2015-11

Factors Associated with High Risk Infant Follow-Up Attendance

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UNIVERSITY OF SAN DIEGO

Hahn School of Nursing and Health Science

DOCTOR OF PHILOSOPHY IN NURSING

FACTORS ASSOCIATED WITH HIGH RISK INFANT FOLLOW-UP ATTENDANCE

By

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A dissertation presented to the

FACULTY OF THE HAHN SCHOOL OF NURSING AND HEALTH SCIENCE

UNIVERSITY OF SAN DIEGO

In partial fulfillment of the

requirements for the degree

DOCTOR OF PHILOSOPHY IN NURSING

November, 2015

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Abstract

Prematurity is a significant public health problem and preterm infants face well described risks of adverse neurodevelopmental outcomes. Bronfenbrenner's bioecological theory of development describes interactions between biological and environmental factors and explains how these interactions can impact development. Systematic follow-up of preterm, high-risk infants is recommended for early identification of problems and provision of interventions and support services. Most research on follow-up attendance has involved small, single sites. A retrospective analysis of population based data available in the California Children's Services High Risk Infant Follow-up Quality of Care Initiative (HRIF-QCI) data system was performed to examine factors associated with attendance at the second recommended visit.

Applying the bioecological theory of development to the high-risk infant population reveals the numerous biologic, family, social, cultural, and political factors that influence development. This theory supports the provision of early intervention services to this population.

Only 74% of those infants seen for the first visit attended the second recommended visit. Infants with birth weights equal to or less than 750 grams were almost twice as likely to attend the visit compared with those with birth weights greater than 1,250 grams. Private insurance, two parents as caregivers, completion of the first visit during the recommended interval and enrollment in early intervention during the first visit were all associated with higher attendance rates. Public insurance, a single parent as caregiver, or maternal race of Black or Asian were all associated with decreased attendance. Infants with maternal race of Black were 45% less likely to attend the second visit, and the factor with the strongest association with lack of visit two attendance.

Rural residence was associated with decreased HRIF attendance (they were 30% less likely to attend) and there were marked differences between the rural and non-rural population, with rural caregivers being younger, less educated, and with lower rates of employment. There were marked differences in attendance rates between different HRIF programs, with risk-adjusted rates ranging from 34.4% to 89.9%.

These findings offer new knowledge into factors associated with HRIF clinic attendance and suggest future research opportunities to improve clinical practice with this population.

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Dedication

This work is dedicated to the infants and families served by the California Children's Services High Risk Infant Follow-up Program. Their resilience, strength, and love teach and inspire me daily.

Acknowledgements

I owe many thanks to the members of my committee. Dr. Mary Barger, chair, offered support, expertise, and her careful reading helped me complete this work. Dr. Barger's sense of humor and skill as an educator made the process fun. Dr. Lois Howland has provided support and encouragement and shared her knowledge. I greatly appreciate her willingness to guide me through analysis and application of a theory. Dr. Susan Hintz is an inspiration. As a clinician and researcher she is a tireless in her efforts to improve care provided to premature infants. I appreciate her assisting me to obtain access to the data used for this project and her support and encouragement along the way.

This research could not have been done without the efforts of Dr. Mihoko Bennett, statistician with the CPQCC/HRIF-QCI project. She provided data tables and figures that were clear and easy to read and became the basis of the two data papers. I owe a debt of gratitude to my fellow HRIF coordinators, and all of the staff working on the HRIF-QCI project.

The faculty and staff of the Hahn School of Nursing and Health Science are incredible. They are always willing to go the extra mile to help students. My classmates, especially the "PHDN 2015" cohort, are wonderful. We have laughed, cried, and celebrated together.

Last, but not least, I owe a debt of gratitude to my husband Rus for his help and love throughout this process. The support and encouragement of Rus, our children, my parents, and friends has helped me achieve my goals.

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Chapter 1

Introduction

Overview

Prematurity is a significant public health issue in the United States. In 2012, 11.5 % of all births in the U.S. were preterm (<37 weeks) with 1.9% (76,041 babies) delivered prior to 32 weeks gestational age. Almost ten percent of these infants (7,231 or 1.4% of live births) were born in the state of California (National Center for Health Statistics, 2013). Adverse neurodevelopmental sequelae of preterm birth include cerebral palsy, mental retardation, learning disabilities, coordination disorders and sensory deficits (Behrman & Butler, 2007). The adverse impact of prematurity persists as children enter kindergarten, with former premature infants having lower scores on IQ tests and achievement tests compared with term controls. Outcomes are worse for those of low socioeconomic status (Taylor et al., 2011; Potharst et al., 2010). It is believed that systematic early follow-up of these high-risk infants will result in earlier identification of physical and developmental problems and earlier referral for needed services, decreasing long term morbidity.

Background and Significance

Theoretical framework

The bioecological model of development proposed by Bronfenbrenner is the framework guiding this study. This theoretical model has been suggested as basis for epidemiological research on child development (Avan & Kirkwood, 2010). The high-risk infant population faces well-documented risks for adverse developmental outcome associated with prematurity and recommended high-risk infant follow-up (HRIF) visits are an attempt to ameliorate this. In the bioecological framework, the individual child brings biologic factors (prematurity in the case of the HRIF population) that interact with the environment to influence development. Bronfenbrenner (1994) places the interactions between the developing human and the immediate environment as the central influence on development. While acknowledging the importance of genetic potential on intelligence, Bronfenbrenner (2005) noted that the interaction between heredity and environment is very important, with an appropriate environment being necessary to allow full expression of an inherited trait. Shonkoff and Marshall (2000) wrote, “the maturation of the central nervous system itself is affected by the experiences that characterize each individual’s personal environment” (p. 50).

Microsystems, consisting of interactions and relations closest to the child, are the central context impacting the child; however they are impacted by concentric systems that include education, resources and culture (Bronfenbrenner, 1979). The systems influencing the child are impacted by time, both individual life events and societal and historical events (Bronfenbrenner, 1999).

Early intervention, offered to former premature infants to improve their developmental outcomes, can influence the development at the level of the microsystem, while the policies that provide early intervention and support for families interact at the mesosystem and macrosystem. Societal influences impact the child indirectly, yet may be significant factors influencing development and must be considered in planning intervention for a child (Sameroff & Fiese, 2000).

Premature Infants are High-Risk

Premature infants are at risk for impairment and measurement of neurodevelopmental outcome is needed to determine success after care in the neonatal intensive care unit (NICU) (Stephens & Vohr, 2009). This is most often measured using a composite of the incidence of cognitive delay, cerebral palsy, and/or vision or hearing impairment called neurodevelopmental impairment (NDI). Risk factors associated with NDI include medical risks such as the following: decreasing gestational age, bronchopulmonary dysplasia or chronic lung disease, intracranial abnormalities including intraventricular hemorrhages grades 3 and 4 and periventricular leukomalacia, infection and male sex (Stephens & Vohr, 2009; Mercier, et al. 2010). There are higher rates of NDI found with decreasing gestational age (Kyser, Morriss, Bell, Klein, & Dagle, 2012) and this has not improved over time (Hintz et al. 2011).

Socioeconomic factors, including poverty and low levels of education in parents, are associated with adverse outcome in the preterm population (Mercier, et al. 2010; Taylor, et al. 2011; Potharst, et al. 2010). Mercier et al. (2010) found an association between caregiver education of high school or less and severe disability at 18 to 24 months corrected age in a multi-site Vermont Oxford Network (VON) study of 3,567 infants.

Early Intervention

Early intervention has been found to have a positive impact on the outcome of preterm infants (Spittle et al., 2010; Spittle, Orton, Anderson, Boyd, & Doyle, 2012; Gianni et al., 2006). Early intervention is theorized to enhance child development by supporting the central relationship of family and child (Guralnick, 2011). This is at the level of the microsystem described by Bronfenbrenner (1979). This evidence supports Bronfenbrenner's (1994) bioecological theory of development, placing the interactions between the growing child and the immediate environment as central to influencing development.

High-Risk Infant Follow-up

Due to the known risks for adverse outcome and positive impact of early intervention, both the American Academy of Pediatrics (Committee on Fetus and Newborn, 2008) and an expert panel (Wang et al., 2006) recommended that preterm infants be enrolled in a specialized clinic to receive comprehensive neurodevelopmental assessments at regularly scheduled intervals. There are follow-up programs associated with NICUs in 91% of teaching hospitals responding to a national survey (Kuppala, Tabangin, Haberman, Steichen, & Yolton, 2012).

In California, these recommendations have become the basis for an organized system of neonatal follow up which includes insurance coverage for eligible infants. California Children's Services (CCS) mandates that all CCS licensed NICUs in California have an organized system to provide HRIF) services to eligible infants. The aim of this statewide program is early diagnosis of conditions requiring ongoing services, providing three visits at specified intervals during the first three years of life. High-Risk Infant Follow-up programs perform assessments, provide comprehensive case management and health education, and make referrals for needed services and early intervention. Referral to HRIF programs and outcome data from visits is reported in a mandatory web-based quality improvement data system, the High-Risk Infant Follow-up Quality of Care Initiative (HRIF-QCI) (Department of Healthcare Services [DHCS], 2013). Data collected includes medical and social risk factors known to be associated with neurodevelopmental outcomes. The HRIF-QCI system offers the opportunity for quality improvement activities for NICUs and HRIF programs, and allows CCS to follow site-specific activities.

Attendance at HRIF Programs

Compliance rates with recommended neurodevelopmental follow-up are low, with 59% complying with one visit at a single center in New York, (Nehra, Pici, Visitainer, & Kase, 2009) and 65% complying with three visits in a Canadian study (Ballantyne, Stevens, Guttmann, Willan, & Rosenbaum, 2012). In an anonymous online survey of U.S. academic institutions with NICU follow up programs, Kuppala et al. (2012) found that 71% of the 128 responding had follow up rates $\leq$ 60% with 47% having follow up rates $\leq$40%. The steepest drop-off in compliance was found to occur between the time of discharge from the NICU and the first recommended visit, with the second highest rate of drop-off in compliance occurring between the first and second clinic visit (Ballantyne et al. 2012). No show rates are high (20%) and create programmatic difficulties (Brockli, Andrews, Pellerite, & Meadow, 2014).

Factors associated with noncompliance include lower socioeconomic status, residing a greater distance from the clinic, and maternal substance use (Harmon, Conaway, Sinkin, & Blackman, 2013; Ballantyne et al., 2012). Those infants and children facing the highest risks due to the combination of prematurity and low socioeconomic status are the least likely to attend clinic visits for assessment and referral for early intervention services. These studies are limited by small sample sizes. Ballantyne et al. (2012) followed 357 infants from three NICUs with two follow-up programs for one year. Harmon et al. (2013) studied a single site with a population of 133 infants, 91 of whom were seen for at least one visit. The study by Ballantyne et al. (2012) was performed in a Canadian population and may not be generalizable to the United States.

Outcome of noncompliant high-risk infants

Evidence is mixed regarding the outcome of high-risk infants who do not comply with neonatal follow-up. Most authors find that those who are noncompliant or followed with difficulty are more likely to have disability (Tin, Fritz, Wariyar, & Hey, 1998; Callanan, et al. 2001; Hille, Elbertse, Gravenhorst, Brant, & Verloove-Vanhorick, 2005). In this context, the need to improve compliance with HRIF visits gains importance, since those who are not seen may have more deficits than those who comply. In contrast, Guillén et al. (2012) and Castro et al. (2004) found those lost to follow-up are less likely to have neurodevelopmental impairment. In a structured telephone survey of 23 noncompliant families, 78% of those interviewed noted their child was doing well at the time of the appointment, so they did not comply with the visit (Harmon et al., 2013). If an infant is not seen, outcome, an important measure of NICU care, is not known.

Rural Residence

California has a large rural landmass with 44 of 58 counties classified as rural by the California State Office of Rural Health (CA Department of Healthcare Services, 2012). These counties have higher rates of poverty and somewhat higher birth rates than urban counties (CA Department of Healthcare Services, Primary and Rural Health Division, 2012). In a voluntary system of regionalized care, many preterm infants in California are cared for in tertiary medical centers, and all of the CCS licensed regional

NICUs and HRIF clinics are located in urban areas (Nowakowsk et al., 2010; DHCS, 2013). After their NICU stay, infants discharged to residences in rural areas often need to travel great distances to attend clinic, decreasing the likelihood that they will come for their appointments. Residence at a greater distance from the clinic is associated with poor compliance for visits (Harmon et al., 2013; Ballantyne et al., 2012). They may receive substandard and fragmented healthcare due to inadequacies in the healthcare system in rural areas (Farmer, Clark, Sherman, Marien, & Selva, 2005). Researchers in Oregon found a higher proportion of children with special health care needs (CSHCN) in rural areas (DeVoe, Krois, & Stenger 2009). High-risk infants are CSHCN. It is not known if residence in a rural region is associated with follow-up appointment attendance.

The second HRIF visit

California Children's Services recommends that the second HRIF visit take place between 12-16 months adjusted age (DHCS, 2013). This is an extremely important period in the development of the child, who is moving out of infancy, becoming a toddler, and developing skills in multiple domains including rapid cognitive changes, independent mobility and ambulation, early speech and language, perfection of a neat pincer grasp and use of objects as tools (Gesell, 1968; Illingworth, 1987; Piaget, 1977). Poor compliance with this visit removes an opportunity to identify children with developmental delays, abnormal neuromuscular findings suspicious of evolving cerebral palsy, poor growth, or other issues that could respond to early intervention. Factors associated with compliance with the second HRIF visit are unknown. Ballantyne et al. (2012) found that the second largest drop-off in compliance with an NICU follow-up clinic in a Canadian population occurred between the first and second recommended visit, however that visit was scheduled much earlier, at six to eight months rather than the 12-16 months recommended by CCS.

Statement of the Problem

Despite known risks for adverse outcomes in preterm infant, attendance at recommended follow-up is low. High-risk infant follow-up programs perform assessments and make referrals and recommendations for services such as early intervention. The theoretical basis for the effectiveness of early intervention is strong. Quantitative research supports improved outcomes for participants in early intervention. At approximately 12 months adjusted age, infants undergo significant developmental changes and it is important to identify difficulties in progress in order to offer timely interventions. Most studies of follow-up are small, single site studies, and there is a paucity of recent literature. Although identifying distance as a factor in clinic attendance, no research has explored the association of rural residence with attendance. There is a gap in the literature and there are no large, population-based studies to better understand factors associated with attendance at the time of the second recommended visit.

Purpose of the Study

The purpose of this study is to determine factors associated with adherence to recommended HRIF clinic visits in California. Experts recommend that high-risk infants be followed systematically in order to identify risks and neurodevelopmental deficits and make appropriate referrals for services to support their growth and development. The HRIF program in California provides comprehensive case management, performs assessments, and makes recommendations and referrals for services. The bioecological theory of development proposed by Urie Bronfenbrenner provides a theoretical basis to support the importance of early intervention and the supports offered by the HRIF program. Compliance with recommended follow-up is low, and this limits the ability of HRIF programs to provide comprehensive services. There is very little information regarding compliance rates for high-risk infants residing in rural geographic areas. In a large geographically diverse state, it is important to determine if differences exist between urban and rural regions in order to identify areas for quality improvement.

Specific Aims of the Three Papers

This dissertation will consist of three papers, each formatted according to the guidelines of potential publications. The specific aims of each paper are as follows:

1) Analyze Bronfenbrenner's bioecological theory of development, applying it to interventions to support the high-risk infant population.

2) Identify factors associated with compliance (attendance) at the second recommended High-Risk Infant Follow-up visit in a population of infants born in California in 2010 and 2011.

3) Examine the pattern of follow-up for infants residing in rural counties compared with those residing in urban counties to determine if differences exist in the same population of infants.

A secondary aim for this analysis was to examine program specific factors and impact on clinic attendance.

Summary

Premature infants are at risk for adverse neurodevelopmental outcomes. The purpose of the HRIF program is to help mitigate risk by early identification of problems and facilitating referrals for services. The concept of development and early intervention for this population should be explored using a theoretical framework that addresses the multiple factors influencing development. Future studies will be informed by knowledge gained from the population based studies as well as the proposed theory analysis.

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Chapter 2:

Bronfenbrenner's Bioecological Theory of Development: Application to the High-Risk Infant Population

Abstract

The bioecological theory of development proposed by Urie Bronfenbrenner can be applied to preterm infants (high-risk infants). Describing interactions between an infant, caregivers, and the environment, this theory explains factors that may lead to adverse neurodevelopmental outcomes in this population. This theory supports provision of early intervention and family supports to promote optimal development as well as encouraging a larger view of societal factors influencing development. The bioecological theory provides context for future research.

Keywords: Child Development, Theory, Premature Infant

Introduction

Premature infants face risks of adverse outcomes and are known as high-risk infants. In addition to risks of medical morbidities, these infants are at high risk for adverse neurodevelopmental sequelae including cerebral palsy, mental retardation, learning disabilities, coordination disorders, and sensory deficits (Behrman & Butler, 2007). Developmental outcome is influenced by multiple factors, both biological and environmental (Hintz, et al 2011; Taylor et al., 2011; Potharst et al., 2010). Child development is described in nursing literature as stages of change and growth over time from birth through adolescence (Child development, 2012). These changes include cognitive, motor, language, and social/emotional components.

A theoretical model is needed to guide research and inform clinical practice (Peterson, 2013). The bioecological theory of development proposed by Bronfenbrenner has been suggested for use in research on child development (Avan & Kirkwood, 2010). The bioecological model has not been applied to the high-risk infant population, where it can be useful in identifying potentially modifiable factors impacting child development.

Components of the Theory

The Developing Human

The infant is central in this theory of development. Specific genetic, psychological, and physiological characteristics of the infant influence development (Bronfenbrenner, 1999; Bronfenbrenner, 2001/2005). These individual characteristics are then influenced by interactions in multiple settings. Bronfenbrenner (1975/2005) argues that it is impossible to examine genetic factors in isolation. Genetic factors are felt to be important but the full expression of genetic potential requires an appropriate environment. Shonkoff and Marshall (2000) posit that neurological maturation is impacted by experiences. Neurologic maturation is part of the biological foundation of development. Magnusson (1995) proposes a continuous feedback loop involving behavior, environment, cognitive function, and physiological processes.

Environment and systems

The bioecological theory of development describes reciprocal interactions between a developing infant and parents, care givers, and others, in an environment of nested, concentric systems (Bronfenbrenner, 1979) as shown in Figure 1. All of these systems are influenced by the passage of time and by historical events such as wars, economic crises, or environmental disasters (Adapted from Bronfenbrenner, 1979, pp16-42). The descriptions below and the figure are not intended to imply stasis. There are continuous bidirectional interactions. The infant is influenced by and influences the parent and other care givers (Bronfenbrenner, 2001/2005). The systems closest to the infant, the microsystem and mesosystem, are dependent on resources from the systems farther away (the exosystem and macrosystem) in order to support the infant's development (Bronfenbrenner, 1999).

Microsystem

The relationships and activities between the infant and others in a single setting make up the microsystem. Examples include direct interactions with parents/guardians at home or interactions between infant and primary nurse in the hospital. The interactions change both infant and adult. The impact of touch on infants is a prime example of this

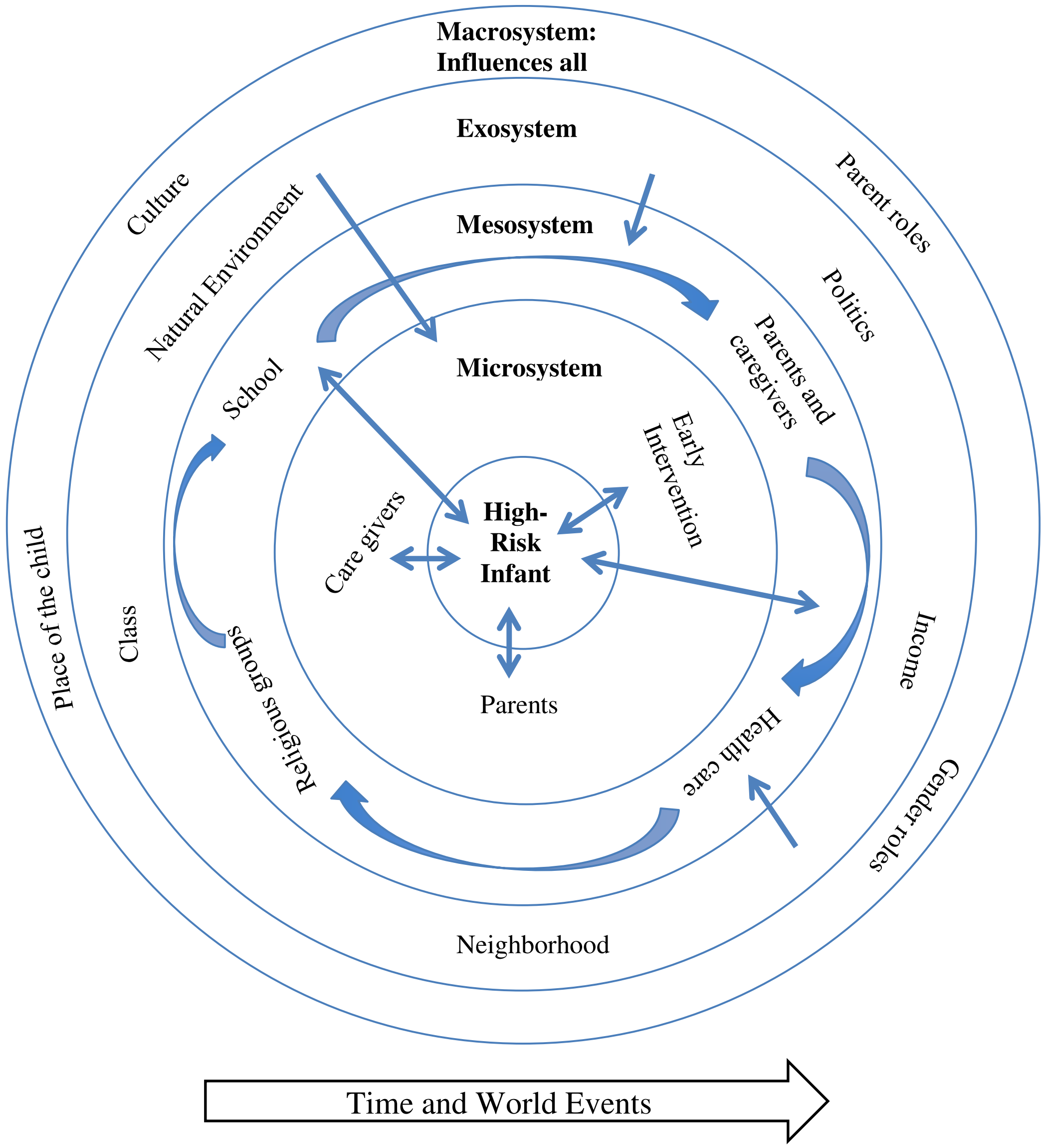

Figure 1. Bioecological Theory Model

reciprocity, skin-to-skin contact promotes growth and physiologic regulation in premature infants. The parents providing the care also experience psychological and physiological changes including increases in oxytocin levels (Champagne, 2014).

Mesosystem

Moving out from the infant, and surrounding the microsystem, is the mesosystem. This refers to interactions in and between two or more microsystems involving the infant. Examples include the interactions between parent, infant and teacher at home and at school; or infant, parent, and nurse in both home and health care settings.

Exosystem

The exosystem surrounds the mesosystem and influences the more central systems without direct interactions with the infant. Changes in parental employment or income, world events such as economic downturns, or governmental policies are examples of factors that influence the infant without direct interaction with the infant.

Macrosystem

Culture, a consistent pattern of behaviors or beliefs across all systems, is referred to as the macrosystem. Child rearing practices and culturally defined parental roles are examples of how culture influences all systems and interactions. As an example, Bronfenbrenner (1970/2005) found that families in the former Soviet Union used group childcare more than families in the U.S. yet spent more time with their children and were more physically affectionate than families in the U.S.

Time

The systems surrounding the infant are all influenced by the passage of time and by world events such as wars, economic down turns, and natural disasters.

Activities and Interactions

In the microsystem, the infant interacts with others and demonstrates ongoing meaningful behaviors. The infant does not develop passively, but must participate in activities (Bronfenbrenner, 1999). All participants in the microsystem carry out behaviors; for example, interactive social games like peek-a-boo. As the infant grows and develops, these activities become more complex and the infant is able to participate in more than one activity at a time. Reading with an infant is an example of this; the adult reads and shows pictures in a book, the infant listens and watches, then is able to point to pictures in a book, then to name pictures and add information to the story. This increase in complexity comes through attention to activities of others and through interactions with those with whom the infant has an emotional attachment (Bronfenbrenner, 1979). Development occurs through a process of reciprocal interactions with persons and the environment surrounding the infant (Magnusson, 1995).

Application to the High Risk Infant Population

The High-Risk Infant

The premature infant has numerous physiologic factors that can adversely influence developmental outcome. Lower gestational age is associated with increased risk of neurodevelopmental impairment, a composite measure of presence of cognitive impairment, cerebral palsy, or vision or hearing deficits (Hintz, et al. 2011; Kyser, Morriss, Bell, Klein & Dagle, 2012). The human brain undergoes multiple changes during gestation. Peak neuronal migration occurs between three to five months of gestation with organization occurring starting at the fifth month of gestation. The peak time of gyral development is during the last three months of gestation (Volpe, 2008). Infants delivered at less than 32 weeks gestation have immature brains. These infants are at risk for intraventricular hemorrhages (IVHs) that arise in the germinal matrix, the source of neuronal and glial cells in the premature brain (Bolisetty et al., 2014). These IVHs, particularly when larger (grades three or four), are associated with cerebral palsy and poor cognitive outcome (Bolisetty, et al., 2014). White matter and cerebellar injury are also associated with neurodevelopmental impairment (Hintz, et al., 2015).

Multiple biologic systems are affected by preterm birth. Preterm infants are at risk for infections that can cause inflammatory responses associated with higher rates of central nervous system injury, bronchopulmonary dysplasia, and poor neurodevelopmental outcome (Adams-Chapman, 2012). Chronic lung disease, defined as receiving supplemental oxygen for 28 days with radiographic evidence of lung changes or requiring oxygen supplementation at 36 weeks gestation, is associated with poor developmental outcome (Mercier, et al. 2010). Surgical repair of a patent ductus arteriosus (Madan, Kendrick, Hagadorn, Frantz, & the National Institute of Child Health, 2009) is also associated with adverse neurodevelopmental outcome. The care provided in the neonatal intensive care unit (NICU), for example oxygen supplementation, is needed to sustain life, may have adverse sequelae for the preterm infant (Behrman & Butler, 2007).

The epigenetics of the stress response has been suggested as an additional cause of adverse neurodevelopmental outcomes. Samra, McGrath, Wehbe, & Clapper (2012) performed a review of the literature including both animal and human models, and found that maternal separation and the adverse, stressful stimuli in the NICU may be related to the known adverse outcomes facing preterm infants.

Microsystem: The family and caregivers

Ongoing interactions within the microsystem are key to promoting development. These include daily caregiving tasks (comforting or feeding) and activities such as reading to or playing with the infant. The biological factors of both parents and infants influence these interactions. An infant with chronic lung disease may have less stamina for interactions; a mother with medical problems may be physically unable to perform caregiving tasks.

There are also psychological factors that can challenge these interactions. Higher rates of maternal depression have been found in mothers of premature infants. A study of mothers attending a high-risk infant follow-up program found over 20% of mothers with symptoms of depression compared with rates of 10-15% in mothers of term infants without health problems (Northrup, Evans, & Stotts, 2013). Maternal depression, particularly if prolonged, has been associated with adverse infant outcomes. Koutra and colleagues (2013) found an association between antenatal and/or postpartum depression and lower cognitive scores in infants tested at age 18 months.

Parental stress is increased in parents of infants in the NICU (Melynk, et al., 2006). There was increased maternal stress at one year for mothers of preterm infants compared with mothers of term infants in a study one study (Gray, Edwards, O'Callgahan, Cuskelly, & Gibbons, 2013). In this study, dysfunctional mother-child interactions were highly associated with maternal stress. Using a standardized instrument, the Parenting Stress Index-Short Form, mothers of premature infants were more likely to report that their child did not meet their expectations when compared with mothers of term infants. This perception may influence how a mother responds to her infant, for example, a highly stressed mother may perceive an infant's initial response to tasting solid food as dislike and make no further attempts to advance the diet, while a mother experiencing less stress may recognize that the infant needs multiple exposures to learn to eat solids and make continued efforts. Fathers have been found to have an increased risk of posttraumatic stress disorder after the infant has been discharged from the NICU

(Shaw et al., 2009). It is not known how this may impact interactions and development in the high-risk infant population.

Mesosystem: Interactions in multiple environments

The mesosystem encompasses the interactions between multiple microsystems involving the infant. These environments are diverse and include home, day care, health care, school, and religious institutions (Bronfenbrenner, 1979). High-risk infants have medical risks and may have frequent interactions with the health care system due to respiratory illnesses or neurologic problems associated with premature birth (Behrman & Butler, 2007). Almost half of children with special health care needs lack a medical home (Singh, Strickland, Ghandour, & van Dyck, 2009). Access to consistent health care providers may improve developmental and medical outcome.

Exosystem

External forces impact interactions in the microsystem and mesosystem. Socioeconomic factors, including poverty and low levels of education in parents, are associated with adverse outcomes in the preterm population (Mercier, et al. 2010; Taylor, et al. 2011; Potharst, et al. 2010). Chaos, a lack of structure in family interactions and routines, is associated with poor developmental outcomes in children and adolescents (Fiese & Winter, 2010). Chaos is often associated with poverty; there are crowded living conditions, parents with irregular work schedules, and there may also be parental substance use.

Sociodemographic risk factors were associated with a less stimulating home environment and poorer cognitive outcome in a population of preterm infants in the Netherlands (Weisglas-Kuperus, Baerts, Smrkovsky, & Sauer, 1993). In this study, a stimulating home environment was found to compensate for biological risks between ages one and three, with improvement in cognitive development measured by standardized tests.

Studies in the U.S. have similar findings. Inadequate income to meet family needs was associated with lower Intelligence Quotient (IQ) scores in a longitudinal study of low birth weight children (Brooks-Gunn, 1995). Those who were poor most of their first four years had lower IQ scores than those who were poor only some of that time (Duncan, Brooks-Gunn, & Klebanov, 1994). Development of language is impacted by poverty in the high-risk infant population. Low socioeconomic status was associated with substantially lower scores on a test of expressive and receptive language in a population of preterm infants matched for biologic risk factors (Wild, Betancourt, Brodsky, & Hurt, 2013).

Macrosystem: Culture and Context

Consistent system structure, patterns of behavior, and beliefs make up the macrosystem. These cultural norms impact all systems including parenting practices and may influence developmental achievement. Mexican American mothers demonstrated fewer instances of praising and encouraging their child during developmental teaching tasks compared with non-Hispanic White mothers (Fuller, Bein, Kim, & Rabe-Hesketh, 2015). In that population, Mexican American children had lags in cognitive development but very good social-emotional development compared with a matched population of non-Hispanic White children. Infants who were <1,500 grams at birth had poorer cognitive outcomes than those who were normal birth weight in both populations.

Parental roles are assigned by culture; mothers (women) are assigned the role of primary care giver of infants and young children across cultures and societies. The role of the father has been found to vary greatly between different cultures (Maccoby, 1987/1990/1995). These differences may influence interactions but the significance of these differences on outcome of the high-risk infant is not known.

Research on adherence with recommended high-risk infant follow-up clinic visits has found much lower rates of follow-up in the U.S. population compared with other countries (Ballantyne, Stevens, Guttmann, Willan, & Rosenbaum 2012). This may reflect the impact of culture on care of these infants.

Application to Supports for High-Risk Infants

The bioecological theory provides a framework to describe points of potential intervention to support the developmental outcome of high-risk infants. The more central systems, interacting directly with the infant, are important areas for direct efforts to support necessary nurturing environments (National Research Council (NRC)/Institute of Medicine (IOM), 2000). Socioeconomic factors (exosystem) and culture (macrosystem) have significant impact on the infant and caregivers. These systems are extremely important, yet identifying modifiable factors and implementing changes in these areas is very challenging.

Microsystem: Early intervention

Prior to NICU discharge, family-centered developmental care may help decrease the stress response and its adverse consequences. Interventions such as infant massage and decreasing maternal separation have been proposed as techniques to support brain development (Samra, et al., 2012).

Post discharge, early intervention is recommended to support the development of the high-risk infant and is theorized to enhance child development by supporting the central relationship of family and child (Guralnick, 2012. A systematic review in the *Cochrane Library* found that early intervention is beneficial, improving cognitive outcome in infancy and at preschool age (Spittle, Orton, Anderson, Boyd, & Doyle, 2012). Early intervention has been associated with improved mobility at 44 months in a Dutch population (Verkerk et al., 2011) and improved language and cognitive scores at age 8 in a U.S. population (Hill, Brooks-Gunn, & Waldfogel 2003).

Microsystem: Parent supports

Parents of high-risk infants have increased risks of mental health problems that impact their relationship with their infant. Melnyk and colleagues (2006) developed an educational and behavioral intervention for parents of infants hospitalized in the NICU; creating opportunities for parent involvement (COPE). The authors found less depression and anxiety two months post discharge in mothers who received the COPE intervention.

A pilot study developed a standardized intervention to reduce symptoms of posttraumatic stress disorder associated with parenting an infant in the NICU (Shaw et al., 2013). This intervention was found to be helpful to mothers.

Mesosystem: Services in multiple settings

High-risk infants receive services in multiple locations including home, school, and varied health care settings. The services received are diverse and include home health nursing, infant education, social work support, and therapies (physical, occupational, and speech). Bronfenbrenner (1979) theorizes that an infant or child will have a more successful transition to new settings if supported by someone who has already interacted with the child in a familiar setting, strengthening the links between the settings. The Infant Health and Development Project (Hill et al., 2003) offered a combination of home visits and center-based services, and this program was successful in improving cognitive outcome in a preterm population.

Exosystem

As described previously, the high-risk infant is adversely impacted by socio-economic factors, their biologic risks compounded by cumulative environmental factors (NRC/IOM, 2000). This NRC/IOM report, *From Neurons to Neighborhoods,* highlights the scientific evidence of the importance of the early environment and experiences on child development. There are successful community based projects that have worked to meet the needs of poor families using approaches that provide supports directly to the family while strengthening their neighborhoods of residence (Austin, Lemon, & Leer, 2005). The Harlem Children's Zone is an example of such a project, providing services staring during pregnancy and including education, health, as well as environmental changes (Komro, Flay, Biglan, & the Promise Neighborhoods Research Consortium, 2011). These projects do not specifically target the high-risk infant population, though infants residing in participating communities will benefit from the services provided.

Macrosystem: Culture and context

Interventions designed to enhance the developing child may have differential impact based on the culture of the child. A Canadian study of an impoverished population found that the duration of participation in a preschool educational program was statistically significant in improving receptive language skills in the poor Aboriginal children enrolled, but not the skills of Canadian or immigrant children (Benzies et al., 2011). This study, informed by Bronfenbrenner's bioecological theory, included parental participation as well as educational services to the children. There was no documentation of the gestational age or birth weight of the participants.

Discussion

The bioecological model provides a useful framework that can guide clinical practice by prioritizing activities that will support the developing infant. Concrete examples include referrals for early intervention services, assisting families in establishing care at a medical home, and offering support to parents with depression or anxiety. All are interventions that will impact the infant and caregivers directly.

This model can also guide future research efforts. Much research on the developmental outcome of high-risk infants has focused on the impact of medical interventions performed during the neonatal course, for example, treatment of patent ductus arteriosus (Schmidt et al., 2001; Chorne, Leonard, Piecuch, & Clyman, 2006) or differing ventilation strategies (Vaucher et al., 2012). The bioecological model can identify non-medical interventions that may improve developmental outcomes in high-risk infants. Previous research has utilized the bioecological theory to inform studies of African American adolescents (Riina, Martin, Gardner, & Brooks-Gunn, 2013) and of the impact of school reform (Guhn, 2009). There is limited literature on use of the bioecological theory as the framework for studies on developmental outcomes of high-risk infants.

The Infant Health and Development Program (IHDP) is an example of a longitudinal, multi-site trial of interventions to improve the development of premature infants after discharge from the NICU. The IHDP focused on context, environment, and interactions to improve development of high-risk infants and Brooks-Gunn (1995) describes the trial as being influenced by the work of Urie Bronfenbrenner. The interventions provided were home visits, intensive center based early childhood education (transportation was provided), and parent meetings. Those participants receiving the most intervention (more attendance at the child development center) had higher intelligence quotient scores at age eight (Hill et al., 2003). This research influenced development at both the microsystem and mesosystem.

The bioecological theory can guide future research. The mean birth weight of infants enrolled in the IHDP was 1,798 grams. Replication of the IHDP trial in an extremely low birth weight (<1,000 gram) population would be a way to explore the impact of these interventions on infants with greater biologic risk. Nurse home visits to pregnant and parenting women, support the mother-child microsystem and have sustained effects on outcome of mothers and children (Olds, et al. 2007; Olds, et al. 2010). Research on the impact of this intervention on outcomes of the high-risk infant population is another opportunity for using the bioecological theory as a framework to guide research. Future studies on interventions to address the mental health issues of mothers and fathers of infants in the NICU should include longitudinal follow-up and developmental outcome measures to measure impact on the high-risk infant.

There are gaps in the literature investigating the effects of the macrosystem on neurodevelopmental outcomes. The impact of culture on the development of high-risk infants is one potential area for study. In 2010, 16.3% of the U.S. population and 37.6 % of the California population was Hispanic (U.S. Census Bureau, 2011). There is an achievement gap between Mexican American children and non-Hispanic White children

(Fuller et al., 2010). This persists into childhood, but high quality early childhood education has been found to narrow this gap (Cabrera, Shannon, Rodriguez, & Lubar, 2009). A study of extremely preterm (<28 weeks gestation) infants found no difference in cognitive outcomes between Hispanic, Black, and non-Hispanic White children after adjusting for medical and socio-economic factors (Duncan et al., 2012). Research has shown that Latino parents, particularly low-income and immigrant parents are viewed as less involved in their children's education due to multiple factors and are less likely to volunteer in the classroom or participate in parent teacher association meetings (Grau, Azmitia, & Quattlebaum, 2009). Poverty is a key factor influencing their lack of involvement. Provision of supplemental education experiences such as reading and other educational activities in a Latino Head Start population was related to family resources including income and parent levels of education (McWayne & Melzi, 2014). Parental involvement is culturally influenced and the concept of *respeto*, a value emphasizing deference and respect to adults, influences proximal processes between parent and child and parent and educational institution (Calzada, Fernandez, & Cortes, 2010). Research is needed to tease out impact of culture compared with socio-economic factors.

In addition to poverty, social policy and political realities are in the exosystem, impacting the lives of high-risk infants and their families. There is a need for research on the impact of social policy on outcome. Social policy had a major impact on services provided to and inclusion of disabled children in preschool programs (Odom et al., 2004). Research is needed to determine the effects of budget cuts and decreased services on the outcome of high-risk infants. Shonkoff (2003) recommends multidisciplinary efforts and increased use of scientific knowledge to inform public policy to address disparities

Development in early childhood has been found to have a profound lifelong impact on individuals and societies (Irwin, L., Siddiqi, A., & Hertzman, C. 2007). Brooks-Gunn (1995) described hearing Bronfenbrenner express hopes that developmental theory would be used to help change the lives of children. There are multiple opportunities for researchers to utilize the bioecological framework to develop and test non-medical interventions aimed at improving the outcome and lives of a very vulnerable group of children.

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Chapter 3:

Factors Associated with Attendance at the Second Recommended High Risk Infant Follow-up Appointment

Abstract

Objectives: Due to risks of adverse outcomes, systematic follow-up is recommended for premature infants, yet attendance rates are low. The purpose of this study was to identify factors associated with attendance at the second recommended High Risk Infant Follow-up (HRIF) visit.

Methods: Population based cohort study, retrospective analysis of prospectively collected data on preterm infants, birth weights <1500 grams born in 2010 and 2011 and enrolled in the California Children's Services HRIF Quality of Care Collaborative data system. Identification of significant factors and backwards stepwise logistic regression to build a full model.

Results: There were 3,494 seen and 1,207 not seen. Factors associated with attendance included birth weight of < 750 g (OR 1.98, 95% CI [1.56,2.52]) and attendance at the first HRIF clinic visit during the recommended interval (OR 1.97, 95% CI [1.62, 2.41). Sociodemographic factors associated with non-attendance included: maternal race of Black (OR 0.55, 95% CI [0.43-0.71]) or Asian (OR 0.77, 95% CI [0.6-0.99]); single parent household (OR 0.77, 95% CI [0.65-0.91]); and public insurance (OR 0.77, 95% CI [0.65, 0.91]). There were significant programmatic differences with risk adjusted follow-up rates ranging from 34.5% to over 100%.

Conclusions for Practice: The smallest infants with the greatest biologic risks were more likely to be seen, however infants at risk due to sociodemographic factors were less likely to be seen. There were striking differences between HRIF programs. This presents an opportunity for collaboration to develop best practices.

Keywords: Follow-up Studies, Premature infants. Ambulatory Care.

Introduction

Prematurity is associated with well described risks of adverse neurodevelopmental outcomes including developmental delays or disability and cerebral palsy.[1,2] These risks increase with decreasing gestational age[3], presence of high grade intraventricular hemorrhages (grades III or IV)[4], and with requiring major surgery during the neonatal period.[5] Sociodemographic factors including poverty[6,7] and maternal education level[8] are also associated with adverse outcomes. American Academy of Pediatrics[9] and expert panel[10] recommendations are that these high-risk infants are enrolled in a specialized program and receive ongoing assessments to facilitate early identification of problems and make appropriate referrals. Despite recommendations, attendance rates at neonatal intensive care unit (NICU) follow-up clinics are low. A survey of U.S. academic institutions with NICU follow-up programs found that 71% of the 128 responding had follow-up rates $\leq$ 60% with 47% having less than 40% follow-up rates.[11]

In California, there is an organized system of neonatal follow-up that includes insurance coverage for eligible infants. California Children's Services (CCS) mandates that all CCS licensed NICUs in California have an organized system to provide high-risk infant follow-up (HRIF) services to eligible infants and documenting referral and visit status in a web-based reporting system.[12] Even with this program in place, only 80% of eligible very low birth weight infants were referred for HRIF services at the time of NICU discharge in a cohort of infants born 2010 and 2011.[13] The aim of the HRIF program is early diagnosis of conditions requiring ongoing services, providing three visits at specified intervals (4-8 months; 12-16 months; and 18-36 months). The second CCS HRIF recommended visit (V2), is to occur between 12-16 months adjusted age,[12] an important time in the development of the child.[14,15,16]

Multiple medical, socioeconomic, and demographic factors are associated with attendance at HRIF clinic visits. Poverty, public insurance, black race, low levels of maternal education, single parent households, distance to clinic, and substance use have all been associated with decreased attendance.[17,18,19] Smaller, more preterm infants and those with a history of chronic lung disease are more likely to attend clinic while larger, more mature infants are less likely to attend HRIF clinic visits.[17,18] Multiple gestation is also associated with decreased attendance.[17]

Many studies of HRIF clinic attendance use a dichotomous measure of attendance; yes or no. Prior studies of HRIF attendance have assessed compliance with at least one visit, finding that 68% attended at least one visit in a single site in Virginia[17] and 59% attending one visit in suburban New York.[18] There is far less information regarding patterns of attendance with a series of recommended visits. In a Canadian study, the steepest drop-off in clinic attendance was found to occur between the time of discharge from the NICU and the first recommended visit, with the second highest rate of drop-off occurring between the first and second clinic visits.[19] That second visit was to occur at 6 to 8 months, much earlier than the timing of the second visit recommended by CCS. There was no differentiation of factors associated with attendance at the different recommended visits.

The aim of this study is to identify factors associated with attendance at the second recommended HRIF visit.

Methods

This is a retrospective analysis of prospectively collected data from the High Risk Infant Follow-up-Quality of Care Initiative (HRIF-QCI) and the California Perinatal Quality of Care Collaborative (CPQCC) data systems. California Children's Services mandates that participating institutions enter referral to HRIF programs and outcome data from visits in a mandatory web-based quality improvement data system, HRIF-QCI.[12] The HRIF-QCI system offers the opportunity for quality improvement activities for NICUs and HRIF programs, and allows CCS to follow site specific activities. It is linked to the CPQCC perinatal data system, a quality improvement database with data for over 90% of the neonates cared for in California NICUs.[20] The CPQCC data system contains demographic, perinatal, and neonatal factors, collected during the NICU stay. At the time of NICU discharge, the infant is referred to an HRIF program in the HRIF-QCI system and baseline data is entered including socio-demographic factors and medical eligibility. The NICU CPQCC identifying number is entered, linking the infant to the perinatal data available in the CPQCC system. At each HRIF visit, additional sociodemographic data is entered into the system as well as results of the assessment and any medical specialty and support services received (including early intervention). If an infant is not seen, data is entered capturing reasons for not being seen. Each NICU is classified based on the level of care provided and the HRIF programs are classified based on their associated NICU. NICUs that do not have their own HRIF program refer infants to another institution at the time of NICU discharge.

This population-based cohort study includes very low birth weight (<1,500 grams) infants born in 2010 and 2011 and entered into the HRIF-QCI data system at the time of NICU discharge and seen for the first HRIF visit. Exclusion criteria are those seen for the first visit but then discharged from the program at that time (not expected to return), those who died, or those residing in a pediatric sub-acute facility.

We obtained data from the both the HRIF-QCI and CPQCC databases. Variables included sociodemographic characteristics, medical risks, and hospital and HRIF program factors. Data obtained from the CPQCC system included classification of hospital as a teaching institution (yes/no), maternal factors: race, age, prenatal care, and some infant factors: congenital anomalies, surgery during NICU stay or requiring high frequency ventilation. Remaining variables were found in the HRIF-QCI data system.

Maternal and family sociodemographic factors included maternal age, race, prenatal care (yes/no), education level (the highest level reported for the caregivers), employment, living arrangement, primary language, distance to the HRIF clinic, and residence in rural areas.

Infant factors evaluated included sociodemographics: gender, race, insurance type, primary caregiver, and multiple gestation, whether the infant was out born, and transfers between NICUs. Medical risk factors included birth weight, gestational age, small for gestational age (SGA) status, congenital anomalies, chronic lung disease (CLD), surgery during the NICU stay, persistent apnea requiring medication at discharge, high frequency ventilation, or persistent pulmonary hypertension requiring inhaled nitric oxide. Neurologic factors included periventricular leukomalacia, intraventricular hemorrhage (IVH) grades 3 or 4, seizures, cerebral thrombosis, and presence of developmental central nervous system abnormality.

We evaluated characteristics at the time of the first visit including results of neurologic assessment, the timing of the visit (in relation to the CCS recommended schedule of 4-8 months), an active child protective services case, enrollment in early intervention, having a primary care provider (PCP) at time of visit, and whether the PCP served as a medical home as defined by the American Academy of Pediatrics.[21]

Neonatal intensive care unit and HRIF program CCS level (regional, intermediate, or community), geographic location (urban vs. rural), and HRIF program volume were all considered, as was teaching hospital status.

Residence and clinic zip codes were used to calculate distance to the HRIF program and broken down into quartiles. Four outliers were removed (all zip codes more than 500 miles from the HRIF program). None of the excluded cases had residence zip codes in California.

We performed bivariate analysis on variables listed above. Initial bivariate analyses using chi-square for categorical variables and t-test for continuous ones identified statistically significant relationships with V2 attendance. Collinearity was evaluated by examining tolerance statistics between variables. With the identified significant factors, we developed three models using backward stepwise logic regression using a cutoff of $p<.15$ for model inclusion. The first model used only significant sociodemographic variables, the second model added infant risk factors, and finally hospital characteristics were added for the full model. We used a mixed effects model in order to better account for the interactions among multiple variables of interest. Analyses were conducted using SAS version 9.4 (SAS Institute, Cary, NC)

This study was approved by the Institutional Review Boards of the University of San Diego and the University of California San Diego.

Results

There were 8,049 eligible infants discharged from participating NICUs, with only 6,442 (80%) referred to the HRIF program. Only 4,863 (75%) of these infants were seen for the first visit. 4,701 met eligibility criteria for this analysis of visit 2 (V2) attendance. As is shown in Figure 1, there were 3,494 seen and 1,207 not seen for the second visit. Only 54% of infants referred to HRIF were successfully seen for V2.

Figure 1. Visit 2 Eligible Population and Attendance

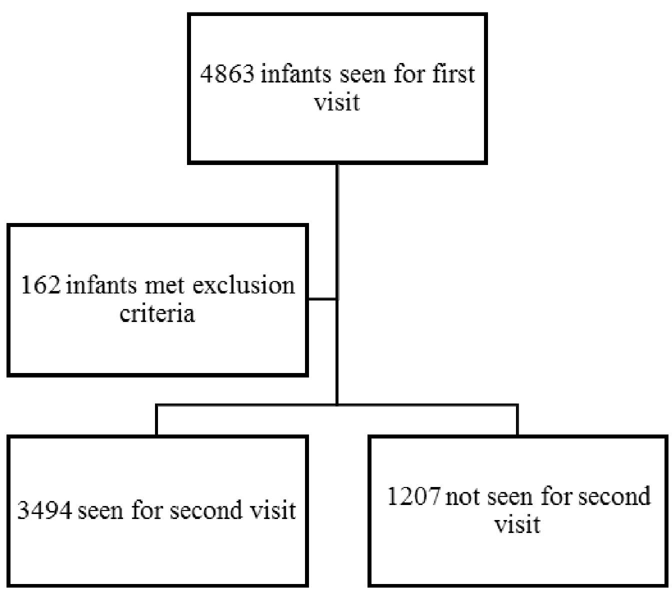


Table 1 shows important demographic characteristics of those seen and those not seen. Bivariate analysis factors associated with decreased second visit attendance included maternal race (black), Medi-Cal insurance, single mother as primary care giver, educational level of less than 9th grade, and residence in a predominantly rural county. There was no statistically significant association between V2 attendance and having an identified primary care provider (PCP) who provided a medical home ($p = 0.84$).

Table 1

Demographics for Those Seen and Not Seen for the Second Recommended High Risk Infant Follow-up Visit

Demographic Factor	**Level**	**Lost to Follow-up N=1207***	**(%)**	**Seen for Visit 2 N=3494***	**(%)**	**p-value if <0.05**
Maternal Age	≤19	115	(9.5)	257	(7.4)	0.0471
	20-29	490	(40.6)	1403	(40.2)	
	30-39	509	(42.2)	1586	(45.4)	
	≥40	92	(7.6)	245	(7)	
Maternal Race	Non-Hispanic Black	194	(16.1)	363	(10.4)	<0.0001
	Hispanic	510	(42.4)	1583	(45.5)	
	Asian	147	(12.2)	417	(12)	
	American Indian/Other	41	(3.4)	102	(2.9)	
	Non-Hispanic White	312	(25.9)	1014	(29.1)	
Caregiver Education	<9th Grade	39	(5.4)	136	(5.8)	0.001
	Some High School	113	(15.5)	308	(13)	
	High School Grad/GED	202	(27.8)	533	(22.6)	
	Some College/Other	179	(24.6)	549	(24.5)	
	College or Graduate Degree	194	(26.7)	807	(34.2)	
Infant Insurance	Medi-Cal**	665	(56.5)	1772	(50.8)	0.0008
	Healthy Families**	10	(0.8)	52	(1.5)	
	Private	485	(41.2)	1566	(45)	
	CCS Coverage***	685	(58.1)	1953	(56)	
Home Language	English	920	(78.9)	2487	(81.8)	0.0002
	Spanish	183	(15.7)	718	(21.3)	
	Other	63	(5.4)	173	(5.1)	
Primary Caregiver	Mother	522	(44)	1204	(35)	<.0.0001
	Father	6	(0.5)	22	(0.6)	
	Both Parents	628	(53)	2124	(61.8)	
	Foster/Adoptive or Other	30	(2.5)	89	(2.6)	
Sex	Male	598	(49.6)	1839	(52.6)	.
Birth Weight (g)	≤500	9	(0.7)	40	(1.1)	<0.0001
	501-750	124	(10.3)	576	(16.5)	
	751-1000	276	(22.9)	859	(24.6)	
	1001-1250	363	(30.1)	957	(27.4)	
	1251-1499	435	(36)	1062	(30.4)	
Gestational Age <32 weeks	Yes	1036	(85.8)	3095	(88.6)	0.0101
Multiple Gestation	Yes	337	(27.9)	970	(27.8)	
Small for Gestational Age	Yes	358	(29.7)	932	(26.7)	0.045
Chronic Lung Disease	Yes	279	(23.3)	975	(28.1)	0.0011
IVH Grade III or IV	Yes	57	(4.9)	201	(5.9)	
Surgery	Yes	144	(11.9)	584	(16.7)	<0.0001
Visit 1 between 4-8 months	Yes	885	(73.3)	3004	(86)	<0.0001
Residence in a Rural County	Yes	398	(33.2)	910	(26.1)	<0.0001
Primary care provider is medical home	Yes	611	(54.9)	1822	(55.2)	

*due to missing data sums of factors may be less

**Public: Medi-Cal (Medicaid) and Healthy Families (S-CHIP)

***CCS coverage available to those with both public and private insurance

There was a significant amount of missing sociodemographic data. Highest educational level was only available for 727 (60%) of the 1,207 not seen for V2 and for 2,362 (67%) of those seen for the second visit. Employment was listed as unknown for 371 (31%) of those not seen and for 855 (25%) of those seen for V2.

In this population, multiple gestation was not associated with decreased attendance, with essentially equal percentages of those not seen compared with seen being singletons (72.1% not seen vs. 72.2% seen), twins (23.1% vs. 23.1%) and 4.8 % of those not seen being triplets or higher order multiples compared with 4.6% of those seen (data not shown).

Outcome of neurologic assessment at the first visit was not associated with second visit attendance with 70.6% of those lost and 70% of those seen having normal neurologic assessments. Of those lost, 26.1% had abnormalities on their neurologic assessment at the first visit, 27.3% of those seen had abnormal assessments.

Those seen for the second visit were more likely have been more premature and smaller (birth weight less than 1,000 grams). Those in the lowest birth weight groups were the most likely to attend with 81.6% of those less than 500 grams and 82.3% of those weighing 501-750 grams attending their second visits. Almost 84% of those born ≤ 24 weeks attended the second visit, while only 68% of those born ≥33 weeks attended. Those attending were more likely to have had neonatal morbidities such as CLD, high grade IVH, neonatal surgery, or requirement for high frequency ventilation.

Residence distance was broken down into quartiles as shown in Table 2. The highest quartile contained 1,167 infants residing between 20.8 and 480.2 miles from the HRIF program. The vast majority of these (99%) residing less than 200 miles and 90%

residing less than 100 miles from the HRIF program (data not shown). Approximately equal percentages of those lost (27.4%) and those seen (25.4%) lived more than 20 miles away from the HRIF program. On bivariate analysis, distance from the HRIF program was significantly associated with decreased attendance for both the second and fourth quartiles.

Table 2
Distance from High Risk Infant Follow-up Program

Quartile	**#Infants**	**Miles**
Lowest	1179	0-4.5
2nd	1160	4.6-9.4
3rd	1166	9.5-20.7
Highest	1167	20.8-480.2
Missing Zip	29	--
Overall	4701	0-480.2

In a mixed model of sociodemographic and infant risk factors, residence distance was associated with decreased odds of attendance for those in the second (23%) or fourth (25%) distance quartiles while residence in the third quartile was not statistically significantly associated with decreased attendance.

In the full model that included sociodemographic factors (see Table 3), infant risk factors, and HRIF program characteristics, distance from HRIF program was no longer retained, probably due to the addition of the rural residence location variable. This variable showed a 21% decreased odds for completing V2, similar magnitude of the quartile distance variable. The other variables from model 2 were retained with only small changes in the magnitude of their effects.

One of the strongest predictors of being lost to follow-up for the second visit was maternal race with Asian, blacks, or other (non-white) having a 23% to 45% decreased odds of attendance. Residence in a rural location was also associated with decreased attendance (OR 0.73 95% CI [0.58,0.91]). Households with a single parent (OR 0.769 95% CI [0.652, 0.907]) and having Medi-Cal as insurance (OR 0.77 95% CI [0.65, 0.91]) were also negatively associated with attendance at the second HRIF visit. Enrollment in early intervention at the time of the first visit was associated with improved attendance (OR 1.34 95% CI [1.12,1.61]). There was a significant amount of missing data related to caregiver level of education and employment, therefore these factors were not included in the final model.

Controlling for other factors, those with birth weight of less than 750 grams were almost twice as likely to return for the second visit (OR 1.98; 95% CI [1.56,2.52]). Other indicators of the severity of neonatal illness such as chronic lung disease, IVH grades 3 or 4, surgery during the neonatal period, requiring high frequency ventilation, were not significantly associated with HRIF visit attendance. Gestational age, highly correlated with birth weight, was not in the final model.

Table 3

Mixed Method Regression Results for Attendance at the Second High Risk Infant Follow-up Visit by Sociodemographic and Infant Factors and Hospital Characteristics for Three Different Models

Risk Factors	**Level**	**Model 1 OR (95% CI)**	**Model 2 OR (95% CI)**	**Model 3 OR (95% CI)**
Sociodemographic Factors				
Maternal age	40+	1.09 (0.8-1.55)	1.10 (0.77-1.59)	
	30-39	1.36 (1.04-1.78)	1.37 (1.04-1.80)	
	20-29	1.34 (1.03-1.74)	1.33 (1.02-1.73)	
	19 or younger	Referent	Referent	
Maternal Race	NH Black	0.58 (0.45-0.74)	0.60 (0.47-0.76)	0.55 (0.43-0.71)
	Hispanic	1.08 (0.90-1.31)	1.1 (0.91-1.33)	1.09 (0.91-1.32)
	Asian	0.73 (0.57-0.93)	0.75 (0.58-0.97)	0.77 (0.6-0.99)
	Am-Indian	1.25 (0.33-4.76)	1.6 (0.41-6.26)	1.5 (0.39-5.84)
	Other	0.68 (0.44-1.05)	0.68 (0.44-1.05)	0.69 (0.45-1.07)
	NH white	Referent	Referent	Referent
Medi-Cal Insurance		0.78 (0.65-0.92)	0.77 (0.65-0.92)	0.77 (0.65-0.92)
Caregiver	Other	1.01 (0.64-1.6)	0.95 (0.6-1.51)	0.95 (0.6-1.51)
	One parent	0.79 (0.67-0.93)	0.79 (0.66-0.93)	0.77 (0.65-0.91)
	Both parents	Referent	Referent	Referent
Distance quartile	Highest	0.75 (0.60-0.93)	0.74 (0.59-0.92)	
	3rd quartile	0.83 (0.67-1.03)	0.83 (0.67-1.03)	
	2nd quartile	0.77 (0.63-0.95)	0.77 (0.62-0.95)	
	1st quartile	Referent	Referent	
Rural Residence				0.73 (0.58-0.91)
Infant risk factors				
Birth Weight	750 or less		2.04 (1.61-2.60)	1.98 (1.56-2.52)
	751-1000		1.31 (1.08-1.59)	1.32 (1.09-1.60)
	1000-1250		1.11 (0.93-1.33)	1.11 (0.93-1.32)
	1251+		Referent	Referent
Sex	Male		1.16 (1.01-1.34)	
Early Start Enrollment at 1st HRIF Visit			1.31 (1.10-1.58)	1.34 (1.20-1.61)
1st HRIF visit at 4-8 mo		1.97 (1.61-2.40)	1.99 (1.65-2.40)	1.97 (1.62-2.41)

Model 1: Sociodemographic factors only
Model 2: Model 1 and infant factors
Model 3: Model 2 and hospital characteristics
Blank cells mean those factors were dropped in the specific since they did not meet model criteria

One of the strongest predictors of attendance at the second visit was completing an HRIF clinic visit during the recommended 4-8 months adjusted age (OR 1.97 95% CI [1.62, 2.41]). Table 4 shows demographic characteristics of those seen during the recommended time period compared with those not seen. There were many similarities between those not seen on time for visit 1(V1) and those not seen at all for the second visit, with maternal race of black and health insurance of Medi-Cal being significantly associated with having the first visit outside the recommended window. Maternal race of Asian was not significantly associated with having a first visit outside the recommended window. Those with a preferred provider organization (PPO) were more likely to attend the first visit on time, 17% of those seen on time had a PPO, while 11.1% of those seen outside the window had a PPO (p=0.0002). There was no statistically significant difference in timing of V1 for other private insurance coverage.

Table 4

Demographics of Visit 1 Attendance During or Outside Recommended Time Period of 4-8 Months

Demographic Factor	Level	Visit one outside 4-8 months N=666*	(%)	Seen for any visit between 4-8 months N=4035*	(%)	p-value if <.05
Maternal Age:	$\leq$19	59	(8.9)	313	(7.8)	
	20-29	277	(41.7)	1616	(40.1)	
	30-39	277	(41.7)	1818	(45.1)	
	$\geq$40	52	(7.8)	285	(7.1)	
Maternal Race	Black	95	(14.4)	462	(11.5)	.0002
	Hispanic	319	(48.3)	1774	(44.1)	
	Asian	83	(12.6)	481	(12)	
	American Indian	4	(0.6)	9	(0.2)	
	Other	21	(3.2)	109	(2.7)	
	Non-Hispanic White	139	(21)	1187	(29.5)	
Infant Insurance	Public **	383	(60)	2116	(52.5)	.0001
	Private: PPO	71	(11.1)	685	(17)	.0002
	Private: HMO/POS/EPO	156	(24.5)	1070	(26.6)	
	CCS Coverage***	378	(59.3)	2260	(56.1)	
Home Language	English	452	(70.3)	2955	(75.7)	
	Spanish	147	(22.9)	754	(19.3)	
	Other	44	(6.8)	192	(4.9)	
Primary Caregiver	One Parent	257	(39.1)	1477	(37.2)	<.0001
	Both Parents	385	58.6	2367	(59.7)	
	Other	15	2.3	104	(2.6)	
Sex	Male	341	(51.2)	2096	(52)	.
Birth Weight (g)	$\leq$500	7	(1.1)	42	(1)	<.0001
	501-750	105	(15.8)	595	(14.7)	
	751-1000	162	(24.3)	973	(24.1)	
	1001-1250	172	(25.8)	1148	(28.5)	
	1251-1499	220	(33)	1277	(31.6)	
Gestational Age <32 weeks	Yes	580	(87.1)	3551	(88)	
Multiple Gestation	Yes	165	(24.8)	1142	(28.3)	
Small for Gestational Age	Yes	199	(29.9)	1091	(27)	.
Chronic Lung Disease	Yes	192	(29.1)	1062	(26.5)	
Intraventricular Hemorrhage Grade III or IV	Yes	35	(5.4)	223	(5.7)	
Surgery	Yes	115	(17.3)	613	(15.2)	
Residence in a Rural County	Yes	172	(25.9)	1136	(28.3)	

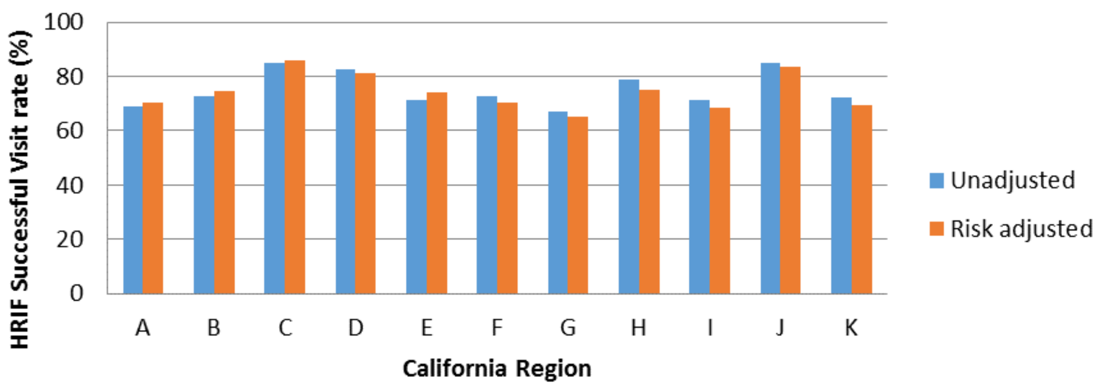
*due to missing data sums of factors may be less

**Public: Medi-Cal (Medicaid) and Healthy Families (S-CHIP)

***CCS coverage available to those with both public and private insurance

There were differences in follow-up rates based on geographic regions as is seen in Figure 2.

Figure 2. Successful Second Visits by Region

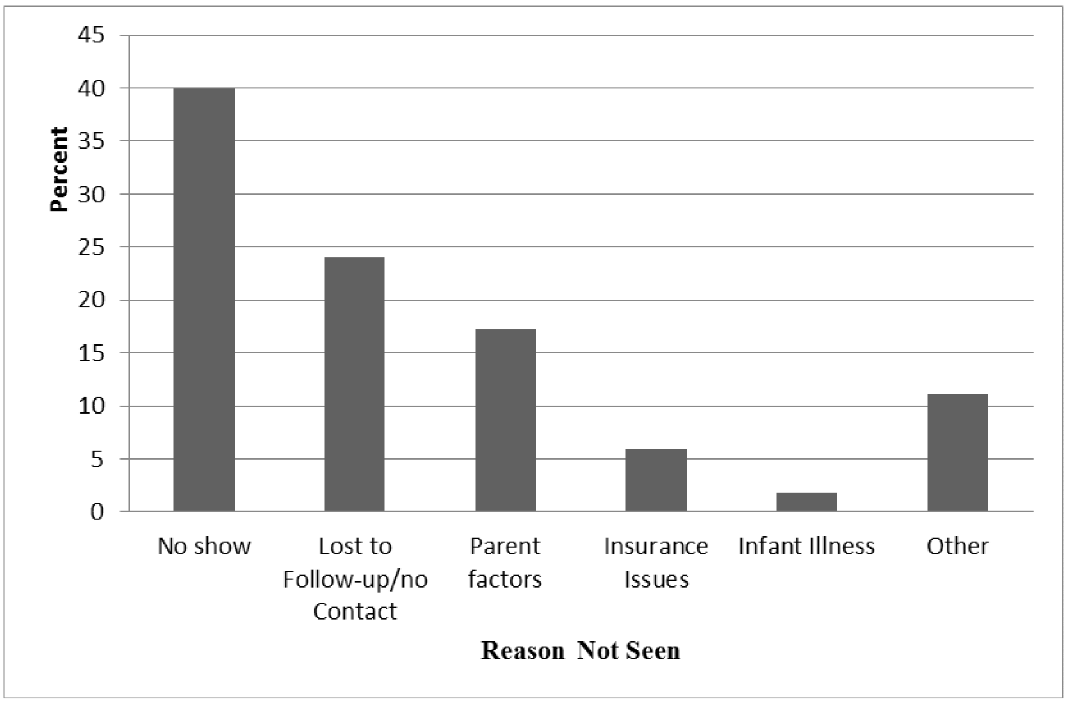


Risk adjusted rates range from a low of 65.2% in a region with 5 HRIF programs with 365 infants to a high of 85.5% in a region with 3 HRIF programs serving 200 infants.

The 66 HRIF programs varied greatly in size, performing between 10 and 235 second visits for infants born in 2010 and 2011. The smallest programs each had 22 infants referred to them and had risk adjusted V2 follow-up rates of 79.7% and 88.2%. The largest program had 323 infants and a risk adjusted rate of 74.6%. Using the final mixed model, the risk adjusted V2 completion rates ranging from a low of 34.5% to a high of 105.2%. Programs in the third (80-130 visits/year) and fourth (140-444) volume quartiles were associated with improved attendance, OR of 2.19 95% CI [1.55, 2.62] for the third quartile and OR of 1.76 95% CI [1.38, 2.24] for the fourth quartile. This was not significant when controlling for other factors in the model.

There were 1,207 not seen for V2. The reason for loss to follow-up was documented for only 953 (79%). As Figure 3 shows, the three leading documented reasons for not attending V2 were being a no show for a scheduled appointment (382), unable to contact (227) and parental reasons (164). Of the 17% not seen for parental reasons the majority were parents refusing (65) or having competing priorities (80) with a few due to lack of transportation (15), cost (15) or parental illness (4). A small percent were not seen due to insurance issues including 4 who were denied coverage from CCS.

Figure 3. Reason Infant not Seen for Second HRIF Visit (N=953)



Discussion

This is a large, multi-site, population-based study, assessing attendance at the second of three recommended visits. The large amount of data available affords an opportunity to explore both patient/family and site specific factors. Overall follow-up rates were low as has been found previously, confirming prior research on smaller populations.[11,17-19] There was an approximately 25% drop-off between referral and first visit attendance and approximately the same drop-off between first visit and second visit attendance. This differs from previous research showing greater drop-off between referral and the first visit compared with first and second visit.[19]

Those who were smaller and at higher risk due to neonatal morbidities and marked prematurity, were more likely to attend the second visit. This is consistent with prior research.[17-19] Available data is limited and factors that may have influenced this are unknown. It is possible that these smallest and less mature infants had more problems[2] and caregivers were more willing to bring them for follow-up. The HRIF programs may have made extra efforts to schedule these highest risk infants or the PCPs may have encouraged follow-up. Though more likely to attend V2, almost 20% of those ≤750 grams did not attend. A study of extremely preterm infants found that approximately 20% of infants born at <25 weeks gestation had minimal or no disability on follow-up.[3] In contrast some research has suggested that those who are not seen or seen only with difficulty are more likely to have disability.[22] It is unknown if those of the lowest weight who were not seen were doing well and parents did not perceive a need for follow-up or if they had deficits or potential disability and were not seen because their families were overwhelmed and did not return to clinic. In the total population studied (all birth weights), there was no association between neurologic outcome (normal vs. abnormal) at the first visit, and attendance at the second visit. The neurologic outcome by gestational age or birth weight was not examined, but could prove informative for future studies.

In prior research,[17] multiples were less likely than singletons to attend follow-up. This was not true in this population. Future research could examine the multiples population more specifically to identify sociodemographic variations and to determine if there are differences in attendance between spontaneous multiples and those conceived with assisted reproductive technologies.

Infants residing in rural counties were less likely to attend the second visit. These infants may receive substandard and fragmented healthcare due to inadequacies in the health care system in rural areas.[23] These infants would benefit from the evaluation and case management services provided through the HRIF program and it will be important to determine mechanisms to meet their needs.

Although only 6.6% of the California population is African American,[24] 11.8% of this population had the mother identified as black and this represented 16.1% of those not seen for the second visit. Black race was the factor most strongly associated with drop-off in attendance, a particularly concerning finding. Past research has associated black race with decreased follow-up compliance and poor neurodevelopmental outcomes.[25,26] Even when they have developmental delays, black infants may not be receiving early intervention services. A study of less than 1,000 gram (ELBW) infants found a significant decrease in eligible black infants receiving early intervention services at 24 months compared with white infants.[27] It is very important that children facing the highest risks be assessed appropriately in order to facilitate referrals for services. The data available does not allow sufficient analysis to determine the root causes of this finding. There may be multiple factors interacting, including socioeconomic factors and institutional factors, influencing drop-off in attendance. There are known disparities in health outcomes for African Americans in the United States.[28] Unrecognized (implicit) bias by health care providers has been found to adversely impact health care and health outcomes for minority populations.[29] There are approximately 1.5 million immigrants from Africa residing in California. Using available data, it is not possible to determine if the mother was an immigrant with potential language and cultural barriers, or if the mother was native born. Future research is needed to explore causal factors and to identify areas for improvement in the care provided to black infants.

The Asian population also had increased risks of non-attendance. This population is quite diverse and includes those with heritage from all parts of Asia, Hawaii, and the Pacific Islands. This population has been found to have significant differences in birth outcomes based on their specific population subgroup.[30] The data available does not allow exploration of the specific Asian sub-groups. Future studies should collect this information.

One interesting finding with implications for HRIF programs is that those infants seen for their first visit during the recommended interval, 4-8 months, were almost twice as likely to attend V2. This finding allows early identification of families at risk for being lost to follow-up. Those seen outside the recommended visit window had similar demographics to those not seen for V2 yet they were successfully seen for the first visit. Future research should focus on teasing out HRIF program specific factors related to scheduling to help identify best practices.

There was a great deal of variation between different HRIF programs. Those programs with the highest volume had improved attendance. Due to limits in the data, there is no information available about the structure of individual HRIF programs. In the future, it will be important to determine if different staffing patterns, or appointment processes are associated with improved attendance in order to make recommendations for best practices.

There were 1,207 (26%) who not seen for the second visit. This high rate existed despite CCS mandates and available CCS insurance coverage for 58% of those not seen. The fact that the infants had been seen for one visit and had scheduled a return visit yet did not attend, requires more detailed exploration. Reasons for loss to follow-up and no-show are unknown for most of the infants who did not return to clinic.

The most frequent known cause for an infant not being seen for V2 was no-show for a scheduled appointment. This no-show rate of 10.9% is lower than the average no-show rate of 20% found in a survey of medical directors of NICU follow-up programs.[31] As prospectively collected data, this rate may be more accurate than that collected by a retrospective survey. A NICU follow-up clinic in Baltimore routinely overbooked the clinic due to a no-show rate of 48.5%. This led to lengthy waiting times for clinic visits and parents stating they would not return to clinic.[32] Due to limits in data collected it is unknown what, if any, clinic-specific factors influence this no-show rate. There may be variations in scheduling the visits, with some programs scheduling months in advance and others closer to the visit date. A lengthy duration between the call for an appointment and the actual appointment date was associated with less attendance in a clinic performing autism assessments.[33] Clinics may have a system for making reminder calls, or may use an automated reminder call system. Reminder calls made personally by clinic staff have been associated with improved attendance compared with automated reminder calls.[34]

Parent and child factors may also be associated with this high loss to follow-up rate. Parental perception that their child was doing well and that follow-up was not needed has been associated with lack of appointment attendance in previous research.[17,26]

Only 2% of those not seen stated that lack of transportation was the cause. It is possible that this underrepresents the impact of transportation issues. The costs of transportation have been reported as a significant factor impacting lack of attendance.[17] Future research should explore both clinic-specific and parent/child specific factors associated with no-shows and failure to return to clinic.

Although the risk for neurodevelopmental impairment is higher for the smaller and less mature preterm infants, larger and more mature premature infants also face the risk of adverse cognitive outcome and poor executive function.[35] This population may also have the most benefit from early intervention.[36] Future areas of research include a focus on identifying potentially modifiable factors to increase follow-up rates in infants with birth weights more than 1,000 grams.

The American Academy of Pediatrics recommends that all infants, children, and adolescents should have a medical home to provide and coordinate comprehensive care.[37] Included in this is the recommendation to facilitate appropriate consultation and referrals. California Children's Services mandates that the HRIF program assist families in establishing care with a primary care provider.[12] Despite these recommendations, the presence of a primary care provider functioning as a medical home did not improve follow-up attendance. Reasons for this are unknown but it is a concerning finding. There may have been poor communication between the HRIF program and the identified primary care provider, or the primary care provider may not perceive an added benefit to the patient from encouraging follow-up attendance.

This study was limited to analyzing data available in the HRIF-QCI data system. It is unknown if race and ethnicity data was collected by self-report, following best practices.[38] Missing demographics regarding parental education level and education meant those factors could not be put into the final mixed model due to small sample sizes. For infants born in 2010 and 2011, this information was captured only at the time of registration into the system. In an effort to improve collection of this information, it is now collected as part of each standard visit.

There was a significant amount of missing data, for 21% of those not seen for V2 there was no data entered regarding the cause. Future additional analysis of missing data may be helpful in identifying factors associated with missing data. If there are programs with less missing data, clarification of their processes may help other programs improve their practices. The HRIF-QCI system has developed a warning system to flag cases to encourage appropriate data entry. The missing data seemed to be missing completely at random without a pattern that would impact analysis of available data.

This is the first time that analysis of the second recommended visit has been performed using population-based data. The results of this study can inform clinical practice as HRIF programs seek methods to improve attendance. This study provides suggestions for future research to identify interventions to improve attendance. Adding a qualitative component and developing a mixed methods research plan could strengthen research in this area. This would allow investigators to explore a broader range of factors associated with clinic attendance, interviewing families, primary care providers, and HRIF team members to elicit perception and information to inform research and interventions.

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Chapter 4:

ASSOCIATION OF RURAL RESIDENCE AND PROGRAM FACTORS WITH HIGH RISK INFANT FOLLOW-UP ATTENDANCE

Abstract

Purpose: Due to risks of adverse neurodevelopmental outcomes, systematic follow-up is recommended for preterm infants. The purpose of this study was to compare patterns of High Risk Infant Follow-up (HRIF) clinic attendance between infants with rural and non-rural residence. A secondary aim was to compare program specific factors associated with attendance.

Methods: A retrospective analysis of prospectively collected data in the California Children's Services mandated HRIF Quality of Care Collaborative Data system. Infants with birth weights of <1500 grams born in 2010 and 2011 were included. Statistical analyses included bivariate analysis and backward stepwise logistic regression.

Findings: Of 3481 seen, 910 (27.1%) were rural, 1198 were not seen, 398 (33.2%) rural. Rural mothers were younger, more likely to be Hispanic, and had lower levels of education. Rural infants were less likely to attend the recommended clinic visit (OR 0.73 95% CI [0.58,0.91]). Rural programs had average follow-up rates of 69.0% compared with 76.1% for urban programs. One rural region was an exception with a follow-up rate of 85%. There were significant differences in risk adjusted follow-up rates between programs, ranging from 34.5% to 89.9%.

Conclusions: Infants with rural residence were less likely to attend the recommended follow-up visit and were more likely to have sociodemographic risks for adverse neurodevelopmental outcome. Overall, rural follow-up programs had lower attendance rates than non-rural programs with one significant regional exception. Additional research is needed to identify specific infant, family, and program factors associated with recommended HRIF visits.

Keywords: Follow-up studies, premature infants, rural population,

Background

Due to the risk for adverse health and neurodevelopmental outcomes, it is recommended that premature infants be systematically followed after discharge from the neonatal intensive care unit (NICU).[1,2] These specialized follow-up visits afford the opportunity for early identification of developmental delays, abnormalities in neurologic function, and unmet medical needs. Patients can then be referred for appropriate medical and early intervention services to improve ultimate outcomes.[3] Previous research has documented very low rates of follow-up with recommended follow-up visits. Poverty, substance use, and residence at a greater distance from the clinic have all been associated with decreased clinic attendance.[4] There is no information available regarding clinic location or the impact of residence in a rural region on attendance at recommended neonatal follow-up clinics. Rural areas have been found to have higher rates of preterm and low birth weight deliveries. A population-based study in Québec found that residence in a rural region was associated with increased risk of delivering a low birth weight infant after controlling for socioeconomic factors.[5] These high-risk infants may receive substandard and fragmented health care due to inadequacies in the healthcare system in rural areas.[6] Children with special health care needs residing in the western part of the United States had higher rates of unmet health care needs compared with other regions.[7]

The State of California has developed a system of follow-up for preterm infants discharged from California Children's Services (CCS) licensed NICUs. Referral for high-risk infant follow-up (HRIF) services is mandated, and infants are registered into a web-based reporting system, High Risk Infant Follow-up Quality of Care Collaborative (HRIF-QCI) to facilitate quality of care activities. This data system links to the California Perinatal Quality of Care Collaborative (CPQCC) data system, a web-based system designed to collect data to measure NICU quality of care. Data is entered on the vast majority (>90%) of infants cared for in California NICUs. Both NICUs and their associated HRIF programs are classified based on the level of service provided in the NICU, with a Regional NICU providing the highest level of care, followed by Community NICUs, with Intermediate NICUs providing the lowest level of care.[8]

At discharge and referral for HRIF services, eligible infants may receive CCS insurance to cover the costs of 3 recommended visits during the first 3 years of life.[9] Services provided by HRIF clinics include physical exams, developmental assessments, case management, and recommendations or direct referrals for needed health care services and early intervention. The 3 HRIF visits are recommended by CCS are to occur at specific intervals that coincide with important phases of development, visit 1 between 4-8 months, visit 2 between 12-16 months and the third visit between 18-36 months. The second visit (V2) is scheduled to occur at the time that the child should be developing important language and motor skills.[10,11] California is a large and diverse state with a large rural landmass. In a voluntary system of regionalized care, many preterm infants in California are cared for in tertiary medical centers, and all of the CCS-licensed regional NICUs and HRIF clinics are located in urban areas.[12,13] Those infants discharged to residences in rural areas often need to travel great distances to attend clinic, possibly decreasing the likelihood that they will attend. The impact of rural residence on HRIF attendance is unknown.

The purpose of this study is to identify patterns of attendance at the second recommended HRIF visit, comparing infants residing in urban regions with those residing in rural areas and identifying differences between the two populations. A secondary aim is to compare successful visits between HRIF programs based on the program type (regional, community, intermediate) and program geographic location.

Methods

Definition of Rural. An initial step required determining which definition of rural would be used for this analysis. The US Census Bureau defines rural as areas not included in urban areas (population ≥50 000), or urban clusters (2500-<50 000).[14] Based on this definition, only 11 of the 58 counties in California are defined as predominately "rural". The National Center for Health Statistics (NCHS)[15] uses a definition based on metropolitan statistical areas (MSA). Counties with MSAs with a population of 50 000 or more are considered metropolitan (or urban). Those with urban cluster populations or less than 49 000 are classified as nonmetropolitan (rural). In this scheme, 21 counties in California are considered rural.

Medical Service Study Areas (MSSAs) are geographical units used to describe population and health care data. These MSSAs do not cross county lines. They are classified by population density as urban (75 000-125 000 in a recognized community or neighborhood with similar sociodemographic characteristics); rural (<250 persons/square mile); and frontier (<11 persons/mile).[16] The California State Office of Rural Health defines counties as being rural[17] if more than 80% of the landmass consists of MSSAs that are either rural or frontier. The State of California Office of Statewide Health Planning and Development (OSHPD) uses MSSAs in planning for health care workforce,[18] and this definition from the Office of Rural Health is the definition of rural utilized for this study.

Data Sources. This is a retrospective review of prospectively collected data from the linked CPQCC and HRIF-QCI data systems. These systems (linked by the CPQCC number identifying the infant in the NICU) collect sociodemographic and medical information at each visit. When not seen for a recommended HRIF visit, data is collected specifying the reason the infant was not seen. Inclusion criteria were those born in 2010 and 2011, birth weight less than1500 grams, and who were seen for the first recommended HRIF visit. Exclusion criteria were those who were discharged after the first visit, those residing in a pediatric sub-acute facility, and those who had expired.

The CPQCC data system provided maternal sociodemographic data: maternal race and prenatal care (dichotomous yes/no for any amount of prenatal care), and infant medical risks: surgery during the neonatal period, presence of congenital anomalies, requirement for high frequency ventilation, late sepsis, and transfer between NICUs. Data in CPQCC is collected during the NICU stay.

The HRIF-QCI system provided remaining data. Sociodemographic factors; infant race, insurance type, caregiver(s) and living arrangement, caregiver educational level and employment information, home language, and zip code. Baseline medical eligibility characteristics included were gestational age <32 weeks, multiple gestation, chronic lung disease, persistent apnea, persistent pulmonary hypertension (PPHN) requiring inhaled Nitric Oxide (iNO), infants who were persistently unstable (i.e.

hypotension requiring pressor support), or with cardio-respiratory depression (i.e. acidotic, low Apgars). Multiple neurologic system factors collected included presence of documented seizures, intracranial hemorrhage (any type), intraventricular hemorrhage (IVH) grade III or IV, cerebral thrombosis or infarction, other CNS or neurologic abnormalities, and retinopathy of prematurity requiring surgery. First visit factors captured included neurologic assessment, timing of the first visit in relation to the recommended time, presence of a child protective services case, and presence of an identified primary care provider (PCP) and a determination if the PCP functions as a medical home as defined by the American Academy of Pediatrics.[19] California Children's Services classifies NICUs based on the staffing, the level of service provided, and the patient acuity. These classifications are regional (highest acuity), intermediate, and community (lowest acuity). The HRIF program is classified based on the associated NICU classification. This classification, status as a teaching hospital (yes/no), the geographic region, and visit volume are all available in the data system.

Zip code was used to determine county of residence and distance from residence to HRIF clinic. Distance quartiles were used in statistical modeling.

In 1979, California developed the Regional Perinatal Programs of California (RPPC) in order to provide comprehensive care to women and infants through coordination between public and private providers in geographic areas.[20] Clinic location was utilized to group programs into RPPC regions.

A publically available data file from OSHPD[21] that crosswalks, zip codes, and MSSAs was utilized in an effort to further refine location data for this project.

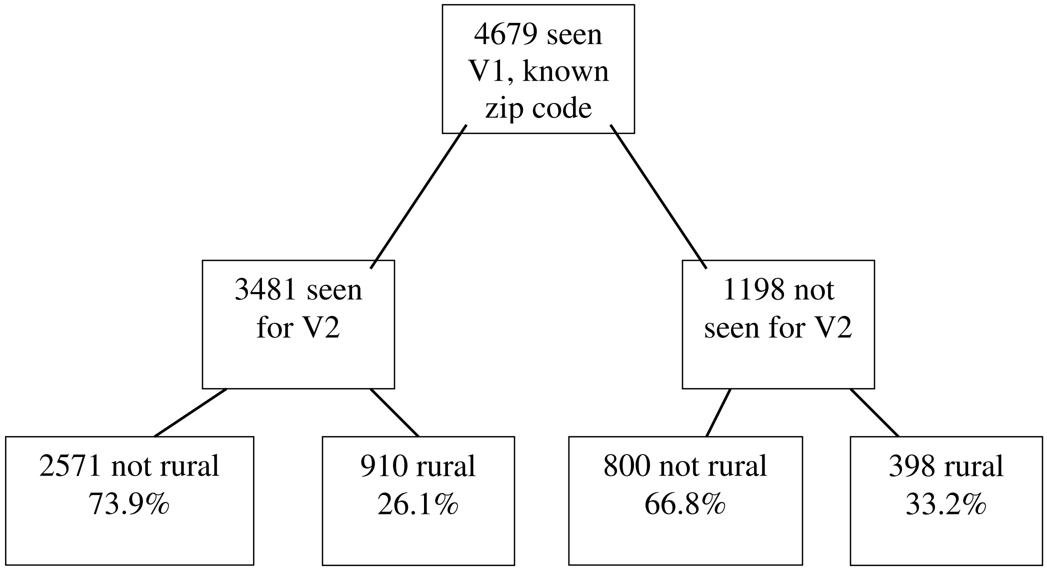
Statistical Analyses. The initial step in analysis was to perform bivariate analysis on variables listed above, comparing rural to non-rural. We used chi-square for categorical variables and t-test for continuous variables to identify factors with statistically significant relationships with attendance at the second HRIF visit. Tolerance statistics were utilized to evaluate collinearity. Mixed effects regression modeling was used to account for interactions among multiple variables and to calculate the odds ratio and 95% confidence interval associated with rural residence controlling for confounders. Backward stepwise logistic regression was used to develop the final model. The cut off for inclusion of a variable in the model was $P < .15$. Analyses were conducted using SAS version 9.4 (SAS Institute, Cary, NC)

Institutional Review Board approval was obtained from two universities in Southern California.

Results:

Forty-four out of 58 counties were classified as rural. There were 4701 infants seen for the first visit and eligible for inclusion in this study. However, residence zip code was missing for 22, leaving a population of 4679 infants. Of these, 3481 (74.4%) were seen for V2 and 1198 (25.6%) were not seen. As can be seen in Figure 1, only 26.1% of those seen had rural residence and 33.2% of those not seen had rural residence (p<.0001).

Figure 1. Residence Location for those Seen and Not Seen for the Second HRIF Visit



Demographic and Medical Factors

As can be seen in Table 1, there were marked sociodemographic differences between the two populations. The rural population was younger (10.9% ≤19 years compared with 6.7% of the non-rural population). Maternal race was more likely to be Hispanic (50.6% vs 42.3% non-rural). Those with rural residence were more likely to have lower levels of education, 233 (26.9%) of the rural population had less than a high school education compared with 360 (16.3%) of the urban population and only 4.4% (38) of the rural caregivers had completed graduate school compared with 256 (11.6%) of the non-rural caregivers. The rural population was less likely to have full time employment, with only 361 (27.6%) of the caregivers working full time compared with 1285 (38.1%) of the non-rural population.

Table 1. Selected characteristics for those with non-rural vs. rural residence

Factor	Level	Non-Rural Residence N=3371	(%)	Rural Residence N=1308	(%)	*P* value if <0.05
Demographics						
Maternal Age	≤19	225	(6.7)	143	(10.9)	< .0001
	20-29	1258	(37.4)	626	(47.9)	
	30-39	1617	(48.0)	471	(36.0)	
	≥40	268	(8.0)	67	(5.1)	
Maternal Race	Hispanic	1421	(42.3)	661	(50.6)	`
	Non-Hispanic White	910	(27.1)	411	(31.5)	
	Non-Hispanic Black	445	(13.3)	109	(8.3)	< .0001
	Asian	478	(14.2)	84	(6.4)	
	American Indian/Other	102	(3.0)	41	(3.2)	
Caregiver Education	<9th Grade	100	(4.5)	75	(8.7)	< .0001
	Some High School	260	(11.8)	158	(18.2)	
	High School Grad/GED	493	(22.4)	237	(27.3)	
	Some College/Other	517	(23.5)	237	(27.4)	
	College Degree	579	(26.3)	122	(14.1)	
	Graduate Degree	256	(11.6)	38	(4.4)	
Caregiver Employment	Full-Time	1285	(38.1)	361	(27.6)	< .0001
	Part-time/Temporary	224	(6.6)	89	(6.8)	
	Not Employed	1012	(30.0)	457	(34.9)	
	Other/Unknown	850	(25.5)	401	(30.7)	
Infant Insurance	Public**	1644	(49.2)	842	(64.5)	< .0001
	CCS Coverage***	1782	(53.4)	846	(64.8)	< .0001
	HMO	888	(26.6)	249	(19.1)	< .0001
	PPO	583	(17.5)	171	(13.1)	.0003
	POS/EPO	81	(2.4)	3	(0.2)	< .0001
	Other/Unknown	79	(2.3)	22	(1.7)	
Home Language	English	2409	(74.1)	982	(77.0)	.0001
	Spanish	643	(19.8)	255	(20.0)	
	Other	198	(6.1)	38	(3.0)	
Primary Caregiver	One parent	1243	(37.5)	502	(38.7)	
	Both Parents	1978	(59.8)	764	(59.0)	
	Foster/Adoptive or Other	79	(2.3)	29	(2.3)	
Sex	Male	1745	(51.8)	681	(52.1)	.
Selected Medical Risk						
Out Born	Yes	607	(18.0)	333	(25.5)	< .0001
Birth Weight (g)	≤500	32	(0.9)	17	(1.3)	
	501-750	497	(14.7)	199	(15.2)	
	751-1000	812	(24.1)	318	(24.3)	
	1001-1250	949	(28.2)	366	(28.0)	
	1251-1499	1081	(32.1)	408	(31.2)	
Gestational Age <32 weeks	Yes	2962	(87.9)	1153	(88.2)	
Multiple Gestation	Yes	973	(28.9)	329	(25.2)	.0115
Small for Gestational Age	Yes	928	(27.5)	355	(27.1)	
Chronic Lung Disease	Yes	869	(26.8)	351	(27.0)	
Cardio-respiratory Depression	Yes	75	(2.2)	52	(4.0)	.0009
Persistently Unstable Infant	Yes	364	(10.8)	103	(7.9)	.0028
IVH grade III or IV	Yes	182	(5.6)	75	(5.9)	

*due to missing data sums of factors may be less**Public: Medi-Cal (Medicaid) and Healthy Families (S-CHIP ***CCS coverage available to those with both public and private insurance

With a few exceptions, there were fewer differences in medical risk factors. There were no statistically significant differences between gestational age, birth weight, or small for gestational age (SGA) status between the two groups. Non-rural infants were more likely to be multiples, 973 (28.9%) compared with 329 (25.3%) of rural infants. Over 25% of rural infants were out-born compared with 18% of the non-rural population. There was no difference in diagnosis of chronic lung disease (CLD). Rural infants were somewhat more likely to have experienced cardio-respiratory depression (4% vs 2.2%), however were less likely to have been described as being persistently unstable (7.9% vs 10.8%).

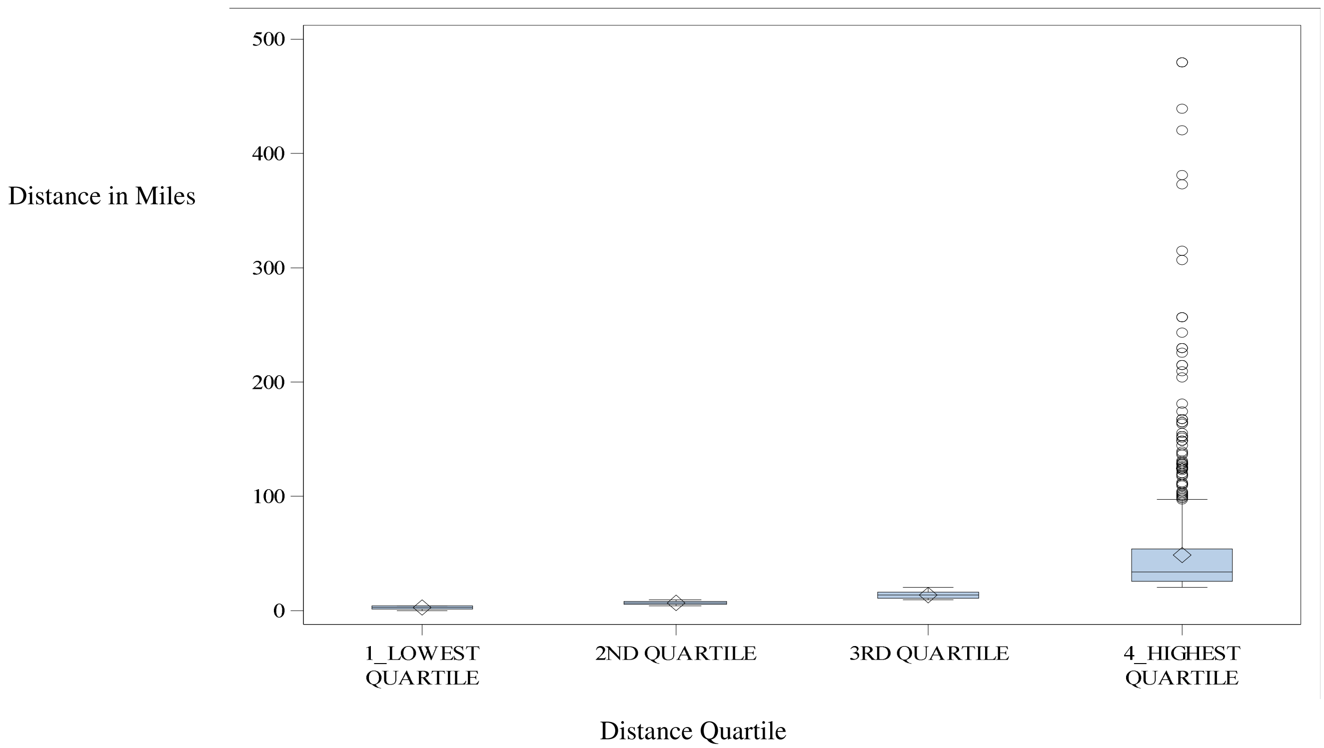
There were differences at the first visit (data not shown); those with rural residence were more likely to have attended the first visit within the recommended time interval of 4 to 8 months (85.3% compared with 81.7% of non-rural infants). They were much more likely to be enrolled in California Early Start (Early Intervention) at the time of the first visit, with 31.3% of those with rural residence enrolled compared with 23.9% of the non-rural residence. There was no statistically significant difference in those having a primary care provider (99.5% compared with 99.7%) or to have the PCP identified as providing a medical home (46.5% in rural areas and 44.2% in non-rural areas).

Distance

Dividing the distance into quartiles, lowest was 0-4.5 miles, second 4.6-9.4 miles, third 9.5-20.7 miles and the largest quartile ranged from 20.8-480.2 miles. There were four cases identified as outliers and removed, none of the removed cases had residence zip codes in California. The lowest quartile, 0-4.5 miles had 28.5% of the non-rural and

16.9% of the rural population. It is not surprising that those in rural residences were more likely to reside at a greater distance from the HRIF program. Of the 1160 infants who resided in the highest distance quartile (≥ 20.8 miles), 653 (56.3% of all in the fourth quartile) were rural. A large number of non-rural infants, 507 (43.7%) resided in the fourth quartile. The majority of all infants residing in the fourth quartile lived within 100 miles of the HRIF program as can be seen in figure 2.

Figure 2. Box Plot of Distance Distribution



HRIF Program

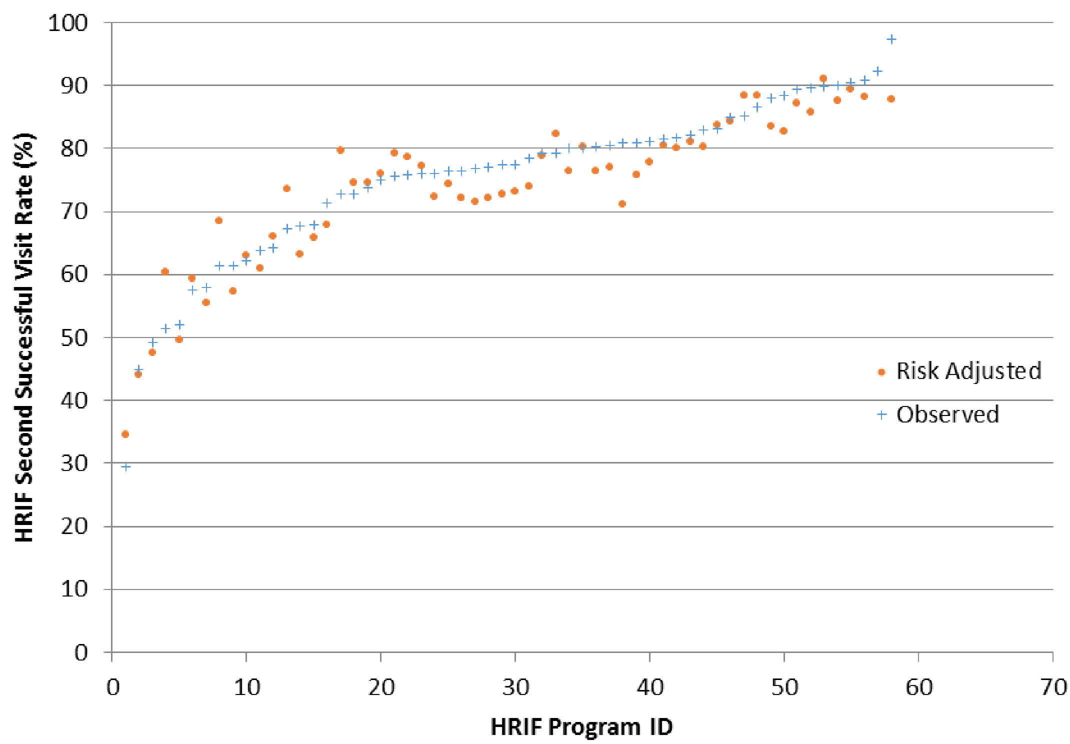
There were a total of 66 HRIF programs with average annual total visits (for all infants, not merely those in the study population) ranging from 4.5 to 443.5. High Risk Infant follow-up program volume was divided into quartiles, shown in Table 2, with those in the third quartile twice as likely to successfully complete V2 compared with those in the lowest quartile.

Table 2. HRIF Program Volume Quartile and V2 Visits

HRIF Volume Quartile	Average Annual Visits	Visit 2 Attendance		Odds Ratio (95% CI)
		N	%	
1	4.5-44.5	211	(63.0)	REF
2	46.5-78	544	(72.5)	1.55 (1.18, 2.04)
3	79.5-138.5	890	(77.50	2.02 (1.56, 2.62)
4	140-443.5	1849	(74.9)	1.76 (1.38, 2.24)

Significant variation in program enrollment and in visit 2 attendance was noted. There were 58 programs with >20 eligible infants enrolled. Program enrollment for these ranged from a low of 22 to a high of 323 eligible infants. Number of visits ranged from 10 to 235. As can be seen in Figure 3, there was significant program variation in both observed and risk adjusted V2 rates.

Figure 3. HRIF Program* specific V2 rate



*Programs with >20 infants enrolled

California Children's Services program classification was associated with V2 attendance on bivariate analysis. There were 20 Regional HRIF programs serving 2373 infants with 72.4% successful V2 follow-up (reference category). The 44 Community HRIF programs served 2249 infants and had a 76.9% V2 follow-up rate (OR of 1.27 95% CI [1.11, 1.45]). The two intermediate programs enrolled 79 infants and had a V2 follow-up rate of 58.2% (OR 0.53 95% CI [0.34, 0.84]). The 11 teaching hospitals enrolled 1022 infants and had a follow-up rate of 69.1% compared with the 51 HRIF programs in non-teaching hospitals that enrolled most of the infants in the state (3369), and had successful follow-up rate of 75.4% (Unadjusted OR 0.73 95% CI [0.62, 0.85]). There were 4 unclassified programs with 310 infants and a 79.7% follow-up rate.

Geographic location of the HRIF program was associated with successful V2 follow-up on bivariate analysis. The 16 rural HRIF programs enrolled 1173 infants with a follow-up rate of 69.0%, compared with 50 urban HRIF programs that enrolled 3528 infants and had a 76.1% V2 follow-up rate (unadjusted OR 0.7 95% CI [0.6, 0.81]).

Effect of Rural Residence

In the final fixed mixed effects model, the following factors were retained. Sociodemographic factors were: maternal race, insurance type, distance quartile, and rural residence. Infant factors were: gender, birth weight, SGA status, CLD, surgery, transfer, persistent apnea, PPHN requiring iNO. Visit one (V1) factors were: the timing of V1 and receiving early start services (yes/no) at the time of V1. High Risk Infant follow-up program factors in the full model were program volume and classification as being in a teaching hospital. After controlling for these factors, infants residing in rural counties were less likely to attend the second visit (OR 0.73 95% CI [0.58, 0.91]). These infants resided in counties with >80% of the landmass considered rural using MSSA classification. Residence was determined by zip code and there is often more than one MSSA classification within a single zip code. There are 102 zip codes containing both urban and rural MSSAs, 3 with both frontier and urban MSSAs, there are 11 with both rural and frontier MSSAs and 4 zip codes containing all 3 types of MSSA. It is not possible to determine a more specific designation for infant residence based on the available data.

Regional Perinatal Programs of California

There were 66 HRIF programs located in 11 different RPPC regions. Two of these regions (with 12 HRIF programs) were the northern and southern regions of a large integrated health maintenance organization, providing both insurance and health care. Their respective V2 follow rates were 85% for the northern region and 73% for the southern region.

The remaining regions are geographically based. Two of the RPPC regions are exclusively rural counties and 3 are exclusively non-rural counties. The 4 remaining regions are a mixture of rural and non-rural counties. As can be seen in Table 3, these 9 RPPC regions had successful V2 follow-up rates ranging from 69%-85%. Region 3, with the highest V2 follow-up rate (85%), contains only rural counties and has 25 birth hospitals and only 3 HRIF programs. Region 6 had the lowest rate (67%), is non-rural, and includes 18 birth hospitals and 5 HRIF programs. Using the mixed effects full model, risk adjusted visit rates ranged from 65.2%-85.8%.

Table 3. Regional Perinatal Programs of California Classification* and V2 Attendance

RRPC Region	Classification:	Birth Hospitals	HRIF Programs	Unadjusted V2 Attendance Visits	%	Risk Adjusted V2 Attendance Visits***	%
1	Mixed	33	3	128	(69)	126	(70.4)
2	Mixed	38	6	284	(72)	280	(74.4)
3	Rural	25	3	212	(85)	200	(85.8)
4	Mixed	20	5	369	(82)	367	(81.1)
5	Rural	30	5	387	(71)	378	(74.2)
6	Non-Rural	42	20	811	(71)	801	(70.3)
7	Non-Rural	18	5	250	(67)	244	(65.2)
8	Non-Rural	18	4	268	(78)	265	(75.0)
9	Mixed	19	3	208	(71)	208	(68.6)

* Excludes two integrated HMO regions
** Percent based on first visits
*** Visit count based on records with complete data used in final model

Discussion

In this population-based study, infants residing in rural counties were less likely to attend the second recommended HRIF visit than infants in non-rural areas. These infants were more likely to have public insurance and parents or primary caregivers with lower levels of education, both markers of poverty. They were more likely to reside at a greater distance from their HRIF clinic than non-rural infants. This finding is in agreement with previous research on a much smaller population that has shown poverty and distance have a negative impact on clinic attendance.[4] The purpose of HRIF programs is to identify infants and toddlers with problems and facilitate referrals for early intervention. Poor, rural infants face the biologic risks associated with prematurity and the social risks associated with poverty. Cumulative risk exposure has been associated with adverse parenting and poor cognitive development in poor infants residing in rural areas.[22] Decreased visit attendance in the high-risk rural population is a concerning finding.

The HRIF programs located in rural counties had lower follow-up rates than those in non-rural counties. Rural hospitals have poorer economic performance than urban hospitals and those with higher proportions of publically insured patients have even worse financial status[23] and these factors may influence the ability of rural HRIF programs to track and provide follow-up services. Rural regions have poor public transportation[24] and families may be unable to get from home to the clinic due to a lack of transportation.

An interesting exception to the findings above was that Region 3, an exclusively rural RPPC region, had the highest overall follow-up rate. Rural counties in California have higher rates of poverty (18.71% below the federal poverty level (FPL) compared with 14.76%) than urban counties.[25] Region 3 consists of 9 rural counties, all with per capita annual incomes less than the California average and an average of 21.7% of the population has income less than the FPL. These 9 counties make up 19% of the landmass of the state and have just less than 9% of the total population.[26] In the future, an examination into the specific practices of the 3 HRIF programs in Region 3 will be important to provide information helpful to develop best practices.

Rural infants were more likely to be out-born, a risk for neonatal morbidity and mortality[27,28] and for increased risks of poor neurodevelopmental outcomes associated with these morbidities.[29] Poverty and low parental education levels are both associated with adverse neurodevelopmental outcomes[28] both factors that were more prevalent in the rural population, further increasing their risks. It is encouraging that a higher percentage of rural infants were enrolled in early intervention at the time of V1, since this is known to have a positive impact on outcome. The fact that more were receiving services at V1 but fewer attended V2 is in contradiction to findings from a previous single site study showing increased follow-up rates when infants are enrolled in early intervention.[30] That study was far smaller, with a total population of 298, and a successful follow-up rate of 59%. The authors did not describe if this was attendance at one or more visits.

Distance from residence to HRIF program was associated with attendance and those in rural residences were more likely to live at a greater distance from their assigned HRIF program. Distance alone, however, cannot account for variations in attendance between the rural and non-rural population. A significant percentage of those residing in the highest distance quartile resided in urban counties and half of those in rural areas resided ≤ 20.7 miles away from their assigned program. Many counties in California are quite large as are many urban areas. The city of Los Angeles is 465 square miles[31] and infants may reside at a great distance from the HRIF program within the city limits. Another potentially confounding factor is that location of the HRIF program is based on zip code of the primary clinic and does not capture outreach clinics in geographic locations that may be much closer to the residence.

There was marked program variation in successful follow-up rates. In order to make sense of the widely divergent risk adjusted follow-up rates, it will be important to obtain program specific information including staffing and scheduling procedures. This information is not captured in the available data system.

Limitations

Analysis was limited to available aggregate, de-identified data. There was a significant amount of missing data particularly related to caregiver level of education, a factor known to be associated with HRIF visit attendance.

The use of county level classification for residence may mean that the classification (rural vs non-rural) was inaccurate for some infants. An infant may reside in a county classified as "non-rural" yet be in an MSSA that is frontier, or the opposite may be true, with an infant residing in a small metropolitan region of an otherwise rural county. Zip code was utilized to assign county of residence. Had zip code been used to assign residence classification (rural vs non-rural) it would have been more specific. However there are more than 100 zip codes containing more than one type of MSSA. Analyzing zip code level data might compromise patient anonymity and would require informed consent. In the future, a prospective study that includes informed consent could be utilized to more specifically determine the association between clinic attendance and residence in each of the 3 MSSA types.

Due to data limits, we were unable to build a regression model for factors associated with visit attendance exclusively for the rural population, and this should be done in the future. Patterns of referral to HRIF programs were not examined. It is not known if the infants were referred and enrolled in the HRIF program associated with the NICU of birth, discharge, or the program closest to their residence. This information could be captured from the HRIF-QCI system. The reasons for referral to a specific HRIF program may have been based on insurance coverage, caregiver request, or NICU choice. Reason for referral to a specific program is not captured in the HRIF-QCI system.

It is unknown what proportion of rural infants was assigned to follow-up with a rural HRIF program.

In summary, adjusting for other risk factors, rural residence was independently associated with decreased attendance at the second recommended HRIF visit. There are marked differences in successful follow-up rates between individual HRIF programs and in California regions. These findings should be utilized as the foundation for future research to examine specific infant, family, and program factors associated with clinic attendance for high-risk infants overall and specifically for the rural population. This will be important in providing guidance to improve access to recommended services for this vulnerable population.

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Chapter 5

Preterm infants are at risk for adverse neurodevelopmental outcomes. The interactions between biologic and social risk factors can increase these risks for the most vulnerable children (Taylor et al., 2011; Potharst et al., 2010). Systematic follow-up of preterm infants is recommended in an effort to identify risks and make referrals for services and supports that are known to improve outcomes (Wang et al., 2006). The California Children's Services (CCS) High Risk Infant Follow-up (HRIF) program seeks to fill this role for eligible premature children in California, recommending three visits at specified intervals (Department of Health Care Services, 2013). The second visit is recommended to occur between 12 and 16 months, a time period associated with achievement of significant developmental milestones (Gesell, 1968/1925; Piaget, Gruber, & Vonèche, 1977). A mandatory web-based data system collects baseline referral data and visit specific data including capturing reasons why an infant was not seen. This system, the High Risk Infant Follow-up Quality of Care Initiative (HRIF-QCI), allows CCS to track program activities and support quality improvement activities for individual neonatal intensive care units (NICUs) and HRIF programs.

Research has shown that follow-up rates are poor, with 59% attending one visit at a single center in New York, (Nehra, Pici, Visitainer, & Kase, 2009) and 65% attending three visits in a Canadian study (Ballantyne, Stevens, Guttmann, Willan, & Rosenbaum, 2012). There has been no population based research on attendance at recommended follow-up visits.

Previous research has shown that residence at a greater distance from the program is associated with decreased HRIF clinic attendance (Harmon, Conaway, Sinkin, &

Blackman, 2013) but there is a gap in the literature regarding the impact of residence in a rural region on clinic attendance.   In at large state like California with a significant rural landmass, the impact of rural residence on attendance at a statewide recommended program is important to understand.

Research requires a theoretical framework. The HRIF program seeks to support the development of infants by early identification of problems and facilitating services to support infants and their families. The bioecological theory of development has been proposed as a valid basis for epidemiological studies involving child development (Avan & Kirkwood, 2010).

Aims

This research had three aims, each to be addressed in separate papers formatted to meet the requirements of potential academic publications. These aims were to:

1) Analyze Bronfenbrenner's bioecological theory of development, applying it to interventions to support the high-risk infant population.

2) Identify factors associated with compliance (attendance) at the second recommended High-Risk Infant Follow-up visit in a population of infants born in California in 2010 and 2011.

3) Examine the pattern of follow-up for infants residing in rural counties compared with those residing in urban counties to determine if differences exist in the same population of infants. A secondary aim for this study was to examine program specific factors and impact on clinic attendance.

The Bioecolocial Theory of Development

The bioecological theory of development developed by Urie Bronfenbrenner (1979, 1994) describes the interactions between biological and environmental factors influencing outcomes for preterm infants. The theory describes bidirectional interactions; parents and caregivers influence the infant and are influenced by the infant (Bronfenbrenner, 2001/2005). This theoretical framework supports the provision of early intervention and other family supports as mechanisms to improve outcomes for preterm infants and their families and has been utilized as the theoretical framework in previous research on the impact of interventions to support the development of preterm infants (Brooks-Gunn, 1995).

The bioecological theory explains biological factors, family and parent-child factors, and the impact of larger cultural, social, and political processes on the developing infant. Applying this theory to interventions for the high-risk, preterm infant identifies why various interventions can be beneficial. Numerous opportunities for future research are also revealed. It will be important to investigate the impact of both biological or medical factors as well as family, environmental, cultural, societal, and political factors on the outcomes of premature infants.

High Risk Infant Follow-up Second Visit Attendance

In the context of Urie Bronfenbrenner's bioecological theory of development, data collected in the HRIF-QCI data system was examined to determine factors associated with attendance at the second recommended HRIF visit for infants born during 2010 and 2011 with birth weight <1,500 grams. We performed a retrospective analysis of prospectively collected data. Infants were eligible if they had attended the first recommended visit and were not residing in a long-term care facility and had not been discharged from the program. We performed bivariate analyses using chi-square for categorical variables and t-test for continuous ones to identify statistically significant relationships with V2 attendance. After identifying significant factors, we developed three models using backward stepwise logic regression using a cutoff of $p<.15$ for model inclusion. The first model used only significant sociodemographic variables, the second model added infant risk factors, and finally hospital characteristics were added for the full model. Mixed effects modeling was used in order to better account for the interactions among multiple variables of interest.

The smallest infants (birth weight less than 750 grams) were almost twice as likely to attend compared with those with birth weights greater than 1,250 grams. Gestational age was dropped from the final mixed model due to high correlation with birth weight; the smallest infants are more likely to be the most preterm. A higher rate of attendance in this population is a reassuring finding, as the smallest and more preterm infants are at increased risk for neurodevelopmental disability (Hintz, et al., 2011; Kyser, Morriss, Bell, Klein, & Dagle, 2012).

Those who attended the first visit during the recommended interval were more likely to attend the second visit, and those seen for visit one outside the recommended time frame were very similar to those who did not attend the second visit. Due to limits in the data, it is not possible to identify potentially causal factors for this finding.

Sociodemographic factors were associated with attendance. Maternal race was a significant factor associated with attendance; infants whose mothers were Black or Asian were far less likely to attend. Those who had Medi-Cal (a marker for poverty in this time period) and infants with a single parent were also much less likely to attend. The socioeconomic factors of poverty and low levels of parental education are associated with adverse outcome in the preterm population (Mercier, et al. 2010; Taylor, et al. 2011; Potharst, et al. 2010). Due to a large amount of missing data, we were unable to assess the association of parent level of education with attendance in this study.

The finding that infants born to Black mothers were almost half as likely to attend the second clinic visit is very concerning. Past research has associated Black race with decreased follow-up compliance and poor neurodevelopmental outcomes (Mercier, et al., 2010; Perenyi, Katz, Sklar, & Flom, 2011). Prior research of extremely low birth weight (<1,000 gram) infants found a significant decrease in eligible Black infants receiving early intervention services at 24 months compared with White infants (Feinberg, Silverstein, Donahue, & Bliss, 2011). These very high-risk infants, who might benefit the most from HRIF services, are less likely to attend the second HRIF visit.

The most frequent known cause for an infant not being seen for the second visit was no show for a scheduled appointment. This overall no-show rate of 10.9% is lower than the average no-show rate of 20% found in previous survey research (Bockli , Andrews, Pellerite, & Meadow 2014). Data used in the present study was prospectively collected and may be more accurate than data collected by survey methodology. Although the overall no-show rate was lower than reported previously, this finding is concerning: a high no-show rate puts a significant burden on clinics and without obtaining information from the parents or guardians,it is impossible to know if there are potentially modifiable factors associated with those who did not attend a scheduled appointment.

The American Academy of Pediatrics (2004) recommends that all infants have a medical home to provide and coordinate comprehensive care, make referrals for appropriate consultation. California Children's Services mandates that the HRIF program assist families in establishing care with a primary care provider (DHCS, 2013). Despite these recommendations, the presence of a primary care provider functioning as a medical home was not statistically associated with improved visit attendance and was not included in the logistic regression models.

The strength of this research is the use of prospectively collected, statewide-level data. This population based analysis of factors associated with visit two attendance confirmed information known from previous research using far smaller populations; socioeconomic factors such as race and poverty are highly associated with clinic attendance. This current research also revealed new information: timing of attendance at the first visit is highly correlated with visit two attendance and that having a medical home is not associated with attendance. The no-show rate in this study was far higher than that in previous studies.

Rural Residence, Program Factors and HRIF Visit Attendance

Using the Bronfenbrenner's theoretical framework as context and the same CCS-HRIF dataset analyzed for overall attendance at the second HRIF visit, we sought to determine if rural residence for the infant was associated with clinic attendance. We also examined program-specific factors for association with clinic attendance. We used the same statistical methodology described above. Residence zip code was used to determine if infant residence was in a rural or non-rural county.

California has a large rural landmass. An initial step in this performing this analysis was determining what definition of rural would be utilized to categorize infant residence. For this study we utilized the definition developed by the California State Office of Rural Health (CA Department of Healthcare Services, 2012). This agency defines counties as rural if more than 80% of their landmass consists of Medical Service Study Areas (MSSAs) that are defined as rural (<250 persons/square mile) or frontier (<11 persons/square mile).

There were significant sociodemographic differences between the rural and non-rural populations. The rural population was younger, maternal race was more likely to be Hispanic, those with rural residence were more likely to have lower levels of education, and less likely to have full time employment. Unsurprisingly, rural infants were more likely to have been out-born, a factor that has been associated with adverse neonatal outcomes (Boland, Dawson, Davis, & Doyle, 2015).

Infants residing in rural residences were approximately 30% less likely to attend the second HRIF visit. These infants were more likely to have public insurance and parents or primary caregivers with lower levels of education, both markers of poverty. They were more likely to reside at a greater distance from their HRIF clinic than non-rural infants. All of these factors have been associated with decreased clinic attendance in previous research (Harmon, et al. 2013). This finding is concerning. Poor, rural infants face the biologic risks associated with prematurity and the social risks associated with poverty. Congruent with the bioecological theory of development, cumulative risk exposure has been associated with adverse parenting and poor cognitive development in poor infants residing in rural areas (Burchinal, Vernon-Feagans, Cox, & Key Family Life Project Investigators, 2008).

There were significant program variations in successful second visit completion with risk-adjusted rates ranging from 34.5% to 89.9%. The available data does not allow further explanation of these significant differences.

Program location was associated with follow-up rates; HRIF programs located in rural areas had follow-up rates of 69% compared with a follow-up rate of 76% for non-rural HRIF programs.

Excluding two regions that represent a self contained health maintenance organization; there are nine geographic regions for the Regional Perinatal Programs of California (RPPC). One of these regions, exclusively rural, had a risk adjusted follow-up rate of 85%, higher than any of the other regions. The region with the lowest follow-up rate (67%) is a non-rural region.

The low follow-up rate for infants with rural residence and low rates of follow-up for HRIF programs located in rural areas add important information to the literature on follow-up of preterm infants after NICU discharge. The contradictory finding of high follow-up rates in one geographic region offers the opportunity for future investigation to determine best practices to serve the rural population.

Implications for Nursing Practice

Nursing practice can be informed by the information gained from this research. Bronfenbrenner's theory shows that infant development is influenced by interactions with parents and caregivers and by the impact of larger social, cultural, environmental, and political factors. Nurses support infant development by supporting parent/child interactions as has been done in the Nurse Family Partnership described by Olds et al. (2007, 2010). As clinicians, nurses can make referrals for early intervention services that are known to improve outcomes for preterm infants (Spittle, et al. 2010; Spittle, Orton, Anderson, Boyd, & Doyle 2012). Maternal depression is associated with adverse developmental outcomes (Koutra, et al. 2013). Nurses can help identify mothers with depression and facilitate appropriate services.

In their roles of clinicians, managers, and coordinators of HRIF programs, nurses can use the information learned about factors associated with visit attendance to develop mechanisms to improve program performance and increase follow-up rates. For example, recognizing that attending the first visit outside the recommended time period is associated with decreased attendance at the second visit, HRIF program staff can institute enhanced measures to track and schedule those patients. Nurses in HRIF programs with low follow-up rates could compare clinic practices with programs with high follow-up rates to identify best practices.

Implications for Future Research

This study provides information on factors associated with attendance at the second HRIF visit using existing data. There are numerous opportunities for future research based on these findings. There were significant differences in follow-up rates based on maternal race and ethnicity. Determining factors associated with attendance within specific racial and ethnic groups could be an important first step in identifying subgroups at the highest risk for non-attendance to develop focused interventions. Rural residence was associated with decreased HRIF visit attendance. Using the existing data, a logistic regression model should be developed to identify factors associated with attendance in the rural population.

There was one RPPC region that had very high follow-up rates, yet was in a rural region served by only three HRIF programs. An investigation into practices of those three programs would be very important to identify potential best practices. Future research could investigate the impact of instituting these practices.

There is significant variation between different programs in their risk adjusted visit rates. Future research should investigate program staffing and practices and attempt to determine what staffing levels or practices are associated with improved attendance.

The no-show rate may have a negative impact on programs by decreasing income from reimbursement. High-Risk Infant follow-up programs strive to meet CCS mandates yet may be adversely impacted by the very high no-show rate. Economic research should be performed to determine the actual costs of meeting CCS requirements.

Future research on HRIF attendance will be strengthened by inclusion of information obtained from parents or guardians, either through surveys and quantitative methodology or by qualitative or mixed methods research. Previous qualitative research found that parents disliked the long waiting times in this clinic, with one family noting that they would not return due to this. Concerns were also raised regarding the cost, one mother noting that her child was doing well and that the benefits of attending clinic did not seem worth the cost (Hussey-Gardner, Wachtel, & Viscardi, 1998).

Alternative methods to provide HRIF services could be the focus of a Patient Centered Outcomes Research Initiative (PCORI) project. Involving parents/guardians in development of a program may improve follow-up rates.

There was not an association between having a medical home and attendance at HRIF visits. This finding requires further investigation and should include input from primary care providers who are in a position to facilitate HRIF clinic attendance through referrals and patient education.

The third recommended HRIF visit should occur during ages 18 to 36 months. It will be important to perform an analysis of factors associated with the third recommended visit especially since it is often the last HRIF visit before the child exits the program. Developmental assessments performed at this time period in extremely low birth weight infants have been found to have predictive value for cognitive outcome at school age (Doyle, Davis, Schmidt, & Anderson, 2012).

Health Policy Implications

Prematurity is a significant public health issue in the United States. In 2012, 11.5% of all births in the U.S. were preterm (<37 weeks) with 1.9% (76,041 babies) delivered prior to 32 weeks gestational age. Almost ten percent of these infants (7,231 or 1.4% of live births) were born in California (National Center for Health Statistics, 2013). The services offered by HRIF programs and early intervention providers are mechanisms to ameliorate some of the adverse outcomes associated with prematurity. Funding for these programs must be adequate to meet the needs of these large numbers. Future policy-focused research could include exploring the impact of changes in funding on services provided and the outcomes of high-risk infants.

Scholarly Trajectory

As a pediatric nurse practitioner and HRIF program coordinator, I have provided services to this population for 16 years. As a clinician, I have seen the impact of prematurity on infants and families and the benefits of early intervention services. As a program coordinator, I have struggled to track and schedule high-risk families. Working in a rural area, I have seen the difficulties facing rural families who reside two hours drive from the closest physical or occupational therapists, or who have no access to a compounding pharmacy for necessary medications. As a member of a research team, I have had the opportunity to perform research assessments and see the data generated become a scholarly work that changes clinical practice.

This current study will inform my continued clinical practice as I work to identify those least likely to come to HRIF visits and try to partner with their families to increase follow-up rates. I will use the information learned to advocate for children and families so that they receive appropriate services and interventions.

As a nurse scientist, I plan to continue to investigate HRIF clinic attendance as a mechanism to provide services to this high-risk population. Working with colleagues in the field, I plan to investigate what parents/guardians need and work in partnership with them to design an HRIF program that meets their needs while gathering information needed for NICUs to perform quality of care activities. Future efforts will be research to explore non-medical factors impacting outcomes in this population, particularly focusing on those with socioeconomic risk factors. I hope to work with a multidisciplinary team to investigate interventions to enhance the development of infants at the highest risk due to combination of biologic and social risks.

Conclusion

Preterm infants face numerous risks for adverse outcomes that are responsive to appropriate interventions. Attendance at HRIF programs is low, and many sociodemographic risk factors associated with adverse outcomes in this population are also associated with decreased clinic attendance. Infants residing in rural areas are less likely to attend clinic and have increased sociodemographic risks for adverse outcomes compared with those in non-rural areas. The bioecological theory of development can be applied to the preterm population and provides context and support for the benefits of HRIF clinics and the various supports and early intervention services available to this vulnerable population. The information gained through this study can inform nursing practice, health policy, and multiple avenues of future research. All are important mechanisms to support preterm infants and their families.

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APPENDIX A

University of San Diego

ENTERED

Institutional Review Board
Project Action Summary

Action Date: September 11, 2014 *Note: Approval expires one year after this date.*

Type: __New Full Review ___New Expedited Review ___Continuation Review _X__Exempt Review ____Modification

Action: _X__Approved ___Approved Pending Modification ___Not Approved

Project Number: 2014-09-008
Researcher(s): Martha Fuller Doc SON
Dr. Mary Barger Fac SON
Project Title: Factors associated with compliance with the second recommended High Risk Infant Follow-up Visit

Note: We send IRB correspondence regarding student research to the faculty advisor, who bears the ultimate responsibility for the conduct of the research. We request that the faculty advisor share this correspondence with the student researcher.

Modifications Required or Reasons for Non-Approval

None

The next deadline for submitting project proposals to the Provost's Office for full review is N/A. You may submit a project proposal for expedited review at any time.

Dr. Thomas R. Herrinton
Administrator, Institutional Review Board
University of San Diego
herrinton@sandiego.edu
5998 Alcalá Park
San Diego, California 92110-2492

Office of the Executive Vice President and Provost
Hughes Administration Center, Room 214
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SIGNATURE PAGE

All applicable signature lines MUST be signed. If any required lines are left blank, the application will be returned to the principal investigator.

9/3/14

Researcher (signature) Hahn School of Nursing and Health Science Date

Martha G. Fuller mfuller@sandiego.edu 619-804-3220
Researcher (printed) REQUIRED: email Phone

9/4/14

Faculty Advisor (signature) Hahn School of Nursing and Health Science Date
(Only required if PI is a **USD Student**)

Mary Barger, Ph.D. mbarger@sandiego.edu 619-260-7757
Faculty Advisor name (printed) REQUIRED: **email** **Phone**

N/A

USD Sponsor (signature) email Phone
(Only required if PI is **NOT a USD student/faculty**. The USD sponsor must be a full-time employee of USD).

N/A

USD Sponsor name (printed) Department/School and Date

9/8/14

School/College IRB Representative Date
(ALL applications must obtain this signature, whether your unit has a designated IRB representative or not. Contact the IRB Chairperson if you need guidance.)

9/10/14

Dean or His/Her Representative (signature) Date

The project described above has been approved by the USD Institutional Review Board.

9/11/14

Chair or Administrator to IRB (signature) Date

APPENDIX B

August 18, 2014

Martha Fuller, MSN, PNP-BC
High Risk Infant Follow-up Program
University of California, San Diego Medical Center
200 W. Arbor Dr., MC 8452
San Diego, CA 92103-8452

Dear Ms. Fuller:

I am writing you regarding your project "Factors associated with compliance with the second recommended High Risk Infant Follow-up visit". The California Perinatal Quality of Care Collaborative (CPQCC), and it's affiliated program, the High Risk Infant Follow-up, Quality of Care Initiative (HRIF-CQI) approves the afore-mentioned project, pending your IRB approval. We also grant you access to the aggregate, de-identified CPQCC data and authorization to use this data for this project, as outlined in CPQCC's Application for Research Collaboration, which can be found on our website at http://cpqcc.org/research/research_collaboration.

Best of luck with your efforts. We look forward to working with you.

Sincerely,

Barbara Murphy, RN, MSN
Administrative Director, CPQCC
Director, Perinatal Programs
Stanford University School of Medicine
Division of Neonatal and Developmental Medicine

APPENDIX C

141093X

UNIVERSITY OF CALIFORNIA, SAN DIEGO
HUMAN RESEARCH PROTECTIONS PROGRAM

TO: Ms. Martha Fuller

RE: Project #141093X
Factors associated with compliance with the second recommended High Risk Infant Follow-up Visit

Dear Ms. Fuller:

Your project has been reviewed by an IRB Chair and/or the IRB Chair's designee and certified as exempt from IRB review under 45 CFR 46.101(b), category 4: Research involving the collection or study of existing data, documents, records, pathological specimens, or diagnostic specimens, if these sources are publicly available or if the information is recorded by the investigator in such a manner that subjects cannot be identified, directly or through identifiers linked to the subjects.

Please note: When a study has been certified as exempt from IRB review, continuing review and approval is not required. Certification of Exemption is effective for the life of the study. However, all modifications to a study that has been certified exempt must be submitted to the IRB for prospective review and certification of exemption prior to implementation. In some circumstances, changes to the protocol may disqualify the project from exempt status.

The research activities described in the application have been determined to meet the criteria for exemption from IRB review. The PI should ensure that the research activities are conducted in compliance with applicable UCSD and Rady Children's Hospital San Diego policies and ethical standards as well as local, state, and federal regulations.

On behalf of the UCSD Institutional Review Boards,

/jc

Anthony Magit, M.D.
Director
UCSD Human Research Protections Program
(858) 657-5100; hrpp@ucsd.edu

Release date: 8/19/2014