspectives, the FQOL outcome measure did elicit a perspective of two family members (parent and child) and the family as a whole, from the one parent's perspective. Although gender or parent role differences may remain unexplained by this approach, understanding the family outcome from the perspective of the one parent in this analysis was congruent with research question exploring outcomes of PDS and FQOL.

The measurement of parent perception of EF may limit study findings. This instrument has been validated and normative data exists for parent report of EF for individuals 5-18 years of age. The use of the 18 year olds normative data to compute T-scores for AYA 19 years and older may under or over represent EF problems in 16% of the sample. However, there was no support for the relationship of EF to age that provides some evidence that use of the normative values for 18 year olds is reasonable.

Family income measurement needs to be further explored. Income did not take into account family size, poverty level or other indicators of socioeconomic status.

Although these data will better inform relationships of factors related to FQOL, further understanding may be enhanced by including other salient concepts. Factors such as performance measures of executive function, and other adolescent and parent beliefs

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such as perceptions of health and future expectations of adolescents should be considered for future studies.

Conclusion

This study provided tentative evidence for understanding patterns of factors associated with outcomes of PDS and FQOL in families with children. A theoretical framework of FQOL explaining the relationships between context factors, process factors and the proximal and distal outcomes was partially supported. Expanded testing of the proposed model is indicated. Implications of this study for parents with adolescents include an understanding that family satisfaction and parent depressive symptoms are important factors related to FQOL in families of AYA with SB. For the comparison subsample, resources and stress were the significant factors. Optimizing outcomes for all families with AYA include attention to strengthening family resources to enhance the quality of their family lives.

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Chapter 5

In parents of children with SB, up to 48% of parents can be at risk for PDS (Ridosh, Sawin, & Klein-Tasman, 2014). Depressive symptoms in current study above the cut score of 13 were reported by 22% of parents with SB and 14% of comparison group. Consistent with a similar finding in a large Canadian population health study, parents of children with health problems (non-specified) were over twice as likely than parents of children without conditions to report high depressive symptoms (*OR* =2.48; 95% CI =1.40, 4.40) (Brehaut et al., 2009).

This study contributes evidence of a unique finding in parents of AYA. In the total sample, resources and PDS were significant and parent depressive symptoms (PDS) partially mediated the relationship of family resources on family quality of life (FQOL), but not in the subsamples. As parents experience depressive symptoms, it is possible that parents with PDS are less able to recognize, build, and/or utilize resources. Family-professional partnership (family relationship with health providers) is the only other partial mediator of service adequacy on FQOL identified in the literature of families with children with intellectual disabilities (Summers et al., 2007). However, in subsample analysis, only parent factors (satisfaction and PDS for those with an AYA with SB and resources and stress for parents without AYA with SB) were significant. These findings support the importance of addressing internal and external family resources especially in parents with depressive symptoms or at risk for depression.

Underlying differences between groups in family resources and parent depressive symptoms may explain why PDS was not significant in comparison group in FQOL regression. PDS and family resources were significantly different by group and variance

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of PDS was larger for the comparison group. Family resources had a lower beta value in the group with SB than comparison group (0.16 versus 0.25) in the second step of the regression analysis. Beta weight for PDS was higher in the group with SB (-0.36 versus -0.12).

For parents in this study presence of SB, family stress and PDS were related to FQOL. The family satisfaction finding is supported by other findings in families with AYA with SB in which family satisfaction and parental hope were predictors of overall FQOL (Sawin, Buran, Brei, & Fastenau, 2003). Other studies have found aspects of family functioning (family satisfaction, family resources, family relationships) consistently correlate with FQOL (Ridosh, Sawin, & Brei, 2013; Werner et al., 2009).

Support from family and professional family centered support were predictors in an early childhood study (Davis & Gavidia-Payne, 2009). One study identified stress of everyday life and presence of SB as related to FQOL in parents of AYA (Sawin, Brei, Buran, & Fastenau, 2002). PDS was only evaluated in one other study of families with AYA with SB and found highly related to FQOL (r = -0.72). Significance of stress as an important variable is consistent in two studies of PDS outcomes (Holmbeck et al., 1997; Kronenberger & Thompson, 1992b).

Findings of this study are consistent with components of the Transactional Stress and Coping Model. According to the model, stress appraisal and expectations of efficacy of locus of control, methods of coping, and supportive, conflicted or controlling family functioning patterns of the individual and family have an impact on adaptation more so than severity of illness or socioeconomic status (Thompson & Gustafson, 1996). Severity of illness was not a variable in the study but presence of SB was significant. Process

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variables took away the explanatory power of income in the second step of all regressions except for comparison group. When families are satisfied with their family functioning, the status of their income does not appear to affect their FQOL. Stress and family function (family satisfaction) were important predictors of FQOL as in the Thompson and Gustafson (1996) model. This model has also been tested in families with children with sickle cell disease, insulin-dependent diabetes mellitus and in families with children with chronic conditions compared to health controls (Hocking & Lochman, 2005; McClellan & Cohen, 2007). Thus, the findings support the possibility that other CHC may also affect FQOL.

Theory

This dissertation generated a theoretical framework of factors related to FQOL from the results of the study (see Figure 6). The framework identifies and explains relationships between variables.

Study results supported the potential of process factors mediating relationship of context factors and outcome. These relationships were similar to the relationships proposed by Thompson and Gustafson (1996) and Sawin et al. (2003) and need to be further tested. However, if the ability of process factors to mediate relationship of context and outcome relationship is supported it further expands the importance of the perception of how the family is doing with the condition, not the condition itself that is critical in FQOL.

Further, results only give weak support to the proposition that PDS mediates relationship between one process variable and FQOL. Only one potential mediation relationship met criteria for Sobel testing and that relationship was only partially

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mediated. It may be that PDS and FQOL are unique outcomes. Further testing of the outcome is needed.

Practice

Practice implications for families of children with conditions include primary and secondary prevention strategies. The following will address nurses' contribution to care of parents and their children—family. While the nurses' role will be addressed, it is important to note approaches would require involvement from multiple disciplines. Primary prevention strategy discussion will include addressing parent depression, and family assessment. Secondary prevention includes screening, early detection and treatment if depression is present and intervention to build family strengths.

Primary Prevention

The goal of primary prevention is to provide services or programs that prevent the occurrence of PDS and prevent problems in FQOL. Well child and community based efforts to strengthen families, enhance family resources and increase skill in dealing with stressful situations can provide parents with tools to prevent PDS and enhance FQOL. Identifying for parents and family members the risks of low income, difficulty with child executive functioning and presence of SB (or other CHC) could have on both their own mental health and the well-being of their family. Raising parent awareness of risk factors and stressors faced by families can enable parents to accept anticipatory guidance and seek help early from their support resources whether from family or support from others such as professionals.

Primary prevention would be services or programs that enhance family strengths and family functions (perceptions skills and abilities that provide protection from PDS

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and enhance FQOL) such as family satisfaction, family resources, and reduction of stress. Family assessment is needed to identify family strengths and deficits. Understanding family functioning will facilitate the contracting process with families to set goals, suggest practical support strategies based on priorities and preferences of the family. Resources can include coordination of care, identification of internal and external support, strategies for effective communication, parenting and/or coping skills. Specific strategies should match family needs as determined by the family and in anticipation of needs based on condition and age of the child to promote self-management, family and community connectedness.

Nurses must be knowledgeable of existing local support resources and their effectiveness to make recommendations. Nurses can serve as a professional resource in community, primary care and or case manager depending on role. Continuous contact with families with children with CHC establishes relationship and facilitates coordination of care. In an effective case management program, the nurse case manager has structured frequent (i.e. bi-monthly) contact with families in their homes if they have complex needs. The nurses provide education, monitoring of child's health and regimen to ensure best practices for condition. The nurse is then an available resource for a parent to call in case of questions during an acute illness, complication, or change in condition. For internal support, nurses' visit in the home is ideal to understand family environment, identify strengths and areas for improvement in care, safety, parenting, and stress management.

When families are ready to contact and engage external resources, a specific resource is a local chapter of an association unique to the condition such as the Spina Bifida Association. This resource can provide specific information for parents and

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children that are developmentally appropriate and include support groups. Local groups of parents of children with conditions also can connect through the Parent to Parent network. This is a volunteer organization whose mission is to match veteran parents of children with disability with a parent in need of support. The organization trains veteran parents to function in a facilitative support role addressing resources. For families with young children, early intervention programs such as Early Head Start, Head Start, and Nurse-Family Partnership programs address parents through interventions specifically addressing parenting and parent-child connections. For families with AYA, parent support groups may be affiliated with a faith community or child's school. Nurses can help parents to identify and access their community resources based on what is important to the family.

Secondary Prevention

Evaluation of families can begin with a preliminary screening of FQOL in clinic settings. The outcome measure for FQOL in this study is a practical 3-item measure that can help providers assess how families are doing with their global overall FQOL. Using a threshold of upper or lower quartiles as an initial screener can help providers identify families who need further assessment. The broader tolerance allows for a parent report less than 50 to trigger other screenings or interventions such as referral for a specific service such as case management. Additional screening could follow with a standardized tool. FQOL may also be an outcome measure to monitor families at times of transition such as during or after hospitalizations. Understanding perceptions of FQOL may help develop and refine dosage of family interventions.

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A structured intervention is proposed to address each component of the findings from this study. Nurses can develop and implement a multipronged intervention called Family PCS (Parent depression, Cognitive restructuring, Social support for FQOL). This would involve development of partnerships with health providers to include advanced practice nurse, physician or mental health provider equipped to provide mental health care treatment and follow-up care. Next, the healthcare team can develop content for sessions with parents to appraise and reframe stress perceptions, establish social support and support from others. The sessions can focus on topics of stress appraisal and cognitive restructuring with practical examples from family circumstances. Parents can identify internal and external supports and set goals to try a new support such as asking a family member to complete a new task, attempt a new leisure activity, or meet someone with a child with same condition for example. These sessions should focus on parent perceptions of being supported, belonging, stress reduction, and effective coping patterns. Goals of treatment would be to enhance family decision-making and adaptability for a sense of mastery. Sense of belonging and helping each other in the family creates better family mutuality in which emotional support, togetherness and cooperation are part of the family process. Trying a new leisure activity and community engagement promotes non-health-related activities to develop positive health behaviors for parent's own physical and emotional health.

Additionally, evidence from this study identified income as a predictor of PDS. Screening should target at-risk families earning low income. Families at-risk include single parents, parents with less education, and change in job status. In these families at risk for low income, first identification and treatment of parent depression should occur,

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and then family resources and stress can be effectively addressed. Although findings suggested family satisfaction is more important than low income in families with AYA with SB, families may have available resources but not recognize or utilize them if PDS is present and not treated. PDS did influence the relationship of resources and FQOL. Before formulating a plan for utilization of resources, a family assessment is needed and helping families maintain economic self-sufficiency should be a goal of care. In particular, screening and family assessment is indicated in at-risk families with an AYA with SB, whether in primary care setting, hospital, clinic, or specialty service provider setting.

This study provides preliminary evidence for health providers to begin to integrate depression screening in parents of adolescents, adult and pediatric primary care and specialty care practice. In the current study all parents with and without AYA with SB were at risk for PDS. However, only 1.4% of adults report depression screening was a part of their primary care visit in 2010 (National Ambulatory Medical Care Survey, 2010, p. 19). Evidence of PDS mediation relationship between parent perception of internal family resources and FQOL supports practice of depression screening with particular attention to parents who have a child or adolescent with a CHC such as SB. Parents of children with SB do have significantly higher PDS. However, this higher rate of PDS was not explained by presence of SB but by income while controlling for other child variables such as child age and parent perception of EF.

Depression screening process can be can be facilitated by nurses in the pediatric primary care setting. A toolkit available through the Commonwealth Fund, Dartmouth Institute's *Parental Depression Screening for Pediatric Clinicians Implementation*

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Manual guides the process by engagement of nurses, health providers, staff and parents during well-child visits. This process resulted in 70% rate of screening and positive depression screen in about 1 in 20 mothers (Olson, Dietrich, Prazar, & Hurley, 2006). Nurses can assist training of staff to prompt purpose for yearly screening during parent check in, remove any barriers to screening and follow up with appropriate resources. Nurses can develop the screening criteria, monitoring, communication plan, and link parents to resources.

Secondary prevention includes diagnosis, screening and early treatment and service support for parent depression. When a parent is diagnosed with depression, it is known this will affect FQOL and a barrier for parents' ability to use available resources. Screening will improve early detection of mild depression to minimize the progression or consequences of undetected depression and cumulative effects of stress. Treating depression is therefore essential to the care of parents with a child with chronic condition such as SB.

Research

Future research should continue to explore factors related to outcomes, conduct psychometric testing of The FQOL Scale, and design intervention research. See Figure 2 for factors associated with PDS and FQOL from syntheses of literature for key variables to explore in future research.

Further identification of factors that contribute to direct and indirect effects on FQOL is needed. Research questions may include the following. Does process mediate the relationship of context to outcome? What other context and process factors such as coping, parental hope or time to pursue leisure activities influence FQOL through PDS?

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Specifically, leisure activities were ranked low in attainment in studies using the FQOL 2006 Survey in US and international samples. The sample from Canada reported a large relationship between leisure and FQOL. This specific variable is one that can be better understood.

Support to investigate family functioning as a potential mediator of parent perception of EF on PDS was found in the regression of total and subsample. Parent perception of EF was significant in the first block, not the second block and the beta value decreased in total sample and in the group analysis. Further analysis is indicated to understand the mediation role of family functioning on PDS. Previous research found the interaction of adolescent neuropsychological functioning and family functioning variable did not moderate PDS (Brei et al., 2013), perhaps it is a mediator or suppressor of PDS. Since PDS is a mediator of FQOL, it will be important to further investigate the relationship of child executive functioning, family functioning and PDS.

The relationship of income as a predictor of PDS is unclear. Since income did not include indicator of family structure or poverty level, other aspects of income may explain relationship. Inclusion of income and other indicators of socioeconomic status in a matched sample are needed. Perhaps a cluster analysis may identify groups of variables stratified by income levels.

Measurement of executive function was limited by the tool used. First, normative values for 18 year olds were used that may not be valid. Second, parents may not be the best reporter of young adults executive functioning. Perhaps other measures of parent perception of EF could be used to address the transition age children who no longer attend a structured school day. Other measures of executive functioning, especially

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clinical evaluation, may better capture specific metacognitive processes in addition to behavioral regulation.

Future research should focus on patterns of relevant factors not explored in this study such as child and family factors from multiple informants when possible. What other factors as reported by adolescent such as health beliefs and future expectations impact PDS and FQOL? AYA report of future expectations was strongly related to FQOL (Ridosh et al., 2013). In a sample of young adults attitude, communication and problem solving, perceived health competence and health status, amount and satisfaction with responsibility taken for self-management related to a single item measure of young adult report of FQOL (Sawin, Whitmore, & Ridosh, 2013). Understanding what works well for young adults can help guide research for families with adolescents. The process category of variables may be enhanced by inclusion of AYA future expectations and other beliefs such as attitude, self-efficacy, perceived health competence, and perceived severity of SB to more fully explain FQOL.

If longitudinal analysis is possible, what are the context and process factors that affect depression and FQOL over time? Only one study examined child adjustment variables and depressive symptoms over a two-year period. Exploring known related variables over time will identify predictors and causal factors important to understand. No studies examined FQOL over time. Therefore longitudinal research using The FQOL Scale as an outcome measure over time will help to evaluate predictors across the life course to better establish best practices for parents of children in specific age groups. This research can guide policies to ensure resources and service adequacy for parents.

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What is the reliability and validity of the FQOL 3-item measure in other samples of parents of children or adolescents with CHC? Psychometric testing of the FQOL Scale is warranted to strengthen construct and discriminate validity of the measure. Confirmatory factor analysis and test-retest are next steps for further development of this measure. Use of the FQOL Scale as an outcome measure in families with children with and without conditions and asking other questions perhaps about differences between simple and complex conditions or inclusion of perceptions of other family members help to develop this measure. Intervention programs for parents with depressive symptoms can use The FQOL Scale as an outcome to evaluate FQOL to determine efficacy of treatment and monitor progress.

Research addressing both domain-specific and overall global FQOL would be fundamental to understanding which domains are important to families and the factors related to them. The domain-specific measures would provide direction for development of interventions, while overall global measure of FQOL determine state of FQOL and monitor progress of interventions. Additionally, both a broad global measure of PDS and specific measures would provide useful data for interventions. Measures such as the global items of patient-reported outcomes measurement information system (PROMIS) (Hays, Bjorner, Revicki, Spritzer, & Cella, 2009) can be compared to other standardized measures. Input from different family members in analysis while maintaining unique perspectives such as cluster analysis. Identifying family member agreement in variables may be useful to identify patterns of variables in family types related to outcomes.

Future descriptive research of predictors of PDS and FQOL will inform development of interventions for families with and without adolescents who have SB to

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improve their health and well-being. Family intervention research is needed to build knowledge of effective ways to provide primary and secondary prevention. Interventions such as Family PCS (Parent depression, Cognitive restructuring, Social support for FQOL) would require an interprofessional team to implement and evaluate. Partnerships between health providers to include advanced practice nurse, physician or mental health provider will ensure strategies address multiple components of education, counseling, treatment and follow-up care. Intervention must specifically address building positive family functioning patterns and building internal and external support structures, and enhancing communication to access and utilize social support systems.

Policy

Sufficient evidence is available to recommend depression screening of parents in primary and secondary care settings. This study provides evidence that PDS is one of the factors that influences quality of life outcome for families. Although family satisfaction was also predictive of FQOL in parents of children with SB, efforts of healthcare providers may prove futile when PDS is present. Internal and external support resources must be in place to experience greater family satisfaction and lower stress then depression. Then management of PDS is critical while continuing to facilitate management of family resources.

The recommendation to screen adults in primary care settings aligns with the U.S. Preventive Services Task Force (USPSTF) recommendation and contributes to an objective of the Healthy People 2020 initiative (U.S. Department of Health and Human Services, 2010; U.S. Preventive Services Task Force, 2009). The USPSTF specifically recommends staff support to respond when screening is positive with diagnosis, treatment

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and referral to mental health services as needed. The Community Preventive Services Taskforce in 2010 recommended a collaborative care model as an evidence based team approach to manage care of the depressed adult in partnership with case managers, primary care providers and mental health specialists. A primary care practice can identify other providers who would collaborate to provide services and disease management can be provided by a nurse case manager.

Reimbursement for parental depression screening, diagnosis and treatment is covered as a result of The Patient Protection and Affordable Care Act (2010) (PPACA) law, effective January 2014. The Affordable Care Act built upon the Mental Health Parity and Addiction Equity Act of 2008 includes mental health benefits, an Essential Health Benefit must be offered by all new small group and individual market plans. This coverage ensures federal parity protection, commensurate with medical and surgical coverage (Beronio, Po, Skopec, & Glied, 2013; The Patient Protection and Affordable Care Act, 2010).

Consistent measurements of depressive symptoms are needed for effective clinical management and research. The review of literature of parent depression identified a variety of measures that made synthesis of findings difficult. Further research will rely on comparable measures for meta-analysis to monitor prevalence, differentiate aggregates at risk and track effectiveness of interventions. While the USPSTF does not recommend one depression screening tool over another, the ability to identify and then monitor progress of treatment will rely on a consistent measureable outcome. The USPSTF recommends two questions as an initial screening (a) “Over the past 2 weeks, have you felt down, depressed or hopeless”; and (b) “Over the past 2 weeks, have you felt little

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interest or pleasure in doing things?" followed by full diagnostic interview using DSM criteria if positive. Identifying presence and severity of symptoms using a standardized valid and reliable tool such as BDI would facilitate screening and monitoring of progress. The BDI is a self-report questionnaire of the last 2 weeks of symptoms best aligned with diagnostic criteria such as sadness, pessimism, loss of pleasure or interest, changes in sleep and appetite, feelings of worthlessness, concentration difficulty, agitation and irritability (American Psychiatric Association, 2013). Primary care can then use this measure to monitor individual patient progress. To monitor population prevalence of depressive symptoms, the Behavior Risk Factor Surveillance System (BRFSS) developed an additional optional module questionnaire in 2006 of the last 2 weeks of symptoms of depression and anxiety (Centers for Disease Control, 2013). This BRFSS data will be invaluable to evaluate disease rates and disparities across the US.

Needs of parents who have children with SB in particular must be addressed in the specialty clinical setting. Sufficient evidence from this study supports the screening of parents to occur in the specialty clinic as way to ensure parents with children with CHC are identified when in contact with providers. Nurses could support practice of attending to parents in the clinic setting as a collaborative model of care. This model would need to include billing for services of the parent in addition to child with a plan for continuity of care when screening is positive. Currently reimbursement mechanism does not exist to reimburse for parent screening in the specialty care clinic and payment for screening is only once a year.

The SBA national resource center currently provides information to parents about depression in the children (Spina Bifida Association of America, 2014). This resource

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can target parents by expanding repository of educational materials raising awareness of risk of depression. The parent resources on the SBA website could include contacts for mental health services. Identification and treatment of depression will enable parents to build and establish the support systems they need within and outside of their family units. It is by preventing and treating depression and strengthening resources, the health of their family will be optimized and their family quality of life enriched.

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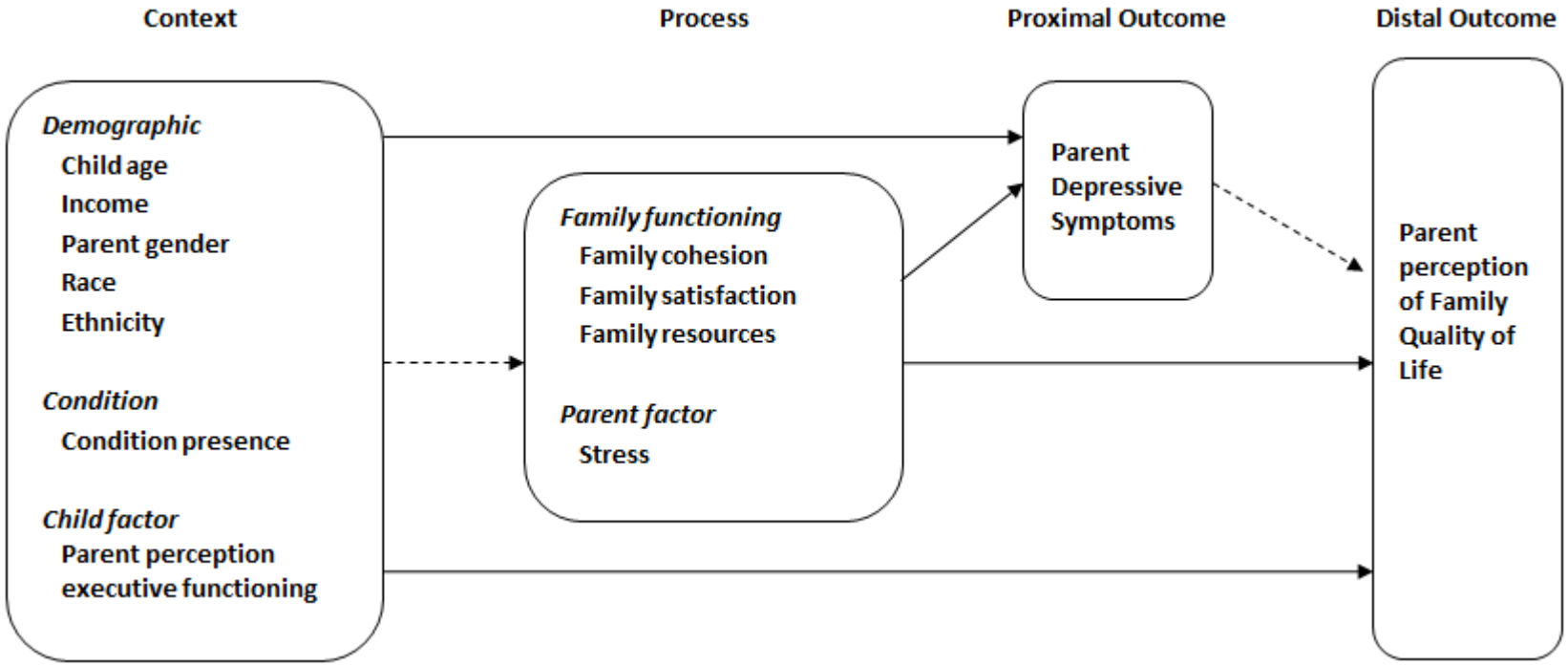

Figure 1. Measurement model: factors related to PDS and FQOL. Factors selected from two syntheses of literature: Depression in Parents of Children with Spina Bifida: a review of literature and Family Quality of Life: a review of literature. Solid lines are relationships between concepts with empirical support. Paths with theoretical support are represented by dotted line.

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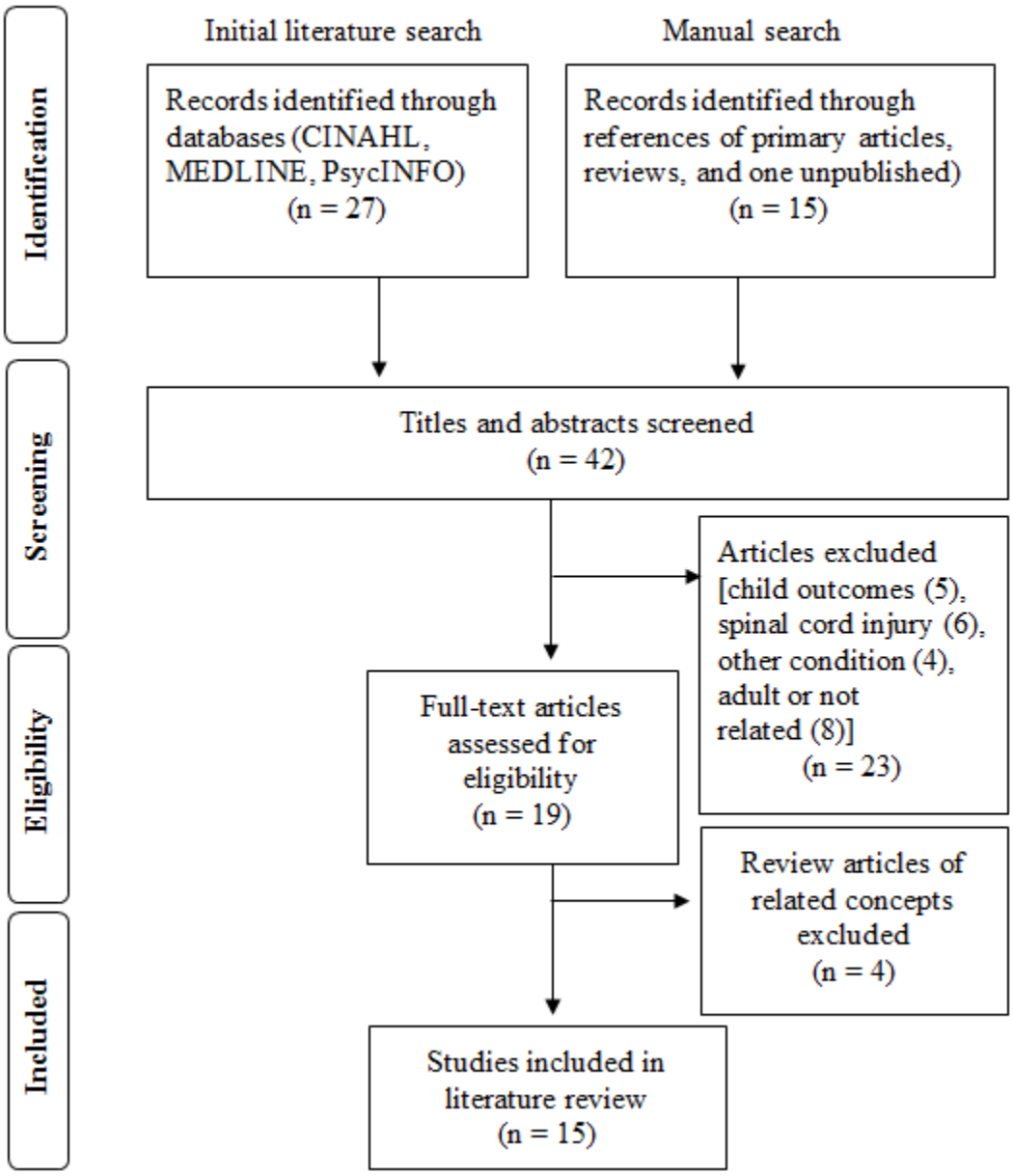

Figure 2. Flow Diagram of Search Strategy for Depression Review of Literature. Adapted from Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). *P*referred *R*eporting *I*tems for *S*ystematic Reviews and *M*eta-*A*nalyses: The PRISMA Statement. PLoS Med 6(6): e1000097. doi:10.1371/journal.pmed1000097.

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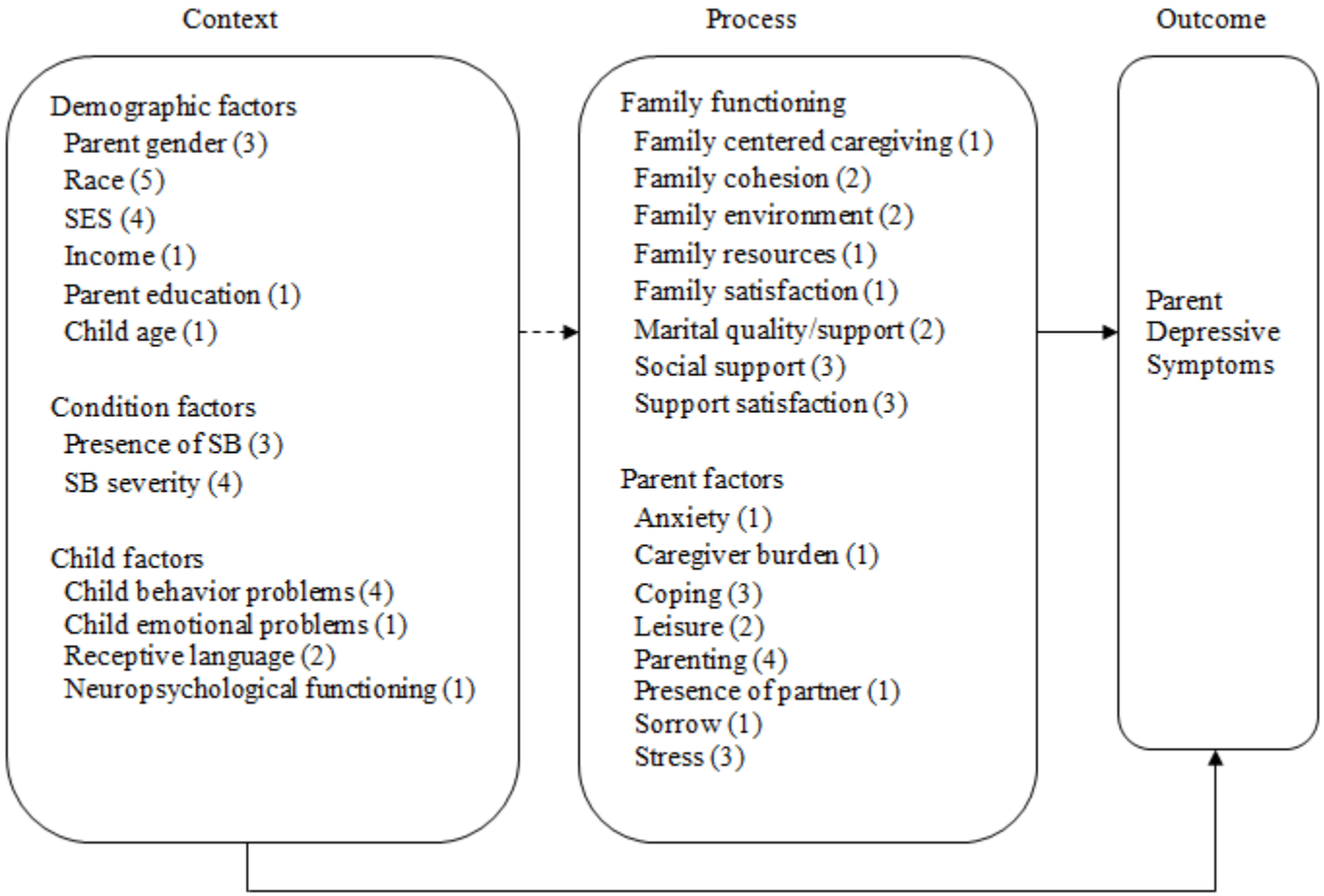

Figure 3. Factors related to PDS Identified in the Synthesis of the Literature. Only significant context and process findings are reported ($p < .05$). Number of studies evaluating concepts are identified.

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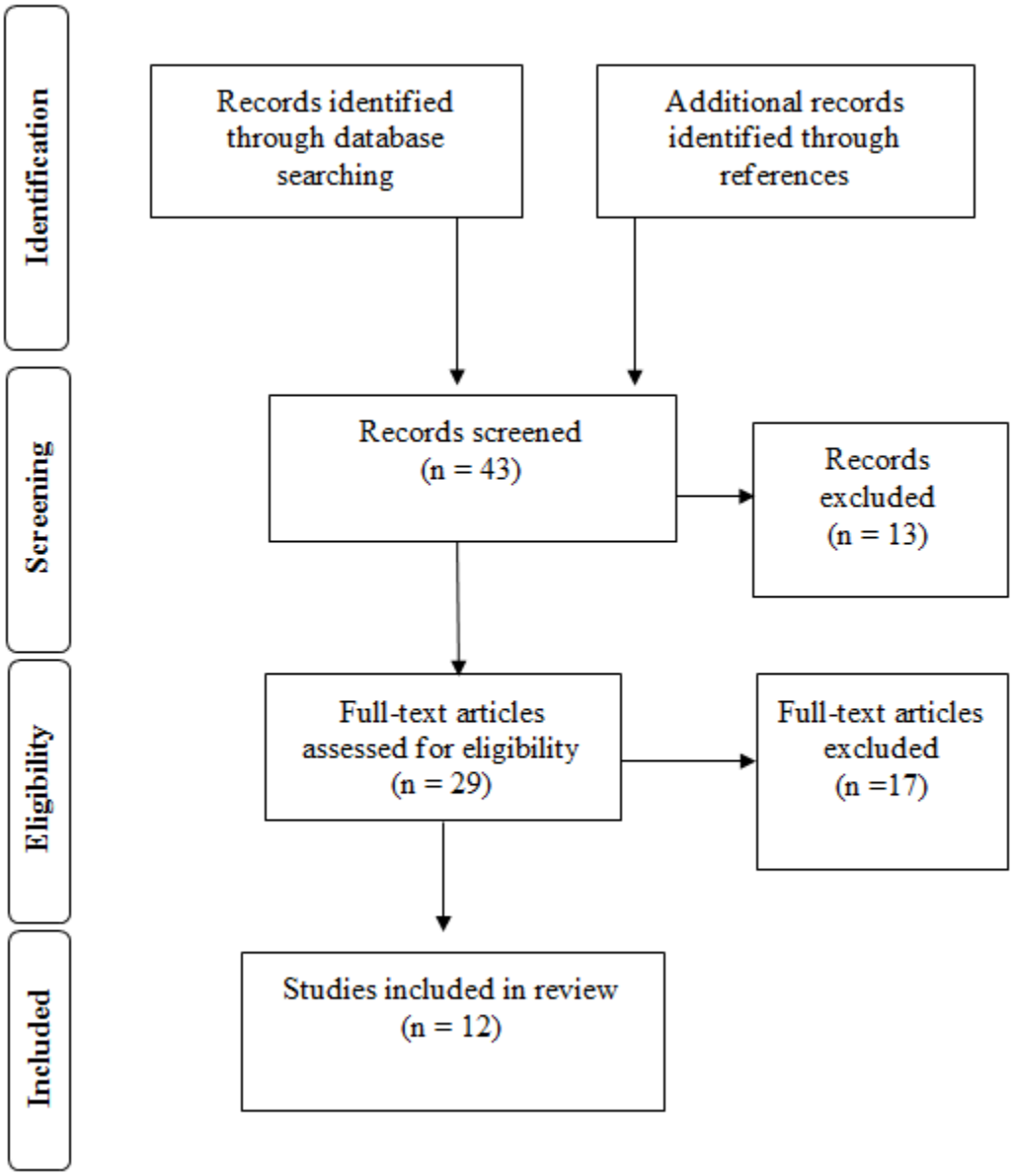

Figure 4. Flow Diagram of Search Strategy for FQOL Review of Literature. Adapted from Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). *P*referred *R*eporting *I*tems for *S*ystematic Reviews and *M*eta-*A*nalyses: The PRISMA Statement. PLoS Med 6(6): e1000097. doi:10.1371/journal.pmed1000097.

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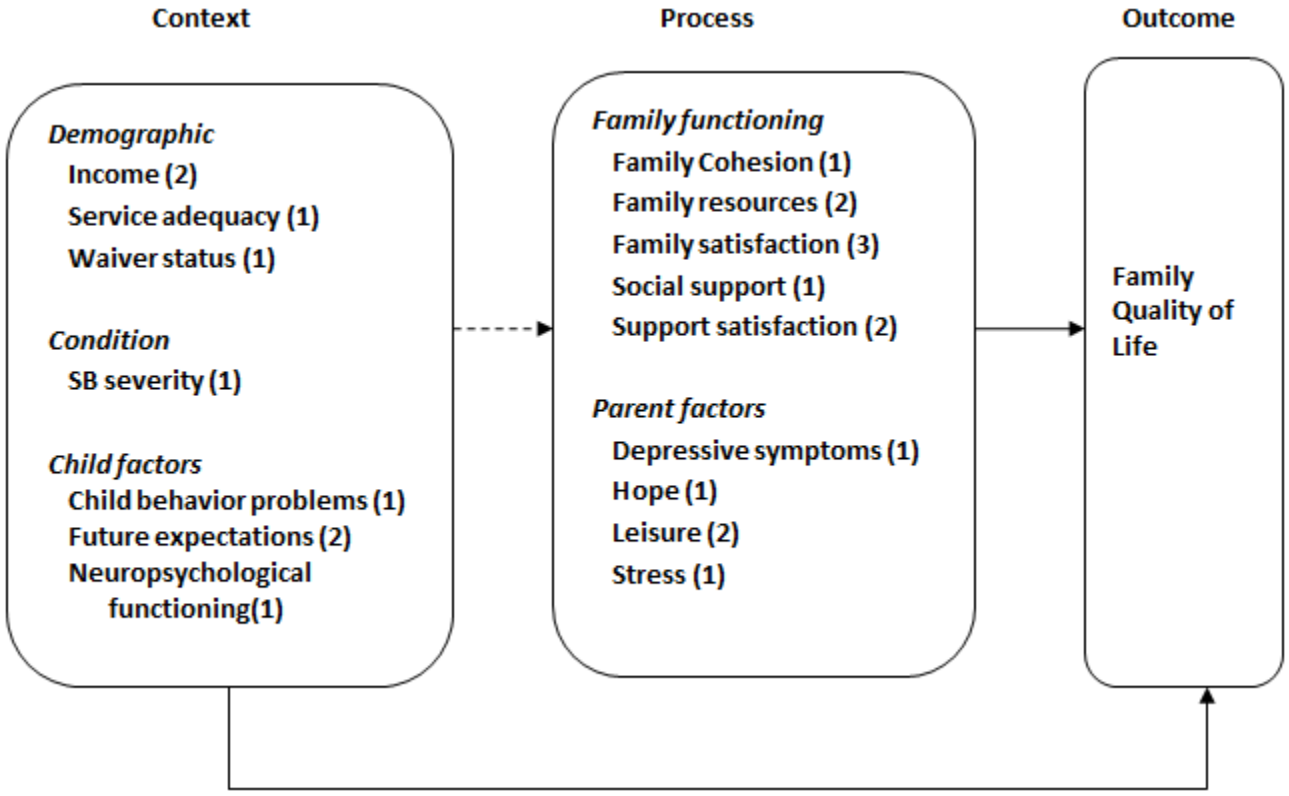

Figure 5. Theoretical Framework of factors related to FQOL. Only significant context and process findings are reported ($p < .05$). Number of studies evaluating concepts is identified.

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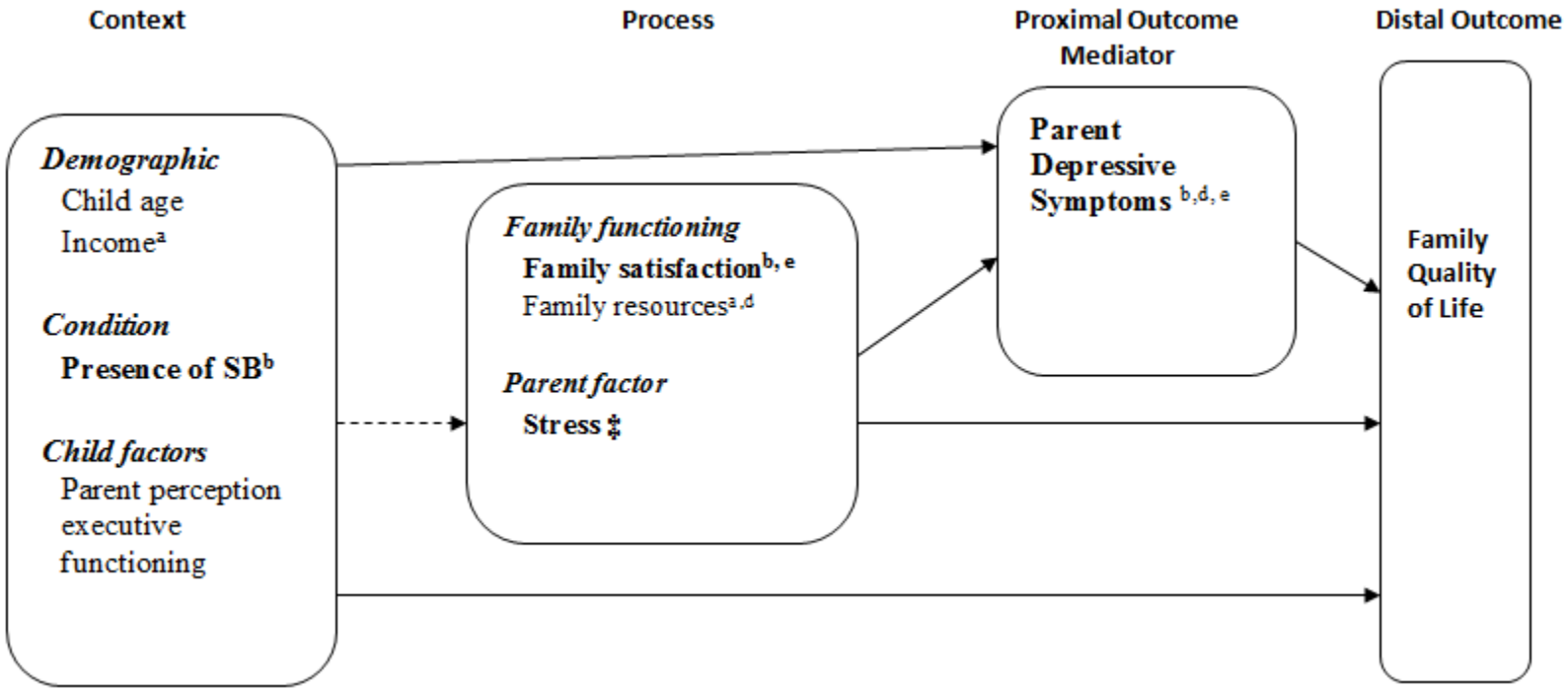

Figure 6. Results: Factors related to FQOL. Factors from results of current study identified. Bold were significant factors related to FQOL in total sample.

a. Factors related to PDS were income, family resources, and stress.
b. Factors related to FQOL were condition, family satisfaction, stress, and PDS.
c. Stress ‡ related to both PDS and FQOL.
d. PDS partially mediated family resources on FQOL.
e. Family satisfaction and PDS were only significant pathways in the final model for subsample with SB related to FQOL.

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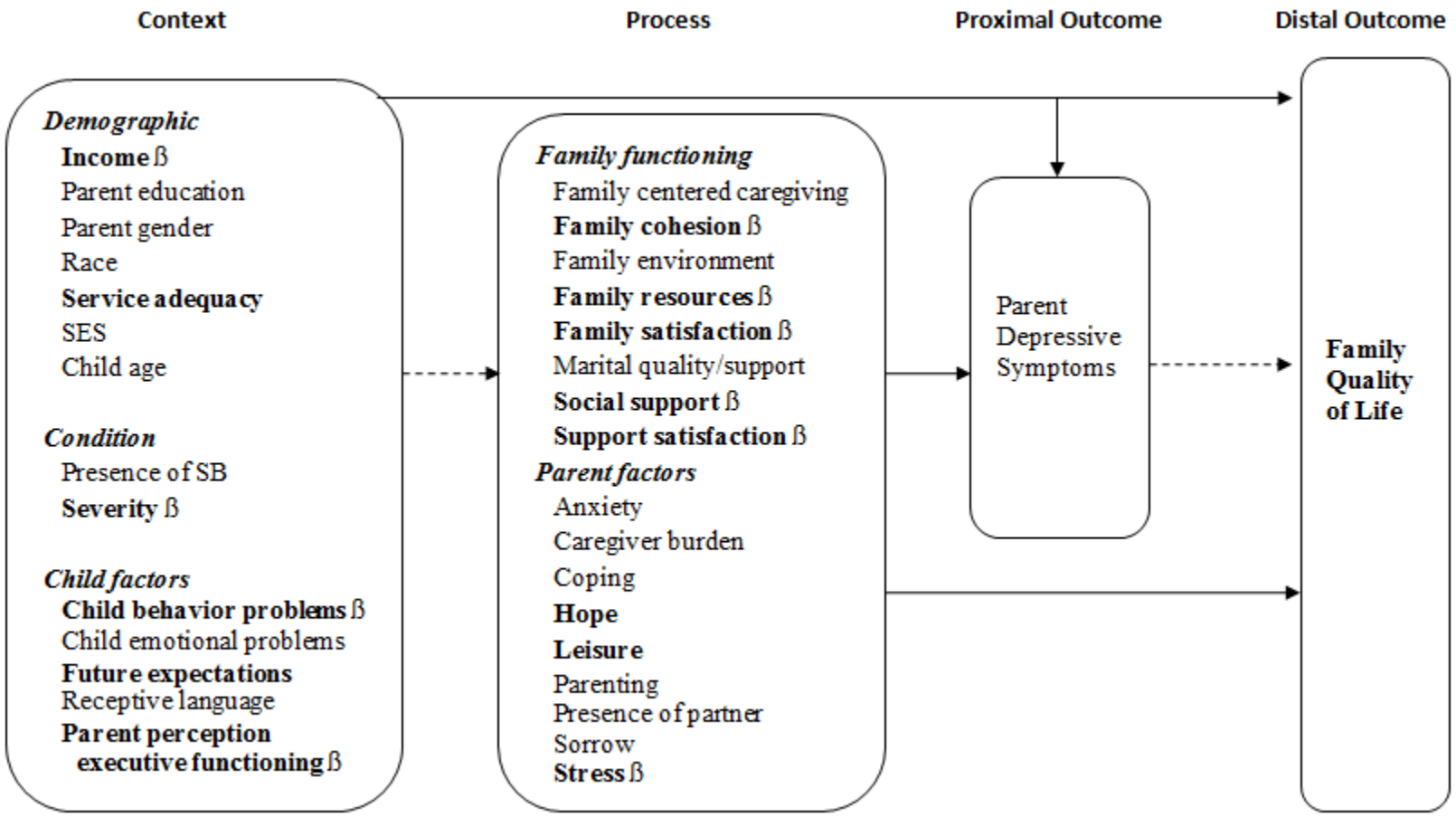

Figure 7. Factors related to PDS and FQOL from the literature. Factors related to PDS, bold factors related to FQOL, ß factors related to both PDS and FQOL. All factors are statistically significant, $p < .05$ level.

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Table 1

Depression in Parents of Children with Spina Bifida

Author(s), Year, & Level of evidence	Research question	Sample & Location	Design & Analysis	Instrument/ Concept	Relevant Findings	Strengths/Limitations
Early studies - before 2005						
Kronenberger, W. G., & Thompson, R. J., J. (1992a). Level of evidence VI	social relationships; marital quality/support, social support, & social coping association with **psychological adjustment** of mothers of children with SB	2 mo - 18 yrs. *N* = 66 mothers US - South, Clinic	Correlational Correlation & regression	Symptom Checklist-90-Revised (SCL-90-R)[a] **psychological distress**	*a.* -almost ½ sample (*n* = 29; 44%) met criteria for poor psychological adjustment *b. 50% variance psychological adjustment* *Context* Demographic - 1 - Mother's race (R^2 = .22) *Process* Family functioning - 3 - marital quality/support (Dyadic Adjustment Scale (DAS) total score) & controlling family environment (Family Environment subscale (FES) Other bivariate findings:- -FES related to outcome (support factor strongest, *r* = -.51, *p* < .001) -Friend coping related to outcome (*r* = .39, *p* < .01) (more emotional regulation using friends)	Weakness – Correlational design does not allow for testing of causation. Self-report data from mother's perspective.
Barakat, L. P., & Linney, J. A. (1992). ‡ Level of evidence IV	relationships of social support & maternal **psychological adjustment**	6-11 yrs. 29 mothers & 9 fathers SB group & 28 mothers & 7 fathers comparison group US- Midwest Clinic	2-group design correlations, multiple regression	Brief Symptom Inventory [b] **psychological distress**	*a.* No group differences in regression results of social support variables related to outcome (maternal adjustment) *b. 42% variance psychological adjustment (SB group) (no significant factors for comparison group)* *Context –none entered in regression* *Process* Family functioning - Social support factors – Available network ($R^2 \Delta$ = .24); number of family members ($R^2 \Delta$= .21); support satisfaction ($R^2 \Delta$ = .17) Other findings:-baseline group differences related to SES, parent education, race, child PPVT-R score, and child classroom placement -other group difference related to child adjustment: SB group lower self-concept & adaptive behavior -comparison group maternal adjustment related to internalizing behavior problems (*r* = -.60)	Weakness – Maternal psychological adjustment had little variance and positive skew--variable was transformed with square root of value Groups differed significantly on SES, parent education, race (SB group 3% and comparison 36% ethnic minority), child PPVT-R score and child classroom placement

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Author(s), Year, & Level of evidence	Research question	Sample & Location	Design & Analysis	Instrument/ Concept	Relevant Findings	Strengths/Limitations
Kronenberger, W. G., & Thompson, R. J. (1992b). Level of evidence VI	stress appraisals relationship to medical severity & stress related to **psychological adjustment**	2 mo - 18 yrs. 66 mothers US Midwest Clinic	correlational correlations, regression	Symptom Checklist-90-Revised (SCL-90-R)[a] **psychological distress**	*a.* almost ½ sample (*n* = 29; 44%) met criteria for poor psychological adjustment *b. 32% variance psychological adjustment* *Context* Demographics - 1 - mother's race (R^2 = .17); *Process* Parent factor - 3 - Parent perceived stress (appraised stress of the child's medical condition) (R^2 = .32); (R^2 Δ 0.15) *Other findings:* -psychological adjustment related to appraised stress (stress items were child medical stress, mother's emotional response to stress, and stressfulness of other life crises -child/medical stress *r* = .39, *p* < .01 & social/non-child stress *r* = .26 *p* < .05.	Weakness – Variable selected for severity of illness to place in regression model was number of shunts, which was 2.8 - low, may not be generalizable. Same data as 1990 and different process factors led to less variance in results.
Barakat, L. P., & Linney, J. A. (1995). ‡ Level of evidence IV	relationships of coping resources & maternal & child adjustment **maternal psychological adjustment**	6-11 yrs. 33 families SB group; 29 comparison group US – Midwest Clinic	2-group design Regression	Brief Symptom Inventory (BSI)[b] **psychological distress**	*a.* No group differences in regression results of social support variables related to outcome (maternal adjustment) *b. 67% variance maternal psychological adjustment (SB group)* *Context (R^2 = .20)* Demographics - PPVT-R, SES, race *Process* --Parent factor - Parent coping (avoidant coping, problem-focused, emotion-focused); (avoidant coping alone explained 47% of variance) – total of 3 forms coping & context factors (R^2 = .67) *Other findings: 44% variance maternal psychological*	Weakness – Maternal psychological adjustment had little variance and positive skew--variable was transformed with square root of value Groups differed significantly on SES, parent education, race (SB group 3% and comparison 36% ethnic minority), child PPVT-R

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Author(s), Year, & Level of evidence	Research question	Sample & Location	Design & Analysis	Instrument/ Concept	Relevant Findings	Strengths/Limitations
					adjustment in comparison group *Context* -Demographics - PPVT-R, SES, race *Process* --Parent factor - Parent coping (avoidant coping, problem-focused, emotion-focused); (avoidant coping alone explained 12% of variance) – total of 3 forms coping and context factors (R^2 = .44)	score and child classroom placement.
Holmbeck, G. N., Gorey-Ferguson, L., Hudson, T., Seefeldt, T., Shapera, W., Turner, T., & Uhler, J. (1997). ‡ Level of evidence IV	Examination of parents of children with SB across areas of functioning (individual, parental, and marital) & predictors of **parental adjustment** in family with or without child with SB.	8-9 yrs. 55 SB group & 55 child matched comparison group, 51 mobility limited, 74% in 2 parent family US Midwest clinic	2 group design MANOVAs for group differences, SCL-90-R, Chi-square for differences between groups	Symptom Checklist-90-Revised (SCL-90-R)$_a$ and Global Severity Index (GSI) **psychological symptoms**	a. 19.2% mothers and 25.6% fathers met criteria in SB group and 11.1% mothers & 16.3% fathers met criteria in comparison for psychological symptoms. *b. Group differences factors in psychological adjustment* *Context* Demographic - Parent gender group differences, fathers reported more PDS *Process* Parent factors - ↓Parental satisfaction (father & mother) ↓Parental Mastery (competence) (mother) Parent factors - ↑Parent perceived stress (mother & father), role restriction (father & mother), social isolation (mother) Parent coping (mother) behavioral disengagement (positive) & adaptability to change (negative); (father) behavioral disengagement (positive) and focus on venting of emotions (positive) Other findings: *Outcome* PDS - *(*psychological adjustment)↑Psychological symptoms (father) No differences in psychological symptoms between parents of CHC and comparison for mothers	Comparison sample was matched.

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Author(s), Year, & Level of evidence	Research question	Sample & Location	Design & Analysis	Instrument/ Concept	Relevant Findings	Strengths/Limitations
King, G., King, S., Rosenbaum, P., & Goffin, R. (1999). Level of evidence VI	Factors predicting parent well-being (3 indicators above) **parent emotional well-being.**	3-5 Ys, *N* = 164 parents Canada (multi-site 6 clinics)	descriptive SEM	Symptom Checklist-90-Revised (SCL-90-R)$_a$ – **psychological distress** Centre for Epidemiological Studies Depression Scale (CES-D)$_d$ - **Depressive symptoms** Stress One-time measure Likert 0-5 (degree caregiving by center affected stress and worry in caring for child in past year or less)	*a. Incidence of PDS not reported.* *b. Structural model – parent (emotional) wellbeing* *Context* Child factor - Child behavior problems (.60 path coefficient) *Process* Family functioning - Social-ecological factors (family functioning, satisfaction social support) (.23 path coefficient); family centered caregiving (-.13 path coefficient) Adequate goodness of fit χ^2 (309) = 634.09, *p* <01; RMSEA = .08; TLI = .83; RNI =.85	Theoretically based study with large multi-site sample
Lemanek, K. L., Jones, M. L., & Lieberman, B. (2000). Level of evidence VI	differences in parent adaptation & condition within SB compared to norm; **psychological distress**	3-16 yrs. *n* = 59 mothers *n* = 19 for comparative data of mother & father	descriptive & comparative t-tests, correlations, ANCOVA, paired comparisons	Symptom Checklist-90-Revised (SCL-90-R)$_a$ **psychological distress**	a. PDS - no differences in maternal rating of psychological distress when compared to norms. Mothers psychological distress lower than fathers but within normal range *b. Correlations with maternal psychological distress* *Context* Child factor -↑ child problem behavior (*r* = .41) *Process* Parent factors –	Weakness-Sampling bias – parents white (93.2%) & mother's SES middle income

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Author(s), Year, & Level of evidence	Research question	Sample & Location	Design & Analysis	Instrument/ Concept	Relevant Findings	Strengths/Limitations
		2 US clinics/ regional medical centers, region not specified			↓parenting competence & satisfaction combined (r = -0.51); ↓parent satisfaction (r = -.58), ↓parenting competence (r = -.26) Other findings-main effect of SB condition severity (F(3, 45) = 5.11 p <.01) on child problem behaviors found between mild and moderate severity of condition but not severe	
Friedman, D., Holmbeck, G. N., Jandasek, B., Zukerman, J., & Abad, M. (2004). ‡ Level of evidence IV	longitudinal examination of child adjustment and parent functioning **psychosocial functioning** and child adjustment. (Parent functioning domains were parenting stress, individual **psychosocial adjustment**, and marital satisfaction)	8-9 yrs. 68 SB group; 68 comparison group US - Midwest Clinic	2-group design hierarchical regression analyses	Symptom Checklist-90-Revised (SCL-90-R)a **psychological distress**	a.19.2% mothers and 25.6% of fathers met criteria (GSI) for severity of psychosocial functioning with one significant group difference (group status and parent functioning). *b.Correlations between condition, child adjustment and parent adjustment (parent functioning)* *Context* Condition (SB group) SB group X child externalizing symptoms b = .229 (time 2) (paternal) Child factor - behavior problems *1. child internalizing symptoms,* (time 2) (maternal) *child externalizing symptoms* (time 1) (maternal) (time 1) (paternal) *Outcome* A change in PDS (Parent functioning of mother and father) is significant from time 1 to time 2 and significantly related to child adjustment (time 1 & 2)	Strength – longitudinal and comparison sample matched Weakness – parent functioning measure composite and difficult to compare across studies

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Depression in Parents of Children with Spina Bifida

Author(s), Year, & Level of evidence	Research question	Sample & Location	Design & Analysis	Instrument/ Concept	Relevant Findings	Strengths/Limitations
Hobdell, E. (2004). Level of evidence VI	describe parental chronic sorrow following birth of child with NTD & explore relationship between chronic sorrow & **depression**	6 months - 6 yrs. *N* = 63 mother-father pairs US - 2 tertiary care pediatric hospitals, region not reported	descriptive ANOVA	Brief Symptom Inventory (BSI)[b] **psychological distress**	*a. 14% of parents met criteria for caseness of PDS* *b. Correlation of parent depression/ chronic sorrow* *Process* Parent factor – 2 measures of chronic sorrow (current) fathers *r* = .34; *r* =- .49 mothers *r* =.22; *r* = - .30 Other findings: -86% parents experience chronic sorrow, mothers more sorrow than fathers	Weakness - positive skew, log 10 transformations reduced skew to non-significant levels;
Vermaes, I. P., Janssens, J. M., Bosman, A. M., & Gerris, J. R. (2005). Level of evidence I	Do parents of children with SB have more psychological distress than controls? Do mothers and fathers differ? Which factors correlate with variations in **psychological adjustment**?	This article reviewed 33 studies and included 15 in meta-analysis portion of the review.	Weighted average effect sizes calculated based on two or more studies; One to four articles supported factors related to parental adjustment. When one study available then correlation coefficient was reported.	Brief Symptom Inventory (BSI) (4 studies) [b]; General health Questionnaire (GHQ) (1 study); Symptom Check List-90R (SCL-90R) (5 studies)[a]/ **psychological distress** Malaise Inventory (4 studies); Langner Symptom Checklist (1 study)/**psychological and physical symptoms**	*a. psychological adjustment* – parent gender and parent status had medium to large effect size (0.73 standard deviations more mothers of children with SB than comparison had psychological distress; parents of children with SB had 0.76 standard deviations more psychological distress than comparison). *b.Effect size results* *Context-* Demographic - ↓socio-economic (race, SES; parent education level & employment) (*r* =-0.13); ↑parent gender (mother) – d_+ = 0.73; ↓family income (*r* = -0.22); Condition - ↑severity – (*r* = 0.14) Child factors - ↑child behavior problems (*r* = 0.37); ↑child emotional problems (*r* = 0.47)	Strength – Cohen's Kappa is reported for process of identification of studies (.82 - .92) Weakness – Review based on condition effect on "adjustment" or "adaptation". These key words were included in search strategy versus inclusion of "depress*" in this lit review. Parents' psychological adjustment is defined as "the adaptive task of managing upsetting feelings aroused by the illness of the child and preserving a reasonable emotional balance" (p. 2). This definition is inconsistent with psychological distress and presence or severity of depressive symptoms.

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Author(s), Year, & Level of evidence	Research question	Sample & Location	Design & Analysis	Instrument/ Concept	Relevant Findings	Strengths/Limitations
					Process – Family Functioning - ↓Positive family environment (r = -0.42), ↓quantity social support (r = -0.28); ↓satisfaction social support (r = -0.28); ↓marital quality & support (r = -0.40) Parent factors - ↑Parent stress (r = 0.63); ↑parent coping (r = 0.38); ↓parenting satisfaction & parental competence (r = -0.41); ↓presence of partner (r = -0.16)	Critical appraisal of the quality of the primary studies was not reported. Duplication of samples used to calculate effect sizes may have introduced error. A small number of studies per concept were used to calculate effect size mostly 2-3. Pooled factors were categorized from a variety of variables-conceptually inconsistent. Outcome measures were conceptually inconsistent.
Later studies - after 2005						
Grosse, S., Flores, A., Ouyang, L., Robbins, J., & Tilford, J. (2009). ‡ Level of evidence IV	Compare time use, health, and **well-being** of caregivers with child/adolescent with SB; compare with parents of comparison group children accounting for level of lesion. **mental health outcomes**	0-17 yrs. *n* = 98 SB group *n* = 49 comparison group US – Arkansas population based registry	2-group design Comparison group by referral with 68% response rate Pearson's Chi square test; t-test; linear regression analysis; logistic regression	2 questions adapted from SF-36 about **depressive symptoms** Quality of Well-Being scale - preference-weighted health-related quality of life.	*a.* PDS - 32% caregiver of children with SB vs. 12% comparison group reported feeling blue more than a little of the time, *b. Group differences on factors related to PDS* *Context* Demographic – child age (<6 years) Condition - severity - lesion level – highest with higher lumbar *Process* Parent factor - leisure days (1 or no days) *Outcome* Group differences from regression PDS significant in sacral and high lumbar SB group vs. comparison group	Strength – SB group was recruited from a population-based registry of birth defects Weakness - Sample not matched. - Comparison group was not representative of population. ---SB group child older by about 2 years and caregivers older by about 3 years; --39% college level of education of children in comparison group was about double the SB children's group; --% married in comparison group was 91.8%, 77.6% in SB group. Reliability of the 2-

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Author(s), Year, & Level of evidence	Research question	Sample & Location	Design & Analysis	Instrument/ Concept	Relevant Findings	Strengths/Limitations
					Other findings: -quality of wellbeing score of SB group (high lumbar group of SB group was significantly lower than comparison group. -poor health significant in caregivers of young children (ages 0-6)	items from the SF-36 is unknown.
Ok, J., & Kurzrock, E. A. (2011). Level of evidence III	Evaluate impact of ACE surgery on • QOL • Child Experience • Impact on family • Social interaction **Mental health** (anxiety, **depression**, worry, & bother)	median age 11 yrs. *N* = 23 families; analysis on 18 completed pre and post-surgery surveys; 72% Caucasian US – West Clinic	descriptive, comparative pre and post-surgery paired analysis (Wilcoxon signed rank test)	Fecal incontinence and constipation on quality of life survey (FICQOL survey)[i] **depressive symptoms**	*a. Incidence of PDS not reported.* *b. Differences between pre-test and post-test* *Context* Condition (child) ↑Sensation & bowel movements into toilet from 45% to 97%. ↓Accidents from 3.9 to 0.3 per week. ↓abdominal pain from constipation ↓ Laxative from 44% to 6%. *Process* (parent) Parent factor – leisure (travel and socialization); ↓bother or anxiety of leaving the house *Outcome* (parent) PDS - caregiver support & emotional impact ↓caretaker anxiety, depression ,worry & bother *Other findings:* Total time for bowel care 45 min.	Strength - comparative based on 2 times of data collection Weakness – small sample no intention to treat analysis
Valença,, M, P, A, Calado,, A, & G. (2012). Level of evidence VI	Investigate burden, QOL, anxiety and **depressive symptoms** of caregivers	0-15 yrs. M 6.2 (4.3) *N* = 43 caregivers Brazil	descriptive t-tests/ Mann-Whitney U test; Pearson's r coefficient & Spearman's r coefficient; ordinary	Medical Outcomes Study Short Form-36 survey (SF-36)[e] Caregiver Burden Scale (CBS)[h] Beck Depression Inventory	a. 44.2% mothers considered depressive (BDI greater than or equal to 10); *b. Correlation with depressive symptoms* *Context* Condition SB with severe motor impairment (67%), sensitivity impairment (95.3%), & fecal incontinence	Weakness – selection bias issue correlation coefficients not reported

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Author(s), Year, & Level of evidence	Research question	Sample & Location	Design & Analysis	Instrument/ Concept	Relevant Findings	Strengths/Limitations
		Clinic	least squares estimation/Heckman method	(BDI)c **Depressive symptoms** Beck Anxiety Inventory (BAI)f	(48.8%) *Process* Parent factor – stress -Caregiver burden (CBS)– positive correlation (except emotional involvement dimension) and anxiety (BAI) *Outcome* PDS - SF-36 (pain, gen health, vitality, social functioning, & mental health) –negative correlation *Other findings*-fecal incontinence, low income, unemployment, and living with partner related to caregiver burden SES - Caregiver unemployed 74.4% and living with a partner	
Ulus et al. (2012) Level of evidence VI	evaluate impact of functional disability on parent **psychological status** and family functioning	7m -12 yrs. M 4.35 yrs. Median 39 months *n* = 54 mothers and 54 fathers of children with SB Turkey	Descriptive Multivariate linear regression analysis/Univariate analysis/Student t-test	Beck Depression Inventory (BDI)c **Depressive symptoms**	a. PDS - mean BDI scores 13.3 (7.52) mothers; 8.2 (5.48) fathers *b. Correlation with depressive symptoms* *Context* Demographic - parent gender - mothers significantly higher in depressive symptoms than fathers *Process* Parent factor – parenting (role (mother); problem solving (father); behavioral control (father)) *Other findings:* -no difference between groups in receiving news of SB diagnosis during pregnancy on depressive symptoms outcomes -no difference between groups in number of children in families and depressive symptoms	Weakness – Parents, who were divorced, separated, or had psychiatric disorders were excluded from the study, which may limit external validity of results. All mothers were unemployed and 55% fathers were government officials. All children had lumbar lesion level Inconsistency in test and table results concerning father general functioning or behavioral control as the significant factor.

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Depression in Parents of Children with Spina Bifida

Author(s), Year, & Level of evidence	Research question	Sample & Location	Design & Analysis	Instrument/ Concept	Relevant Findings	Strengths/Limitations
Brei, T., J. , Woodrome, S. E., Fastenau, P. S., Sawin, K. J., & Buran, C. F. (2013) Level of evidence VI	Examine relationship of risk and protective factors and PDS.	12 - 21 yrs. *N* = 50 parent and AYA US – Midwest	descriptive Correlation, Hierarchical Multiple Regression	Generalized Contentment Scale (GCS)$_g$ **Depressive symptoms**	*a.* 48% of parents depressive symptoms *b.57% of variance in PDS* *Context* Child factor *1.* Neuropsychological functioning (Mental processing, attention, oculomotor skills, & executive function) (*r* = .26 -.46) negative correlation (strongest is executive functioning) *Process* Family functioning - family protective factors (family cohesion, family satisfaction, family resources (mastery and esteem)) (*r* = .40 - .76) negative correlation (strongest is family satisfaction); *Composite of NP functioning and family protective factor Other findings:- mean normal IQ, -NP measures .75 - 1 SD less than norm	small sample

Note. Findings are significant at p ≤ .05 unless otherwise specified. ‡ 2-group studies. Levels of evidence are I systematic review/meta-analysis; II randomized controlled trials; III controlled trials without randomization; IV case-control/cohort studies; V systematic reviews of descriptive studies; VI single descriptive study; VII opinion of authorities or reports of expert committees (Melnyk & Fineout-Overholt, 2011). a. Symptom Checklist-90-Revised (SCL-90-R) – measures current psychological distress (90 items) using Likert 0-4 scale. *9 symptom dimensions:* *Somatization, Obsessive-compulsive, Interpersonal sensitivity, *Depression, *Anxiety, Hostility, Phobic anxiety, Paranoid ideation, Psychoticism. *Global Severity Index (GSI) – overall psychological distress level (sum of score for all items/number of items answered). b. Brief Symptom Inventory $_b$ (Short form developed from Symptom Checklist-90-Revised) (53 items) using Likert 0-5 scale Measures psychological distress. *9 symptom dimensions:* Somatization, Obsessive-compulsive, Interpersonal sensitivity, Depression, Anxiety, Hostility, Phobic anxiety, Paranoid ideation, Psychoticism. Global Severity Index (GSI)-overall psychological distress level. c. Beck Depression Inventory (BDI) 21 categories of symptoms measures behavioral manifestation of depression. d. Centre for Epidemiological Studies Depression Scale (CES-D) (20 item) – measures frequency & duration of cognitive, affective and behavioral symptoms. e. Medical Outcomes Study Short Form-36 survey (SF-36) (36-item) measures Quality of Life one of 8 domains measures mental health. f. Beck Anxiety Inventory (BAI) 21 symptoms measures common symptoms of anxiety. g. Generalized Contentment Scale (GCS) (25 item) measures degree, severity, magnitude of non-psychotic depressive symptoms. h. Caregiver Burden Scale (CBS) (22-item) measures one of 5 dimensions measures emotional involvement. i. Fecal incontinence and constipation on quality of life survey (FICQOL survey) (51 item) measures aspects of daily life when bowel incontinence & bowel care have significant impact subscale 8-items on caregiver support & emotional impact measured depressive symptoms.

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Table 2

Summary of depression prevalence in parents of children with SB, context factors and process factors variable related to PDS

Author (year)	Prevalence	Context			Process	
		Dem	Condition	Child Factors	Family Functioning	Parent Factors
Kronenberger (1992a)	44% mothers	race			family environment (controlling); marital quality/support	
Kronenberger (1992b)	44% mothers	Race				parent stress (condition)
Hobdell (2004)	14% parents					chronic sorrow
Barakat (1992)‡		race, SES		receptive language	social support & support satisfaction	
Barakat (1995)‡		race, SES		receptive language		parent coping (avoidant)
Holmbeck (1997)‡	19.2% mothers/ CHC; 11.1% mothers/no CHC; 25.6% fathers/ CHC;16.3% fathers/no CHC	parent gender	condition presence (SB)			parenting (competence, role restriction, satisfaction, social isolation); parent coping (behavioral disengagement/adaptability to change) & venting emotions; stress
King (1999)				child behavior problems	family cohesion; social support, support satisfaction, family centered caregiving	
Lemanek (2000)				child behavior problems		parenting (competence & satisfaction)
Friedman (2004)‡	19.2% mothers; 25.6% fathers		condition presence (SB)	child behavior problems		
Vermaes (2005)		parent gender; race; SES; parent education level & employment; family income;	condition severity	child behavior problems child emotional problems	family environment (positive), quantity social support; satisfaction social support; marital quality & support	parent stress; parent coping; parenting (competence & satisfaction); presence of partner
Grosse (2009)‡	32% parents/CHC; 12% no CHC	child age	condition presence & severity (lesion level)			leisure (days)

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Table 2

Summary of depression prevalence in parents of children with SB, context factors and process factors variable related to PDS

Author (year)	Prevalence	Context			Process	
		Dem	Condition	Child Factors	Family Functioning	Parent Factors
Ok (2011)			condition severity, (sensation & BM accidents, pain, laxative)			leisure (travel & socialization/ leaving the house)
Brei (2013)	48% parents			Neuro-psychological functioning	family cohesion, family satisfaction, family resources	
Valença (2012)	44.2% mothers	SES	condition severity (severe motor impairment, sensitivity, fecal incontinence)			caregiver burden & anxiety
Ulus (2012)		parent gender				parenting (role, problem solving, behavioral control)

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Table 3

Summary of Domain-Specific and Overall measures of FQOL

Scale / Author	Domains-Specific Measures	Overall Measures		Summary of Psychometric Properties
	Domains	Overall Sum of domains	Overall Global	
Beach Scale/ Hoffman et al., 2006[1]	Physical/Material well-being Family interaction Parenting Disability-related Support Emotional well-being	X		25-item scale measures satisfaction in five domains Good internal reliability reported for the five subscales (α = 0.70 - 0.90) and total scale (α = 0.88). Response pattern was 1 (*very dissatisfied*) to 5 (*very satisfied*). Confirmatory factor analysis supported a good fit for a model with five subscales and a second order overall FQOL factor (χ^2 (270) = 439.24, *p* <001, CFI = 0.92, RMSEA = 0.05). Convergent validity with 2 domain subscales-The Family Interaction subscale of the Beach Scale related to Family APGAR[2], (*r* = 0.68). The Family Resource Scale[3] related to Physical/Material Well-being subscale (*r* = 0.60). Test-retest reliability for satisfaction subscales across domains showed significant correlations between time points (*r* = 0.60 - 0.77), time between test and retest was 3 months.
FQOL Survey – 2006 Brown et al., 2006[3]	Family Relationships Influence of values Health Careers Community Support from services Support from others Leisure Finances	X	X	54-item survey. Overall global items are "*Overall, how would you describe your family's quality of life?*" and "*Overall, how satisfied are you with your family's quality of life?"* [4] Response pattern for overall global item was 5 (*excellent*) to 1 (*poor*). Response pattern for satisfaction question was 5 (*very satisfied*) to 1 (*very dissatisfied*).Confirmatory factor analysis supported a good fit for a model with nine domains and overall latent FQOL factor. Internal reliability for each domain was good in families in Nigeria (α = 0.73 – 0.83) and fair to good in 3-countries sample (α = 0.53 - 0.83) (Isaacs et al., 2012). Although several domains had problems (health α = 0.53; support from service α = 0.67), an overall latent FQOL factor was supported in this analysis. Additionally test-retest and convergent validity were not reported [6].
Single item/Sawin et al., 2002[5]			X	*"How would you rate your family's quality of life?"* The response pattern was zero (poor) to 100 (excellent). Single item measure has support in literature.
FQOL 3-item Scale/Ridosh et al., 2013[6]			X	3-item scale, "*How would you rate your quality of life?*"; "*How would you rate your teen's quality of life?*" *"How would you rate your family's quality of life?*" The response pattern was zero (poor) to 100 (excellent). A principal component factor analysis with Varimax rotation (*N* = 43) supported a single factor with an eigenvalue greater than 1. The factor loadings were .91 for FQOL, .91 for parent's quality of life and .80 for teen's quality of life[7]. Internal reliability was strong (α = 0.84). Similar results were found when the factor analysis was repeated using a sample of parents of adolescents with and without SB (*N* = 240). A single scale with high factor loadings (0.86-0.94) and strong internal reliabilities (α = 0.86-0.90) were supported.

Note. 1. Hoffman, L., Marquis, J., Poston, D., Summers, J. A., & Turnbull, A. (2006). Assessing family outcomes: Psychometric evaluation of the Beach Center Family Quality of Life Scale. *Journal of Marriage and Family, 68*(4), 1069-1083. doi: 10.1111/j.1741-3737.2006.00314.x. 2. Austin, J. K., & Huberty, T. J. (1989). Revision of the family APGAR for use by 8-year-olds. *Family Systems Medicine, 7*(3), 323–327. doi: 10.1037/h0089774. 3.Dunst, C. J., & Leet, H. E. (1985). *Family Resource Scale: reliability and validity*. Asheville, NC: Winterberry Press. 4. Brown, I., Brown, R. I., Baum, N. T., Isaacs, B. J., Myerscough, T., Neikrug, S., . . . Wang, M. (2006). *Family Quality of Life Survey: Main caregivers of people with intellectual or developmental disabilities.* Toronto, ON, Canada: Surrey Place Centre. 5. Isaacs, B., Wang, M., Samuel, P., Ajuwon, P., Baum, N., Edwards, M., & Rillotta, F. (2012). Testing the factor structure of the family quality of life survey. *Journal of Intellectual Disability Research, 56*(1), 17-29. doi: 10.1111/j.1365-2788.2011.01392.x. 6. Sawin, K. J., Brei, T. J., Buran, C. F., & Fastenau, P. S. (2002). Factors associated with quality of life in adolescents with spina bifida. *Journal of Holistic Nursing, 20*(3), 279-304. doi: 10.1177/089801010202000307. 7. Ridosh, M., Sawin, K., J., & Brei, T., J. (2013, March). *Risk and protective factors associated with adaptation in parents of adolescents and young adults with spina bifida*. Paper presented at the MNRS 37th Annual Research Conference, Chicago, IL.

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Table 4

Psychometric Properties of FQOL Measures

Authors	Instrument	Evidence of Validity	Evidence of Reliability	Strengths/Weaknesses
Summers (2007)	Beach FQOL Scale	*Content* Literature review in qualitative data	*Internal consistency -Cronbach's* Family interaction α= 0.92 Parenting α= 0.88 Emotional well-being α= 0.80 Physical material well-being α= 0.88 Disability-related support α= 0.92	19% response rate
Davis (2009)	Beach FQOL Scale	*Content* Literature review		16% response rate
Jackson (2010)	Beach FQOL Scale			The instrument was modified by omitting question related to adult with disabilities; modification included impact of deafness on family life, child outcomes and desired family support.
Eskow (2011)	Beach FQOL Scale	*Content* Literature review		28.8% response rate; 80% male and 20% female; children in waiver group were older
Hu (2012)	Beach FQOL Scale	*Construct – confirmatory factor analysis* Children with ID in China sample—importance rating & satisfaction rating acceptable-good fit similar five-factor structure of FQOL construct to US sample; factor loadings ranged from 0.45 - 0.83 except satisfaction in physical well-being domain (0.20 - 0.65); *Content – analytical critique* Pilot tested Chinese version of Beach Center FQOL Scale and made changes based on interview to ensure instrument is culturally sensitive, then 3 bilingual experts translated back to English;	*Internal consistency -Cronbach's* sub-scales α 0.73 - 0.84 and overall scale α = 0.93	return rate of 89.1% fathers and mothers respondents, initial response rate 72% /skewed distribution of family income (low income); no data of family dynamics, family support services and family coping
Ajuwon (2012)	FQOL-2006			Qualitative findings add context to family experience beyond questions of instrument /sample included those receiving services

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Table 4

Psychometric Properties of FQOL Measures

Authors	Instrument	Evidence of Validity	Evidence of Reliability	Strengths/Weaknesses
Werner (2009)	FQOL-2006	*Content* Literature review	*Internal consistency -Cronbach's* Reliability reported on six dimensions across the nine life domains were found to be moderate Importance α = 0.55; Opportunities α = 0.56; Initiative α = 0.71; Attainment α = 0.57; Stability α = 0.78; Satisfaction α = 0.64	small sample, sample recruited from 2 sites, which differed in age and living situation (residential placement or home) of participants Low internal consistency on dimensions (importance, opportunities, attainment, and satisfaction)
Neikrug (2011)	FQOL-2006	*Content* Theoretical domains and dimension in literature	*Internal consistency -Cronbach's alpha* For 9 domains were α = .77 - 0.88 except for overall health domain with internal consistency α = 0.33; Total instrument had high internal consistency α = 0.92	translated to Hebrew by professional translator not part of research team pretested for modifications; not random sample, did not report qualitative findings of instrument
Clark (2012)	FQOL-2006	*Content* Literature review		survey instrument translated and back translation done (details of changes not available); short form did not allow for data to add meaning or context to responses; sample gender of child not accounted for 38 boys and 16 girls; Eighteen of the 52 families in the current study reported that they had live-in paid caregivers or extended family members that provided care and support for their family member with a disability, reducing responsibility left to the primary caregiver.
Rillotta (2012)	FQOL-2006	*Content* Literature review	*Cronbach's alpha* Importance α = 0.24, Attainment α = 0.69, Opportunities α = 0.79, Stability α = 0.45, Satisfaction α = 0.82, Initiative α = 0.48	low to moderate internal consistency across dimensions (importance, stability, initiative)

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Table 4

Psychometric Properties of FQOL Measures

Authors	Instrument	Evidence of Validity	Evidence of Reliability	Strengths/Weaknesses
Sawin (2002)	Single item measure	*Content* Literature review		
Ridosh (2013)	3-item measure	*Construct – factor analysis* Single factor in US sample with AYA with SB Inter-item correlations were between 0.47 -0.78; factor loadings were 0.91 for FQOL, 0.91 for parent's quality of life & 0.80 for teen's quality of life	*Cronbach's alpha* internal reliability α=0.84	

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Table 5

Sample Characteristics

Author Year	Sample Size	Location	Sample Characteristics
Davis 2009	64	Australia	Mean age 51.98 months (9.65), range 36-72 months, child gender 43 males/21 females; received services in early intervention program between 2 - 60 months; diagnoses autism (34), speech/language impairment (28), DD (19), physical disability/CP (9), Down syndrome (1), Fragile X (1), Dandy-Walker (1), Dravet syndrome (1); 48% described severity of delay as moderate
Rillotta 2012	150	South Australia	Age range 2-46 years and had ID or autism; mean 17.3 years; 64.3% male, 35.7% female; 2 parent home 66.7%
Hu 2012	442	China	Age of child 0 - > 18 with majority between 7 – 17 years old; child with ID living in urban and suburban Beijing; stratified sampling method;
Neikrug 2011	103	Israel	Mean age 10.86, range 1 -31 years old; 81% mothers 4 % fathers other unknown; 7 single parent homes (others 2-parent); child gender 70% male;; 19% DD, 3%CP, 32%PDD, 8.7% Downs', 3.9% Rett, 28% other; convenience sample
Clark 2012	52	Malaysia	Mean age 7.54 (3.99) range from 2 -18 years; 43 respondents were mothers; 33 were in 2 parent families children had DD/ID, 2 families had 2 children with disability, 49 of 54 children lived at home, diagnoses were ID, Down's syndrome, cerebral palsy, autism, & others; random selection receiving services,
Ajuwon 2012	80	Nigeria	Mean age 12.3 (7.85); main caregivers of school-aged children & youth with ID; 82% 2 parent home; 78% children lived with family; 35% unknown diagnosis, 30% CP, 15% Downs' Syndrome, 12.5 Autism
Werner 2009	35 family members	Toronto Canada	Mean age 25.43 (14.58); range 3- 59; majority families with member with autism;; 60% lived in residential group homes, 40% lived with family; 24 mothers, 7 fathers, 3 siblings, 1 mother and sister participated together; 26 families were 2 parent homes.
Jackson 2010	207	US - 42 states	Mean age 44 months (SD 16.58); range 2 - 72 months (6 years); deaf or hear of hearing and receiving services;
Eskow 2011	waiver group 228; registry group 627	US – Maryland	Ages 3 years – adult; child with autism
Sawin 2002	60	US Midwest	Mean age 16.2; range 12 - 21; parents 73% married
Summers 2007	180	US Midwest	Age range birth to 5 years
Ridosh 2012	43	US Midwest	Mean age 17 years; multi-site sample AYA with SB, 58% female 42% used wheelchairs 72% married

Note. AYA is adolescents and young adults.

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Table 6

Overall FQOL Scores

Author (year)	Instrument	FQOL Scores		
Beach FQOL Scale		Mean	SD	Range (possible)
Summers (2007)	Beach FQOL	3.99	0.64	0-5
Jackson (2010)	Beach FQOL	DS		
Davis (2009)	Beach FQOL	3.74	0.69	0-5
Eskow (2011)	Beach FQOL (waiver/registry group)	3.90/ 3.56	0.61/ 0.72	0-5
Hu (2012)	Beach FQOL	DS		0-5
	Summary Mean Overall Score	3.80	0.67	0-5
FQOL-2006				
Werner (2009)	FQOL-2006 single item (satisfaction)	3.71	NR	0-5
Neikrug (2011)	FQOL-2006	DS		
Rillotta (2012)	FQOL-2006 single item (satisfaction)	3.90	0.91	0-5
Clark (2012)	FQOL-2006	DS		
Ajuwon (2012)	FQOL-2006	DS		
	Summary Mean Overall Score	3.80	0.91	0-5
Single items				
Sawin (2002)	single item	72.50	21.60	0-100
Ridosh (2013)	3 item FQOL scale	80.51	15.62	0-100
	Summary Mean Overall Score	78.00	18.61	0-100

Note. DS is domain specific, mean of FQOL not reported. NR is not reported. The overall score for the Total Beach Score and the FQOL-2006 were created by the investigator.

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Table 7

The Beach FQOL Scale Domain Scores

Domains	Summers (2007) *N* = 180	Davis (2009) *N* = 64	Jackson (2010) *N* = 207	Eskow (2011) waiver/registry *n* = 288 / *n* = 627	
	Domain Mean Score (SD)				Summary Mean Score[1]
Physical/ Material well-being	4.21 (0.73)	4.03 (0.78)	4.38 (0.65)	4.09 (0.71)/ 3.83 (0.78)	4.11 (0.73)
Family interaction	4.06 (0.76)	NR	4.27 (0.76)	4.07(0.74)/ 3.78 (0.84)	4.05 (0.78)
Parenting	4.07 (0.71)	NR	4.33 (0.79)	3.93 (0.74)/ 3.69 (0.78)	4.01 (0.76)
Disability-related Support	4.13 (0.73)	NR	4.22 (0.79)	3.89 (0.71)/ 3.45 (0.87)	3.92 (0.78)
Emotional well-being	3.43 (1.00)	3.10 (1.05)	3.65 (0.94)	3.43 (0.89)/ 2.81 (1.07)	3.28 (0.99)

Note. NR is not reported. 1. The summary scores created by the investigator. Domain means placed in rank order highest to lowest.

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Table 8

FQOL-2006 Domain Scores in the Satisfaction and Attainment Dimensions

Domains	Ajuwon (2012) Nigeria *N* = 80	Neikrug (2011) Israel *N* = 103	Clark (2012) Malaysia *N* = 52	Rillotta (2012) South Australia *N* = 150	Werner (2009) Toronto Canada *N* = 35	
	Domain Mean Score (SD)					Summary Mean Score[1]
Satisfaction Dimension						
Family Relationships	4.31 (0.72)	4.01 (0.99)	4.23 (0.65)	4.36 (0.90)	3.91 (0.92)	4.16 (0.84)
Influence of values	4.22 (0.60)	3.82 (0.90)	4.14 (0.58)	4.17 (0.70)	3.73 (0.72)	4.02 (0.70)
Health	3.90 (0.87)	3.86 (0.95)	3.98 (0.64)	3.78 (0.82)	3.57 (0.78)	3.82 (0.81)
Careers	3.81 (0.86)	3.70 (1.06)	3.86 (0.85)	3.94 (0.80)	3.70 (1.16)	3.80 (0.95)
Community	3.68 (0.87)	3.32 (1.01)	4.00 (0.64)	3.71 (0.83)	3.40 (0.85)	3.62 (0.84)
Support from services	3.06 (1.12)	2.91 (1.13)	4.10 (0.67)	3.54 (1.07)	3.84 (0.68)	3.49 (0.94)
Support from others	3.18 (1.00)	3.11 (1.15)	3.73 (0.70)	3.59/3.75† (1.12/1.11)	3.37 (0.84)	3.46 (0.99)
Leisure	3.04 (1.08)	3.25 (1.05)	3.76 (0.80)	3.78 (0.86)	3.43 (0.98)	3.45 (0.95)
Finances	3.43 (0.90)	3.45 (1.11)	3.53 (0.90)	3.30 (1.02)	3.37 (0.97)	3.42 (0.98)
Attainment Dimension						
Family relationships	4.68 (0.57)	4.06 (0.96)	4.00 (0.98)	4.34 (0.63)	3.91 (1.09)	4.20 (0.85)
Health	4.44 (0.74)	3.91 (0.76)	4.04 (0.91)	4.08 (0.69)	3.57 (0.77)	4.01 (0.77)
Influences of Values	4.59 (0.69)	3.65 (1.13)	4.06 (0.95)	3.91 (1.07)	3.73 (1.05)	3.99 (0.98)
Careers	4.04 (1.04)	3.58 (1.13)	3.58 (1.16)	3.43 (1.43)	3.70 (1.33)	3.67 (1.22)
Finances	3.59 (1.02)	3.30 (0.96)	3.69 (0.83)	3.05 (1.15)	3.37 (1.08)	3.40 (1.01)
Community	3.69 (1.05)	2.86 (1.08)	3.71 (0.99)	3.18 (0.94)	3.40 (0.97)	3.37 (1.01)
Leisure	2.70 (1.18)	3.39 (1.03)	3.38 (1.02)	3.47 (0.86)	3.73 (0.88)	3.33 (0.99)
Support from services	2.39 (1.36)	2.79 (1.06)	3.39 (0.92)	3.17 (1.34)	3.84 (1.18)	3.12 (1.17)
Support from others	2.55 (1.25)	2.62 (1.18)	2.63 (1.13)	2.08 (1.28)/ 2.77 (1.33)†	3.37 (1.10)	2.67 (1.21)

Note. † Practical/emotional support from others. 1. The summary scores created by the investigator. Domain means placed in rank order highest to lowest.

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Table 9

Summary of context factors and process factors related to FQOL

1st Author (year)	Instrument measuring FQOL	CHC	Total variance	Context		Process	
				Demographic /Condition	Child factors	Family functioning	Parent factors
Sawin (2002)	single item global FQOL	SB	R^2 = 0.50		future expectations (r = 0.33)	family satisfaction (together with parental hope)(R^2 = 0.50); other correlations family factors (activity, mastery, esteem, cohesion, satisfaction) (r = 0.41 -0.60)	parental hope (r = 0.54) (together with family satisfaction) (R^2 = 0.50); condition stress (r = -.30); everyday stress (r = -.47);
Ridosh (2013)	3-item FQOL scale	SB		income (r = 0.42)	neuropsychological functioning (r = - 0.33), future expectations (r = 0.61)	family satisfaction (r = 0.60); family resources (r = -0.62)	Parent depressive symptoms (PDS) (r = -.72)
Summers (2007)	Beach FQOL Scale	ID	Direct effect of model 0.34	service adequacy (t-value = 4.74)		support satisfaction (family-professional partnership) (partial mediator) (Sobel test statistic 2.14, p = .031)	
Davis (2009)	Beach FQOL Scale	ID	R^2= 0.42	(controlling for income)	child behavior problems (R^2 = 0.07)	Social support (family support) (R^2 = 0.17); support satisfaction (professional support)(R^2 = 0.10)	
Eskow (2011)	Beach FQOL Scale	ID	Partial eta squared 0.036	waiver status (F(6, 758) = 11.28) (controlling for age and income)			
Hu (2012)	Beach FQOL Scale	ID	R^2= 0.016	income & severity of condition (R^2 = 0.016)			
Werner (2009)	FQOL-2006	ID		health of the family (r = 0.48)		Family satisfaction (family relationships) (r = 0.45)	leisure (r = 0.66)
Domain specific frequencies							
Jackson (2010)	Beach FQOL Scale	hearing impaired		community inclusion (mean 3.88) (satisfaction low); finances (mean 3.95) (satisfaction low)		support to relieve stress (item on emotional well-being scale) (mean 3.35) (satisfaction low); services from local agencies (satisfaction low 3.83)	time to pursue interests (mean 3.34) (satisfaction low)

Note. All factors significant at p < .05.

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Table 10

Characteristics of the Sample

Variable	*Total*		*Subsample with SB*		*Comparison Subsample*	
	N	*%*	*n*	*%*	*n*	*%*
Group			112	54	97	46
AYA age						
12 – 15 years	121	58	67	60	54	56
16 -18 years	56	27	29	26	27	28
19 – 25 years	32	15	16	14	16	17
Gender (child)						
Female	113	54	57	51	55	43
Male	97	46	55	49	42	57
Combined family income*						
Less than $20,000	24	12	20	18	4	4
$20,000 – $50,000	57	27	35	31	22	23
$50,000 or over	126	60	56	50	70	72
Gender (parent)						
Female	196	94	105	94	90	93
Race (parent)						
Black	22	11	6	5.4	16	17
Caucasian	179	86	101	90.2	78	80
Other	7	3.5	4	3.6	3	3.1
Ethnicity						
Hispanic	6	3	4	3.6	2	2.1

Note. Demographic variables were tested for significant differences between subsamples using Chi Square statistic. Income significantly different by subsample. * χ^2 (207) = 16.67, $p < .001$

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Table 11

Descriptive Statistics for Continuous Variables

	Total			*Subsample with SB*			*Comparison Subsample*			
	M	*SD*	*Range*	*M*	*SD*	*Range*	*M*	*SD*	*Range*	α
Context										
Parent perception EF-BRI (T-scores)	54.18	10.84	37-96	56.70	11.97	37–96	51.27	8.55	37-71	.93
Parent perception EF MCI (T-scores)	56.88	11.89	37 - 86	61.23	11.98	37-86	51.85	9.63	37-73	.96
Process										
Family Cohesion	40.28	5.64	25–50	40.28	5.52	25–50	40.36	5.80	26-49	.83
Family Satisfaction	4.13	0.62	1.8–5.0	4.10	0.66	1.8-5.0	4.17	0.58	2.2-5.0	.84
Family Resources	3.13	0.46	1.78–4.0	3.03	0.51	1.78-4.0	3.24	0.36	2.2-3.9	.91
Parent Stress	53.33	26.32	0 -100	55.61	27.8	0-100	50.70	24.4	5-100	NA
Outcomes										
PDS	7.98	7.75	0- 46	9.11	8.67	0-46	6.67	6.33	0-28	.88
FQOL	85.62	13.23	27-100	82.47	14.8	26.7-100	89.25	10.1	47-100	.88

Note. Total sample $N = 209$; Subsample with SB $n = 112$; Comparison Subsample $n = 97$

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Table 12

Correlations for Factors Related to PDS and FQOL in the Total Sample

	1	2	3	4	5	6	7	8	9	10	11	12	13	14
Context Variables														
1. AYA age	1													
2. Income	-.097†	1†												
3. Parent gender	.124†	.081†	1†											
4. Race	.015†	.205**†	.208**†	1†										
5. Ethnicity	.031†	.033†	.311**†	.259**†	1									
6. Presence of SB	.086†	.252**†	.037†	-.160*†	-.045†	1†								
7. Parent perception EF BRI	-.107	-.201**†	.096†	.158*†	-.100†	-.223**†	1							
8. Parent perception EF MCI	-.049	-.200**†	.084†	.250**†	-.083†	-.391**†	.698**	1						
Process Variables														
9. Family cohesion	-.176*	.204**†	-.087†	-.067†	-.030†	.019†	-.111	-.230**	1					
10. Family satisfaction	-.123	.172*†	-.074†	-.061†	-.024†	.027†	-.252**	-.365**	.631**	1				
11. Family resources	-.097	.328**†	-.008†	-.184**†	.078†	.216**†	-.453**	-.455**	.428**	.573**	1			
12. Stress	.002	-.232**†	-.029†	-.013†	-.085†	-.101†	.177*	.249**	-.222**	-.250**	-.458**	1		
Proximal Outcome														
13. Parent Depressive Symptoms	.151*	-.324**†	-.054†	.088†	-.126†	-.133†	.304**	.320**	-.255**	-.335**	-.514**	.398**	1	
Distal outcomes														
14. Family Quality of Life	-.050	.283**†	.046†	-.085†	.053†	.264**†	-.334**	-.397**	.342**	.515**	.552**	-.416**	-.535**	1

Note. Pearson reported for all continuous variables correlations; †Spearman's rho reported for correlation with a categorical variable; * Correlation is significant at the 0.05 level (2-tailed); **Correlation is significant at the 0.01 level (2-tailed).

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Table 13

Multiple Hierarchical Regression: Factors Related to PDS

Model summaries	ΔR^2	*ß*	*t*	*p*
Context Block 1 (R^2 = .255)	.255*			
AYA age		.140	2.248	.026*
Presence of SB		.025	.369	.712
Income		-.346	-5.314	<.001*
Parent perception EF BRI		.104	1.194	.234
Parent perception EF MCI		.188	2.085	.038*
Context and Process Block 2 (R^2 = .378)	.124*			
AYA age		.106	1.817	.071
Presence of SB		.024	0.369	.712
Income		-.254	-4.077	<.001*
Parent perception EF BRI		.042	.496	.620
Parent perception EF MCI		.065	.746	.457
Family cohesion		.026	.343	.732
Family satisfaction		-.051	-0.617	.538
Family resources		-.277	-3.334	.001*
Stress		.181	2.812	.005*

Note. * $p < .05$. Dependent variable: Parent Depressive Symptoms.

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Table 14

Multiple Hierarchical Regression: Factors Related to FQOL

Model summaries	ΔR^2	*ß*	*t*	*p*
Context Variables Block 1 (R^2 = .220)	.220*			
AYA age		-.058	-.919	.359
Presence of SB		.081	1.161	.247
Income		.206	3.085	.002*
Parent perception EF BRI		-.081	-.906	.366
Parent perception EF MCI		-.266	-2.888	.004*
Context and Process Block 2 (R^2 =.438)	.218*			
AYA age		-.003	-.056	.955
Presence of SB		.123	2.022	.045*
Income		.089	1.509	.133
Parent perception EF BRI		-.049	-.617	.538
Parent perception EF MCI		-.049	-.588	.557
Family cohesion		-.016	-.226	.822
Family satisfaction		.315	4.036	<.001*
Family resources		.183	2.307	.022*
Stress		-.204	-3.322	.001*
Full Model, Block 3 (R^2 =.485)	.047*			
AYA age		.026	.482	.631
Presence of SB		.130	2.215	.028*
Income		.020	.335	.738
Parent perception EF BRI		-.038	-.493	.622
Parent perception EF MCI		-.031	-.389	.698
Family cohesion		-.009	-.132	.895
Family satisfaction		.301	4.014	<.001*
Family resources		.107	1.365	.174
Stress		-.154	-2.566	.011*
Parent Depressive Symptoms		-.274	-4.197	<.001*

Note. * $p < .05$. Dependent variable: Family Quality of Life

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Table 15

Independent Samples Test

		Levene's Test for Equality of Variances		t-test for Equality of Means						
									95% Confidence Interval of the Difference	
		F	*Sig.*	*t*	*df*	*Sig. (2-tailed)*	Mean Difference	Std. Error Difference	Lower	Upper
Parent perception EF BRI	Equal Variances assumed	12.09	.001	3.719	207.00	< .001	5.43	1.46	2.55	8.31
	Equal variances not assumed			3.807	200.05	**< .001***	5.43	1.43	2.62	8.24
Parent perception EF MCI	Equal variances assumed	7.26	.008	6.178	207.00	< .001	9.39	1.52	6.39	12.38
	Equal variances not assumed			6.275	205.89	**< .001***	9.39	1.50	6.44	12.34
Family Cohesion	Equal variances assumed	.15	.698	-0.107	207.00	.915	-.08	.78	-1.63	1.46
	Equal variances not assumed			-0.107	199.41	.915	-.08	.79	-1.64	1.47
Family Satisfaction	Equal variances assumed	2.02	.157	-0.744	206.00	.458	-.06	.09	-.23	.11
	Equal variances not assumed			-0.750	205.98	.454	-.06	.09	-.23	.10
Family Resources	Equal variances assumed	14.01	< .001	-3.476	207.00	.001	-.21	.06	-.34	-.09
	Equal variances not assumed			-3.563	198.65	**< .001***	-.21	.06	-.33	-.10
Parent Stress	Equal variances assumed	3.33	.070	1.346	207.00	.180	4.91	3.64	-2.28	12.09
	Equal variances not assumed			1.359	206.96	.176	4.91	3.61	-2.21	12.02
Parent Depressive symptoms	Equal variances assumed	3.70	.056	2.289	207.00	**.023***	2.44	1.06	.34	4.54
	Equal variances not assumed			2.340	201.44	.020	2.44	1.04	.38	4.49
Family Quality of Life	Equal variances assumed	9.47	.002	-3.814	207.00	< .001	-6.78	1.78	-10.29	-3.28
	Equal variances not assumed			-3.915	196.88	**< .001***	-6.78	1.73	-10.20	-3.36

Note. Bold and * significant difference $p < .05$.

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Table 16

Correlations for SB and Comparison Subsamples

	1	2	3	4	5	6	7	8	9	10	11	12	13
Context variables													
1. AYA age	**1**	**-.175**	**.145**	**-.007**	**.207***	**-.065**	**.009**	**-.147**	**-.032**	**-.113**	**-.059**	**.171**	**-.060**
2. Income†	-.118	**1**	**.163**	**.144**	**.031**	**-.256***	**-.217***	**.187***	**.223***	**.329***	**-.285***	**-.418***	**.341***
3. Parent gender†	.047	-.026	**1**	**.280***	**.381***	**-.027**	**-.024**	**-.028**	**.081**	**.097**	**-.022**	**-.119**	**.111**
4. Race†	.082	.382*	.177	**1**	**.334***	**.097**	**.113**	**.009**	**.076**	**.002**	**.048**	**-.031**	**.095**
5. Ethnicity†	-.176	.088	.240*	.201*	**1**	**-.273***	**-.253***	**.034**	**.044**	**.189***	**-.117**	**-.119**	**.109**
6. Executive functioning-BRI	-.152	.038	.255*	.145	.188	**1**	**.656***	**-.047**	**-.216***	**-.414***	**.197***	**.372***	**-.285***
7. Executive functioning–MCI	-.086	.008	.260*	.269*	.162	.714*	**1**	**-.152**	**-.333***	**-.358***	**.282***	**.291***	**-.310***
Process Variables													
8. Family Cohesion	-.215*	.267*	-.107	-.159	-.070	-.213*	-.384*	**1**	**.568***	**.426***	**-.259***	**-.199***	**.301***
9. Family Satisfaction	-.262*	.159	-.239*	-.149	-.133	-.300*	-.445*	.714*	**1**	**.570***	**-.312***	**-.315***	**.532***
10. Family Resource	-.121	.195	-.165	-.254*	-.090	-.429*	-.504*	.480*	.605*	**1**	**-.514***	**-.487***	**.505***
11. Parent stress	.104	-.127	-.039	-.124	-.076	.089	.152	-.178	-.148	-.340*	**1**	**.416***	**-.375***
Proximal Outcome													
12. PDS	.151	-.272*	-.013	.048	-.133	.068	.273*	-.355*	-.365*	-.522*	.346*	**1**	**-.559***
Distal Outcome													
13. FQOL	-.084	.107	-.024	-.060	-.020	-.292*	-.385*	.454*	.508*	.570*	-.479*	-.423*	**1**

Note. Pearson Correlation reported for continuous bivariate correlations. †Spearman Rho reported when one variable is categorical; Group with SB correlations bold. *Correlation is significant at the $p < 0.05$ level (2-tailed).

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Table 17

Factors Related to FQOL in Subsample with SB

Model summaries	ΔR^2	*ß*	*t*	*p*
Context Block 1 (R^2 = .178)	.178*			
AYA age		-.024	-.271	.787
Income		.272	2.886	.005*
Parent perception EF – BRI		-.074	-.608	.544
Parent perception EF – MCI		-.202	-1.719	.089
Context and Process Block 2 (R^2 =.391)	.213*			
AYA age		-.026	-.314	.754
Income		.152	1.767	.080
Parent perception EF – BRI		-.048	-.422	.674
Parent perception EF – MCI		-.034	-.315	.753
Family Cohesion		-.053	-.544	.588
Family Satisfaction		.372	3.438	.001*
Family Resources		.166	1.438	.154
Parent stress		-.127	-1.340	.183
Full Model, Block 3 (R^2 =.471)	.081*			
AYA age		.030	.384	.702
Income		.077	.934	.353
Parent perception EF – BRI		.029	.274	.785
Parent perception EF – MCI		-.051	-.505	.614
Family Cohesion		-.034	-.376	.707
Family Satisfaction		.341	3.360	.001*
Family Resources		.099	.903	.368
Parent stress		-.044	-.481	.632
Parent Depressive Symptoms		-.361	-3.912	<.001*

Note. * $p < .05$. Dependent variable: FQOL

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Table 18

Factors Related to FQOL in Comparison Subsample

Model summaries	ΔR^2	*ß*	*t*	*p*
Context Block 1 (R^2 = .172)	.172*			
AYA age		-.119	-1.229	.222
Income		.093	.973	.333
Parent perception EF – BRI		-.059	-.432	.667
Parent perception EF – MCI		-.356	-2.612	.011*
Context and Process Block 2 (R^2 =.486)	.314*			
AYA age		.055	.658	.512
Income		-.054	-.667	.506
Parent perception EF – BRI		-.027	-.238	.812
Parent perception EF – MCI		-.037	-.304	.762
Family Cohesion		.126	1.101	.274
Family Satisfaction		.214	1.702	.092
Family Resources		.248	2.267	.026*
Parent stress		-.345	-4.164	<.001*
Full Model, Block 3 (R^2 =.494)	.008			
AYA age		.059	.714	.477
Income		-.086	-1.001	.319
Parent perception EF – BRI		-.064	-.549	.585
Parent perception EF – MCI		-.004	-.035	.972
Family Cohesion		.125	1.096	.276
Family Satisfaction		.211	1.684	.096
Family Resources		.202	1.734	.087
Parent stress		-.326	-3.872	<.001
Parent Depressive Symptoms		-.117	-1.152	.252

Note. * $p < .05$. Dependent variable: FQOL

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Publications

Ridosh, M., Braun, P. Roux, G., Bellin, M., Sawin, K. (2011). Transition in young adults with spina bifida: a qualitative study. *Child: Care, health and development*. *37*(6), 866-874. doi: 10.1111/j.1365-2214.2011.01329.x

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