UNDERSTANDING WELL-BEING AMONG CHILDREN BORN EXTREMELY PRETERM & THEIR CAREGIVERS: A MULTI-METHOD STUDY

Crisma Jazmin Emmanuel

A dissertation submitted to the faculty at the University of North Carolina at Chapel Hill in partial fulfillment of the requirements for the degree of Doctor of Philosophy in Nursing Science in the School of Nursing.

Chapel Hill
2021

Approved by:
Hudson P. Santos Jr.
Michael O’Shea
Kathleen Knafl
Sharron Docherty
Eric Hodges
ABSTRACT

Crisma Jazmin Emmanuel: Understanding Well-Being Among Children Born Extremely Preterm & Their Caregivers: A Multi-Method Study  
(Under the direction of Hudson P. Santos Jr.)

Background: Approximately 30-40% of extremely premature (EP) infants will develop a neurodevelopmental disorder. Individuals with neurodevelopmental disorders, and their caregivers, report lower quality of life and higher rates of stress as compared to the general population. Adverse outcomes which are further compounded by prevailing inequities in environment that disproportionately affect racial/ethnic minorities. Yet, despite the multiple risks associated with EP birth, there is still the possibility for positive health and well-being. However, literature focused on identifying and exploring potentially protective socio-environmental factors remains limited.

Method: This was a multi-method, three-chapter dissertation. Paper 1 was a secondary analysis of existing ELGAN data whose purpose was to identified specific individual, and familial, characteristics at age 10 that was associated with positive health outcomes at age 15. Paper 2 was a qualitative meta-synthesis whose aim was to synthesize qualitative studies exploring how families from a racial/ethnic minority background in the United States experienced well-being and responded to challenges faced while caring for a child diagnosed with three selected neurodevelopmental disorders. Finally, paper 3 was a qualitative descriptive
study focused on the perception, and experiences, of primary caregivers of extremely preterm children.

Results: Well-being was identified throughout this dissertation as a complex, personal experience for primary caregivers of EP children with/without neurodevelopmental disorders; with what was emphasized as being essential to well-being varying by the neurodevelopmental health of the child.

Conclusion: While well-being is a unique, personal experience, resilience in the face of multiple barriers is a necessity to develop well-being. Facilitators to well-being that were identified were: adaptation to an EP/neurodevelopmental diagnosis, becoming a resilient advocate and primary caregivers’ reliance on their spiritual/religious beliefs.
ACKNOWLEDGEMENTS

I would like to thank everyone both at the UNC School of Nursing and at home for all your support and guidance throughout this journey. First and foremost, I would like to thank my committee chair, and mentor, Dr. Hudson Santos who encouraged me and believed in me throughout this process. You did such a wonderful job balancing both practicality with empathy and kindness, and I learned so much about how to be a researcher and mentor from you. I would also like to thank Dr. Michael O'Shea for welcoming me into the ELGAN research team and giving me the opportunity to learn and work with such wonderful researchers, staff, and group of people. Specifically, thank you to the ELGAN moms, hearing your journeys was an inspiration. In addition, thank you to Dr. Kathy Knafl who has supported me throughout this dissertation journey- thank you for showing me just how important family research truly is. Also, thank you to Dr. Sharron Docherty and Dr. Eric Hodges for your guidance, critique, and encouragement throughout this process. Finally, but certainly not least, I would like to thank my classmates, the Hillman program and specifically Dr. Cheryl Jones, whose positivity and belief were essential through the ups and downs of the PhD program.

At home, I would like to thank my husband for always pushing me to be my best, even when I would have preferred to stay in my safety bubble. And lastly to my son and daughter, who are the inspiration for both this dissertation and everything I do. You have taught me to see the beauty in the diversity that exists in the human experience. Something I hope to continue and appreciate as you both grow.
TABLE OF CONTENTS

LIST OF TABLES .............................................................................................................. x

LIST OF FIGURES ........................................................................................................... xi

LIST OF ABBREVIATIONS ............................................................................................... xii

CHAPTER I: A MULTI-METHOD STUDY OF EXTREMELY PRETERM CHILDREN ....................... 1

Introduction ........................................................................................................................ 1

Background ....................................................................................................................... 2

Theory Background .......................................................................................................... 5

Aims ................................................................................................................................ 7

Summary of Chapters 2-5 ............................................................................................... 8

REFERENCES .................................................................................................................... 11

CHAPTER II: INDIVIDUAL AND FAMILIAL CHARACTERISTICS TO POSITIVE CHILD HEALTH OUTCOMES AMONG ADOLESCENTS BORN EXTREMELY_PRETERM ................................................................. 16

Introduction ..................................................................................................................... 16

Background ...................................................................................................................... 16

Methods ............................................................................................................................ 18

ELGAN Study Participants & Procedures ......................................................................... 18

Characteristics from Age 10 years .................................................................................. 18

Statistical Analyses .......................................................................................................... 20

Results .............................................................................................................................. 21
| **Introduction** | 65 |
| **Background** | 65 |
| **Methods** | 67 |
| **Study Design** | 67 |
| **Sample** | 67 |
| **Setting** | 67 |
| **Recruitment** | 68 |
| **Ethical Considerations** | 68 |
| **Data Collection** | 68 |
| **Data Analysis** | 69 |
| **Results** | 70 |
| I. Familial Impact to Health & Well-Being | 71 |
| II. Contributors & Barriers at the Community level | 76 |
| **Discussion** | 79 |
| **Research & Clinical Implication** | 82 |
| **Strengths & Limitations** | 83 |
| **Conclusion** | 83 |
| **REFERENCES** | 87 |
| **CHAPTER V: DISCUSSION & CONCLUSION** | 92 |

Chapter 2: “Individual and Familial Characteristics and Positive Child Health Among Adolescents Born Extremely Preterm” ................................................................. 93

Chapter 3: “Family members’ experience of well-being as racial/ethnic minorities raising a child with a neurodevelopmental disorder: A Qualitative Meta-Synthesis” ................................................................................................................................. 94
Chapter 4: “Mothers Experience of the Socioenvironmental Effects to Well-Being for Adolescents’ Born Extremely Preterm: A Qualitative Descriptive Study” ................................................................. 96

Well-Being & Its Facilitators ........................................................................................................ 98

Strengths & Limitations ............................................................................................................... 100

Research & Clinical Implications ............................................................................................... 101

Conclusion .................................................................................................................................. 102

REFERENCES ............................................................................................................................. 104

APPENDIX A ............................................................................................................................. 111
LIST OF TABLES

Table 2.1: Demographic Characteristics of Participants.......................................................... 25
Table 2.2 Potentially Protective Individual & Family Characteristics ........................................ 26
Table 3.1: CASP Appraisal of Included Studies........................................................................ 53
Table 3.2: Description of Included Studies............................................................................. 55
Table 3.3: Analytical & Descriptive Themes & Condensed Codes............................................. 60
Table 4.1: Semi-Structured Interview Guide............................................................................. 84
Table 4.2: Description of Caregiver/Child Demographic Characteristics 7............................ 85
LIST OF FIGURES

Figure 1.1: The Multi-Hit & Resilience Framework .......................................................... 7
Figure 2.1: Percentile Distribution of Youth’s Global Communication Score.................. 25
Figure 3.1: PRISMA Diagram of Reviewed Studies ............................................................ 52
# LIST OF ABBREVIATIONS

<table>
<thead>
<tr>
<th>Abbreviation</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>ASD</td>
<td>Autism Spectrum Disorder</td>
</tr>
<tr>
<td>ADD/ADHD</td>
<td>Attention Deficit Disorder/Attention Deficit Hyperactivity Disorder</td>
</tr>
<tr>
<td>CCC-2</td>
<td>Children’s Communication Checklist-2</td>
</tr>
<tr>
<td>CSI-4</td>
<td>Child Symptom Inventory Checklist-4</td>
</tr>
<tr>
<td>CLD</td>
<td>Chronic Lung Disorder</td>
</tr>
<tr>
<td>DSM-IV</td>
<td>Diagnostic and Statistical Manual of Mental Disorders, 4th edition</td>
</tr>
<tr>
<td>ELGAN Cohort</td>
<td>Extremely Low Gestational Age Newborn Cohort</td>
</tr>
<tr>
<td>EP</td>
<td>Extremely Preterm</td>
</tr>
<tr>
<td>GA</td>
<td>Gestational Age</td>
</tr>
<tr>
<td>GCC</td>
<td>General Communication Composite</td>
</tr>
<tr>
<td>ID</td>
<td>Intellectual Disability</td>
</tr>
<tr>
<td>LPA</td>
<td>Latent Profile Analysis</td>
</tr>
<tr>
<td>PROMIS</td>
<td>Patient-Reported Outcomes Measurement Information System</td>
</tr>
<tr>
<td>SES</td>
<td>Socioeconomic Status</td>
</tr>
<tr>
<td>UNC</td>
<td>University of North Carolina at Chapel Hill</td>
</tr>
<tr>
<td>U.S.</td>
<td>United States</td>
</tr>
</tbody>
</table>
CHAPTER I: A MULTI-METHOD STUDY OF EXTREMELY PRETERM CHILDREN

Introduction

Despite dramatic improvements in mortality, neurological disorders remain a significant concern for children born extremely prematurely (born <28 weeks gestation); with up to 40% of these children being diagnosed with a neurodevelopment disorder (Hirschberger et al., 2018; Joseph et al., 2016; Luu et al., 2017; Serenius, 2016; Synnes et al., 2018). Neurodevelopmental disorders are a significant public health problem, associated with lifetime costs of about one to two million United States (U.S.) dollars (Buescher et al., 2014; Rogge & Janssen, 2019) and lower quality of life for the child and their family as compared to neurotypical individuals (Benavente-Fernandez et al., 2020; Botting et al., 2016).

Within the preterm population, low socioeconomic status (SES) has been identified as a risk factor for the development of neurodevelopmental disorders (Constantino et al., 2020; Dubois et al., 2020; Forrest et al., 2018), including deficits in executive functioning, verbal reasoning, and academic performance (Dubois et al., 2020; Forrest et al., 2018; Hamed et al., 2015). In contrast, high SES has been identified as a potential protective factor; with preterm infants from higher SES demonstrating greater gains in cognitive scores compared to preterm infants from lower SES (O'Shea et al., 2009; Sprafkin, 2002). Currently, new studies have identified preterm children who do not develop adverse outcomes despite the biological insult(s) associated with preterm birth (Bangma et al., 2018; Jacqueline T. Bangma et al., 2019).
However, research exploring protective socio-environmental factors that promote these positive outcomes remains extremely limited. Within the Healthy People 2030 framework, one of the major plans of action is to stimulate research that promotes positive health and well-being across the lifespan for individuals with and without disabilities (Ochiai et al., 2021; U.S. Department of Health and Human Services). For this dissertation we defined well-being as 1) an absence of disease and 2) a person’s health and life satisfaction in three domains—physical, mental, and social (Jacqueline T. Bangma et al., 2019; Vik & Carlquist, 2018). Therefore, this dissertation advances existing knowledge of how socio-environmental factors at the individual, family, and community level have the potential to promote well-being among children at high risk for adverse developmental and health outcomes.

The objective of this chapter is to 1) Provide a brief background on neurodevelopmental disorders and positive child health and well-being among vulnerable populations 2) Describe research needs and how this dissertation addresses those needs 3) Describe the theoretical underpinning of this dissertation 4) Identify the aims of this dissertation and 5) Provide an overview of chapters two through five of this dissertation.

**Background**

Approximately 30-40% of extremely premature (EP) infants will develop a neurodevelopmental disorder (Hirschberger et al., 2018; Joseph et al., 2016; Luu et al., 2017; Serenius, 2016; Synnes et al., 2018). In this dissertation, we defined neurodevelopmental disorders as a group of conditions producing impairments of personal, social, academic, or occupational functioning (American Psychiatric Association, 2013). Unlike improved mortality rates, the prevalence of neurodevelopmental disorders has remained relatively unchanged among EP children (Luu et al., 2017; Rogers & Hintz, 2016); with decreasing gestational age associated
with a higher risk of adverse neurodevelopmental outcomes (Larsen et al., 2021; Serenius, 2016). In general, children with neurodevelopmental disorders have higher rates of physical and psychological co-morbidities—such as neurological and gastric conditions, anxiety, and depression (Gordon-Lipkin et al., 2018; Houghton et al., 2018; Lai et al., 2019; Muskens et al., 2017; Reale et al., 2017; Singh et al., 2019). In addition, individuals with neurodevelopmental disorders, and their caregivers, report lower quality of life and higher rates of stress as compared to the general population (Cappe, 2017; Ncube et al., 2018; van Heijst & Geurts, 2015).

Further compounding adverse outcomes, being a racial/ethnic minority in the U.S. results in greater risk for preterm birth (Beck et al., 2020; Janevic et al., 2018). It is important to note that race is a social construct that refers to a person’s externally perceived group membership (Burris et al., 2019). Thereby disparities in outcome between racial/ethnic groups in the U.S are attributable primarily to prevailing inequities in environment that disproportionately affect racial/ethnic minorities (Burris et al., 2019; Burris et al., 2020). Non-Hispanic Black Americans, for example, have a two-fold greater risk of preterm birth as compared to White Americans (Beck et al., 2020; Manuck, 2017). A compounding effect that follows the individual across the lifespan; racial/ethnic minorities with a neurodevelopmental disorder experience delayed diagnosis and less access to specialized care (Bishop-Fitzpatrick & Kind, 2017; Magaña et al., 2016). In this way, risks associated with being born extremely preterm are compounded by the socioenvironmental risk factors associated with being a racial/ethnic minority in the U.S. (Beck et al., 2020; Bishop-Fitzpatrick & Kind, 2017; Magaña et al., 2013; Manuck, 2017).

Notably, despite elevated risk some EP children, including racial/ethnic minorities, do not develop a neurodevelopmental disorder or adverse health condition. A 2018 study from the Extremely Low Gestational Age Newborn (ELGAN) Cohort shows that about 32% of the
ELGAN children (284 of 889) had positive health at age 10 (Bangma et al., 2019); defined as not having any major health disorders, including neurodevelopmental, physical, or mental health disorders (Bangma et al., 2019). Potentially protective factors associated with these positive outcomes include perinatal or early childhood variables, such as gestational age and specific medical procedures (Bangma et al., 2019). Yet, beyond proxies for SES (e.g., income, maternal education), researchers have not fully explored how socio-environmental aspects at the family and community level can be potentially protective to positive health and well-being among these high-risk populations (Bangma et al., 2019; Joseph et al., 2018).

Among full-term infants, higher parent-child interactions and social support throughout childhood have been known to act as protective factors for child health and development, even among infants exposed to low SES, food insecurity and single marital status (McDonald et al., 2016). However, considering the biological insults associated with preterm birth, it remains largely unexplored whether these factors will also be protective among extremely preterm children. In the general population, for example, early intervention programs that target children from lower SES have seen improved development and health outcomes, with participants demonstrating higher rates of educational achievement, health, and quality of life (Campbell et al., 2014; Hajizadeh et al., 2017). However, among preterm infants, these interventions have not been as consistently effective, possibly due to the increased number of biological and environmental risks associated with preterm birth (Benavente-Fernández et al., 2019; McCormick et al., 2006). Thus, research targeting the general child population cannot be generalized to extremely preterm infants who experience a different developmental trajectory.

Definitions for positive health and well-being among EP children have focused largely on the absence of neurodevelopmental and/or health conditions (Bangma et al., 2018; Forrest et al.,
This approach fails to explore how caregivers and/or family members define positive child health and well-being, making it difficult to understand how caregivers or families with EP children, particularly caregivers of children who do develop a neurodevelopmental disorder, conceptualize positive health and well-being.

**Theoretical Background**

This dissertation is based on an adapted version of the multi-hit hypothesis (also known as the cumulative stress hypothesis) (Kaindl et al., 2009; Sarkar et al., 2019). We propose that early neurodevelopmental disorders among extremely preterm children are precipitated by perinatal neurodevelopmental insults (i.e., hits), making the child more vulnerable to additional insults from the environment (Davis et al., 2016; Knudson, 1971; Picci & Scherf, 2015). Within preterm research, the multi-hit hypothesis has been used to explain brain injury among preterm infants- where antenatal factors act as the initial hits that make the brain sensitized to additional postnatal hits that then result in brain injury (Barnett et al., 2018; Davis et al., 2016; Leviton et al., 2013). We wanted to expand this hypothesis to also include potentially protective socio-environmental factors.

The central idea for this study is that multiple insults before and after birth make preterm children vulnerable to the development of a neurodevelopmental disorder. These neurodevelopmental disorders and insults, in turn, result in poorer health and well-being. In contrast, more positive socio-environmental conditions act as potentially protective factors to positive health and well-being, even when the EP child has a neurodevelopmental disorder.

Prior research supporting these ideas within the preterm population have identified SES to act as both a risk and protective factor to neurodevelopment, with an effect size similar in magnitude to that of neonatal brain injury (Benavente-Fernandez et al., 2020; Benavente-
Fernández et al., 2019). Benavente-Fernández (2019), identified higher SES as an effect modifier of the association between neonatal brain injury and worse neurocognitive outcomes. In the ELGAN cohort, children whose mothers were identified as having social disadvantage at birth, indexed by maternal education, had worse neurocognitive and academic outcomes at age 10, independent of gestational age (GA) (Joseph et al., 2018). In addition, advancements in maternal education between birth and the 10-year visit were associated with improvements in inhibitory control and processing speed (Joseph et al., 2018). However, differences in other socio-environmental factors, such as receiving behavior therapy, have had mixed results as compared to the general population (Benavente-Fernández et al., 2019). We hypothesize that preterm children have distinct neuronal vulnerabilities from early insults/hits and are more sensitized to early socio-environmental risk factors and are less responsive to improved SES later in childhood. While the role of SES has been explored, how caregivers believe social-environmental factors result in improved cognitive outcomes, and the process by which they do so, remains largely unexplored. Figure 1.1 shows the proposed relationship within the adapted multi-hit hypothesis:
Figure 1.1: The Multi-Hit & Resilience Framework

The arrows represent the EP child’s development across childhood. The lines with the captions of first and additional hit(s) represents the insults that are believed to affect EP/preterm infants. Within the figure, the stars represent potentially protective factors to both child health and well-being. Finally, within the blue arrow, potential socio-environmental factors that act as protective or risk factors at the individual, family and community level are depicted.

Aims

The object of this dissertation was to identify and describe the effect socio-environmental factors play in extremely preterm children’s positive child health and well-being by achieving the following aims:

1) Explore potentially protective factors beyond the perinatal/early childhood period for EP children and their effect on overall child well-being and neurodevelopment (Chapter 2).

2) Describe how families in high-risk populations experience and develop well-being when their child has a neurodevelopmental condition (Chapter 3)
3) Explore and describe primary caregivers’ experience of socio-environmental factors that affect their EP child’s, and their own, well-being (chapter 4).

**Summary of Chapters 2-5**

Chapters 2, 3 and 4 are the three manuscripts that correspond with each of the three aims of this dissertation.

Chapter 2: “Individual and Familial Characteristics to Positive Child Health Outcomes Among Adolescents Born Extremely Preterm” (Aim 1).

Chapter two is a quantitative, secondary analysis whose purpose was to identify specific individual, and familial, characteristics at age 10 associated with positive child health outcomes at 15 years of age. We evaluated characteristics at age 10 to achieve our first aim- which was to identify protective, or risk, factors beyond the perinatal/early childhood period that could be potentially protective to positive child neurodevelopment and well-being. Currently, research has focused on early childhood factors that have the potential to improve EP well-being, with research focused on factors and socio-behavioral interventions beyond this period having mixed results in effectiveness (Benavente-Fernandez et al., 2020; McCormick et al., 2006). Therefore, by achieving this aim we would provide researchers and clinicians with more information about specific characteristics of the socio-environment that have been identified as potentially capable of promoting increased well-being for the EP populations beyond the early childhood period—allowing for more targeted interventions across childhood.

Chapter 3: “Family members’ experience of well-being as racial/ethnic minorities raising a child with a neurodevelopmental disorder: A Qualitative Meta-Synthesis” (Aim 2).

While we were able to identify individual and familial characteristics that have the potential to promote EP well-being in chapter 2, we based our definition of well-being from the
clinical/research perspective. How well-being is defined by families remained largely unexplored. Therefore, chapter three is a qualitative meta-synthesis whose aim was to systematically synthesize qualitative studies exploring how families from a racial/ethnic minority background in the United States 1) experienced well-being and 2) responded to challenges they faced while caring for a child diagnosed with three selected neurodevelopmental disorders: Autism spectrum disorder, Attention Deficit Hyperactivity Disorder, and Intellectual Disability. It has been submitted to the Journal of Research in Nursing & Health. We chose to focus on what we identified as vulnerable families at the greatest risk for adverse well-being—specifically racial/ethnic minorities with an already diagnosed neurodevelopmental disorder. By focusing on this population, we were able to identify how well-being is experienced, and maintained, in the face of multiple socio-environmental risk or protective factors that have the potential to affect EP children’s well-being.

Chapter 4: “Caregivers’ Perception of Socio-Environmental Characteristics & Their Extremely Preterm Child’s Well-Being” (Aim 3).

However, despite EP children being at a substantially higher risk of neurodevelopmental disorders, we were unable to identify any information on how prematurity affected well-being by participants in chapter 3; with no information provided on prematurity. Therefore, chapter four is a qualitative descriptive study whose aims were to 1) describe primary caregivers’ perspective of how social-environmental, and family, characteristics affect their child’s, and their own, well-being and 2) describe primary caregiver’s perspective on their role in their child’s development and well-being. Twenty caregivers of EP children participating in ELGAN at one research site were interviewed. The Journal of Pediatric Nursing Research is the proposed journal for submission of this manuscript.
Chapter 5: Discussion & Conclusion

Chapter 5 provides a discussion of results and implications from the three manuscripts discussed in chapters 2-4. In addition, strength and limitations of the dissertation study are discussed as well as research/clinical implication of findings.
REFERENCES


CHAPTER II: INDIVIDUAL AND FAMILIAL CHARACTERISTICS TO POSITIVE CHILD HEALTH OUTCOMES AMONG ADOLESCENTS BORN EXTREMELY PRETERM

Introduction

Individuals born preterm have an increased risk of chronic health and/or developmental disorders; with prevalence of adverse outcomes as high as 78% among individuals born extremely preterm (< 28 weeks gestation) (Crump et al., 2019; Luu et al., 2017). Nonetheless, despite the biological insults they encounter in the perinatal period, individuals born extremely preterm (EP) can develop positive health (Bangma et al., 2018; Bangma et al., 2019). With positive health being defined as both 1) an absence of disease, and 2) positive cognitive and social development and emotional well-being (Bangma et al., 2019). Among diverse participants in the Extremely Low Gestational Age Newborn (ELGAN) Cohort study of individuals born before 28 weeks of gestation, up to 35% were free of 11 major medical diagnoses (e.g., cerebral palsy, autism) at age 10 (Bangma et al., 2018). However, limited research has focused on individual or familial characteristics that promote these positive health outcomes among EP children (Benavente-Fernández et al., 2019; Forrest et al., 2018).

Background

Current research has identified characteristics that are associated with adverse health and cognition among preterm children during early childhood including: 1) testing positive for internalizing and/or externalizing symptomology, and 2) communication deficits (Bangma et al., 2019; Benavente-Fernandez et al., 2020; Botting et al., 2016; Forrest Claire et al., 2020; Joseph et al., 2016). Unfortunately, whether an absence of these characteristics results in improvements
in health has remained largely unexplored (Bangma et al., 2019; Benavente-Fernandez et al., 2020). Instead, individuals with these features are described as less likely to be resilient and have worst psycho-social outcomes during adolescence and adulthood (Botting et al., 2016; Dubois et al., 2020). Therefore, for this study we wanted to begin to address this gap by identifying whether the absence of these characteristics would result in more positive health outcomes.

Beyond individual characteristics, research on familial factors that promote better health and cognition remains limited; with most findings focusing on familial factors during the perinatal or early childhood period (McDonald et al., 2016; Soni et al., 2021). While there is consensus that certain cognitive features remain stable after certain periods of childhood development, whether health outcomes (general health and neurocognitive outcomes) can be affected by better familial factors beyond early childhood remains largely unexplored. By addressing this gap, interventions beyond early childhood can be developed; something that is significant, considering the average age of diagnosis for certain developmental disorders, particularly for vulnerable populations, occurs past the ideal, early childhood intervention period (Constantino et al., 2020; Hamed et al., 2015).

Therefore, for this study we set out to identify modifiable individual and familial characteristics at age 10 that are associated with positive child health outcomes (global health and neurocognitive outcomes) at 15 years of age. We hypothesized that selected individual and familial characteristics can result in potentially better child health and a reduced risk for adverse neurocognitive outcomes.
Methods

ELGAN Study Participants & Procedures

The Extremely Low Gestational Age Newborn Cohort (ELGAN) is a longitudinal, observational study (data collected at ages 2, 10 and 15 years of age) focused on neurological, positive health, psychiatric and behavioral outcomes (O'Shea et al., 2009). During the years 2002-2004, women delivering before 28 weeks’ gestation in 11 cities in 5 states were asked to enroll in the first phase of the study. A total of 1,506 infants, born to 1,249 mothers, were enrolled and 1,198 survived to age 10 years. Of these, 966 were recruited at age 10 and 889 participated. At age 15 years, 694 completed data collection, which included both participants who participated at age 10 and those from earlier cohorts. All procedures for this study were approved by the institutional review boards of all participating institutions.

At age 15, child measures were selected to address the ELGAN study’s focus on psychiatric, cognitive, behavioral, and neurological outcomes at age 10. Most participants were assessed in a single day, but, when necessary, a second day was used. While the child was tested, the parent or caregiver, hereafter referred to as parent, was interviewed and completed questionnaires regarding the child’s medical and neurological status in addition to behavioral outcomes.

Characteristics from Age 10 years

For this study, we assessed the effect of three individual level characteristics 1) an absence of internalizing symptoms 2) an absence of externalizing symptoms and a 3) absence of clinically significant impairments in communication. Both internalizing and externalizing symptoms were evaluated using the parent-completed Child Symptom Inventory Checklist-4 (CSI-4) (Sprafkin, 2002). The CSI-4 is a 97-item scale that evaluates the occurrence of
behavioral and emotional symptoms corresponding to disorders based on the Diagnostic and Statistical Manual of Mental Disorders, 4th edition (DSM-IV) to include: disruptive behavior disorders, such as attention deficit hyperactivity disorder (ADHD); Autism; and anxiety and mood disorders (Sprafkin, 2002). The CSI-4 has satisfactory psychometric properties with respect to reliability and validity (Sprafkin, 2002). To aid in classification of psychiatric co-occurrence we used the CSI-Positive Screens (diagnostic model based on symptom counts) rather than Symptom Severity scores (continuous quantitative model).

To measure the absence of communication deficits, the child had to have a score of greater than 80 on the Children’s Communication Checklist-2 (CCC-2), indicating no clinically significant impairments in communication (Norbury et al., 2004). The CCC-2 is a 70-item questionnaire which was completed by the child’s parent, with the General Communication Composite (GCC) providing a standardized summary score of the first eight subtests (Bishop, 2006; Norbury et al., 2004).

In addition to individual characteristics, three familial characteristics were measured: 1) absence of major illness in the family 2) absence of any job loss for either parent at any point in the child’s life 3) absence of parental separation. All three variables were dichotomous with a yes/no response obtained from a socioeconomic questionnaire completed by the child’s parent.

Health Outcomes at age 15 years

Global Health

Data was collected about the child’s health through the parent/guardian proxy Global Health Patient-Reported Outcomes Measurement Information System (PROMIS) measure and child/teacher proxy Pediatric Global Health PROMIS measure (Broderick et al., 2013) The Global Health PROMIS measure includes seven items assessing general, physical, mental and
social health. Internal consistency was 0.88 for the child-report form and 0.84 for the parent form (Christopher B. Forrest et al., 2014). The Global health PROMIS measure has been found to be a good representation of an individual’s overall assessment of their view of their own/the child’s health (C. B. Forrest et al., 2014). Therefore, to characterize global health outcomes, global health t-scores was constructed, with higher scores indicating better health (Broderick et al., 2013; Christopher B. Forrest et al., 2014).

To characterize neurocognitive outcomes, we used latent profile analysis (LPA) classification (previously reported in an ELGAN study) (Kuban et al., 2016) based on similarities in level of performance across all WASI-II Verbal and Nonverbal scales and the National Institutes of Health Toolbox Cognition Battery subtests (Weintraub et al., 2013). For this study, we separated latent profiles into a binary variable where one was normal/low normal cognition and zero was impaired cognition.

**Statistical Analyses**

We evaluated whether the selected individual and familial characteristics (collected at age 10) were associated with improved overall global health t-scores and cognitive outcomes (at age 15). Descriptive statistics, including frequencies and proportions, were used to describe the sample and determine the number of families and individuals who experienced the selected individual/familial characteristics.

For Global Health t-score outcomes, a Generalized Linear Model (GLM) was used to determine how selected characteristics affected global health t-scores. For cognitive outcomes, binomial logistic regression models were utilized to estimate odd ratios (ORs) with 95% confidence intervals (CI). Adjustments for maternal age, race/ethnicity, child’s sex, and birth weight z-scores as potential cofounders were done for both GLM and logistic regression. We
selected these factors for adjustment due to prior research indicating their effect on both
development and child health in the preterm/EP population (Bangma et al., 2019; Beck et al.,
2020; Fall et al., 2015). All analysis was conducted using SAS software. A p value of p<0.05
was considered for statistical significance.

Results

Of the 693 study participants at age 15, 52% were male and 48% were female. For age
15, 67% of their mothers identified as White, 24% identified as Black, and 9% identified as
Hispanic. For maternal education, 37% had less than 12 years of education, while 40% had
greater than 16 years of education at age 15 years of the child. In addition, 32% of the
participants’ families qualified for public insurance. Of the ELGAN children in this sample, 21%
had experienced white matter damage identified on neonatal cranial ultrasounds (See Table 2.1).

Individual & Family Level Characteristics

For individual level characteristics, the majority had a global communication score
greater than 80—indicating no communication deficits (Figure 2.1). Additionally, 88% and 75%
of the children had no internalizing or externalizing symptoms, respectively. At the family level,
a total of 76% of parents reported no history of parental separation, 65% reported no history of
major illness in the family, and 72% reported no parental job loss.

General Health Outcomes: Parent Proxy Measure

Based on parent/guardian proxy measures, the absence of internalizing symptoms at 10
years was associated with an increase in global health t-scores of 7.23 (p<0.01). In addition, an
absence of externalizing symptoms and communication deficits at 10 years was associated with
an increase in global health t-scores of 5.65 (p<0.01) and 6.51 (p<0.01), respectively. For
familial level characteristics, absence of parental job loss was associated with a significant
increase in global health t-scores of 1.72 (p=0.04). In addition, absence of parental separation was also significantly associated with an increase in global health t-scores of 2.00 (p=0.03). However, there was no significant associated between absence of major illness and the child’s global health t-scores (See Table 2.2).

**General Health Outcomes: Child/Teacher Proxy Measure**

For child/teacher proxy measures, absence of internalizing and externalizing symptoms was found to be significantly associated with an increase in global health t-scores of 2.53 (p=0.04) and 1.80 (p=0.04), respectively, but not absence of communication deficits. For family level characteristics, child/teacher proxy measures only identified an absence of job loss to be significantly associated with an increase in global health t-scores of 1.72 (p=0.04) (See Table 2.2).

For neurocognitive outcomes, only an absence of externalizing symptoms [OR= 2.00; CI: 1.22, 3.36)] and communication deficits [(OR= 4.7; CI: 2.7, 8.2)] were associated with better odds for better LPA outcomes. No significant association with cognitive outcomes was identified for any of the three selected familial factors (See Table 2.2).

**Discussion**

Prior studies have focused on perinatal and early childhood individual/familial traits, identifying potentially protective factors such as higher gestational age (Bangma et al., 2019) and maternal education (Joseph et al., 2018) to preterm/EP’s development and health. The current study expanded this research to analyze individual/familial level characteristics beyond early childhood that are potentially protective to extremely preterm youth’s health— allowing for more targeted interventions across childhood. A vital implication when diagnosis, and the resulting behavioral interventions, are often delayed to later childhood for certain children,
particularly those from vulnerable racial/ethnic minority populations (Hamed et al., 2015; Magaña et al., 2013; Magaña et al., 2016).

Our findings, indicating an absence of internalizing and/or externalizing symptoms at age 10 were associated with better odds for better global health at age 15 for EP adolescents, were consistent with prior research identifying the association between internalizing and/or externalizing behaviors and levels of resilience and health among preterm children (Flouri et al., 2019; Papachristou & Flouri, 2020; Poehlmann-Tynan et al., 2015). Also, like prior preterm studies, cognitive outcomes were more stable at this later point in childhood, with none of the family, and only two of the individual level characteristic being significantly associated with cognitive outcomes (Linsell et al., 2015; Rogers & Hintz, 2016). However, this study did identify an absence of externalizing symptoms and communication deficits were significantly associated with improved cognitive outcomes for EP children—indicating a need for further analysis to understand how improvements in communication and externalizing symptoms have the ability to affect various components of cognitive function.

The strengths of this study include the large sample of extremely preterm youth with diverse sociodemographic backgrounds. However, a limitation of the study is the difficulty determining whether our potentially protective traits are protective or are simply promoting factors associated with positive outcomes. Future studies would benefit from a comparison group that would allow for further analysis of these potentially protective factors. In addition, while we mainly operationalized positive child health as absence/reduction of disorders, the way parents/caregivers view well-being and positive health needs further study; particularly for vulnerable populations, such as racial/ethnic minorities at higher risk for adverse outcomes (Adams et al., 2021; Beck et al., 2020).
Our findings are consistent with those of prior studies in highlighting the importance of mental health and communication abilities to overall child health and development for extremely preterm children (Botting et al., 2016; Poehlmann-Tynan et al., 2015). In addition, it is important to note that family level characteristics that were identified to be associated with better global health differed between parent proxy and child/teacher proxy reports, indicating a difference in view between parents and their child/child’s teacher. However, the absence of parental job loss was associated with better global health t-scores by both parent and child/teacher proxy global health measures; results which are consistent with prior studies highlighting how higher socioeconomic status can be potentially protective to EP child health (Benavente-Fernandez et al., 2020; Benavente-Fernández et al., 2019; Joseph et al., 2016).

**Conclusion**

Overall, these findings indicate that behavioral treatment and a positive family environment could have the ability to promote positive child health and cognitive outcomes among preterm adolescents. However, more research is needed in conceptualizing caregivers’ views of positive child health, and familial characteristics they consider crucial for child health; particularly for vulnerable sectors of the preterm population, such as racial/ethnic minorities (Adams et al., 2021; Beck et al., 2020).
Figure 2.1: Percentile Distribution of Youth’s Global Communication Score

Table 2.1: Demographic Characteristics of Participants

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Number (Percentage)</th>
<th>Age 10 Sample (N=889)</th>
<th>Age 15 Sample (N=693)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Newborn Sample (N=1506)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sex – n (%)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>799 (53.0%)</td>
<td>455 (51.2%)</td>
<td>358 (51.7%)</td>
</tr>
<tr>
<td>Female</td>
<td>707 (47.1%)</td>
<td>434 (48.8%)</td>
<td>335 (48.3%)</td>
</tr>
<tr>
<td>Maternal race – n (%)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Black</td>
<td>427 (28.9%)</td>
<td>227 (25.6%)</td>
<td>158 (23.7%)</td>
</tr>
<tr>
<td>White</td>
<td>866 (58.5%)</td>
<td>562 (63.4%)</td>
<td>449 (67.2%)</td>
</tr>
<tr>
<td>Other</td>
<td>187 (12.7%)</td>
<td>98 (11.1%)</td>
<td>61 (9.1%)</td>
</tr>
<tr>
<td>Maternal ethnicity – n (%)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hispanic</td>
<td>179 (12.0%)</td>
<td>86 (9.7%)</td>
<td>63 (9.1%)</td>
</tr>
<tr>
<td>Non-Hispanic</td>
<td>1313 (88.0%)</td>
<td></td>
<td>627 (90.9%)</td>
</tr>
<tr>
<td>Maternal education (years) – n (%)</td>
<td>800 (90.3%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>----------------------------------</td>
<td>------------</td>
<td></td>
<td></td>
</tr>
<tr>
<td>≤12</td>
<td>367 (41.3%)</td>
<td>367 (41.3%)</td>
<td>249 (37.3%)</td>
</tr>
<tr>
<td>&gt;12 - &lt;16</td>
<td>210 (23.6%)</td>
<td>210 (23.6%)</td>
<td>153 (22.9%)</td>
</tr>
<tr>
<td>≥16</td>
<td>312 (35.1%)</td>
<td>312 (35.1%)</td>
<td>266 (39.8%)</td>
</tr>
<tr>
<td>White matter damage – n (%)</td>
<td>347 (23.9%)</td>
<td>188 (21.2%)</td>
<td>142 (20.5%)</td>
</tr>
<tr>
<td>Public insurance – n (%)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>No</td>
<td>575 (64.7%)</td>
<td>575 (64.7%)</td>
<td>452 (67.7%)</td>
</tr>
<tr>
<td>Yes</td>
<td>314 (35.3%)</td>
<td>314 (35.3%)</td>
<td>216 (32.4%)</td>
</tr>
</tbody>
</table>

Table 2.2 Potentially Protective Individual & Family Characteristics

<table>
<thead>
<tr>
<th>Potentially Protective Factors to General Health</th>
<th>Adjusted OR (95% CI)</th>
<th>Unadjusted OR (95% CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. GCF ≥ 80</td>
<td>1.6 (1.0, 2.7)</td>
<td>1.9 (1.2, 3.1)*</td>
</tr>
<tr>
<td>2. No internalizing symptoms</td>
<td>2.2 (1.3, 2.9)*</td>
<td>2.2 (1.3, 2.9)*</td>
</tr>
<tr>
<td>3. No externalizing symptoms</td>
<td>1.9 (1.3, 3.7)*</td>
<td>2.0 (1.3, 3.0)*</td>
</tr>
<tr>
<td>4. No parental separation</td>
<td>1.1 (0.7, 1.7)</td>
<td>1.2 (0.7, 1.8)</td>
</tr>
<tr>
<td>5. No major illness in family</td>
<td>1.5 (1.0, 2.2)</td>
<td>1.5 (1.0, 2.2)</td>
</tr>
<tr>
<td>6. No parental job loss</td>
<td>1.0 (0.6, 1.5)</td>
<td>1.1 (0.7, 1.6)</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Potentially Protective Factors to Mental Health</th>
<th>Adjusted OR (95% CI)</th>
<th>Unadjusted OR (95% CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. GCF ≥ 80</td>
<td>4.8 (2.8, 7.7)*</td>
<td>4.6 (2.8, 7.7)*</td>
</tr>
<tr>
<td></td>
<td>LPA (95% CI)</td>
<td>LPA (95% CI)</td>
</tr>
<tr>
<td>-----------------------------</td>
<td>-------------</td>
<td>-------------</td>
</tr>
<tr>
<td>2. No internalizing symptoms</td>
<td>3.2 (1.9, 5.6)*</td>
<td>3.2 (1.9, 5.6)*</td>
</tr>
<tr>
<td>3. No externalizing symptoms</td>
<td>3.3 (2.7, 5.0)*</td>
<td>3.4 (2.3, 5.0)*</td>
</tr>
<tr>
<td>4. No parental separation</td>
<td>1.6 (1.1, 2.4)*</td>
<td>1.8 (1.2, 2.6)*</td>
</tr>
<tr>
<td>5. No major illness in family</td>
<td>1.2 (0.9, 1.8)</td>
<td>1.3 (0.9, 1.8)</td>
</tr>
<tr>
<td>6. No parental job loss</td>
<td>1.1 (0.8, 1.6)</td>
<td>1.2 (0.8, 1.7)</td>
</tr>
</tbody>
</table>

**Potentially Protective Factors to LPA**

<table>
<thead>
<tr>
<th></th>
<th>LPA (95% CI)</th>
<th>LPA (95% CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. GCF ≥ 80</td>
<td>4.7 (2.7, 8.2)*</td>
<td>5.6 (3.32, 9.28)*</td>
</tr>
<tr>
<td>2. No internalizing symptoms</td>
<td>1.1 (0.55, 2.30)</td>
<td>1.1 (0.57, 2.25)</td>
</tr>
<tr>
<td>3. No externalizing Symptoms</td>
<td>2.0 (1.22, 3.36)*</td>
<td>2.2 (1.37, 3.57)*</td>
</tr>
<tr>
<td>4. No parental separation</td>
<td>1.2 (0.7, 2.1)</td>
<td>1.0 (0.6, 1.6)</td>
</tr>
<tr>
<td>5. No major illness in family</td>
<td>1.2 (0.7, 2.0)</td>
<td>1.3 (0.8, 2.1)</td>
</tr>
<tr>
<td>6. No Parental job loss</td>
<td>1.4 (0.8, 2.3)</td>
<td>1.6 (1.0, 2.6)</td>
</tr>
</tbody>
</table>

* Statistical significance at p<0.05
REFERENCES


CHAPTER III: FAMILY MEMBERS’ EXPERIENCE OF WELL-BEING AS RACIAL/ETHNIC MINORITIES RAISING A CHILD WITH A NEURODEVELOPMENTAL DISORDER: A QUALITATIVE META-SYNTHESIS

Introduction

Neurodevelopmental disorders affect anywhere from 1-18% of the population worldwide and are associated with lifelong disabling effects (Cleaton & Kirby, 2018; Zablotsky et al., 2019). These disorders manifest in early childhood and are characterized by deficits in personal, social, and academic functioning (American Psychiatric Association, 2013). Within the wide diagnostic category of neurodevelopmental disorders as defined by the DSM-V, Autism Spectrum Disorder (ASD), Attention Deficit Hyperactivity Disorder (ADHD) and intellectual disability (ID) share a core set of symptoms—specifically impairments in communication, social skills, attention, and executive functioning (American Psychiatric Association, 2013; Thapar et al., 2017). It is important to note that ASD and ADHD themselves are not considered a disability; rather, the associated symptoms and common co-morbidities are what restrict individuals’ ability to fully function in their social setting (Miller & Rosenbaum, 2016; Ripamonti, 2016).

In addition to the core symptomology, individuals with ASD, ADHD and ID share similar adverse health outcomes—demonstrating higher rates of anxiety, depression, and suicidal ideation (Yoshimasu et al., 2019; Zahid & Upthegrove, 2017), lower quality of interaction with healthcare providers (Bishop-Fitzpatrick & Kind, 2017; Epstein et al., 2014), and lower quality of life compared to their neurotypical peers (Biggs & Carter, 2016; Lee et al., 2016).
Unsurprisingly, families of individuals diagnosed with these specific neurodevelopmental disorders also share common experiences such as social isolation, stigmatization, and the added stress associated with managing their child’s behavioral symptoms (Cappe, 2017; Walton, 2019). Unfortunately, there has been limited attention in understanding these experiences from the perspective of racial/ethnic minority families who have historically faced additional barriers to quality care (Alvarado & Modesto-Lowe, 2017; Bishop-Fitzpatrick & Kind, 2017). In the face of such challenges, the well-being of family members, and families, is an important consideration. Therefore, we aimed to conduct a meta-synthesis that explored how racial/ethnic minority families experienced well-being while caring for a child with ASD, ADHD and/or ID.

**Background**

Considerable attention has been paid to the adverse impact that raising a child with a neurodevelopmental disorder has on family members and caregivers- including poorer quality of life and higher rates of stress (Cappe, 2017; Walton, 2019). However, some research studies have demonstrated that families can achieve well-being while raising a child with a neurodevelopmental disorder; with caregivers often reporting high levels of personal enrichment (Bourke-Taylor et al., 2012; Ooi et al., 2016). For this synthesis, we defined a caregiver to be the primary individual, who has a personal relationship with the child, providing unpaid care with activities of daily living, medical tasks and other responsibilities associated with the child’s neurodevelopmental disorder (Family Caregiver Alliance, 2021). In addition, we conceptualized well-being as an outcome indicator of resilience, defined as a dynamic process that allows an individual or family to cope with adversity, change and opportunity in a manner that results in optimum family functioning and family members’ well-being (Richardson, 2002; Walsh, 2003). Well-being reflects a person’s life-satisfaction and health in three domains- physical, mental, and
social (HealthyPeople2020, 2014; Vik & Carlquist, 2018). Relevant examples for family members in this population would include feeling full of energy (physical well-being), a sense of optimism (mental well-being) and receiving quality support from family and friends (social well-being) (Ochiai et al., 2021).

General protective factors that have been associated with higher caregiver and child well-being include perceived social support, caregiver empowerment, and a positive perception of a child’s abilities and contributions to the family (Bourke-Taylor et al., 2012; Halstead et al., 2018). Unfortunately, most of this research has been conducted with White participants, with very low representation from participants from a minority racial/ethnic background. This deficit makes it difficult to determine whether these protective factors are the same for racial/ethnic minority groups who face additional adversities, such as discrimination and community stigma (Alvarado & Modesto-Lowe, 2017; Bishop-Fitzpatrick & Kind, 2017).

For the purpose of this review, racial/ethnic minority groups are based on the U.S. National Institute on Minority Health and Health Disparities (NIMHD) research framework to include: Blacks/African Americans, Hispanic/Latinos, Asians, American Indians/Alaska Natives and Native Hawaiians/Other Pacific Islanders (Alvidrez, Castille, Laude-Sharp, Rosario, & Tabor, 2019). Members of racial/ethnic minorities with a neurodevelopmental disorder in the U.S. are more likely to experience delayed diagnosis, lower access to general and specialty healthcare, and worse overall health outcomes (Bishop-Fitzpatrick & Kind, 2017; Magaña et al., 2016); adverse impacts which are also seen among members of their family.

It is important to note that race is a social construct that refers to a person’s externally perceived group membership (Burris et al., 2019). Therefore, adverse risk factors are mainly associated with the prevailing inequities within the U.S. for racial/ethnic minorities (Burris et al., 2019).
Asian and African American parents of children with ASD, for example, had higher levels of parenting stress compared to White parents; in contrast, Hispanic parents had one of the lowest levels of parenting stress (Alvarado & Modesto-Lowe, 2017; Kim et al., 2020; Williams et al., 2019). Such differences in health outcomes are difficult to interpret without further understanding of how well-being is experienced by caregivers, and family members, from a racial/ethnic minority background in the U.S.

For this study, we focused on these three neurodevelopmental diagnoses due to the shared core symptomology and family experiences associated with these conditions (American Psychiatric Association, 2013). While we realize that factors may differ between racial/ethnic populations, being a racial/ethnic minority in the U.S. carries a unifying experience with common adversities and adverse healthcare outcomes (Alvarado & Modesto-Lowe, 2017; Bishop-Fitzpatrick & Kind, 2017; Magaña et al., 2016). While several meta-syntheses have focused on synthesizing the experience of raising a child with ASD, ADHD or ID in the U.S. (Corcoran et al., 2015; Corcoran et al., 2017a, 2017b; Griffith & Hastings, 2014; Ooi et al., 2016), we were not able to identify any meta-synthesis that explored how U.S. families or caregivers from racial/ethnic minority populations experience and maintain well-being. Therefore, an increased understanding is needed of the role race/ethnicity plays in the process of attaining well-being for minority caregivers and family members.

The aims of our study were to conduct a meta-synthesis of qualitative studies exploring how caregivers, and families, from a racial/ethnic minority background in the U.S. 1) experienced well-being and 2) responded to challenges they faced while caring for a child diagnosed with ASD, ADHD and/or ID.
Methods

For this study, we conducted a meta-synthesis to integrate multiple qualitative findings in a manner that both attempts to preserve original meaning and develop innovative new insights (Mohammed et al., 2016; Sandelowski & Barroso, 2007). Specifically, we utilized a thematic synthesis for its ability to harvest thematic findings and synthesize, interpret, and create a deeper understanding of a challenging area under investigation; this methodology was finalized after an initial review of the final sample report. Therefore, our methodological procedures were 1) systematic literature search, 2) quality appraisal, 3) extraction of relevant findings 4) analysis of findings and 5) synthesis of findings (Thomas & Harden, 2008). We used the “Enhancing transparency in reporting the synthesis of qualitative research (ENTREQ) statement” as a guide (Tong et al., 2012).

Literature Search

A systematic literature search was conducted using four electronic databases- PubMed, PsychINFO, Scopus and Sociological Abstracts. A combination of MeSH, free-text and subject terms were utilized. A search string was developed for PubMed and adapted for the other databases with the assistance of a medical research librarian, who also provided guidance in the selection of electronic databases (Appendix A). The search terms were aimed at a broader, more general, experience of living with a neurodevelopmental disability, from which our identified aims could be met. To increase thoroughness, we also identified additional papers by going through the references of identified studies and including any relevant articles into our review. Finally, we went through the references of previously published meta-syntheses that focused on ASD, ADHD and/or ID to search for any relevant articles. The initial literature search took place
between November and December of 2019 and was regularly updated until October of 2021 when the manuscript was submitted.

**Study Selection**

We selected peer-reviewed articles that either 1) described how families raising a child with a neurodevelopmental disorder (specifically, ASD, ADHD and/or ID) experienced well-being, or 2) described how they responded to challenges they dealt with while caring for their child. We included both qualitative and mixed-method studies that used a qualitative method of data collection and synthesis. We incorporated data from caregivers, and their family members to give a sense of the family experience. Studies were excluded if they focused on families who did not identify as belonging to a racial/ethnic minority population as defined by the NIMHD research framework. However, studies that included both minority and non-minority participants were included if the data was separated by race/ethnicity, or most participants were racial/ethnic minorities. In addition, we excluded articles with a purely quantitative design, conference abstracts, dissertations, and commentaries. Literature reviews, meta-syntheses, intervention studies, discussion papers and book chapters were also excluded. No limitations on publication dates were placed nor were articles excluded based on quality assessment alone.

Figure 3.1 shows a flow diagram of our search process. Articles were imported into Covidence software, and 2,559 articles were identified after removal of duplicates. We assessed title and abstracts and then full-text articles based on eligibility criteria outlined above. Reasons for exclusion were recorded during full-text screening (i.e., wrong population and not addressing our study’s aims).
Quality appraisal Process

To ensure rigor and quality of the included studies, the Critical Appraisal Skills Programme ("CASP Checklists," 2018) was utilized by one reviewer and reviewed by the second reviewer for accuracy and consensus. The CASP has been utilized in other meta-syntheses (Schulman-Green et al., 2016) and is a well-recognized tool for its ability to evaluate the design and rigor of qualitative studies in a systematic and thorough manner ("CASP Checklists," 2018). Studies were not excluded based on quality; a decision based on recommendations from prior studies reporting that exclusion poses a risk to the comprehensiveness of the meta-synthesis (Sandelowski & Barroso, 2007). However, all included studies ranged from medium to high quality contributions (Table 3.1).

Process for Data Extraction & Analysis

Studies were organized based on type of diagnosis and date of publication. Data was extracted by the first author and reviewed for accuracy and thoroughness by a second author. Extracted information included: the aim(s), study design, neurodevelopmental diagnosis, race/ethnicity of participants, sample size, sample characteristics, data collection and analysis methods of included studies. In addition, results, key conclusions, and author stated limitations were extracted.

To address our research questions, we utilized Thomas and Harden’s (2008) thematic synthesis methodology. Data for analysis were based on unchanged texts extracted from the headings ‘results/findings’ of each primary study. One author then free coded line by line utilizing Atlas.ti. These line-by-line codes were then categorized into descriptive codes and collapsed successively based on similarities in concept through discussion with a second author into 15 broader codes. Data under these broader codes were further explored by the two authors
and then grouped by common concepts and ideas through an iterative process into six descriptive and three analytical themes in answer to the research aims. An audit trail was provided and periodically reviewed by a second author throughout this process to ensure rigor.

**Results**

**Study Characteristics**

There was a total of seven qualitative studies included in the final review, with a total of 85 racial/ethnic participants across the studies (numbers ranged from 3-27 participants). Four of the six articles focused on Latino/Hispanic participants, while only one study each focused on Southeastern Asian American participants, African American participants, and a mix of African American and Hispanic participants. Most of the participants were mothers/female caretakers of children whose ages ranged from three to nineteen years of age. In terms of diagnosis, five studies focused on children with an ASD diagnosis (Blanche et al., 2015; Lobar, 2014; Lovelace et al., 2018; Luong et al., 2009), while two focused on children diagnosed with ADHD (Araujo et al., 2017; Perry et al., 2005). No study included participants whose children were diagnosed solely with ID. Six of the seven studies did not provide degree of severity of the child’s diagnosis nor whether children were born prematurely. Six of the studies conducted semi-structured interviews and one conducted a focus group; methods ranged widely from qualitative descriptive to ethnomethodology and narrative inquiry. The study characteristics of included studies are presented in Table 3.2.

**Themes**

Due to the uneven number of racial/ethnic representation within the studies in this synthesis, it was not possible to analyze nuanced differences between racial/ethnic groups.
Therefore, unless specifically noted, the results presented apply to the general experience of being a racial/ethnic minority in the U.S.

A total of three analytical themes were developed—“moving toward well-being as a caregiver,” “family & culture: impact on well-being,” and “community and culture: impact on well-being.” These analytical themes were comprised of six descriptive themes that described participants’ experiences of well-being and facilitators, or barriers, to well-being. Table 3.3 provides a summary of analytical themes, descriptive themes, and categorized codes. In addition, Figure 3.2 provides a visual summary of participants’ view of factors from three levels (caregiver, family, and community) that acted as facilitators or barriers to family well-being.

I. Moving toward well-being as a caregiver.

Moving toward well-being as a caregiver was an overarching theme which conceptualized how caregivers, as individuals, developed and maintained well-being. Within this theme, two descriptive themes were identified: 1. experiencing well-being and 2. maintaining well-being—a cyclical process. It is important to note that while participants emphasized that well-being was possible, there seemed to be numerous obstacles unique to racial/ethnic minorities. Therefore, participants within this synthesis often emphasized adverse experiences versus states of well-being; with many focusing on increased rates of stress, and feelings of frustration and loneliness.

Experiencing well-being

Overall, there were subtle differences in how well-being was experienced between participants raising a child diagnosed with ASD versus ADHD. Participants raising a child diagnosed with ASD defined well-being as a dynamic process that culminated when the parent/caregiver shifted their focus from “fixing their child” to accepting their child’s differences
while nurturing a relationship with their child. This involved readjusting their expectations of their child, themselves, and their family (Blanche et al., 2015; Lobar, 2014; Luong et al., 2009). The following quote exemplifies a mother’s changed orientation to parenting her child with ASD:

I just wanted to see my son happy. I wanted to be around him, be in the moment, and enjoy him while he is still small… (Luong et al., 2009).

Families of children with ADHD also emphasized child well-being but additionally identified the importance of adopting a medical model where ADHD was viewed as a disease to be managed through medication or changes in parental behavior (Araujo et al., 2017; Perry et al., 2005). Both groups, however, emphasized the importance of recognizing and honoring the child’s unique capabilities (Blanche et al., 2015; Lobar, 2014; Luong et al., 2009; Perry et al., 2005). In addition, participants of both groups emphasized the importance of spiritual/religious beliefs in living peacefully and having hope for the future as exemplified by this quote from a mother of a child with ASD:

This is a disorder that obviously there is no cure for it. Not that there can’t be healing. I do believe in the power of God, and there may be healing...I have hope, and I tell him [my son], “You’re gonna get married, and you’re gonna go to college and have children and have a career” (Blanche et al., 2015).

This quote captures how acceptance and hope for the future are often balanced through the participant’s religious/spiritual beliefs.
Maintaining Well-being: A Cyclical Process

Well-being was described by both groups as a cyclical process in which the participants went through stages of adaptation throughout their child’s life—often culminating in the adoption of the role of resilient advocate (Blanche et al., 2015; Lobar, 2014; Lovelace et al., 2018; Luong et al., 2009; Perry et al., 2005). Prior to, and immediately after, the diagnosis participants described themselves as having a sense of urgency to identify the cause of behavioral symptoms, obtain a diagnosis, and initiate behavioral therapies for their child (Lobar, 2014; Lovelace et al., 2018; Luong et al., 2009; Perry et al., 2005). This urgency resulted in some mothers initially giving up their own jobs and deprioritizing personal goals to focus on their children’s daily and therapeutic care (Lobar, 2014; Luong et al., 2009). For some families these changes resulted in exhaustion—particularly in the mother, who often bore the additional brunt resulting from changes in routine and responsibilities (Blanche et al., 2015; Johnson et al., 2020; Lovelace et al., 2018; Luong et al., 2009). This exhaustion was resolved in several ways including: adopting a more realistic schedule, establishing routines, and accepting more respite and community care (Blanche et al., 2015; Lovelace et al., 2018; Luong et al., 2009). The following quote from the mother of a child with ASD exemplifies this experience:

I wanted my son to relax and not work all the time. I didn’t want to push him anymore because it only makes him tired and me tired. I don’t expect him to be a normal kid. I just wanted him to be safe and happy (Luong et al., 2009).

Another way well-being was maintained was in adopting the role of resilient advocate, which involved the ability of participants, mostly mothers, to persevere in accessing services and accommodations for their child and family despite multiple challenges, community stigma, and racial or ethnic discrimination (Araujo et al., 2017; Blanche et al., 2015; Lobar, 2014; Lovelace
et al., 2018; Luong et al., 2009; Perry et al., 2005). Becoming an advocate, however, was a skill that needed to be continually developed. Parents described an evolutionary process where they went from 1) very limited, or no, amount of knowledge about their child’s condition; 2) having the knowledge but not yet being able to advocate for their child; to 3) being able to utilize their knowledge with their child’s healthcare and educational teams to get necessary services and accommodations best fitted for their child and family (Blanche et al., 2015; Johnson et al., 2020; Lobar, 2014; Lovelace et al., 2018; Luong et al., 2009; Perry et al., 2005). One example of this was in the selection of therapeutic interventions. Participants often had to adopt a more realistic schedule to address burn-out and achieve personal and family well-being. This required participants to prioritize the type and frequency of their child’s therapeutic treatment, something which required both knowledge and the ability to advocate for themselves, and their family, in the face of multiple challenges from extended family members and healthcare professionals (Lovelace et al., 2018; Luong et al., 2009). The following quote from the mother of a child with ASD exemplifies this experience:

people will try to talk to me like I don’t know what I’m talking about, and they will make suggestions. . .Like I had a BSC [behavioral specialist consultant] at one agency and um she was just very condescending when she spoke to me and I had to tell her, “with all due respect, this does not work with my child, I’ve tried this” (Lovelace et al., 2018).

In addition, as resilient advocates from a racial/ethnic minority background, caregivers had to learn how to access services and address barriers to their child’s medical/behavioral care in the face of racial/ethnic discrimination, language barriers, and financial constraints (Araujo et al., 2017; Blanche et al., 2015; Johnson et al., 2020; Lobar, 2014; Lovelace et al., 2018; Luong et al., 2009; Perry et al., 2005). It is important to note, however, that while parents emphasized the
importance of being treated as members of their child’s healthcare team, their desired involvement did not always look the same. While some emphasized the ability to lead and manage their child’s treatment (Lovelace et al., 2018; Luong et al., 2009), others wanted healthcare providers to adopt that role (Perry et al., 2005).

II. Family & Culture: Impact on Well-Being

Participants’ families, and their cultural backgrounds, played a large role in the way caregivers experienced and attempted to maintain well-being. Within this analytical theme, there were two descriptive themes: 1) Familial View of Diagnosis and 2) Family Norms & Structure.

Familial View of Diagnosis

Often the type and degree of aid caregivers received depended on how families viewed the child’s diagnosis (Lobar, 2014; Lovelace et al., 2018). Many participants reported being told by their family members to “wait it out,” before seeking a diagnosis for their child or to “pray the condition” away (Araujo et al., 2017; Blanche et al., 2015; Lovelace et al., 2018). In other instances, extended family members recommended spanking or disciplining the child to address behavioral symptoms as exemplified by this quote from a mother of a child with ASD:

Yeah, I don’t really visit them [extended family] anymore. I’ll visit my grandma too, but [she says], “You gotta give her some little spanking so she could understand.” No, no, no, no, no. Oh, whatever. I’m not going to go there anymore (Blanche et al., 2015).

This often conflicted with the way participants viewed their child’s treatment and often led to a sense of isolation from extended family members (Araujo et al., 2017; Blanche et al., 2015; Lovelace et al., 2018; Perry et al., 2005), forcing many participants to rely on more formal sources of aid, such as the school and behavioral therapists (Luong et al., 2009; Perry et al.,
The following quote illustrates how mothers often relied on more formal resources to provide support for their child:

After the fourth grade, the teacher and I realized that he was not reading right, that he was getting behind slowly... We made a plan and decided to push him to see how much he improved. So, we worked together…(Perry et al., 2005).

However, whether this reliance was beneficial depended greatly on the teacher and school system. With many parents stating that teachers, and school staff, needed to receive more education about neurodevelopmental disorders to be a reliable source of support. In addition, learning to navigate the school system was often a difficult, and ongoing, process (Johnson et al., 2020; Perry et al., 2005).

**Family Norms & Structure**

In addition, for many of these families, there was a patriarchal system in place that often resulted in the mother being responsible for both the care of the child and ensuring smooth day-to-day functioning of the household. Some mothers expressed exhaustion, and occasionally resentment, toward their spouse who, many mentioned, offered insufficient aid (Araujo et al., 2017; Blanche et al., 2015; Johnson et al., 2020; Luong et al., 2009) as expressed by a mother of a child with ASD:

We’re gonna eat. She’s [child] gonna have a meltdown; she’s gonna throw stuff everywhere. We’re just gonna have to deal with it. My husband cannot deal with that. It just . . . freaks him out (Blanche et al., 2015).

Unfortunately, the support from extended family members that would have traditionally been offered was often absent due to a variety of factors, including extended family members living in another country, the stigma associated with having a child diagnosed with a neurological
disorder, being blamed for causing the disorder, or differences in opinion in how the child should be disciplined (Araujo et al., 2017; Blanche et al., 2015; Lobar, 2014; Lovelace et al., 2018; Luong et al., 2009; Perry et al., 2005). For single parent households, the exhaustion associated with being the primary caregiver was compounded by the fact that they also had to adapt to multiple, often competing, roles including being the primary financial provider, caregiver, and resilient advocate (Lovelace et al., 2018; Perry et al., 2005).

III. Community and Culture: Impact on Well-Being

Culture played a large role in how participants and the family unit interacted with members of their community, which in turn, affected their overall well-being. Within this analytical theme, there were two descriptive themes: 1) Coping with Community Stigma & Discrimination and 2) Facilitators to Well-Being in the Community.

Coping with Community Stigma & Discrimination

Many participants identified the stigma associated with their child’s condition within their community. With reactions ranging from labeling the child as *loco*, “crazy” to a misbehaving child in need of spanking and/or discipline (Araujo et al., 2017; Blanche et al., 2015; Lobar, 2014; Lovelace et al., 2018; Luong et al., 2009; Perry et al., 2005). This led participants to develop specific coping strategies to address their community’s stigma, including avoiding community functions and turning to members of their spiritual/religious organizations for acceptance and support (Araujo et al., 2017; Blanche et al., 2015; Lobar, 2014; Lovelace et al., 2018; Luong et al., 2009; Perry et al., 2005).

In addition to the stigma experienced within their immediate cultural/ethnic community, participants described experiencing both stigma and racial/ethnic discrimination from professional members of their extended community, particularly healthcare providers, and
teachers across their child’s life (Araujo et al., 2017; Blanche et al., 2015; Johnson et al., 2020; Lobar, 2014; Lovelace et al., 2018; Perry et al., 2005). African American participants, specifically, reported learning to “code-switch,” with their child’s healthcare and educational providers, including changing the way they spoke and behaved to access services and be viewed as valuable members of the treatment team (Lovelace et al., 2018). This difficult process was described by an African American mother of a child with ASD in this way:

   It was a struggle for me because I didn’t know how to go in there and say I’m not okay with this. I couldn’t breathe going into that meeting, and then on top of that meeting is the principal, the assistant principal, and all of his teachers (Lovelace et al., 2018).

This phenomenon was not identified in the other racial/ethnic participant sample, making it difficult to determine whether it is unique to the African American community or was missed in the other racial/ethnic groups.

**Facilitators to Well-Being in the Community**

While all the included articles described the barriers associated with unique community, and cultural, dynamics among racial/ethnic minorities, very limited attention was focused on the way the participant’s racial/ethnic community contributed to well-being. Of the few facilitators that were identified as contributing to well-being at the community level more formal sources of support were mentioned, particularly within their religious community (Araujo et al., 2017; Blanche et al., 2015; Lobar, 2014; Lovelace et al., 2018; Luong et al., 2009; Perry et al., 2005).

For those who reported relying on their religious/spiritual community, believing there was a spiritual plan for their family and integration into social activities in their church were particularly helpful in developing well-being (Blanche et al., 2015; Lobar, 2014; Luong et al.,
The following quote from the mother of a child with ASD exemplifies this experience:

I know the Lord has a plan for his life, and I know that He created who he is for a reason. He has a purpose. (Blanche et al., 2015)

It is important to note, however, that the way religion was utilized greatly affected whether it was a facilitator or barrier to well-being. While spirituality/religious beliefs were often seen as a source of comfort for caregivers, extended family or members of the community also utilized their religious/spiritual beliefs to blame or stigmatize the child and/or caregiver (Luong, 2009; Lovelace, 2018).

**Discussion**

This review synthesized the way in which racial/ethnic minority family members in the U.S. experienced and developed well-being while raising a child with ASD and/or ADHD. Compared to other literature focused on mainly White U.S. participants, well-being was experienced similarly by racial/ethnic minority families; with both syntheses emphasizing the importance of adapting to new roles and demands, developing an appreciation for the child’s diversity, and finding peace in the present (Lashewicz et al., 2017; Ooi et al., 2016). In addition, advocacy was seen by both minority and White caregivers as both a source of coping and a role they adopt- with both this synthesis and others highlighting the ongoing process of developing advocacy skills and becoming a resilient advocate (Boshoff et al., 2016; Corcoran et al., 2017; Laugesen & Groenkjaer, 2015).

Unfortunately, an increase in levels of stress, fatigue, and negative emotional outcomes was also seen in this and other literature focused on caregivers; with both White and racial/ethnic
minorities emphasized the increased levels of stress associated with raising a child with a neurodevelopmental disorder in our current society—where obtaining a diagnosis is associated with numerous barriers, and public stigma is still so prevalent (Dababnah et al., 2018; Laugesen & Groenkjaer, 2015; Magaña et al., 2013).

Differences, however, included the additional barriers racial/ethnic minorities faced to achieve well-being, including racial discrimination, more severe forms of family and community stigma, and a reduced support network—often because of being immigrants or first-generation families living in the U.S. In comparison, results from other syntheses of U.S. participants rarely, if ever, mentioned racial/ethnic discrimination as a barrier to well-being, even when racial/ethnic minorities were included in the sample (Corcoran et al., 2015; DePape & Lindsay, 2015; Laugesen et al., 2016). This difference emphasizes how vital research focused on racial/ethnic minorities is to obtain a more detailed understanding of racial/ethnic minority experiences.

Religion, and particularly church membership, was seen by many of the racial/ethnic minority participants as a source of social support for caregivers that increased their social and overall well-being. However, the way family often utilized religion/spiritual beliefs was seen as a barrier to well-being. In research focused on mainly White caregivers, spirituality/religion was minimally discussed as a method of coping (Superdock et al., 2018); racial/ethnic minorities were more likely to utilize religion as a coping method than White caregivers (Corcoran et al., 2015; Lashewicz et al., 2017; Twoy et al., 2007).

Limitations

Through this review we provided an overview of how some racial/ethnic U.S. minority families experienced and maintained well-being in the context of having a child with a neurodevelopmental disorder. Although we worked with an experienced research librarian to
identify all articles meeting our inclusion criteria, these criteria allow for the possibility of failing to include articles in which well-being may have been incidentally identified or which had a diverse group of participants but no clear separation of results by race/ethnicity. Nonetheless, our systematic approach to identifying eligible studies allowed for a thorough search of the existing literature.

In addition, it is important to note that due to the limited number of minority groups within this study—with four of the six articles focusing on Hispanic/Latinx participants, it is not possible to identify clear differences in the way specific racial/ethnic groups experienced well-being or addressed barriers to well-being; therefore, while we noted whether an experience was only identified in specific racial/ethnic groups, we were unable to fully explore the significance of these differences. As previously mentioned, race is a social construct that refers to group membership; therefore, it is to be expected that there be differences in experiences between race/ethnicities (Burris et al., 2019; Burris et al., 2020).

Additionally, very limited attention was focused on the strengths associated with being a member of a racial/ethnic minority family, or community, with most of the articles focusing on barriers to well-being at the family and community level (Araujo et al., 2017; Blanche et al., 2015; Lobar, 2014; Lovelace et al., 2018; Luong et al., 2009; Perry et al., 2005). Whether this deficit was because of the focus of the interview questions, or an actual reflection of participants’ experiences, remains unknown.

Finally, while we are aware that premature birth is associated with substantially increased risk of a neurodevelopmental disorder, few meta-syntheses have explored whether prematurity affects how families raising a child with a neurodevelopmental condition experience well-being
(Luu et al., 2017; Synnes et al., 2018). Therefore, more research is necessary to identify the unique role prematurity has on family well-being.

**Conclusion**

A total of three themes were identified in this meta-synthesis. Each of these themes included the idea that to develop well-being these families faced additional barriers, including racial/ethnic discrimination and stigma within their family and cultural community. While some strategies were mentioned in how well-being was developed, many of these studies focused on barriers to well-being. Thus, it is difficult to determine whether the few strategies mentioned do, in fact, result in higher overall well-being. Thus, there is a vital need for further, and more developed, research of how well-being is experienced and developed among racial/ethnic minorities raising a child with ASD, ADHD and/or ID.
Records identified through database searching (n = 3,639)

Additional records identified through other sources (n = 0)

Records after duplicates removed (n = 2,559)

Records screened (n = 2,559)

Records excluded (n = 2,510)

Full-text articles assessed for eligibility (n = 49)

Full-text articles excluded, with reasons (n = 42)
- Wrong Population: 31
- No Qualitative Methods Used: 5
- Not Focused on Well-Being: 3
- Duplicate: 2

Studies included in qualitative synthesis (n = 7)

Figure 1.1: PRISMA Diagram of Reviewed Studies
Table 3.1: CASP Appraisal of Included Studies

<table>
<thead>
<tr>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>1) Was there a clear statement of the aims of the research?</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
</tr>
<tr>
<td>2) Is a qualitative methodology appropriate?</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
</tr>
<tr>
<td>3) Was the research design appropriate to address the aims of the research?</td>
<td>Yes</td>
<td>Cannot Tell</td>
<td>Yes</td>
<td>Cannot Tell</td>
<td>Yes</td>
<td>Cannot Tell</td>
<td>Yes</td>
</tr>
<tr>
<td>4) Was the recruitment strategy appropriate to the aims of the research?</td>
<td>Yes</td>
<td>Cannot Tell</td>
<td>Yes</td>
<td>Cannot Tell</td>
<td>Yes</td>
<td>Yes</td>
<td>Cannot Tell</td>
</tr>
<tr>
<td>5) Was the data collected in a way that addressed the research issue?</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
</tr>
<tr>
<td>6) Has the relationship between researcher and participants been adequately considered?</td>
<td>Yes</td>
<td>No</td>
<td>Yes</td>
<td>Yes</td>
<td>No</td>
<td>No</td>
<td>Yes</td>
</tr>
<tr>
<td>7) Have ethical issues been taken into consideration?</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
</tr>
<tr>
<td>8) Was the data analysis sufficiently rigorous?</td>
<td>Yes</td>
<td>No</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
</tr>
<tr>
<td>9) Is there a clear statement of findings?</td>
<td>Yes</td>
<td>No</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
</tr>
<tr>
<td>10. How valuable is the research?</td>
<td>High quality</td>
<td>Medium quality</td>
<td>High quality</td>
<td>Medium quality</td>
<td>Medium quality</td>
<td>Medium quality</td>
<td>Medium quality</td>
</tr>
<tr>
<td>----------------------------------</td>
<td>--------------</td>
<td>----------------</td>
<td>--------------</td>
<td>----------------</td>
<td>----------------</td>
<td>----------------</td>
<td>----------------</td>
</tr>
<tr>
<td>Author (Year)</td>
<td>Aim</td>
<td>Study Design (Data Analysis)</td>
<td>Description of Participants</td>
<td>Description of Participants’ Child(ren)</td>
<td>Data Collection</td>
<td>Study Findings</td>
<td></td>
</tr>
<tr>
<td>---------------</td>
<td>-----</td>
<td>-----------------------------</td>
<td>-----------------------------</td>
<td>----------------------------------------</td>
<td>----------------</td>
<td>----------------</td>
<td></td>
</tr>
</tbody>
</table>
| Perry et al. (2005) | To explore how Latino parents managed their child’s ADHD within the sociocultural context of their everyday lives. | Grounded Theory (Open coding utilized) | 24 Latino Families -20 Mothers -6 Fathers -1 Grandmother | -Dx† (Severity): ADHD (Not Stated) -Age Range 6-19 (Median age 11yrs.) | Semi-Structured Interviews | 5 Categories Identified  
  - Finding Out About ADHD  
  - Taking On a Biomedical Meaning  
  - Living Between Two Cultures  
  - Caring for a Child with ADHD  
  - Looking Toward the Future with ADHD |
| Luong et al. (2009) | To focus on Southeast Asian American parents and their lived experience in caring for a child with ASD; focused on parent’s views and perspective on the effect of autism on families, their support system, and their coping style. | Not Stated (Identifiable as thematic and utilized inductive analysis) | 9 Southeast Asian American Families (All first generation) -8 Mothers -1 Father | -Dx† (Severity): ASD (Moderate to Severe) -Age Range 3-10 yrs. | Semi-Structured Interviews | 2 Themes Identified  
  - Coping Styles (9 Phases)  
    - Denial/Passive Coping  
    - Empowerment  
    - Redirecting Energy  
    - Shifting of Focus  
    - Rearranging Life & Relationships  
    - Changed Expectations  
    - Social Withdrawal  
    - Spiritual Coping  
    - Acceptance  
  - Support System |
<table>
<thead>
<tr>
<th>Study</th>
<th>Purpose</th>
<th>Participants</th>
<th>Diagnosis (Severity)</th>
<th>Semi-structured Interviews</th>
<th>Categories Identified</th>
</tr>
</thead>
</table>
| Lobar et al. (2014) | To establish data for a larger study on the context of family adjustment when faced with having a child who may have, or has, ASD within the context of the families’ worldview and belief system. | 9 Hispanic families (12 total families): 8 Mothers, 1 Grandmother, 2 Husbands of interviewed mothers | ASD (Not Stated-All verbal) | 5-15yrs. | 7 Categories Identified:  
  - Seeking Diagnosis  
  - Engaging in Routines to Control Behavior  
  - Finding Therapies  
  - Finding School Accommodations  
  - Educating Others  
  - Rising to Challenges  
  - Finding the Role of Spiritual & Religious Belief |
| Blanche et al. (2015) | To understand the caregiving experiences of Latino families with children with ASD, including daily activities, coping strategies, and service utilization. | 13 Latino Families: 12 Mothers, 3 Fathers | ASD (Not Stated) | 3-8 yrs. | 4 Themes Identified:  
  - Dealing with the Diagnosis  
  - Dealing with Stigma & Isolation from Family & Community  
  - Understanding the Role of Mothers in Changing Family Routines  
  - Utilizing Services |
| Araujo et al. (2017) | To understand the caregiving experiences of Latino families with children with ASD, including daily care. | (line-by-line coding with case comparison) 13 Latino caregivers: 11 Parents, 1 Father, 1 Grandmother | ADHD (Not Stated) | 7-10 yrs. | 3 Themes Identified:  
  - Acculturation  
    - Latino Families’ Social Relations & Activities  
    - Family Migration to the U.S.  
    - Difficulties Being Latino in the U.S.  
  - Family Dynamics  
    - Latino Family Relations |
<table>
<thead>
<tr>
<th>Study</th>
<th>Research Focus</th>
<th>Methodology</th>
<th>Sample Characteristics</th>
<th>Themes Identified</th>
</tr>
</thead>
</table>
| Lovelace et al. (2018) | To explore the impact of race on the lived experiences of three African American families with boys with ASD. | Descriptive Qualitative Methodology (Thematic Analysis) | 3 African American Families: -3 Mothers | 4 Themes Identified  
- Racial Discrimination  
- Complex Collaboration  
- Family Dynamics  
- Community Dynamics |
| Johnson et al. (2020)  | To explore the experience of accessing autism diagnosis and resources in a Midwest urban public school district for diverse mothers of children with autism spectrum disorder. | Narrative Inquiry Methodology (Thematic Analysis) | 9 Families: -5 African American Mothers -2 Hispanic Mothers | 3 Themes Identified  
- Late Diagnosis  
- Negative Communication Experiences  
- Coping Strategies |
Table 3.3: Analytical & Descriptive Themes & Condensed Codes

<table>
<thead>
<tr>
<th>Analytical Themes</th>
<th>Descriptive Themes</th>
<th>Condensed Codes (After Several Iterations)</th>
</tr>
</thead>
<tbody>
<tr>
<td>I. Moving Toward Well-Being as a Caregiver</td>
<td>1. Experiencing Well-Being</td>
<td>1. Adapting to Diagnosis</td>
</tr>
<tr>
<td></td>
<td></td>
<td>3. Spirituality</td>
</tr>
<tr>
<td></td>
<td></td>
<td>4. De-prioritization of Self</td>
</tr>
<tr>
<td>II. Family &amp; Culture: Impact on Well-Being</td>
<td>1. Familial View of Diagnosis</td>
<td>5. View of Diagnosis</td>
</tr>
<tr>
<td></td>
<td>2. Family Norms &amp; Structure</td>
<td>6. Patriarchal Family Dynamics</td>
</tr>
<tr>
<td></td>
<td></td>
<td>7. Religious Beliefs</td>
</tr>
<tr>
<td></td>
<td></td>
<td>8. Disciplining</td>
</tr>
<tr>
<td></td>
<td></td>
<td>9. Limited/No Family Support</td>
</tr>
<tr>
<td></td>
<td></td>
<td>10. Single Parenthood</td>
</tr>
<tr>
<td></td>
<td>2. Facilitators to Well-Being in the Community</td>
<td>12. Stigma</td>
</tr>
<tr>
<td></td>
<td></td>
<td>13. Discrimination</td>
</tr>
<tr>
<td></td>
<td></td>
<td>14. Difficulty Accessing Services</td>
</tr>
<tr>
<td></td>
<td></td>
<td>15. Healthcare &amp; Academic Institutions</td>
</tr>
</tbody>
</table>
Figure 3.2: Facilitators & Barriers to Family Well-Being
REFERENCES


CHAPTER IV: CAREGIVERS’ PERCEPTION OF SOCIO-ENVIRONMENTAL & FAMILIAL CHARACTERISTICS & THEIR EXTREMELY PRETERM CHILD’S WELL-BEING

Introduction

Despite dramatic improvements in mortality, adverse neurodevelopmental and long-term negative health outcomes remain a significant concern for extremely premature (EP) children (born <28 weeks gestation) (Joseph et al., 2016; Kuban et al., 2016; Luu et al., 2017; Serenius, 2016). Compared to those born full-term, EP children are at a higher risk for neurodevelopmental and psychiatric disorders (Crump et al., 2019; Luu et al., 2017; Sarkar et al., 2019), and have overall higher rates of physical comorbidities, such as respiratory and cardiometabolic conditions (Crump et al., 2019; Luu et al., 2017). Yet, recent studies have identified children that are able to stay free of adverse outcomes despite the biological insults associated with EP birth (Bangma et al., 2018; Bangma et al., 2019). Additionally, quality of life for some individuals born EP has been found to be comparable to those born full term (Roberts et al., 2013; Saigal, 2016).

Background

Currently, most research in the EP population has focused on identifying early childhood risk factors for adverse health and well-being (Barnett et al., 2018; Doyle et al., 2015; Janevic et al., 2018). Of the research that does explore protective factors, there has been a primary focus on early childhood development, with most studies utilizing quantitative data exploring overarching protective factors, such as higher socioeconomic status (SES) (Bangma et al., 2019; Benavente-Fernández et al., 2019), higher maternal education (Joseph et al., 2018) and higher infant
gestational age (Bangma et al., 2018; Bangma et al., 2019). However, there is limited contextualization and understanding of caregivers’ perceptions of how these socio-environmental, and familial, factors can affect child health and family well-being across childhood.

For this paper, we conceptualized well-being as an outcome indicator of resilience. Resilience is defined as a dynamic process that allows an individual or family to cope with adversity, change and opportunity in a manner that results in optimum family functioning and well-being (Richardson, 2002; Rutten et al., 2013; Walsh, 2003). Well-being reflects a person’s health and life satisfaction in three domains—physical, mental, and social (Ochiai et al., 2021; Vik & Carlquist, 2018).

Of the research that focuses on protective socio-environmental factors for EP individuals, in-depth family and community dynamics are often difficult to fully describe due to both their complexity and researchers’ pre-conceived beliefs of what constitutes significant factors. Therefore, we aimed to look at this phenomenon through the perspective of primary caregivers of EP individuals. While traditionally a parent, we defined a primary caregiver as anyone who took primary responsibility for the preterm individual’s ongoing care across childhood.

Currently, several researchers have been able to identify how vital primary caregivers are to an EP child’s well-being across childhood—often acting as both the EP individual’s caretaker and advocate. In addition, caregivers are often dramatically affected by the birth of an EP child, often viewing preterm birth as a traumatic and stressful experience (Gonçalves et al., 2020; Holditch-Davis et al., 2015).

Therefore, the aim of this paper was to 1) describe primary caregivers’ perspective of how family, and social-environmental characteristics affect their child’s, and their own, well-
being 2) to describe primary caregiver’s perspective on their role in their child’s development and well-being.

**Methods**

**Study Design**

We used a qualitative descriptive design (Doyle et al., 2020; Sandelowski, 2010) for this study. This design allowed us to obtain a subjective, rich description from primary caregivers in a manner that allowed for researchers to stay close to the data, and allow for mixing of data (Sandelowski, 2000). Something which was vital since this was an ancillary study of the Extremely Low Gestational Age Newborn (ELGAN) cohort study (O'Shea et al., 2009). ELGAN is a longitudinal, observational study (data collected at ages 2, 10 and 15 years of age for children born extremely preterm) focused on neurological, positive health, psychiatric and behavioral outcomes.

**Sample**

A purposive sample from the ELGAN cohort study was recruited to achieve maximum variation—specifically in neurodevelopmental and racial/ethnic backgrounds (Palinkas et al., 2015; Patton, 1990). We selected this sampling method to both describe variations in experiences and explore core elements and shared outcomes (Patton, 1990). Therefore, our criteria were: 1) a primary caregiver of a child currently enrolled in the ELGAN cohort study; 2) identified as the primary caregiver from the child’s early childhood to present 3) able to give informed consent.

**Setting**

This study took place in the southeastern United States. Due to COVID restrictions, we were unable to conduct interviews in participants’ homes as originally planned. Instead, primary
caregivers were interviewed via either a phone or zoom call depending on participant’s preference.

**Recruitment**

The primary investigator (PI) conferred with the clinical site coordinator at the University of North Carolina at Chapel Hill’s (UNC-CH) ELGAN Cohort study site to identify primary caregivers who met the inclusion criteria. Primary caregivers were informed about the study by the clinical site coordinator through either a routine ELGAN research visit or through a phone call. Primary caregivers who agreed to receive more information provided the study coordinator with a phone number and time for the PI to call to provide additional information. The PI then offered a verbal description of the study’s aims in addition to an emailed written document that provided a detailed description of the study’s aims and procedures. Caregivers who agreed to participate scheduled an interview visit with the PI. Participants were compensated with a gift card for their time.

**Ethical Considerations**

This study was approved by the institutional review board (IRB) at UNC-CH. The IRB granted approval to waive signed written informed consent, citing this study as a minimal risk to participants. Therefore, prior to the start of the interview, the PI emailed and explained the written-informed consent statement and verbal informed consent was obtained from all participants.

**Data Collection**

Semi-structured interviews were conducted via a secure zoom link or via telephone based on participant’s preference, at a day and time convenient to the participant. An interview guide was developed based on input and feedback from clinicians and researchers familiar with both
the ELGAN population and family research (see Table 4.1). In addition, our conceptualization of resilience and well-being informed the development of the interview guide. All the interviews were conducted by the PI, who is a bi-cultural and bi-lingual (English and Spanish), researcher with extensive training in child development and family health. Based on participant preference, 19 interviews were conducted in English, and one was conducted in Spanish using a professionally translated copy of the interview guide. Interviews lasted from 30 minutes to two hours, and an average of 30-45 minutes. 19 interviews were audio-recorded with the participant’s consent and transcribed verbatim by the PI. One participant did not want to be audio-recorded, but consented to being interviewed, with the interviewer taking detailed notes and verbatim quotes from the participant. All data was de-identified and saved on an encrypted drive. Audio recordings were deleted after transcripts were validated for accuracy. Accuracy of transcripts were validated by the PI listening to taped interviews twice and adding corrections/additions to transcripts as needed. A field journal of each step during the data collection was maintained for capturing analytical insights- field notes were reviewed and used during analysis to further analyze interview data.

Data Analysis

Dedoose (v9.0.7, SocioCultural Research Consultants, LLC, Los Angeles, CA), a qualitative data analysis software program, was utilized to assist with data organization and analysis. Inductive content analysis was conducted (Elo & Kyngäs, 2008). The PI read and re-read through all transcriptions to gain familiarity with the data set. The PI then completed initial coding of transcripts and field notes. Codes were defined as segments of text based on similar meaning or words (Elo & Kyngäs, 2008). A codebook was then developed based on caregivers’ own words and iteratively refined by the PI under the mentorship of a second researcher. Codes
were then iteratively organized into categories, which were defined as codes with similar meaning but with a higher level of abstraction than codes. The study’s research aims and the NIMHD model, which categories socio-environmental factors into specific levels, were then utilized to further analyze and organize categories into final sub-themes and overarching themes. This methodology is based on prior literature stating that even while utilizing inductive content analysis, the study’s aims, and prior theoretical models, can still contribute to data analysis (Armat et al., 2018; Elo & Kyngäs, 2008). Rigor was maintained by having the PI maintain an audit trail of memos and the PI peer debriefing with a co-researcher. Additionally, the PI was blinded to diagnoses until after data was analyzed. Finally, the PI and a second researcher reviewed the subthemes and themes to come to a final consensus (Morse, 2015).

Results

In total, 23 primary caregivers were contacted and 20 primary caregivers, all who identified as the children’s mother, agreed to participate in the study. Code saturation was observed after one fourth of the interviews were reviewed; however, meaning saturation of general experiences was only achieved after interviewing all participants (Hennink et al., 2016). It is important to note that while we included children with a range of neurodevelopmental, and health, outcomes, meaning saturation only related to the general experience of mothers raising an extremely premature child. Therefore, we were unable to compare experiences by variations in health and/or well-being.

Overall, the age of the mother’s children varied from 16-18 years of age at the time of data collection. In addition, racial backgrounds, as well as number and type of comorbidities of their EP child, were varied (see Table 4.2). There were two major themes identified: 1) Familial Impact to Health & Well-being 2) Contributors & Barriers at the Community level. The first
theme focused on environmental and social factors at the family level and consisted of three subthemes: a) *The Central Role of Motherhood*; b) *The Child’s Contribution to Health & Well-being*; and c) *The Family Unit*. The second theme, which focused on contributors and barriers at the community level consisted of two subthemes: a) *Formal Sources of Support*; and b) *Informal Sources of Support*.

I. Familial Impact to Health & Well-Being

A. *The Central Role of Motherhood*

Throughout each interview, mothers emphasized the central role they played in their EP child’s ongoing development and well-being. Of the 20 mothers interviewed, all identified themselves as the primary caregiver throughout their child’s life, having responsibility of most of the ongoing caregiving and day-to-day household tasks. Without appropriate levels of support, this heavy reliance on the mother often resulted in feelings of exhaustion and exacerbation of pre-existing, or new, mental, or physical health conditions. One mother, for example, described her experience as follows:

They [family members] wanted to be around her [the child], they just didn’t want to, um, touch her, because they thought she would break or, um, they—she would have a [respiratory] episode. And every—and if someone was holding her and they—and she, um, had a-an episode, then they would immediately hand her to me. Including my husband, including my— yeah. Everyone, I was literally stuck to her 24/7. For two years. I gained a lot of weight

A mother’s perspective which summarizes how all-consuming caring for a preterm child can be and the difficulties in obtaining childcare during early infancy when the EP child’s health conditions are still being identified and managed. It is important to note, however, that as children grew into adolescence, most (9 out of 10) reported resolving or learning to adapt to their child’s physical health conditions. For mothers reporting little social and, or, financial support,
however, high levels of stress were still common, particularly for those whose child had ongoing, or unmanaged, neurodevelopmental or health conditions.

While all mothers emphasized the importance of their role as caregivers, what this entailed varied by the child’s abilities. Mothers of children with mainly neurotypical abilities emphasized teaching their child independence, the need for normalcy, and transitioning into adulthood. In contrast, mothers of children with moderate to severe neurodevelopmental conditions emphasized the need to adapt to their child’s abilities, which often involved planning for ongoing care beyond childhood. A quote from a mother of a child with an intellectual disability, for example, summarizes how mothers matched their own expectations to their child’s unique capabilities:

My fear is how he going to be when he gets a little bit older. He wants to go to college. He said he-’I'm going to college’; he'll say ‘I'm going to state.’ He got high expectations for himself, and I do not discourage that. But I know he's going to have some, limitations-he cannot do that. So, he might have to do a tech. Now this is going on right now. Now as he gets older—but right now, I don’t see that. He's just going to have to have baby steps in life.

In addition, advocacy skills and the importance placed on advocating varied among mothers. Mothers described advocacy as a method of obtaining services, particularly within the school system, for their child. However, whether, or how, this was achieved varied. Mothers with a professional background in an academic setting, who identified a need for services, seemed better able to navigate the school system and work alongside school personnel to create personalized education plans. In contrast, mothers who identified a need for services, but were unfamiliar with the school system expressed frustration at the numerous barriers they experienced accessing services. It is important to note, however, that mothers whose child had milder neurodevelopmental symptoms varied in whether they believed their child needed
services; with many emphasizing the need for *masking*, which emphasizes the importance of appearing and acting as neurotypical as possible.

Unfortunately for families whose children still had developmental or learning differences, this emphasis on normalcy occasionally resulted in failing to identify or use resources used by those EP children with more severe diagnosis or symptoms- such as behavioral counseling, academic accommodations, or vocational rehabilitation services. The quote below summarizes how mothers struggle to balance these opposing needs:

I was kind of thankful that she didn’t have that label in school. Because she was able to stay in the mainstream classes even though she had an IEP [Individual Education Plan] and the PEP [Physical Education Program], um, she still was able to stay with her class. Cause it-it’s definitely a struggle… whenever you’re trying to incorporate a child with issues into a school and trying to make sure their life at the school is as normal as possible. Um, and the-but you want them to get all the help they can get while they’re at the school so they can be as normal as they can.

Individual coping factors mothers identified for personal well-being included their spiritual beliefs, which were utilized to explain both incidences of positive and adverse health and neurodevelopment. When asked about factors that contributed to her child’s positive health, one mother stated:

It was like a struggle because I had to watch her [inhales deeply] leave and come back, leave and come back, you know, like she going to die and came-come back. I’m watching her with these blood transfusions. So, it was a bit stressful on me, but [inhales deeply] by me believing in God I knew she was going to be alright, with the pull through, with my faith. I had to keep my faith in her.

Mothers typically described their relationship with their child as a central component to their, and their child’s, well-being. Mothers who felt they had a healthy, strong relationship with their child focused on their child’s positive personal characteristics, such as their inherent strength, kindness, and unique intelligence. In contrast, mothers who expressed having an overly dependent, or distant, relationship with their child often emphasized their child’s personal
weaknesses—such as difficulty with social interactions; often also expressing feelings of worry for the future.

**B. The Child’s Contribution to Well-Being**

It is important to note that despite any physical diagnosis their child was still living with, most mothers (19/20 mothers) described their child as physically healthy. While commonly initiated by mothers, or family members, the child’s participation in sports or outdoor activities was seen by most mothers to contribute greatly to their child’s overall well-being. In addition to sports, having hobbies in general that allowed the child to develop their own interests gave the child a sense of independence and self apart from the caregiver. Even for children with a neurodevelopmental disorder, having a hobby such as anime, cooking or electronic video games allowed the child to begin taking steps toward independence.

Although most mothers described their child as physically healthy, they varied in their description of the child’s overall well-being, with experiences ranging from loneliness and depression to the child being described as a happy and well-adjusted adolescent. Having age-appropriate interpersonal skills was seen as vital by most mothers to the promotion of psychological health and overall well-being in their child. Youth who were described as able to appropriately engage with peers were viewed as having more success in having their emotional and social needs met. In contrast, when mothers described the child as having reduced interpersonal skills, they also expressed concern about the child’s limited peer network, noting the child relied heavily on the mother or siblings for social engagement and support. One mother summarized her concern and frustration about her child’s limited social interactions, and its connection to a sedentary lifestyle in the following quote:
But if he go with friends, they, you know, we got kids around here, some of them might be his age some not, he’ll do it but other than that [NAME OF CHILD] stay right here under this house, up under me. He won’t care nothing about going outside playing. He'll go out for a little while and he’ll come right back in this house.

An effect which not only affects the child’s social well-being but has implications for the child’s long-term physical health.

C. The Family Unit

Mothers described the family unit as a key influence in the development of their EP child’s overall well-being, with family functioning and child well-being being interdependent. Having a supportive partner, even if they were divorced, seemed to greatly promote overall health and well-being in both the child and mother. Co-parenting was experienced by 9 out of the 20 participants as a state where caregiving and family planning was seen as a shared responsibility. In contrast, the 11 mothers who described themselves as the sole caregiver, with little to no aid or input from the other parent, described feelings of resentment toward the other parent.

In addition to co-parenting, mothers described how spending time together and engaging in shared hobbies greatly contributed to family functioning and well-being. When family members had a shared interest in sports, they, or parents/siblings, emphasized the EP child’s ongoing involvement in sports and physical activity. In contrast, for families who did not have shared interests or family activities, the child was responsible for initiating their physical activities and children with little interest in doing so remained sedentary. In addition to sports, shared family values such as being socially engaged with their neighborhood or religious community seemed to result in children who were more socially engaged. In contrast, families who were isolated or had little contact with extended family or members of their community
seemed to have a reduced social network, often resulting in the mother turning to more formal resources.

Finally, the family’s economic resources greatly affected the child’s, and mother’s, overall well-being. Economic resources were described as material resources or support from their job and/or extended family. Mothers who described themselves as economically stable expressed how relieved they were at being able to take time off without fear of losing their job, particularly during the first few months of birth when their child was often medically frail. In addition, extended family members often provided the family unit with financial, or material, support, as exemplified by this quote:

Because, you know, not only are you dealing with those emotional, emotional stresses. (Inhales breath) but you’re dealing with the financial side of it, too. You know, um, I didn’t work, my husband was the only one working. So that was a challenge, um, just figuring out everything. You know, we sold everything down to the fact that we didn’t sell our house, that was the only thing we had left. Um, we kept our house. And my mom loaned me a vehicle. So, my vehicle we could concentrate only on the kids [twins].

Highlighting how essential extended family support was for families with more limited financial resources.

II. Contributors & Barriers at the Community level

The participant’s community was divided into formal and informal sources of support and, on occasion, barriers. Formal sources of support that mothers described as contributing to the child’s, and family’s, wellbeing included the healthcare and school systems and religious institutions. Informal sources of support included friends and the family’s social and/or physical community.
A. Formal Sources of Support

Conflicting accounts were given in terms of the contribution the healthcare, and school system, had to the child’s well-being. While both were seen as vital in their child’s physical and psycho-social development, there were many barriers experienced by caregivers in accessing services provided by these institutions. In terms of the healthcare system, while the NICU experience was mainly described as positive, mothers’ descriptions of healthcare provided at the community level were more varied. Of the four participants who received care from rural healthcare providers, three expressed frustration with the care they received, and one specifically identified adverse outcomes for their child because of providers’ limited knowledge of preterm infant care. She expressed her experience in the following quote:

Well, at first, there were issues with the healthcare team, because, um, I-we were going to my older daughter’s doctor’s office. And I had specifically asked them beforehand if they had any experience with preemies and stuff like that. . and they said yes. And, um, whenever it came down to it, and she ha-had, she had pneumonia. She-she’d gotten pneumonia one time, um, not-it was like, uh, three months after she came home. Um, I called their emergency line and they just kept telling me to. . keep switching off her, um, Motrin and Tylenol and stuff and I was telling her she wasn’t, um, breathing right and stuff, and then I just took her to the hospital. I just took her to the emergency room. And if I hadn’t, the-the doctors there told me that if I hadn’t had done that, she could have been dead within 24 hours. And so, the fact that the doctor’s office that I had started her with didn’t recognize that with her being a preemie and having respiratory issues and everything. I feel like they fell down on their duties.

A quote which highlights a harrowing experience that generally contrasted with mothers who continued to receive care in the tier one urban hospital in which their child was born. For these mothers, ongoing satisfaction with their child’s medical care seemed to be the norm.

Like healthcare systems, the school system was seen as a core institution in the child’s and family’s life. Types of support seemed to vary by school system and the mother’s ability to successfully advocate for her child’s needs. It is important to note that a mother’s ability to advocate within the school system did not seem to be solely reliant on her educational
background, with some highly educated participants still having difficulty advocating in their child’s school. As a result, some participants described their frustration with the school failing to recognize their child’s academic needs, despite a formal medical diagnosis. One participant, for example, highlighted how difficult it can be for caregivers to ensure their child is receiving the appropriate level of accommodations:

I try to tell them even with an IEP, I try to tell him, they not doing like they should be with him in school. So I have been going toe-to-toe with them teachers…so we had to do a conference call which I didn't like, cause I don't have to tell you, but you want to ask questions and. . that's when I start most of my concern and my anger towards them how they was doing him…

In addition to the school and the healthcare system, mothers emphasized the importance early intervention programs played in their child’s development—specifically, daycare, pre-Kindergarten programs and behavioral therapies, such as speech and occupational therapy. Based on their accounts, mothers believed that through these programs, their child was able to begin reaching milestones much earlier than they had anticipated or been told. In addition, daycare and pre-Kindergarten programs allowed their child to socialize and begin developing interpersonal skills—something which mothers emphasized is vital to EP youth.

B. Informal Sources of Support

In addition to formal sources of support, informal sources of support for mothers, and the family, included having friends or extended family that lived in proximity and who had children that could socialize with their EP children. Mothers emphasized how their family and friends provided a safe environment for their child to practice socializing without fear of being ostracized. In addition, the family’s social community often provided both material and immaterial resources to the child and family, such as childcare, someone to talk to, or even financial help. For families who had a very limited social support network, mothers often
expressed feeling overwhelmed and lonely. Thus, an extended social support network of family and friends was described as vital to the well-being of both the child and the caregiver.

In addition to the social environment, mothers described how the neighborhood in which the child lived played a crucial role in child and family well-being. While most participants described their physical neighborhood as safe, high crime activity often resulted in the mother being fearful and engaging in protective measures such as limiting physical activities outside of the home or the formation of peer relationship with kids in the neighborhood. While effective in keeping the child physically safe, it often resulted in the child becoming more isolated and sedentary.

Religious institutions provided both formal and informal sources of support for the caregiver, child, and family. While formal resources included religious and spiritual guidance and support, informal sources of support included providing the caregiver and child with a place to form friendships and a social support network.

**Discussion**

For this paper we explored the caregivers’ perspective about aspects of their social, community and family environment which they believed affected their child’s development and overall well-being. While prior studies have explored caregiver experiences during early childhood (Granero-Molina et al., 2019; Schuetz Haemmerli et al., 2020), very limited information currently exists that looks beyond this developmental period to adolescence. In addition, by giving mothers the opportunity to share their experiences, we were able to look at more in-depth conditions and dynamics that affect extremely preterm children’s overall well-being; something which has remained limited, with most studies focusing on general factors,
such as maternal education or gestational age (Bangma et al., 2018; Bangma et al., 2019; Benavente-Fernández et al., 2019).

Our findings indicated that the role of motherhood is essential to EP children’s development and well-being. However, it is important to note that we were only able to interview mothers, therefore, a full understanding of the role the father, or significant other, plays are not possible with the current data. Overall, mothers emphasized their role as the primary caregiver, and advocate, of their EP child. However, this heavy reliance often resulted in maternal exhaustion and exacerbation of pre-existing, or new, health conditions. These results align with findings that indicate the increased rate of stress mothers experience during their EP child’s early childhood (O'Donovan & Nixon, 2019; Schuetz Haemmerli et al., 2020). The adverse effect to physical and mental health, however, has not been as well-studied and was a significant finding in this study, indicating a vital need for clinicians and healthcare professionals to provide ongoing support to this vulnerable population.

In addition, while mothers emphasized their role as caregiver and advocate, the type of care or advocacy provided varied based on the child’s development and health. For children with no, or milder, developmental conditions, the need for normalcy was emphasized by mothers—with most behavioral interventions occurring early in childhood and decreasing, or disappearing, as the child’s health needs decreased. This occasionally resulted in EP children with learning or developmental differences having limited academic or behavioral resources to rely on later in childhood—a deficit which for some resulted in difficulties in school or job maintenance once they graduated high school. This aligns with literature indicating that compared to term-born adults, adults born preterm have been found to be less likely to be
employed or have higher educational qualifications, even after controlling for maternal socioeconomic status (Bilgin et al., 2018; Crump et al., 2019).

For EP youth, participation in sports or outdoor activities was expressed by participants as essential to the development of their child’s physical health, being seen as directly benefiting their child’s weight and immune system. It is important to note, however, that while occasionally self-motivated, many of these EP youth’s involvement in physical activities was initially motivated by the caregiver, or a family member, with many caregivers emphasizing the importance of a specific sport or outdoor activity. This aligns with literature in the general population indicating that participation in organized physical activity early in life is associated with higher rates of physical activity in adolescents (Spiegler et al., 2019). Interestingly, however, most of the EP youth seemed to be self-motivated to pursue their own hobbies, which allowed them a sense of self. These findings parallel literature among individuals with disabilities that emphasize how vital having a hobby can be in developing a sense of self and increasing self-esteem (Patterson, 2001; Specht et al., 2002).

At the community level, the healthcare and school system were seen as both a facilitator and barrier to well-being for EP individuals, with many participants finding it difficult to obtain services without extensive knowledge of both systems. These results coincide with literature indicating how complex it can be to access services within the school and healthcare system for caregivers (Boshoff et al., 2016; Magaña et al., 2013). Finally, a family’s social network and physical environment were seen as vital to individual and family well-being. While a supportive and extensive social network provided families with both emotional and material sources of support, participants with limited, or unsupportive, social networks felt isolated and often
overwhelmed by their child’s developmental or health needs—particularly during their child’s early childhood when their medical needs tended to be the highest.

Like social networks, the physical environment in which the family lived greatly affected individual and family well-being. While most families lived in safe neighborhoods, for those who did not, the threat often increased stress and made them take vigilant measures to ensure their child’s safety—which often ran counter to what they believed would be best for their child’s health—including limiting outdoor activity or limiting their child’s peer network.

**Research & Clinical Implication**

We were able to identify conditions and dynamics at the family and community level that have the potential to affect an extremely preterm child’s development and well-being. However, more research is needed to explore how these experiences vary based on racial/ethnic background. While finances were briefly mentioned, access to many of the helpful factors mentioned, such as early childcare and access to outdoor areas to be physically active, are greatly associated with a family’s financial resources. In addition, more research is essential in determining how a family’s racial/ethnic background affect their EP child’s development and health.

Healthcare providers caring for these families must realize that not only are EP youth affect by prematurity, but caregivers are also deeply impacted both emotionally and physically by the preterm experience. Therefore, it is essential that maternal and family care is also emphasized.
**Strengths & Limitations**

This qualitative descriptive study addressed the limited knowledge that currently exists about conditions and dynamics at the family and community level that affect EP children’s development and well-being. Limitations of this study include the inability to fully explore how gender, race and/or culture affect these experiences. Literature among the EP population has identified how race and/or ethnicity impacts the care and services the EP child receives both in the NICU and in follow-up care. For this study, we did not include enough participants from a black or Hispanic/Latino background to fully explore this phenomenon. In addition, all participants were mothers, a limitation which fails to fully describe the experience of fatherhood. Therefore, more research is needed in fully exploring how well-being at the family and community level is affected by gender, race and/or ethnicity.

**Conclusion**

Overall, mothers of extremely preterm children are vital in the extremely preterm child’s well-being. While there are individual level characteristics that contribute to well-being, a support structure at the family and community level is essential to the child and mother’s well-being. Specifically, families with limited material and/or immaterial resources rely on formal institutions, such as the school and healthcare system for resources. Therefore, it is vital that personnel in these institutions ensure that their services are easily accessible for this vulnerable population group.
<table>
<thead>
<tr>
<th>Questions</th>
<th>Focus</th>
</tr>
</thead>
<tbody>
<tr>
<td>1) Can you describe your child’s current health?</td>
<td>General Health</td>
</tr>
<tr>
<td>2) Can you describe their Emotional health?</td>
<td>Emotional Health</td>
</tr>
<tr>
<td>3) Can you tell me about your child’s current wellbeing/Quality of Life?</td>
<td>Well-Being</td>
</tr>
<tr>
<td>4) Thinking back on your child’s life, can you think of experiences he/she had that were especially important turning points in shaping their health?</td>
<td>Development of Health</td>
</tr>
<tr>
<td>5. What about turning points in shaping their wellbeing/quality of life?</td>
<td>Development of Well-Being</td>
</tr>
<tr>
<td>6) Looking back on your child’s life, what stands out as especially important in contributing to his/her health</td>
<td>Contributions to General Health</td>
</tr>
<tr>
<td>7) What stands out as especially important in contributing to his/her wellbeing/quality of life?</td>
<td>Contributions to Well-Being</td>
</tr>
</tbody>
</table>
Table 4.2: Description of Caregiver/Child Demographic Characteristics

<table>
<thead>
<tr>
<th>Caregiver</th>
<th>Child/Children’s Race/Ethnicity</th>
<th>Child/Children’s Diagnosis(es)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Mother</td>
<td>White</td>
<td>None</td>
</tr>
<tr>
<td>2. Mother</td>
<td>African American</td>
<td>None</td>
</tr>
<tr>
<td>3. Mother</td>
<td>African American</td>
<td>Asthma, Speech Delay, Learning Disability &amp; Intellectual Disability</td>
</tr>
<tr>
<td>4. Mother</td>
<td>African American</td>
<td>Asthma</td>
</tr>
<tr>
<td>5. Mother</td>
<td>White</td>
<td>Asthma, ADD/ADHD†, Autism/Pervasive Developmental Disorder, Developmental Delay, Speech Delay, Learning Disability &amp; Intellectual Disability</td>
</tr>
<tr>
<td>6. Mother</td>
<td>White</td>
<td>ADD/ADHD†, Autism/Pervasive Developmental Disorder, Tics, Learning Disability &amp; Intellectual Disability</td>
</tr>
<tr>
<td>7. Mother</td>
<td>White</td>
<td>Developmental Delay, Speech Delay &amp; Learning Disability</td>
</tr>
<tr>
<td>8. Mother</td>
<td>White</td>
<td>ADD/ADHD†, Developmental &amp; Speech Delay</td>
</tr>
<tr>
<td>9. Mother</td>
<td>Black</td>
<td>ADD/ADHD†, Cerebral Palsy &amp; Oppositional Defiant</td>
</tr>
<tr>
<td></td>
<td>Mother</td>
<td></td>
</tr>
<tr>
<td>---</td>
<td>--------------</td>
<td>----------</td>
</tr>
<tr>
<td>10.</td>
<td>White</td>
<td>Asthma, CLD‡/Respiratory Disorder, ADD/ADHD†, Developmental Delay, Speech Delay &amp; Learning Disability</td>
</tr>
<tr>
<td>11.</td>
<td>White</td>
<td>ADD/ADHD†, Developmental Delay, Speech Delay &amp; Learning Disability</td>
</tr>
<tr>
<td>12.</td>
<td>Black</td>
<td>Asthma</td>
</tr>
<tr>
<td>13.</td>
<td>Black</td>
<td>Speech Delay</td>
</tr>
<tr>
<td>14.</td>
<td>White</td>
<td>None</td>
</tr>
<tr>
<td>15.</td>
<td>White</td>
<td>Asthma, CLD‡/Respiratory Disorder, Cerebral Palsy, Developmental Delay, Speech Delay, Learning Disability &amp; Epilepsy/Seizures</td>
</tr>
<tr>
<td>16.</td>
<td>Black</td>
<td>Asthma, ADD/ADHD†, Developmental Delay &amp; Speech Delay</td>
</tr>
<tr>
<td>17.</td>
<td>Hispanic/Latino</td>
<td>None</td>
</tr>
<tr>
<td>18.</td>
<td>White</td>
<td>CLD‡/Respiratory Disorder, Developmental Delay, Speech Delay &amp; Intellectual Disability</td>
</tr>
<tr>
<td>19.</td>
<td>White</td>
<td>None</td>
</tr>
<tr>
<td>20.</td>
<td>White</td>
<td>Twin 1: Asthma, CLD‡/Respiratory Disorder &amp; Developmental Delay</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Twin 2: ADD/ADHD† &amp; Learning Disability</td>
</tr>
</tbody>
</table>

†ADD/ADHD: Attention Deficit Disorder/Attention Deficit Hyperactivity Disorder  ‡CLD: Chronic Lung Disorder
REFERENCES


Currently, up to 40% of Extremely preterm (EP) children will develop a neurodevelopmental disorder (Hirschberger et al., 2018; Joseph et al., 2016; Luu et al., 2017; Serenius, 2016; Synnes et al., 2018). Yet, despite the multiple risks associated with EP birth, there is still the possibility for positive health and well-being (Bangma et al., 2018; Bangma et al., 2019). How socio-environmental characteristics affect the trajectory toward positive outcomes, however, remains largely unexplored. Therefore, the purpose of this dissertation study was to identify and describe socio-environmental influences and their effect on EP children’s health and well-being. There were three aims for this dissertation. Aim 1 was to identify potentially protective, or risk, characteristics beyond the perinatal/early childhood period for EP children and their effect on overall positive child health and well-being (Chapter 2). Aim 2 was to describe how vulnerable racial/ethnic minority families experience and develop well-being when their child has a neurodevelopmental condition (Chapter 3) and Aim 3 was to explore socio-environmental characteristics that affect child health and well-being from the perspective, and experience, of caregivers of EP children (chapter 4). For this chapter, I will review chapters 2, 3 and 4, discuss the strengths and limitations of this dissertation, and provide a synthesis of research and clinical implications.
Chapter 2: “Individual and Familial Characteristics and Positive Child Health Among Adolescents Born Extremely Preterm”

The goal of this chapter was to identify modifiable individual and familial characteristics at age 10 potentially associated with positive child health outcomes at 15 years of age. Data on 694 extremely preterm children enrolled in the Extremely Low Gestational Age Newborn (ELGAN) Cohort Study were analyzed for association between potentially modifiable characteristics at the individual and familial level that occurred at age 10 and positive child health outcomes at 15 years of age. Positive child health for this study was defined as 1) an absence of disease and 2) positive cognitive, social, and emotional well-being. Currently, most of the research has focused on risk factors at the perinatal or early childhood period. By focusing on potentially protective characteristics that occur at age 10, there is the potential for developing interventions later in childhood that promote positive child health among EP youth.

Results for this study indicated that for individual characteristics, the absence of internalizing and/or externalizing symptoms was associated better global health t-scores. For familial factors, absence of parental separation and job loss was also associated with better global health t-scores. In terms of neurocognitive outcomes, absence of externalizing and/or communication deficits were associated with increased odds of having better neurocognitive outcomes.

These outcomes indicate how specific individual, and familial characteristics, later in childhood still have the potential to positively affect child health and well-being. An absence of parental separation, for example, was associated with higher global health t-scores; while we cannot prevent parents from becoming separated, this indicates the need for developing interventions focused on the family unit. These findings corroborate with current literature.
indicating the vital role the family unit has on child health (Boström & Nilsagård, 2016; Wang et al., 2015) In addition, specific individual characteristics—such as an absence of internalizing and/or externalizing symptoms influence overall general health; reinforcing prior research highlighting how vital ongoing psycho-behavioral interventions can be for the overall health of EP individuals (Mathewson et al., 2017; Soni et al., 2021).

Overall, this chapter was an innovative step in identifying and describing how characteristics of EP children and their families still have the potential to affect well-being for EP youth. Unfortunately, a limitation of this study is how well-being was defined—mainly being based on an absence of risk factors, versus factors that can promote well-being. In addition, we were unable to explore how primary caregivers, or the family unit, develop well-being in the face of multiple barriers, particularly for racial/ethnic minorities who face higher degree of societal inequities in the U.S. as compared to White Americans (Beck et al., 2020; Janevic et al., 2018; Magaña et al., 2013; Magaña et al., 2015)—deficits addressed in chapter 3 and 4 of this dissertation.

Chapter 3: “Family members’ experience of well-being as racial/ethnic minorities raising a child with a neurodevelopmental disorder: A Qualitative Meta-Synthesis”

Chapter 3 addressed aim 2 to explore how caregivers, and the family unit, from a racial/ethnic minority background in the U.S. 1) experienced well-being and 2) responded to challenges they faced while caring for a child diagnosed with a neurodevelopmental disorder. For this study a qualitative meta-synthesis was conducted based on a thematic synthesis methodology (Thomas & Harden, 2008). Our methodological procedures were: 1) systematic literature search, 2) quality appraisal, 3) extraction of relevant findings 4) analysis of findings and 5) synthesis of findings (Thomas & Harden, 2008).
While much research has focused on prevention of neurodevelopmental disorders (Lea et al., 2017; Mathewson et al., 2017), we wanted to focus on vulnerable families at high risk for adverse well-being. Therefore, this chapter focused on addressing Healthy People 2030’s objective to improve health and well-being for children with lifelong chronic conditions (Ochiai et al., 2021). For this study, we focused on racial/ethnic minority primary caregivers caring for children with one of three neurodevelopmental disorders: Autism Spectrum Disorder (ASD), Attention Deficit Hyperactivity Disorder (ADHD), and/or Intellectual Disability. We selected these three conditions because of their high prevalence among EP children (Hirschberger et al., 2018; Johnson & Marlow, 2017) and the shared core symptomology, and caregiver/family experience, associated with these conditions (American Psychiatric Association, 2013).

For this synthesis, three analytical themes were developed “moving toward well-being as a caregiver,” “family & culture: impact on well-being,” and “community and culture: impact on well-being.” These analytical themes were comprised of six descriptive themes that described participants’ experiences of well-being and facilitators, or barriers, to well-being. While participants emphasized that well-being was possible, there seemed to be numerous obstacles unique to racial/ethnic minorities in comparison to White caregivers/family members providing care for children with ASD and/or ADHD. Therefore, participants within this synthesis often emphasized adverse experiences versus states of well-being; with many focusing on increased rates of stress, and feelings of frustration and loneliness. Facilitators to well-being occurred at the caregiver, family, and community level. At the caregiver level, facilitators to well-being included 1) Adapting to the diagnosis, which often involved finding a sense of peace and hope for the future 2) Becoming a resilient advocate, and 3) Spirituality, namely their own spiritual/religious beliefs. In contrast, a barrier to well-being at the caregiver level was de-prioritization of
self—which involved caregivers giving up their own job/careers and de-prioritizing their own needs.

Unfortunately, at the family and community level, limited information was available on facilitators to well-being, with much of the focus being on barriers. Of the facilitators mentioned, religious institutions were seen as a facilitator, allowing youth and caregivers the opportunity for social interactions. However, this dissertation took a significant step in highlighting the need for further research focused on how racial/ethnic families and communities can positively contribute to caregiver/family well-being. Another contribution of this synthesis, which is addressed in Chapter 4, was in emphasizing the limited research that exists focused on the effect prematurity has on families raising a child with a neurodevelopmental condition (Luu et al., 2017; Synnes et al., 2018).

Chapter 4: “Mothers Experience of the Socioenvironmental Effects to Well-Being for Adolescents’ Born Extremely Preterm: A Qualitative Descriptive Study”

The aim of this qualitative descriptive study was to explore primary caregivers’ perceptions and experiences of socio-environmental, and familial, qualities that affected 1) their child’s development and well-being from early childhood to adolescence, and 2) their own well-being as the primary caregiver. Participants consisted of 20 mothers who identified as their child’s primary caregiver from birth to adolescence. The age of their children varied from 16-18 years of age at the time of data collection. Racial background as well as number, and type, of comorbidities of the EP child were varied. Of the twenty mothers, 5% of them identified as Hispanic/Latina, 35% as Black/African American, and 60% as White/Caucasian. Additionally, 10% of the EP children were solely diagnosed with a physical condition, 25% had no identified comorbidities, and 65% had a neurodevelopmental condition.
Overall, mothers emphasized the central role of motherhood—with all 20 mothers identifying themselves as the primary caregiver throughout their child’s life. Being a primary caregiver included being responsible for most of the ongoing caregiving and day-to-day household tasks. Without appropriate levels of support, this heavy reliance on the mother often resulted in feelings of exhaustion and exacerbation of pre-existing, or new, mental, or physical health conditions. In total there were two themes identified—“Familial Impact to Health & Well-being,” and “Contributors & Barriers at the Community level.”

The first theme, “Familial Impact to Health & Well-being,” consisted of three subthemes: a) The Central Role of Motherhood; b) The Child’s Contribution to Health & Well-being; and c) The Family Unit. Under the first subtheme, mothers described both their responsibilities as their child’s primary caregiver and facilitators or barriers to their/family unit’s well-being—such as finances, spiritual beliefs, and their view of their child’s strengths and abilities. For the second subtheme, mothers described facilitators or barriers to well-being that were affected by their child’s actions and traits. Facilitators included the child participating in sports/outdoor activities and having their own hobbies. Barriers identified included lacking age-appropriate interpersonal skills and having a limited social network. At the family unit, mothers emphasized the importance of co-parenting and how their relationship with the child’s biological parent could be both a facilitator or barrier to their own, and their child’s, well-being.

For the second theme, “Contributors & Barriers at the Community Level,” two subthemes emerged: a) Formal Sources of Support; and b) Informal Sources of Support. Formal sources of support included the healthcare and school system and religious institutions. Informal sources of support included friends and the family’s local and social community. Conflicting accounts were given in terms of the contribution of the healthcare and school system to the children, and family
unit’s, overall well-being. While both were seen as vital in the child’s physical and psycho-social development, there were many barriers experienced by caregivers in accessing services provided by these institutions. Informal sources of support included having friends or extended family that lived in proximity and who had children that could socialize with their EP children. Mothers emphasized how their family and friends provided a safe environment for their child to practice socializing without fear of being ostracized. In addition, the family’s social community often provided both material and immaterial resources to the child and family, such as childcare, someone to talk to, or even financial aid.

Well-Being & Its Facilitators

Well-being was identified throughout this dissertation as a complex, personal experience for primary caregivers of EP children with/without neurodevelopmental disorders; with what was emphasized as being essential to well-being varying by the neurodevelopmental health of the child. However, much like how well-being was conceptualized in chapter 2, well-being seemed to be an outcome indicator of resilience, defined as a dynamic process that allows an individual or family to cope with adversity, change and opportunity in a manner that resulted in optimum family members’ well-being and family function (Richardson, 2002; Walsh, 2003). To experience well-being, primary caregivers had to 1) adapt to the child’s condition—first, their extreme prematurity and/or chronic neurodevelopmental condition(s), 2) face and address multiple challenges—which varied from financial constraints to community stigma, and 3) learn to identify and utilize resources within themselves, their family, and their community.

In addition to conceptualizing and describing the experience of well-being, we identified facilitators or barriers to well-being at the individual, family, and community level. Facilitators to well-being at the individual-child level were consistent with other literature that emphasizes
the importance of positive emotional health and interpersonal skills (Botting et al., 2016; Poehlmann-Tynan et al., 2015). In chapter 2, the absence of internalizing and/or externalizing symptoms at age 10 was associated with higher odds of better global health in EP youth at age 15. In addition, both chapter 2 and chapter 4 emphasized the importance of interpersonal skills—children with no language impairments having higher odds of emotional health in chapter 2 and mothers in chapter 4 highlighting how the ability to communicate resulted in their child having improved peer relationships.

In addition to the individual-child level, there were multiple facilitators to well-being at the individual-caregiver level that were identified in chapter 3 and 4, with caregivers emphasizing the importance of adapting and being at peace with their child’s condition (EP and/or neurodevelopmental disorders) and the developmental ramifications of the condition. Spiritual and/or religious beliefs were often utilized by mothers/primary caregivers to reach this point. Interestingly, the importance of advocacy varied between mothers whose child was born EP with no neurodevelopmental disorder and those whose children had a neurodevelopmental disorder. In chapter 2, mothers/primary caregivers emphasized the importance of adopting the role of resilient advocate— which involved the ability of participants, mostly mothers, to persevere in accessing services and accommodations for their child and family despite multiple challenges, community stigma, and racial or ethnic discrimination. A process that required ongoing development. In contrast, mothers of EP youth in Chapter 4 varied in the emphasis they placed on advocacy skills. While some mothers emphasized the importance of becoming an advocate, others acknowledged the importance but did little to develop advocacy skills—instead emphasizing the importance of teaching their child to appear as normal as possible despite differences in internalized social/cognitive abilities; a phenomenon seen in the Autism
community termed *masking/camouflaging* (Cook et al., 2018). Like prior literature on this phenomenon, *masking/camouflaging* was seen as a solution by mothers but opened the possibility for problems for EP youth. Mothers of EP youth described that *masking* allowed their child to avoid being stigmatized and fit in with their peers. Problems, like those seen in autism literature (Cook et al., 2018; Halsall et al., 2021) of *masking/camouflaging*, however, included mothers describing how difficult it was for their child to keep up in their mainstream classes, particularly with limited academic resources.

Facilitators at the family level were discussed both in chapter 3 and 4. While chapter 3 found mixed opinions among racial/ethnic minority caregivers, chapter 4 emphasized the importance of co-parenting and having shared interests. Finally, facilitators at the community level conveyed the importance of both formal and informal sources of support that provided acceptance and a safe environment for their child and their family.

**Strengths & Limitations**

Strengths of this dissertation included the diverse demographic background of the sample utilized in chapter 2 and 4. This diversity allowed us to incorporate the perspective of racial/ethnic minorities and individuals from varied racial and medical backgrounds. This was essential since we were looking at how socio-environmental traits could influence well-being; this allowed us to incorporate a variety of socio-environmental traits. In addition, we were able to focus on facilitators to well-being, comprehensively comprised of physical, emotional, and social welfare. This is an innovative perspective and addresses the current limitations in research, which has largely focused on socio-environmental risk factors during the perinatal and early childhood period. In addition, this dissertation allowed for the analysis of socio-environmental characteristics beyond the early childhood period that have the potential for positive well-being.
This is vital, particularly for high-risk population groups such as racial/ethnic minorities, considering the delay in diagnosis and early intervention that currently exists among individuals with neurodevelopmental disorders. In addition, in chapter 2 we focused on racial/ethnic minority caregivers, a vulnerable population with higher rates of prematurity, a higher chance of delayed diagnosis and more difficulty accessing services ((Beck et al., 2020; Magaña et al., 2016).

Limitations of this dissertation include the fact that most of the participants within chapter 2, 3 and 4 were mothers acting as their child’s primary caregiver. This makes it difficult to generalize findings to fathers or other types of primary caregivers. Unfortunately, input from EP youth was absent; something which makes it difficult to develop a full understanding of the facilitators or barriers to well-being for EP children. In addition, further analysis of data is necessary to understand how facilitators to well-being identified in chapter 4 impact well-being, with chapter 2 focusing mainly on general individual and socio-environmental factors. Finally, we were unable to identify the unique experiences among racial/ethnic participants in chapter 4 that were identified in chapter 2—perhaps indicating more focus needed to be placed on socio-environmental factors, such as race, within the interview guide.

**Research & Clinical Implications**

Currently, there is limited research focused on socio-environmental factors that have the potential for improved well-being for EP children at high risk of a neurodevelopmental disorder. Without the ability to fully understand characteristics that impact well-being, it is difficult to identify resources and behavioral modifications primary caregivers, and the family unit, can make to improve EP development and well-being. While most research has focused on early childhood factors, current research indicates that being a racial/ethnic minority carriers a risk of
receiving a delayed diagnosis of up to two to three years delay; a delay which puts racial/ethnic minority children at a disadvantage and makes it more difficult for them to access specialized behavioral interventions until later in childhood (Magaña et al., 2013; Magaña et al., 2016; Magaña et al., 2015). While we cannot immediately change the socio-environmental risks, we are able to identify characteristics throughout childhood that have the potential to promote well-being. Therefore, more research is vital in identifying characteristics at the family and community level that have the potential for improved development and well-being for EP children.

Like other research findings, this dissertation highlighted the fact that healthcare institutions often are not seen as a facilitator to well-being—particularly for already vulnerable racial/ethnic minorities. Nurses and healthcare practitioners, in general, need to emphasize the role primary caregivers play in promoting child well-being. They also should give primary caregivers the tools and resources to be able to develop into resilient advocates in the face of numerous barriers. Due to the time constraints placed on nurses and healthcare providers, sufficient time is not always available to assist families in their role as advocates. These time constraints must be considered when allocating time and resources to extremely preterm, and/or racial/ethnic minorities at high risk of a neurodevelopmental disorders.

Conclusion

In conclusion, this dissertation was able to describe and explore how well-being was experienced by extremely preterm, and racial/ethnic minority, children at high risk of a neurodevelopmental disorder. While well-being is a unique, personal experience, resilience in the face of multiple barriers is a necessity to develop well-being. Facilitators to well-being that
were identified were adaptation to an EP/neurodevelopmental diagnosis, becoming a resilient advocate and primary caregivers’ reliance on their spiritual/religious beliefs.
REFERENCES


APPENDIX A

1. PubMed


2. PsychInfo

(DE (intellectual disability OR intellectual disabilities OR mental retardation) OR (attention deficit hyperactivity disorder or attention deficit disorder) OR (autism spectrum disorders or autism or autistic)) AND (DE "Resilience (Psychological)" OR DE "Well Being" OR DE "Happiness" OR adaptation OR adapt OR resilience OR resiliences OR "quality of life") AND SU (qualitative research OR qualitative OR themes OR phenomenology OR "grounded theory" OR interview)

3. Scopus

(TITLE-ABS-KEY ("autism spectrum disorder" OR autism OR autistic OR "intellectual disability" OR "intellectual disabilities" OR "mental retardation" OR "attention deficit hyperactivity disorder" OR "attention deficit disorder") AND TITLE-ABS-KEY (resilience OR resilient OR resiliency OR "well-being" OR adaptation OR adapt OR "quality of life" OR happiness) AND TITLE-ABS-KEY ("qualitative study" OR "qualitative research" OR interview OR qualitative OR themes OR phenomenology OR "grounded theory"))

4. Sociological Abstracts

(MAINSUBJECT.EXACT.EXPLODE("Autism") OR MAINSUBJECT.EXACT.EXPLODE("Attention Deficit Disorder") OR TI,AB(autism OR autistic OR “attention deficit” OR “intellectual disability” OR “intellectual disabilities” OR “mental retardation”)) AND (MAINSUBJECT.EXACT.EXPLODE("Well Being") OR MAINSUBJECT.EXACT.EXPLODE("Resilience") OR MAINSUBJECT.EXACT.EXPLODE("Happiness") OR MAINSUBJECT.EXACT("Adjustment") OR TI,AB (well being OR well-being OR resilience OR resiliences OR resilient OR adaptation OR adapt OR happiness)) AND (MAINSUBJECT.EXACT.EXPLODE("Qualitative Methods") OR TI,AB(qualitative OR themes OR phenomenology OR "grounded theory" OR interview))