SUPINE SLEEP POSITIONING AMONG HIGH-RISK U.S.-BORN INFANTS:
THE PERSISTENT PRETERM/TERM AND NON-HISPANIC BLACK/NON-HISPANIC
WHITE RACIAL DISPARITY

by

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Supine Sleep Positioning Among High-Risk U.S.-Born Infants: The Persistent Preterm/Term and Non-Hispanic Black/Non-Hispanic White Racial Disparity

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ABSTRACT

Statement of the Problem: Of the nearly 4,000,000 infants born in the US each year, approximately 10% are born preterm, defined as gestational age less than 37 weeks. Preterm infants are 2 to 3 times more likely to suffer sudden unexpected infant death (SUID), defined as “the death of an infant less than 1 year of age that occurs suddenly and unexpectedly, and whose cause is not immediately obvious prior to an investigation”. Compared to non-Hispanic white (NHW) infants, preterm and term non-Hispanic Black (NHB) infants suffer higher rates of SUID. During the first year of life, one of the most effective and modifiable parental behaviors to reduce the risk for SUID is adhering to safe infant sleep practices, including supine sleep positioning (SSP). However, despite the higher risk for SUID, mothers of preterm infants and NHB mothers are less likely to adhere to safe sleep practices compared to mothers of term infants and NHW mothers. While trends in SSP among term infants are known, data on preterm infants are lacking, particularly stratified by race/ethnicity. In addition, several states have identified improving safe infant sleep as a public health priority and have developed related policies and programs. Yet research is lacking on if and how these state-level policies and programs impact the behavior of mothers of preterm infants, and whether they impact differently diverse racial groups.

Objectives: 1) Assess the racial disparity in SSP between NHB and NHW mothers of early preterm (<34 weeks) and late preterm (34-36 weeks) infants, 2) Measure the impact of
states’ prioritization of safe infant sleep in their 2010 Title V Block Grant needs assessment on SSP rates for mothers of NHW and NHB infants, and 3) Investigate whether the comprehensive safe infant sleep initiative implemented by Massachusetts led to improved SSP among mothers of preterm and term infants.

Methods: For aim 1, multivariate multilevel linear models were used to compare the SSP rate change between early preterm and late preterm infants for NHB and NHW groups. For aims 2 and 3, adjusted SSP percents were first calculated through generalized linear models for control and intervention groups. In the second stage, a piecewise regression approach was taken, comparing the SSP rate change from the pre-intervention to the post-intervention periods for control and intervention groups. For all multivariate models, covariates included several maternal sociodemographic characteristics.

Results: The NHB-NHW racial disparity in SSP persisted from 2000 to 2015 with NHB infants having lower SSP percentages compared to NHW infants for early preterm and late preterm groups (p< 0.0001). For the early preterm group, NHB infants had a lower increase in the adjusted rate of SSP compared to NHW infants (p=0.002). For the late preterm group, there was no difference in the rate of SSP increase between NHB and NHW infants (p=0.43).

When SSP rate was compared for states that did (intervention group) or did not (control group) prioritize SSP as part of their Title V needs assessment from 2005 to 2015, for NHW infants, SSP improved from 62.5% to 82.1% in intervention states and from 68.6% to 78.9% in control states. For NHB infants, SSP improved from 39.8% to 62.3% in intervention states and from 46.3% to 58.3% in control states, respectively. After adjustment for maternal characteristics, there was no difference in the rate of SSP change from the pre- to post-intervention periods for either NHW or NHB infants in intervention and control groups.
When Massachusetts, which had implemented a comprehensive SSP program was compared to Vermont and Maine, which undertook limited efforts, for full term infants, SSP improved from 77.4% to 86.7% and from 81.8% to 84.8% for intervention and control states, respectively. For preterm infants, SSP improved from 58.1% to 85.5% and from 78.6% to 91.1% for intervention and control states, respectively. After adjustment for maternal characteristics, there was no difference in the rate of SSP change from the pre- to post-intervention periods for either full term or preterm groups between intervention and control groups.

Conclusions and Significance: While SSP has improved for full term and preterm infants, the racial disparity between NHW and NHB persisted. Title V prioritization of safe infant sleep did not directly impact SSP improvement rate and did not reduce the inequities between NHW or NHB infants. Massachusetts’ safe sleep effort did not significantly improve the SSP rate among full term and preterm infants and disparities in SSP persisted by gestational age.

The form and content of this abstract are approved. I recommend its publication.

Approved: Angela Sauaia
DEDICATION

This dissertation is dedicated to my husband, Stephen, and my daughters, Lucy and Sonia, who fill me with the most profound love and inspiration. My life is so indescribably full because of you.

This dissertation is also dedicated to my parents, Jae-Hun and Ok-Joo Hwang. With two young children, you left all that was familiar in South Korea and immigrated to a new land with very little, except the hope that life would be better for your children. It is because of every sacrifice you made that I stand here.
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CHAPTER I
INTRODUCTION

The Magnitude of the Problem

Of the nearly 4,000,000 infants born in the US each year, approximately 10% are born preterm, defined as gestational age less than 37 weeks.\(^1\) While the greatest risk for morbidity and mortality occurs in the immediate neonatal period and is associated with large resource utilization in the neonatal intensive care unit (NICU), preterm infants continue to be at higher risk for adverse health outcomes after hospital discharge compared to their full-term counterparts.\(^2\)\(^-\)\(^4\) In particular, preterm infants are 2 to 3 times more likely to suffer sudden unexpected infant death (SUID), defined as “the death of an infant less than 1 year of age that occurs suddenly and unexpectedly, and whose cause is not immediately obvious prior to an investigation”.\(^5\) The overall rate of SUID is 91.4 per 100,000 live births\(^6\) and among preterm infants, 291.7 per 100,000 live births.\(^7\) While the years following the Back to Sleep Campaign in 1994 initially saw a decrease in SUID, since 2000, rates have remained stagnant.\(^6\)

While SUID is a significant problem overall, this is compounded by significant racial and ethnic disparities in both preterm birth, a risk factor for SUID and in actual SUID rates in the U.S. For preterm birth, in 2015, Non-Hispanic Black (NHB) mothers were 1.5 times more likely to deliver prematurely compared to Non-Hispanic White (NHW) mothers at 13.4% and 8.9%, respectively.\(^8\) When stratified into early preterm (before 34 weeks) and late preterm (34-36 weeks) gestational age periods, NHB women were more than twice as likely to deliver early preterm (4.8% and 2.3%, respectively) and 1.3 times more likely to deliver late preterm infants (8.6% and 6.5%, respectively).\(^8\) Furthermore, compared to NHW infants, preterm and term NHB
infants suffer higher rates of SUID. Among preterm infants, the SUID rate for NHB infants is nearly 2.5 times that for NHW infants.  

**Interventions**

During the first year of life, one of the most effective and modifiable parental behaviors to reduce the risk for SUID is adhering to safe infant sleep practices, including supine sleep positioning (SSP).  However, despite the higher risk for SUID in the preterm population, data suggest that mothers of preterm infants are less likely to adhere to safe sleep practices compared to mothers of term infants. Moreover, compared to NHW mothers of preterm infants, NHB mothers are less likely to adhere to safe infant sleep practices such as supine sleep positioning. The precise reasons for these disparities in SSP between preterm and term infants and among racial/ethnic groups of preterm infants have not been rigorously studied.

Some studies suggest that the infant sleep practices modeled by medical providers during the prolonged hospitalization of preterm infants in neonatal intensive care units do not follow recommended sleep practices, thus adversely impacting parental behavior after hospital discharge. The racial disparity in sleep practices in the general infant population has been well studied, and attributed to variation in receipt of infant sleep advice from friends, family, and medical providers. In addition, there are significant differences in the level of trust in medical providers’ recommendations about infant sleep practices across racial/ethnic groups. It is, however, unclear if the magnitude of the racial disparity among preterm infants (who are at higher risk for SUID) differs from term infants and what are reasons for non-adherence among different racial groups.

Several states have identified improving safe infant sleep as a public health priority and have developed related policies and programs. Yet research is lacking on if and how these state-
level policies and programs impact the behavior of mothers of preterm infants, and whether they impact differently diverse racial groups.

To address these gaps in the literature, the objectives of this dissertation are to: 1) assess the racial disparity in SSP between NHB and NHW mothers of early preterm (<34 weeks) and late preterm (34-36 weeks) infants, 2) measure the impact of states’ prioritization of safe infant sleep in their 2010 Title V Block Grant needs assessment on SSP for NHW and NHB mothers, and 3) investigate whether the comprehensive safe infant sleep initiative implemented by Massachusetts led to improved SSP for mothers of preterm and term infants.

**Literature Review**

**Introduction**

Approximately 4 million infants are born in the United States (U.S.) each year and nearly 24,000 die in their first year of life for an infant mortality rate of about 6 per 1,000 live births.\(^1,22\) The leading causes of infant mortality (death within the first 365 days after birth) are related to congenital malformations or birth defects and prematurity or low birthweight status, most of which occur in the neonatal period (4 per 1,000 live births), defined as within the first 28 days of life.\(^22\) Beyond this point, in the post-neonatal period, the leading cause of infant death is due to sudden unexpected infant death (SUID). SUID typically encompasses three diagnoses: sudden infant death syndrome (SIDS), accidental suffocation and strangulation in bed (ASSB), and unknown.\(^23\) The distinction across these three diagnoses is often not clear\(^{24,25}\) and thus most current literature related to post-neonatal infant mortality, uses the overarching term of SUID in referring to deaths attributed to suffocation due to unsafe sleep practices or which ultimately are not known even after thorough investigation. Formally, the Centers for Disease Control and Prevention (CDC) defines SUID as “the sudden death of an infant less than 1 year of age that cannot be explained after a thorough investigation is conducted, including a complete autopsy,
examination of the death scene, and a review of the clinical history”. There are approximately 3700 SUIDs in the U.S. each year. While the 1990’s saw a dramatic decline in SIDS due to the Back-to-Sleep Campaign, there has been little improvement in SUID rates since 2000. There is robust literature focused on the risk factors and protective factors against SUID with a large body of work devoted to understanding the association of infant sleep practices and SUID. The majority of the epidemiologic, prospective cohort, and intervention studies of infant sleep practices have focused on the healthy term population, overlooking the unique risk profile of preterm infants and adherence to safe sleep practices in the preterm population. Moreover, there are robust data on racial disparities in infant sleep practices among the general infant population, but data are lacking for the preterm population, despite their higher risk for death.

**Epidemiology of SUID**

In 2015, nearly 3,700 infants suffered SUID in the U.S. The CDC estimates that among these deaths, approximately 1,600 were attributed to SIDS, 1,200 to unknown causes, and about 900 to ASSB. It is widely accepted that there is significant overlap in these three diagnoses due to a number of factors. First, the precise cause of SIDS is still not known. While the biologic mechanism of impaired responsiveness to hypoxia and hypercarbia in affected infants is widely accepted, it is not known why some infants fail to arouse when challenged with these hypoxic and hypercarbic episodes. While the precise cause of SIDS remains elusive, the risk factor profile represented in the Triple Risk Model, shown below, is widely accepted. This model posits that some infants have inherent characteristics that increase their risk for SIDS and that during a critical developmental period in their infancy (1-4 months of age) in the setting of an exogenous stressor, these infants will die.
A second reason for the uncertainty around specific estimates of SUID, is due to variability in declaring the cause of an infant death across local jurisdictions and states. Depending on the state or jurisdiction of the chief medical examiner, different criteria may be applied to define SIDS. For instance, states that participate in the CDC SUID Case Reporting System may use very specific investigation protocols and reporting mechanisms that allow for consistent differentiation of SIDS, ASSB and unknown, while other states, may leave this designation to the discretion of the medical examiner or state fatality review team.\textsuperscript{27-30}

As the importance of understanding the precise causes of SUID has gained attention in the U.S., a diagnostic shift occurred in the epidemiology of SUID. Since 2000, while the SIDS rate has been declining, deaths due to unknown causes and ASSB have been increasing, leading to a stagnant overall combined SUID rate.\textsuperscript{7,23,26}

Source (https://www.cdc.gov/sids/data.htm).
Figure 1.2 Infant death due to SUID, SIDS, ASSB, and unknown cause

**Risk factors**

Infant characteristics present at delivery as well as infant care practices by parents affect the risk for SUID. Infants are more vulnerable if they were exposed to nicotine in-utero, born low birth weight or preterm, or if they have a disruption in their serotonin metabolism and utilization in the brain (which can only be found post-mortem). While these infant characteristics cannot be modified postnatally, there are modifiable risk factors related to infant care practices after birth including adherence to safe infant sleep practices, continued breastfeeding, removing second hand smoke exposure, avoiding excessively warm environmental temperatures, room-sharing but not bed-sharing among infants and their caregivers, and the use of pacifiers.

**Safe infant sleep practices**

The American Academy of Pediatrics (AAP) defines safe infant sleep practice and environment as being positioned supine in a separate sleep space, on a flat firm surface, without
elevation of the head, without any unsafe objects such as blankets or bumper pads, and being put to sleep with a pacifier.26

Population-based epidemiologic studies as well as case-control studies of SUID have demonstrated that non-supine sleep positioning, co-sleeping with a caregiver, and use of bumper pads, blankets and other items that can cause suffocation significantly increased the risk for SUID. Among these practices, the best studied is supine sleep positioning (SSP). It is hypothesized that when infants with certain vulnerable characteristics are placed in the prone position, due to their muscular and motor immaturity, they are unable to lift and reposition their heads during episodes of hypoxia and hypercarbia.35,36

Figure 1.3 Hypothesized mechanism of death in prone sleep position

Studies have shown a significant association between prone positioning and SUID and a greater risk for death compared to supine positioning, with odds ratios ranging from 2.6-13.9.10,37-39 In addition, positioning the infant on the side has also been shown to be associated with SUID. Oyen, et al. undertook a case-control study of 244 SIDS cases and 869 controls and found that the odds ratios for prone and side sleeping compared to supine positioning were 13.9
(95% CI 8.2-24) and 3.5 (95% CI 2.1-5.7), respectively. In addition, these investigators calculated the population attributable fraction of prone and side sleeping to be 18.5% and 26.0%, respectively.\textsuperscript{10}

The factors that foster or hinder parental adherence to SSP continue serve as a focus of research for many maternal and child health investigators. A theoretical model adopted by the leading researchers in this field to understand maternal decision-making related to infant care practices is the Theory of Planned Behavior (TPB).

The Theory of Planned Behavior developed by Icek Ajzen in the 1980’s states that an individual’s intention to perform a certain behavior is determined by the attitude toward the behavior, the subjective norm of that person, and the perceived behavioral control.\textsuperscript{40}

\begin{figure}[h]
\centering
\includegraphics[width=0.5\textwidth]{theory_of_planned_behavior.png}
\caption{Conceptual model for the theory of planned behavior}
\end{figure}

Using the TPB framework, Colson, et al. investigated mothers’ choice of infant sleep position among healthy, mainly full term, infants as part of the Study of Attitude and Factors Effecting infant care practices (SAFE).\textsuperscript{17} Among the over 3200 mothers enrolled from 32 hospitals across the U.S., investigators found that mothers who reported that the infant sleep position was not up
to them (perceived control) were more likely to intend to place their infants in the prone (or non-supine) position. Mothers who reported positive attitudes about prone positioning (agreeing with the survey statements that prone positioning would be healthy, pleasant, good, safer, more comfortable, and reduce the risk for choking in their infants) were more likely to select this sleep position for their infants. Authors found that of all the sociodemographic, psychological, and clinical characteristics, subjective social norms and attitudes were most significantly associated with adherence to SSP.

While robust studies such as SAFE exist for the healthy infant population, the facilitators and barriers to adhering to SSP in the preterm group have not been studied. However, we do have some data on the prevalence of SSP in preterm infants during hospitalization and after discharge home. In addition, some studies suggest that parental behavior in the preterm population could be influenced by what is observed and modeled in the NICU during the prolonged hospitalization of preterm infants.

One study that investigated SSP among preterm and term infants analyzed PRAMS data from 2000 to 2011 and found that the prevalence of SSP declined with decreasing GA such that 59.7% of infants ≤ 27 weeks were placed in SSP compared to 66.8% of term infants. This study, however, did not investigate trends over time in SSP by GA. While authors did demonstrate significant racial/ethnic disparities for all GA categories, the change in disparities over the study period and the differences in the magnitudes of disparities among these GA groups found over time were not assessed. Vernacchio, et al. analyzed a cohort of low birth weight infants (< 2500 grams) from Massachusetts and Ohio from 1995-1998 and found that very low birth weight (<1500 grams) infants were the most likely to be placed in the prone position. While prone positioning declined among the entire cohort low birth weight infants from 19.9% to 11.4%

41
during the study period, among very low birth weight infants, the decline was replaced by an increase in side sleeping.\textsuperscript{14}

More recent studies on sleep practices among preterm infants have been done in the context of local hospital quality improvement (QI) initiatives to improve safe sleep practices in the neonatal intensive care unit (NICU). As recommended by the AAP, these neonatal QI projects sought to implement safe sleep practices in the NICU weeks or months prior to discharge in medically stable infants so that appropriate practices and sleep environments could be modeled to family for an extended period of time before hospital discharge. Investigators from the Children’s Memorial Herman Hospital in Houston, Texas implemented a multi-faceted initiative to integrate safe sleep practices in their neonatal intensive care unit (NICU).\textsuperscript{42} With respect to SSP, they found that at baseline, only 39\% of infants who were medically stable to engage in safe sleep practices, were positioned supine. Following their effort, SSP improved to 85\% and parental compliance with safe sleep practices after hospital discharge improved from 23\% in the pre-intervention period to 82\% in the post period. Researchers from Boston, MA undertook a similar approach to implement safe infant sleep practices into the routine care of preterm infants, first, in two NICUs in Massachusetts, and then to all NICUs throughout the state.\textsuperscript{43,44} When the QI effort was first undertaken in two NICUs, investigators found that among infants eligible for safe sleep practices, only 25.9\% of infants were in a safe sleep position or environment at baseline. In the post-intervention period, adherence improved to nearly 80\%.\textsuperscript{43} Given the success of this work, the QI initiative was expanded to all level III NICUs in Massachusetts. From July 2015 to June 2017, overall compliance with safe sleep practices (of which supine positioning is one component) improved from 47\% to 75\%.\textsuperscript{44} However, since post-discharge data were not collected for these Massachusetts-based efforts, it is unclear if these
hospital-based initiatives led to improved adherence to SSP in the home and thus population-based data on parental adherence to SSP among preterm infants continue to be lacking.

**Epidemiology of preterm birth**

Nearly 10% of the nearly 4 million infants born in the U.S. are born preterm, defined as delivery at less than 37 completed weeks gestational age. Among preterm infants, the majority are late preterm infants (34-36 weeks gestational age) at nearly 70% of all preterm births.\(^1\) While health care costs among pediatric patients pale in comparison to those for the adult U.S. population, neonatal intensive care is the one of the biggest drivers of pediatric medical care, and preterm birth is a major driver of the increasing costs of care for complicated neonates.\(^{45,46}\) A significant portion of these resources are utilized during the initial birth hospitalization when the risk for morbidity and mortality are the greatest. However, even after hospital discharge, preterm infants are at greater risk for poor medical and developmental outcomes compared to term counterparts.\(^4,47-50\) With respect to SUID, even the moderately and late preterm infants are more than two times more likely to suffer SUID compared to term infants. Those born less than 28 weeks are at nearly three times greater risk for SUID.\(^5,7,51,52\) In Massachusetts, while preterm infants comprised 8.9% of all births, they made up 26.7% of all SUID cases. Similarly for low birth weight infants, while they comprised 7.5% of all births in the states, they made up 26.7% of all SUID cases.\(^53\) Despite the higher risk for SUID in the preterm population, few studies have analyzed data related to maternal adherence to SSP stratified by gestational age. The majority of epidemiologic studies on safe sleep practices enrolled primarily healthy term infants from well-baby nurseries across the U.S. In studies where mothers were randomly selected and surveyed by telephone, the majority had delivered a term infant, which is understandable, given that nearly
90% of U.S. births are 37 weeks or greater in gestational age.\textsuperscript{15,54,55} Thus, there is a paucity of data on infant sleep practices among the preterm population despite their higher risk for SUID.

**Racial/ethnic disparities in SUID and SSP**

Racial and ethnic disparities in infant mortality and in SUID rates have persisted for decades. In a recently published analysis, CDC epidemiologists analyzed 1995-2013 US period-linked birth-infant death data to assess SUID rates by the following racial/ethnic groups: non-Hispanic white (NHW), non-Hispanic black (NHB), Hispanic, American Indian/Alaska Native, and Asian/Pacific Islander.\textsuperscript{7} During the study period, SUID rates were consistently highest for American Indian/Alaska Natives and NHBs while lowest for Hispanics and Asian/Pacific Islanders. When stratified by GA categories, NHB infants had the highest rate of SUID for all GA groups. For instance, shown below are SUID rates (expressed as deaths per 100,000 live births) for NHW and NHB infants in the early (1995-1997) and later (2011-2013) periods stratified by GA categories. This study was one of the first publications to analyze SUID data over time by GA and by race/ethnicity.

Table 1.1 SUID Rates per 100,000 Live Births by Gestational Age: United States, 1995-1997 Compared With 2011-2013

<table>
<thead>
<tr>
<th>Gestational Age</th>
<th>Non-Hispanic White</th>
<th>Non-Hispanic Black</th>
</tr>
</thead>
<tbody>
<tr>
<td>&lt; 28</td>
<td>308.7</td>
<td>235.7</td>
</tr>
<tr>
<td>28-31</td>
<td>307.6</td>
<td>206.6</td>
</tr>
<tr>
<td>32-33</td>
<td>255.9</td>
<td>169.2</td>
</tr>
<tr>
<td>34-36</td>
<td>171.7</td>
<td>148.6</td>
</tr>
<tr>
<td>37-38</td>
<td>96.4</td>
<td>94.2</td>
</tr>
<tr>
<td>39-41</td>
<td>73.2</td>
<td>65.7</td>
</tr>
</tbody>
</table>

Given that SSP is a significant protective factor against SUID and preterm and NHB infants are at greater risk for SUID than term and NHW infants, understanding the relationships among SSP, gestational age, and race is needed. The earlier mentioned study of SSP among preterm and term infants reported that less than half of all NHB infants from 2000-2011 were positioned supine compared to about two-thirds of NHW infants across all gestational age categories.\textsuperscript{41} Adherence to SSP among Hispanic mothers ranged from 52\% to 61\% across GA categories. Given that all years of the study period were analyzed in aggregate for this population-based analysis, there is a persistent lack of data on trends in SSP of preterm infants over time, stratified by race and ethnicity. In a more recently published report from the Centers for Disease Control and Prevention of 32 states and New York City in 2015, 37.6\% of NHB mothers placed their infants in the prone position to sleep compared to 16.1\% of NHW mothers.\textsuperscript{56} This analysis did not stratify infants by gestational age and thus, again, adherence to SSP among preterm infants is not well understood.

**Public health initiatives to improve safe infant sleep practices and reduce SUID**

Reducing infant mortality has been a priority at state and national levels for decades. Given that two-thirds of infant deaths occur in the first month of life, mainly attributed to prematurity, a significant amount of attention has been given to strategies to reduce preterm birth and low birthweight. However, both broad public health and clinical efforts have only been successful in initially reducing the birth of late preterm infants from 7.51\% in 2007, the highest rate in decades, to 6.82\% by 2014\textsuperscript{1}, and in fact, there was an increase in late preterm births in 2015 and 2016.\textsuperscript{8,57} The rates of very preterm infants and extremely low birth weight (ELBW; < 1000 grams) and very low birth weight (VLBW; < 1500 grams) births have not improved to the same extent.\textsuperscript{1} In addition, since 2000, post-neonatal death due to SUID, the leading cause of infant
death after the first month of life, has been stagnant.\textsuperscript{23} Recognizing that reducing SUID rates through maternal behavior change in infant care practices may be a more achievable goal compared to preterm birth with its broad array of social and medical risk factors, several national public health efforts were developed and incrementally adopted by states over the past decade, several which will be discussed in detail.

The Eunice Kennedy Shriver National Institute of Child Health and Human Development (NICHD) has been at the forefront of the public health effort to improve safe sleep practices and reduce SUID. This was, in part, due to the passage of the SIDS Act of 1974 when Congress first recognized SIDS as significant public health problem and directed the NICHD to lead research efforts on SIDS within the U.S. Public Health Service. In 1989, the NICHD developed the definition of SIDS which is still currently used, that is, “the sudden death of an infant younger than 1 year of age, which remains unexplained after a thorough case investigation, including performance of a complete autopsy, examination of the death scene, and review of the clinical history”. The NICHD funded several of the national studies, mentioned earlier, focused on understanding the risk factors for SIDS. As these and other international studies demonstrated that stomach sleeping was a major risk factor for SIDS, the NICHD launched their Back-to-Sleep Campaign in 1994, a multifaceted public health campaign to educate parents, providers, and childcare centers about the protective effect of the supine sleep position. Hailed as one of the most successful public health efforts, adherence to supine sleep positioning increased by nearly 50\%\textsuperscript{15} and similarly, death due to SUID decreased by nearly 50% by 2000.\textsuperscript{23}

\textbf{Efforts targeted at reducing racial/ethnic disparities}

Given the stark racial/ethnic disparities in SUID and unsafe sleep practices, the NICHD developed initiatives targeted to the highest risk racial/ethnic groups. In 2000, they tailored their
work to the African-American community and developed “Babies Sleep Safest on Their Backs: A Resource Kit for Reducing the Risk for SIDS in African American Communities”. In 2002, the NICHD collaborated with members of the Indian Health Service (IHS) and other American Indian and Alaska Native organizations to develop continuing education programs for nurses in their communities to reduce the SIDS risk. Prior studies have demonstrated that American Indian/Alaska Native infants in the Aberdeen area had a lower risk for SIDS if public health nurses had visited these families during and after pregnancy. Then in 2005, the NICHD led the Healthy Native Babies Project Workgroup to create culturally appropriate educational and messaging materials to target the five HIS regions with the highest SIDS rates. Finally, in 2013, recognizing that other sleep practices, like bed-sharing, were also significant risk factors for SUID, the NICHD launched their Safe to Sleep Campaign to broaden the initial work of the Back to Sleep Campaign to include other risk factors.

**Improving data systems**

While the NICHD focused their efforts around public health messaging and education, others focused their efforts on improving data collection related to SUID. In 2009, the CDC created a multistate SUID case registry in an attempt to standardize the collection and categorization of data from SUID cases in order to develop a consistent approach to distinguishing the specific types of SUIDs. Participating states work with their local child death review teams to review data from death scene investigations by law enforcement, autopsy findings from medical examiners and coroners, as well as clinical information from pediatric and obstetric providers. These data were then shared with the CDC through the National Center for Child Death Review (NCCDR) Web-based Case Reporting System. In this way, the CDC
hoped to better quantify and describe how unsafe sleep practices contribute to infant death so that stakeholders could then develop appropriate intervention initiatives to reduce SUID rates.

**Recent national effort to reduce infant mortality**

From 2000, the infant mortality rate remained at 6 per 1000 live births, with little improvement despite wide ranging efforts focused on reducing preterm birth and SUID. Thus in 2012, the Maternal and Child Health Bureau (MCHB) of the Health Resources and Services Administration (HRSA) partnered with the National Institute for Children’s Health Quality (NICHQ) in developing an infant mortality reduction program utilizing the science of quality improvement and collaborative learning to accelerate improvements in infant mortality. This effort was first targeted to the southern U.S. states in Regions IV (AL, FL, GA, KY, MS, NC, SC, TN) and VI (AR, LA, NM, OK, TX) where infant mortality rates were 1.5 to 2 times higher than the national average. In 2013, six other Midwestern states joined the effort and by 2014, nearly all states and U.S. territories were participating. The initiative was called the Collaborative Improvement and Innovation Network to Reduce Infant Mortality (IM CoIIN). It adopted the science of the collaborative innovative network (CoIN) developed by Dr. Gloor with the following key elements: 1) a cyber-team where most of the collaborative work is distance-based; 2) innovation and communication occurs rapidly and across all levels; and 3) the work is characterized by meritocracy, transparency, and openness with contributions from all participants. IM CoIIN took this approach, and also added the quality improvement framework to allow for rapid cycle identification of problems with prompt testing of interventions and review of process and outcome measures. The goal was for states to develop Plan-Do-Study-Act (PDSA) cycles lasting 12-18 months. IM CoIIN identified six strategic areas of focus and states selected the topics most relevant to their population. The six focus areas included: 1)
SIDS/SUID/Safe sleep – improve safe sleep practices, 2) maternal smoking cessation – reduce smoking before, during and/or after pregnancy, 3) preconception/interconception health – promote health birth spacing and reduce unintended pregnancy, 4) social determinants of health – incorporate evidence-based policies and programs to improve social determinants of health and equity in birth outcomes, 5) prevention of preterm and early term births – increase use of 17-hydroxy-progesterone to prevent preterm labor and reduce early elective deliveries, and 6) risk-appropriate perinatal care – increase the delivery of higher-risk infants and mothers at appropriate level facilities. From September 2012 to December 2013, the regionally focused phase took place and from September 2013 to September 2017, the national effort occurred.

**Maternal and child health Title V program**

As these safe sleep-focused public health efforts were underway, states were also continuing to identify priority areas related to maternal and child health as part of the Title V Maternal and Child Health Services Block Grant Program. Title V is one of the largest federal block grant programs, providing support for nearly 54 million women, infants, and children in U.S. states and territories. Each year federal dollars are distributed to individual states by taking into account the proportion of low-income children in a state compared to the total number of low-income children in the entire U.S. States and territories must match every $4 of federal dollars with at least $3 of state or local money. In order to receive this federal funding, states must complete comprehensive needs assessments every five years where they identify areas of priority to which resources will be directed and outcomes measured. While the recent changes to the measurement of processes and outcomes are not relevant for the analysis presented in this dissertation, they do illustrate the degree to which the Title V program seeks to hold states accountable. In 2015, under the leadership of Michael Lu, the Associate Administrator for
Maternal and Child Health at HRSA, a new three-tiered performance measure framework was introduced in order to allow states to better demonstrate the impact of Title V on their selected health priorities, and included national outcomes measures (NOAs), national performance measures (NPMs) and evidence-based strategy measures (ESMs)\(^5\). Prior to this new measurement framework, it was not known if the priority areas selected by states and to which resources were directed actually demonstrated direct improvement in their process and outcome measures. For instance, as will be highlighted in the second aim of this dissertation, for states that selected safe infant sleep in their 2010 needs assessment, it is unknown if the safe sleep programs and policies then led to improvements in safe infant sleep practices or whether changes in adherence patterns were due to other factors. Moreover, there are almost no data on whether this prioritization led to varying magnitudes of impact for preterm and term infants. Given the importance of SSP in preventing SUID along with higher risk for death among preterm and NHB infants, it would be important for states to understand whether their prioritization of safe infant sleep leads to improved compliance with SSP by race and/or preterm status.

**Problem Statement, Research Questions, and Hypotheses**

Despite the higher risk for SUID among preterm and NHB infants, and the known protective effect of SSP, the changing prevalence and racial disparities in adherence to SSP after hospital discharge among mothers of preterm infants is not well known. Moreover, it is not known if the degree of racial disparity in SSP present for the term population also exists in the preterm group. In addition, the effect of state policies and program related to safe infant sleep on maternal behavior has not been previously characterized for the preterm population compared to term infants. Finally, it is not known if such statewide efforts reduce racial disparities in SSP.
Tremendous resources are utilized in the care of preterm infants during the first few days and weeks following their birth\textsuperscript{46,59}; numerous studies have demonstrated improved mortality rates of preterm infants in past decades with advancements in respiratory and nutritional care practices in the immediate postnatal period. Rigorous randomized controlled trials and long term longitudinal follow-up studies have been undertaken to study specific medical interventions on short- and long-term health as well as developmental outcomes of preterm infants. However, the adherence to health promoting behaviors after hospital discharge among caregivers of preterm infants requires additional study. It is well recognized that preterm infants continue to have significantly higher risk for poor health outcomes beyond the neonatal period, with continued respiratory, cardiovascular, and nutritional compromise.\textsuperscript{2,47,60} Even late preterm infants who are 1-3 weeks younger than their full-term counterparts, are two times more likely to be re-hospitalized in the short-term and in the longer term, have higher rates of adverse developmental and financial outcomes.\textsuperscript{61,62} While some of these longer term adverse outcomes are likely due to the medical complications suffered during NICU hospitalization such as intraventricular hemorrhage, chronic lung disease, pulmonary hypertension, severe infections like necrotizing enterocolitis, there exist modifiable caregiver behaviors that can reduce the risk for adverse outcomes such as SUID. Thus understanding the changing prevalence of SSP by gestational age and the impact of state policies on maternal adherence to SSP may delineate opportunities for interventions to improve the health of the highest risk infants. Furthermore, if the results of this thesis demonstrate significant racial disparity in SSP among preterm infants despite the prolonged hospitalization in the NICU with greater opportunities for parental education and engagement about safe infant sleep practices, then NICU providers will have to think critically about how to engage more effectively with these families.
Recognizing the higher risk for SUID among preterm and NHB infants, the protective effect of SSP against sleep-related infant death, and the lack of data on the relationship of gestational age and race with SSP, the aims of this thesis are the following:

Aim 1

To assess the NHB-NHW racial disparity in SSP in mothers of early preterm and late preterm infants from 2000-2015.

Hypothesis 1

The racial disparity in SSP among mothers of early and late preterm infants decreased during the study period

Aim 2

To measure the impact of states’ prioritization of safe infant sleep in their 2010 Title V Block Grant needs assessment on SSP for NHW and NHB mothers.

Hypothesis 2

States that prioritized safe infant sleep in the 2010 Title V MCHB Block Grant needs assessment had greater SSP improvement rates among NHW and NHB mothers of infants compared to states that did not explicitly or indirectly prioritize safe infant sleep.

Aim 3

To estimate the effect of Massachusetts’ safe infant sleep programs and policies on maternal SSP report for preterm and term infants.

Hypothesis 3

Compared to Maine and Vermont, Massachusetts had greater improvement in maternal report of SSP for preterm and term infants in the years following their multi-faceted effort to reduce SUID.
Significance of the Study

Despite the long history of public health efforts as well as research devoted to infant sleep practices, significant gaps in the literature still exist. First, while we better understand the racial disparities in infant mortality and safe sleep practices in the healthy term population, we know little about whether these disparities vary by gestational age, and among preterm infants, the magnitude of the racial disparity. Given that NHB mothers are at two to three times higher risk for preterm birth, leading to a greater proportion of NHB preterm infants compared to their NHW counterparts, it is important to understand if these disparities in adverse health persist beyond the birth hospitalization and first month of life. For instance, we know that among term infants, NHB infants are more likely to be placed to sleep with unsafe practices and die from SUID than NHW infants. Yet we do not know whether the same racial disparities affect preterm infants. If we demonstrate that among infants of the same gestational age, significant racial/ethnic disparities persist in SSP, then we can provide important evidence to NICU providers, primary care physicians, and parents that certain racial/ethnic preterm infants and families will require additional engagement and education during the prolonged birth hospitalization as well as after discharge home.

Since the passage of the SIDS Act in 1974, there have been many public health efforts to improve safe sleep practices. While the 1990’s saw a dramatic decline in stomach sleeping with near 50% reduction in SIDS, there has been little improvement since 2000. In response, some states have prioritized safe sleep in their Title V needs assessment and/or have joined the IM CoIIN effort focused on safe infant sleep. While most states do collect data on infant sleep practices and report trends over time, these data are not usually stratified by gestational age, and thus we do not know if the impact of these public health efforts differed for preterm and term
populations. Measuring the differential impact of safe sleep programs and policies across gestational age categories as well as by race will provide evidence for whether more targeted interventions for preterm infants and particularly, NHB mothers of preterm infants will be needed to improve their adherence to safe sleep practices.
CHAPTER II

METHODS

Conceptual model

The conceptual model developed for this thesis draws upon the theory of planned behavior and the social ecological model of health behavior. The Theory of Planned Behavior (TPB) developed by Icek Ajzen in the 1980’s states that an individual’s intention to perform a certain action is determined by the attitude toward the behavior, the subjective norm of that person, and the perceived control over that behavior.\(^{40}\) The social ecological model of health promotion and behavior change developed by McLeroy, et al. posits that both individual and broader social environmental factors influence the health behaviors adopted by individuals.\(^{63}\) Influential factors can be considered to reside in intrapersonal, interpersonal, organizational, community, and public policy domains.

Figure 2.1 Conceptual model of theory of planned behavior combined with social ecological model of health promotion and behavior change
Shown above is a proposed conceptual model that illustrates a host of factors from the level of the individual to the broader social environment, which may affect the domains of the TPB (attitudes, perceived subjective norms, and perceived behavior control) of a mother’s intention and subsequent compliance with supine sleep positioning. Included in this model is the idea that this process of decision making is not static or limited to one moment in time, but is, in fact, an ongoing iterative process from the pre-pregnancy to postnatal periods and includes the experience of preterm birth and prolonged hospitalization. Highlighted in this conceptual model are the three main exposures or factors that I hypothesized impact maternal adherence to SSP. Intrapersonal attributes of maternal race and the infant’s gestational age were considered in order to investigate racial disparities in SSP among preterm infants. The broader context was considered to test the effectiveness of several states’ adoption of infant sleep-related programs and policies on reducing racial disparities in SSP among preterm infants.

**Study Design, Methods, And Statistical Approach**

**Aim 1**

Assess the NHB-NHW racial disparity in SSP among mothers of early preterm and late preterm infants from 2000-2015.

**Hypothesis 1**

The NHB-NHW racial disparity among mothers of preterm infants in SSP decreased during the study period.

**Data Sources**

The data source for this study was the Pregnancy Risk Assessment Monitoring System (PRAMS), a surveillance system established by the Centers for Disease Control and Prevention (CDC) and administered by state public health departments. PRAMS was designed to monitor
selected self-reported maternal behaviors and experiences among women who recently delivered a live-born infant in the prior 2-4 months, with a maximum allowable recall of 9 months postpartum. Using standardized data collection methods, monthly stratified samples were selected from recent birth certificates. Surveys were obtained from mothers using a mixed-mode data collection method with mailed questionnaires and telephone follow-up for non-respondents. The CDC linked survey data to birth certificate data and applies weights for sample design, nonresponse, and noncoverage.64

Time Period and Response Rates: For this study we analyzed PRAMS state-specific data from 2000-2015. The following response rates were reported by the CDC for all states included in the PRAMS dataset: 2000-2006 ≥70%, 2007-2011 ≥ 65%, 2012-2014 ≥ 60%, 2015 ≥55%.

States included in the study: There was yearly variation in states’ inclusion in PRAMS due to non-participation as well as inability to meet threshold response rates in particular years. Among the 41 sites that contributed data to the national PRAMS data system, we analyzed data from 16 states with near complete data from 2000-2015, defined as 2 years or less of missing PRAMS data, which included Alaska, Arkansas, Colorado, Hawaii, Illinois, Maine, Maryland, Michigan, New Jersey, New York, Nebraska, Oklahoma, Utah, Vermont, Washington, and West Virginia.

Exclusions: We excluded surveyed women: 1) who were not of NHW or NHB race/ethnicity; 2) with unknown plurality; 3) whose infants were delivered at home, in a clinic, in-route to a hospital, or in free-standing birth centers, in order for the preterm groups to be more comparable in their interaction with hospital systems; 4) whose infants had died, or were not living with them at the time of the survey; 5) whose infants were born at 37 weeks or greater; and 6) with missing or unknown values for the SSP outcome or any co-variates. We excluded
mothers of infants who died were excluded since mothers are asked about SSP in the present tense, thus this question was not pertinent for mothers of non-survivors.

Outcome

Data on SSP were obtained from PRAMS. Mothers were asked: “In which position do you usually put your infant to sleep (side, back, and/or stomach)”. Responses were then categorized as supine (back) or non-supine (which included a combination of sleep positions). First trimester prenatal care use as well as method of delivery (vaginal or cesarean section), infant sex, and infant birth weight were obtained from birth certificates.

Exposures

Maternal race/ethnicity was categorized as non-Hispanic white (NHW) and non-Hispanic black (NHB) as performed by the CDC’s National Center for Health Statistics. Mothers of other races/ethnicities were excluded given that historically the greatest SUID disparity has been experienced by NHB and NHW infants. While we recognized that the percentage of infants born to women of Hispanic ethnicity is increasing, we concluded that the tremendous heterogeneity of this group and the lack of data on maternal country of origin in our dataset would not allow for robust analysis of disparities among Hispanic mothers.

Covariates

Using clinical estimates of gestational age (GA) from linked birth certificates, GA was categorized using the definitions created by participating states: ≤33 (early preterm), 34-36 (late preterm), and 37-42 (term) completed weeks.

Maternal demographic characteristics included data obtained from birth certificates (maternal age, education, and marital status). A history of previous live birth and insurance prior to pregnancy were obtained from PRAMS.
Analysis

We assessed maternal and infant characteristics in the earliest years (2000-2002) and in the latest years (2013-2015) of the study period to understand whether socio-demographic changes may have also been occurring as prevalence of SSP were changing over time. Prevalence estimates were calculated based on population-based survey weights provided by PRAMS using generalized linear models for correlated data via the GLIMMIX procedure in SAS.65-67

To examine the SSP prevalence among early and late preterm infants, we calculated the weighted prevalence by GA category. Weighted multi-level models were used to compare changes in unadjusted prevalence by GA category, while accounting for correlation across years within each sampling frame, by testing the interaction between time and GA category. We next adjusted the prevalence for maternal age, education, marital status, previous live birth, insurance status before pregnancy, receipt of prenatal care in the first trimester, and plurality, based on documented associations in the literature.13,15,68 We utilized robust standard errors to address correlation within states.

Separate multivariable multi-level models were developed for the early preterm and late preterm groups as shown below with the first level accounting for individual mothers and the second level for the sampling units nested within states.

Model specification

- Level 1:
  \[ Y_{ij} = \beta_0 + \beta_1 X_{ij} + \beta_2 Black_{ij} + \beta_3 Year_{ij} + \beta_4 Black_{ij} \times Year_{ij} + e_{ij} \]

- Level 2:
  \[ \beta_0 j = \gamma_i + \mu_{ij} \]

- Full specification:
  \[ Y_{ij} = (\gamma_i + \mu_{ij}) + \beta_1 X_{ij} + \beta_2 Black_{ij} + \beta_3 Year_{ij} + \beta_4 Black_{ij} \times Year_{ij} + e_{ij} \]

\( i = \) individuals
\( j = \text{sampling units} \)

\( Y = \text{Logit of Supine sleep positioning (SSP)} \)

\( \text{Black}=\text{non-Hispanic Black maternal race/ethnicity (0=NHW, reference group)} \)

\( \text{Year}=\text{study year} \)

\( \gamma = \text{mean estimate for the parameter} \)

\( \mu = \text{random effect for the sampling unit} \)

vector \( X \) contains demographic characteristics

**Aim 2**

To measure the impact of states’ prioritization of safe infant sleep in their 2010 Title V Block Grant needs assessment on the SSP change for NHB and NHW mothers of infants.

**Hypothesis 2**

States that prioritized safe infant sleep in the 2010 Title V MCHB Block Grant needs assessment had greater SSP improvement rates for both NHW and NHB mothers of infants compared to states that did not explicitly or indirectly prioritize safe infant sleep.

**Approach**

The Title V Maternal and Child Health Block Grant Program requires state grantees to complete comprehensive needs assessments every five years. Title V funds are then used to address these priorities in the states. Two states, West Virginia and Oklahoma, specifically listed safe infant sleep, of which SSP is a critical element, in their list of ten priorities in 2010, and also contributed to PRAMS data for the majority of the study period. States that did not either directly or indirectly prioritize safe infant sleep with available PRAMS data served as the comparison group and included Arkansas and Utah. For this aim, intervention and control groups were not further stratified into preterm and full term groups due to the decreased
statistical power to detect significant SSP rates changes by race/ethnicity as the aim 1 population of 16 states was reduced to 2 intervention and 2 control states.

**Data source**

The primary outcome of maternal adherence to SSP was obtained from PRAMS data from West Virginia, Oklahoma, Arkansas, and Utah. The cohort was defined as in aim 1 (see above). Data on selection of priorities for the 2010 needs assessment for all U.S. states and territories were obtained from Michael Kogan, PhD, Director, MCHB Office of Epidemiology and Research. States that explicitly listed safe infant sleep in their priorities were initially selected. States in the comparison group were selected if there was no explicit mention of infant sleep or the possibility of including infant sleep under broader umbrellas such as infant mortality or childhood injury prevention. In addition, among intervention and control group, states were only selected for final analysis if PRAMS data from 2005 to 2015 were available, with the exception of 1 year of missing data. The complete list of 2010 priorities for selected states are listed below.

**Analysis**

Using state-specific sampling weights, we used a two stage multilevel modelling approach to develop adjusted models for NHB and NHW mothers separately. In the first stage, we calculated the adjusted population prevalence of SSP with 95% confidence intervals (95% CI), for the intervention (Oklahoma and West Virginia) and control states (Arkansas and Utah) for each year of the study period from 2005 to 2015. Given that the needs assessment occurred in 2010, this year was considered the washout year with the pre period defined as 2005-2009 and the post period defined as 2011-2015. We then compared maternal demographic and infant birth characteristics between intervention and control groups.
Table 2.1 Title V priorities for intervention and control states in 2010 needs assessment

<table>
<thead>
<tr>
<th>STATE</th>
<th>PRIORITY</th>
</tr>
</thead>
<tbody>
<tr>
<td>Oklahoma</td>
<td><strong>Improve infant safe sleep practices.</strong></td>
</tr>
<tr>
<td></td>
<td>Reduce the prevalence of tobacco use among the MCH population.</td>
</tr>
<tr>
<td></td>
<td>Reduce unwanted, unplanned pregnancies.</td>
</tr>
<tr>
<td></td>
<td>Reduce motor vehicle injuries among children and youth.</td>
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<tr>
<td></td>
<td>Improve access to comprehensive health services for the MCH population.</td>
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<tr>
<td></td>
<td>Reduce infant mortality.</td>
</tr>
<tr>
<td></td>
<td>Improve transition services for CSHCN.</td>
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<tr>
<td></td>
<td>Reduce the prevalence of obesity among the MCH population.</td>
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<tr>
<td></td>
<td>Improve preconception health for females and males of reproductive age.</td>
</tr>
<tr>
<td></td>
<td>Improve the system of child care for families of CSHCN.</td>
</tr>
<tr>
<td>West Virginia</td>
<td><strong>Reduce the infant mortality rate, focusing efforts on black infants and Sudden Unexplained causes</strong></td>
</tr>
<tr>
<td></td>
<td>Reduce accidental deaths among youth 24 years of age or younger</td>
</tr>
<tr>
<td></td>
<td>Reduce smoking among adolescents</td>
</tr>
<tr>
<td></td>
<td>Decrease smoking among pregnant women</td>
</tr>
<tr>
<td></td>
<td>Reduce the incidence of prematurity and low birth weight</td>
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<tr>
<td></td>
<td>Decrease the incidence of fatal accidents caused by drinking and driving</td>
</tr>
<tr>
<td></td>
<td>Assure that children and adolescents access preventive dental services</td>
</tr>
<tr>
<td></td>
<td>Increase the percentage of adolescents who wear seat belts</td>
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<tr>
<td></td>
<td>Reduce obesity among the state’s population</td>
</tr>
<tr>
<td></td>
<td>Maintain and/or increase the number of specialty providers in health shortage areas</td>
</tr>
<tr>
<td>Arkansas</td>
<td>Reduce obesity and overweight among school-aged children.</td>
</tr>
<tr>
<td></td>
<td>Improve oral health in children and women.</td>
</tr>
<tr>
<td></td>
<td>Improved communication between the Title V CSHCN program and the CSHCN population</td>
</tr>
<tr>
<td></td>
<td>Reduce smoking among women of childbearing age.</td>
</tr>
<tr>
<td></td>
<td>Improved training and program development for the Title V CSHCN workforce</td>
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<tr>
<td></td>
<td>Reduce births to older teens.</td>
</tr>
<tr>
<td></td>
<td>Improve trauma care for children.</td>
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<tr>
<td>Utah</td>
<td>Decrease the percent of adolescents who feel so sad or hopeless almost every day for two weeks or more in a row during the last 12 months.</td>
</tr>
<tr>
<td></td>
<td>Increase the percentage of Medicaid eligible children (1-5) receiving any dental service.</td>
</tr>
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<td></td>
<td>Reduce the percentage of live births born before 37 completed week’s gestation.</td>
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<tr>
<td></td>
<td>Increase percentage of students who were physically active at least 60 minutes per day on 5 or more days of the past 7 days.</td>
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<td></td>
<td>Decrease the incidence of tobacco use among adolescents.</td>
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<td></td>
<td>Increase early and appropriate developmental screening for Utah children (Birth – 5).</td>
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<tr>
<td></td>
<td>Increase percent of children with special health care needs in the rural areas of the state receiving direct clinical services through the state CSHCN program.</td>
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<tr>
<td></td>
<td>Increase the percentage of children (birth – 17) eligible for Medicaid DM who are eligible for SSI.</td>
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<tr>
<td></td>
<td>Increase the percentage of women of childbearing age taking multivitamins with optimum level of folic acid.</td>
</tr>
<tr>
<td></td>
<td>Reduce the percentage of primary Cesarean Section deliveries among low-risk women giving birth for the first time.</td>
</tr>
</tbody>
</table>
In the second stage, using generalized linear mixed models, we developed piecewise regression models with year as continuous variable and specified the pre/post intervention periods. We tested the 3-way interaction of year, intervention, and period to assess whether the rate of SSP change differed significantly from the pre to post periods between intervention and control groups for NHW and NHB mothers. We utilized this mixed model approach to account for correlation across years within sampling groups in SSP.

**Specification**

Shown below is second stage multivariable model where $\beta_6$ represents the impact of the intervention on the exposed group in the post period compared to the control group.

- $Y_{ij} = \beta_{0j} + \beta_1 X_{ij} + \beta_2 Year_{ij} + \beta_3 Intervention_{ij} + \beta_4 Post_{ij} + \beta_5 Intervention_{ij} \times Post_{ij} + \beta_6 Intervention_{ij} \times Post_{ij} \times Year_{ij} + e_{ij}$
  
  $i = \text{individuals}$

  $j = \text{sampling units}$

  $Y = \text{Supine sleep positioning (SSP)}$

  $Intervention=\text{Oklahoma and West Virginia (Intervention states) vs Arkansas and Utah (Control states)}$

  $Post=\text{post period of 2011-2015 vs pre-period of 2005-2009}$

  $Year=\text{study year}$

  $\beta_{0j} = \gamma_i + \mu_{ij}$

  $\gamma = \text{mean estimate for the parameter}$

  $\mu = \text{random effect for the sampling unit}$

  vector X contains demographic characteristics
Aim 3

To estimate the effect of Massachusetts’ safe infant sleep programs and policies on maternal adherence to SSP for full term and preterm infants.

Hypothesis

Compared to Maine and Vermont, Massachusetts had greater improvement in SSP for full term and preterm infants in the years following their multi-faceted effort to reduce SUID.

Data source

PRAMS data for Massachusetts, Maine, and Vermont were the focus for this aim. Details about PRAMS has been provided in Aim 1. While Maine has been collecting PRAMS data since the inception of this surveillance system, Vermont and Massachusetts joined the PRAMS effort in 2001 and 2007, respectively. Our study included PRAMS data from these states from 2007-2015 with the following response rates, as set by the CDC: 2007-2011 ≥ 65%, 2012-2014 ≥ 60%, 2015 ≥55%. While there is yearly variation in states’ inclusion in PRAMS due to nonparticipation as well as inability to meet threshold response rates in particular years, data from Maine, Vermont, and Massachusetts were available for the entire study period for analysis.

Interventions

Massachusetts, Maine, and Vermont undertook different approaches to improving infant sleep practices during the study period, allowing for natural experimental analyses to measure the effect of states’ policies and practices to changes in SSP by state. Starting in 2011, Massachusetts undertook a multifaceted effort to improve safe infant sleep practices and later joined the IM CoIIN effort, adopting a rigorous quality improvement framework in scaling up their work. Some components of the safe sleep work included the following:
• Developed, implemented and disseminated a MDPH Safe Sleep policy to align with the 2011 American Academy of Pediatrics (AAP) guidelines for infant safe sleep practices.

• Promoted the adoption of an infant safe sleep policy, consistent with the AAP and the MDPH, among several MA state agencies that were involved in the care of infants such as the Department of Child and Families, Department of Early, Education and Care, Office of Child Advocate, State Child Fatality Review Team, Department of Housing and Community Health Development and The Children Trust.

• Developed and implemented, in collaboration with the WIC Program, new messaging, materials and a train-the-trainer presentation, which were disseminated to all WIC statewide programs.

• Collaborated with the MA Department of Early Education and Care’s Community Family Engagement program to provide education and training on infant safe sleep in November 2012. In 2014, assisted with the development and the dissemination of Request for Qualifications for trainers to become approved safe sleep trainers across the Commonwealth.

• Trained home visiting nurses from the Welcome Family Program, a branch of the state home visiting program, on safe sleep practices in August and September 2014.

• Trained 18 Early Intervention senior staff on infant sleep practices in November 2014, who in return trained over 1400 Early Intervention staff members, as well as provided information and resources to over 30,000 families enrolled in EI to improve safety and prevention of infant fatalities.
In 2014, Massachusetts Governor’s office and the Executive Office of Health and Human Services convened a task force with membership from multiple state agencies. They had the following activities:

- Public Awareness: posters on public transportation, billboards
- Resources for Physicians: Partnering with the Massachusetts Chapter of the American Academy of Pediatrics and Massachusetts Hospital Association
- Expanded website with easy to access and understand information, and links to find out more: www.mass.gov/safesleep

In Maine, the safe sleep effort began in March of 2010 with a presentation to the state Perinatal Nurse Managers of Maine group, a voluntary collaborative of the nurse leaders of birthing hospitals in Maine. At that time, birthing hospitals were encouraged to use the NICHD safe sleep educational materials. In addition, an educational video created by safe sleep advocates in Maine and the First Candle safe sleep video were distributed to the birthing hospitals over the course of 2010. Since 2012, educational lectures on safe infant sleep have been given by safe sleep experts at hospitals, community partner agencies, Maine Families visiting program, Public Health Nursing, and Office of Child and Family services.
In more recent years, Maine has become a Cribs for Kids state, with Maine Medical Center being the state host for this chapter. Maine’s network of home visiting programs has provided crib distribution and safe sleep education to families upon referral, recently formalizing this distribution protocol, and expanding to include participants of the Child Abuse Prevention Councils (in all of Maine’s 16 counties), and any Birthing Facility that wants to distribute directly to families, as long as safe sleep education is also provided at the time of portable crib delivery. The Children’s Trust provides the storage, distribution and tracking of the crib distribution, including follow up surveys with the recipients.

While Maine did join the IM CoIIN effort focused on smoking and safe sleep, additional resources were not available to support the work and thus the robust quality improvement approach taken by Massachusetts was not adopted in Maine.

In contrast, in Vermont, the safe infant sleep efforts have only recently gotten underway. For instance, in Spring 2018, the Vermont Department of Public Health will begin a public messaging effort focused on infant safe sleep for health care providers, community organizations, and parents. Unlike Massachusetts, Vermont has not engaged in a safe sleep related activity for the IM CoIIN effort.

For this aim, racial/ethnic disparities could not be assessed due to the very small number of non-White mothers in these New England states, particularly in Maine.

Analysis

We used a two stage multi-level modelling approach to develop adjusted models for full term and preterm groups separately. In the first stage, using state-specific sampling weights, we calculated the adjusted population prevalence of SSP with 95% confidence intervals (95% CI), for intervention (MA) and control (ME and VT) groups for each year of the study period of
2007-2015, controlling for maternal sociodemographic factors: maternal age, education, marital status, previous live birth, insurance status before pregnancy, and plurality. Given that MA initiated their multifaceted safe infant sleep work in 2011, this year was considered the washout year. The pre-period was defined as 2007 to 2010 and the post-period was defined as 2012-2015. We compared maternal demographic and infant birth characteristics between intervention and control groups in the pre- and post-periods.

In the second stage, using generalized linear mixed models, we developed piecewise regression models with year as continuous variable and specified the pre/post intervention periods. We tested the 3-way interaction of year, intervention, and period to assess whether the rate of SSP change differed significantly from the pre to post periods between intervention and control groups. We utilized this mixed model approach to account for correlation across years within groups in SSP.

**Specification**

Shown below is the second stage multivariable mixed model where $\beta_6$ represents the impact of the intervention on the exposed group in the post period compared to the control group.

- $Y_{ij} = \beta_0 + \beta_1 X_{ij} + \beta_2 Year_{ij} + \beta_3 Intervention_{ij} + \beta_4 Post_{ij} + \beta_5 Intervention_{ij} \times Post_{ij} + \beta_6 Intervention_{ij} \times Post_{ij} \times Year_{ij} + e_{ij}$

  $i =$ individuals

  $j =$ sampling units

  $Y =$ Supine sleep positioning (SSP)

  $Intervention =$ Massachusetts vs Maine/Vermont

  $Post =$ post period of 2012-2015 vs pre-period of 2007-2010
Year = study year

\[ \beta_{0j} = \gamma_i + \mu_{ij} \]

\( \gamma \) = mean estimate for the parameter

\( \mu \) = random effect for the sampling unit

vector \( X \) contains demographic characteristics
CHAPTER III

RESULTS

Introduction

This dissertation follows the three-paper structure and thus, this chapter presents the results of each aim as three distinct papers to be published in peer-reviewed journals.

Results of Dissertation

Aim 1

Introduction

In the United States, nearly 3500 infants die from sudden unexpected infant death (SUID), defined as “the death of an infant less than 1 year of age that occurs suddenly and unexpectedly, and whose cause is not immediately obvious prior to an investigation”.22,23 Non-Hispanic Black (NHB) infants are more likely to suffer SUID compared to non-Hispanic White (NHW) infants, and are over-represented in SUIDs.26,69 From the most recently available linked infant birth and death certificates in 2013, NHB infants comprised 15% of all U.S. births, but comprised 29% of all SUIDs.70,71 In addition to race/ethnicity, preterm birth (<37 weeks gestational age) is an independent risk factor for SUID, with preterm infants being 2 to 3 times more likely to die from SUID compared to term infants.5 In the preterm population, NHB infants are again at significantly greater risk for SUID compared to NHW infants, with more than a two-fold greater risk.7,69

The specific cause of SUID is unknown but the profile of risk factors has been well studied.26,27,35,72 During the first year of life, one of the most effective and modifiable parental behaviors that may reduce the risk for SUID is adhering to safe infant sleep practices, including supine sleep positioning or back-sleeping.26 The American Academy of Pediatrics (AAP) defines
safe infant sleep practice and environment as being positioned supine in a separate sleep space, on a flat firm surface, without elevation of the head, without any unsafe objects such as blankets or bumper pads, and being put to sleep with a pacifier. For the healthy term population, research on the racial/ethnic disparity in adherence to safe sleep practices is robust, but for preterm infants who are at much higher risk for SUID, less is known. Preterm infants are hospitalized after birth for significantly longer periods of time compared to term infants, ranging from weeks to months in the neonatal intensive care unit (NICU). Understanding the racial disparity in sleep practices in this highest risk group may highlight the need to improve parental engagement strategies about safe infant sleep during the prolonged birth hospitalization to increase adherence after hospital discharge.

To address this deficit, the objective of this study was to assess the NHB-NHW racial disparity in maternal report of SSP among U.S. born early (gestational age: <34 weeks) and late (gestational age: 34-36 weeks) preterm infants from 2000-2015. We hypothesized that SSP among NHB mothers of early and late preterm infants was persistently lower than NHW mothers during the study period, similar to trends in the term population.

**Methods**

**Data Sources**

The data source for this study was the Pregnancy Risk Assessment Monitoring System (PRAMS), a surveillance system established by the Centers for Disease Control and Prevention (CDC) and administered by state public health departments. PRAMS was designed to monitor selected self-reported maternal behaviors and experiences among women who recently delivered a live-born infant in the prior 2-4 months, with a maximum allowable recall of 9 months postpartum. Using standardized data collection methods, monthly stratified samples were
selected from recent birth certificates. Surveys were obtained from mothers using a mixed-mode data collection method with mailed questionnaires and telephone-follow-up for non-respondents. The CDC linked survey data to birth certificate data and applies weights for sample design, nonresponse, and noncoverage.\textsuperscript{64}

**Time Period and Response Rates**

For his study we analyzed PRAMS state-specific data from 2000-2015. The following response rates were reported by the CDC: 2000-2006 ≥70\%, 2007-2011 ≥ 65\%, 2012-2014 ≥ 60\%, 2015 ≥55\%.

**States included in the study**

There was yearly variation in states’ inclusion in PRAMS due to non-participation as well as inability to meet threshold response rates in particular years. Among the 41 sites that contributed data to the national PRAMS data system, we analyzed data from 16 states with near complete data from 2000-2015, defined as 2 years or less of missing PRAMS data, which included Alaska, Arkansas, Colorado, Hawaii, Illinois, Maine, Maryland, Michigan, New Jersey, New York, Nebraska, Oklahoma, Utah, Vermont, Washington, and West Virginia. Cohort flow diagram (Figure 1) shows all exclusions.

**Outcome**

Data on SSP were obtained from PRAMS. Mothers were asked: “In which position do you usually put your infant to sleep (side, back, and/or stomach)”. Responses were then categorized as supine (back) or non-supine (which included a combination of sleep positions). First trimester prenatal care use as well as method of delivery (vaginal or cesarean section), infant sex, and infant birth weight were obtained from birth certificates.
Exposures

Maternal race/ethnicity was categorized as non-Hispanic white (NHW) and non-Hispanic black (NHB) as performed by the CDC’s National Center for Health Statistics. Mothers of other races/ethnicities were excluded. While we recognized that the percentage of infants born to women of Hispanic ethnicity is increasing, we concluded that the tremendous heterogeneity of this group and the lack of data on maternal country of origin in our dataset would not allow for robust analysis of disparities among Hispanic mothers. Moreover, historically, the greatest racial/ethnic disparity in infant sleep practices has been among NHB and NHW mothers.

Covariates

Using clinical estimates of gestational age (GA) from linked birth certificates, GA was categorized using the definitions created by participating states: ≤33 (early preterm), 34-36 (late preterm), and 37-42 (term) completed weeks.

Maternal demographic characteristics included data obtained from birth certificates (maternal age, education, and marital status). A history of previous live birth and insurance prior to pregnancy were obtained from PRAMS.

Analysis

We assessed maternal and infant characteristics in the earliest years (2000-2002) and in the latest years (2013-2015) of the study period to understand whether socio-demographic changes may have also been occurring as prevalence of SSP were changing over time. Prevalence estimates were calculated based on population-based survey weights provided by PRAMS using generalized linear models for correlated data via the GLIMMIX procedure in SAS.65-67
To examine the SSP prevalence among early and late preterm infants, we calculated the weighted prevalence by GA category. Weighted multi-level linear models with the logit link function were used to compare changes in unadjusted prevalence by GA category, while accounting for correlation across years within each sampling frame, by testing the interaction between time and GA category. We next adjusted the prevalence for maternal age, education, race/ethnicity, marital status, previous live birth, insurance status before pregnancy, receipt of prenatal care in the first trimester, and plurality, based on documented associations in the literature.\textsuperscript{13,15,68}

To examine the NHB-NHW racial disparity in SSP in early preterm and late preterm infants, we used weighted multi-level linear mixed models to compare changes in prevalence by race over time by testing the interaction between time and race within each GA category, adjusting for the same covariates as above.\textsuperscript{13,16,41} We utilized robust standard errors to address correlation within states.

Results

As shown below, 50,457 (weighted N = 773,383) mothers met eligibility criteria and were included in our study cohort.

Compared to mothers in 2000-2002, NHW and NHB mothers in 2013-2015 were more likely to be 25 years or older, have more than 12 years of education, and be unmarried. NHW and NHB mothers had higher percentages of early preterm births and lower late preterm infants in 2013-2015 compared to the earlier study period. For both time periods, compared to NHW mothers, NHB mothers were younger, less education, unmarried, uninsured before pregnancy, and more likely to experience early preterm birth (Table 3.1).
Figure 3.1 Cohort selection flow diagram
Table 3.1 Characteristics of mothers of in the early (2000-2002) and late (2013-2015) study periods for non-Hispanic Black (NHB) and non-Hispanic White (NHW) subgroups

<table>
<thead>
<tr>
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<tbody>
<tr>
<td></td>
<td>Non-Hispanic White</td>
<td>Non-Hispanic Black</td>
</tr>
<tr>
<td><strong>Age (years)</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>&lt; 20</td>
<td>8322 (7.6%)</td>
<td>5581 (19.7%)</td>
</tr>
<tr>
<td>20-24</td>
<td>23616 (21.7%)</td>
<td>9456 (33.4%)</td>
</tr>
<tr>
<td>25-34</td>
<td>58007 (53.2%)</td>
<td>10571 (37.4%)</td>
</tr>
<tr>
<td>≥ 35</td>
<td>19017 (17.5%)</td>
<td>2688 (9.5%)</td>
</tr>
<tr>
<td><strong>Education (years)</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>0-8</td>
<td>1534 (1.4%)</td>
<td>427 (1.5%)</td>
</tr>
<tr>
<td>9-11</td>
<td>32003 (29.4%)</td>
<td>10174 (36.0%)</td>
</tr>
<tr>
<td>12</td>
<td>28261 (25.9%)</td>
<td>6329 (22.3%)</td>
</tr>
<tr>
<td>13-15</td>
<td>10595 (9.7%)</td>
<td>7404 (26.2%)</td>
</tr>
<tr>
<td>≥ 16</td>
<td>36570 (33.6%)</td>
<td>3962 (14.0%)</td>
</tr>
<tr>
<td><strong>Marital status</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Married</td>
<td>84136 (77.2%)</td>
<td>8365 (29.6%)</td>
</tr>
<tr>
<td>Other</td>
<td>24826 (22.8%)</td>
<td>19931 (70.4%)</td>
</tr>
<tr>
<td><strong>Previous live birth</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Yes</td>
<td>55940 (51.3%)</td>
<td>18851 (66.6%)</td>
</tr>
<tr>
<td>No</td>
<td>53021 (48.7%)</td>
<td>9445 (33.4%)</td>
</tr>
<tr>
<td><strong>Insurance status before pregnancy</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>No Insurance</td>
<td>28016 (25.8%)</td>
<td>14607 (51.6%)</td>
</tr>
<tr>
<td>Medicaid</td>
<td>1025 (0.9%)</td>
<td>1794 (6.4%)</td>
</tr>
<tr>
<td>Non-Medicaid</td>
<td>79921 (73.3%)</td>
<td>11894 (42.0%)</td>
</tr>
<tr>
<td><strong>Gestational Age</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Early preterm</td>
<td>23513 (21.6%)</td>
<td>8574 (30.3%)</td>
</tr>
<tr>
<td>Late preterm</td>
<td>85449 (78.4%)</td>
<td>19722 (69.7%)</td>
</tr>
<tr>
<td><strong>Birthweight (grams)</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>&lt; 1500</td>
<td>8012 (7.4%)</td>
<td>4361 (15.4%)</td>
</tr>
<tr>
<td>1501-2000</td>
<td>13638 (12.5%)</td>
<td>4738 (16.7%)</td>
</tr>
<tr>
<td>2001-2500</td>
<td>31233 (28.7%)</td>
<td>8854 (31.4%)</td>
</tr>
<tr>
<td>2501-3000</td>
<td>35730 (32.7%)</td>
<td>6606 (23.3%)</td>
</tr>
<tr>
<td>&gt; 3000</td>
<td>20349 (18.7%)</td>
<td>3737 (13.2%)</td>
</tr>
<tr>
<td><strong>Infant Sex</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>56469 (51.8%)</td>
<td>13744 (48.6%)</td>
</tr>
<tr>
<td>Female</td>
<td>52493 (48.2%)</td>
<td>14552 (51.4%)</td>
</tr>
</tbody>
</table>
In the unadjusted and adjusted analyses (adjusted shown), the SSP prevalence significantly improved from 2000 to 2015 for early and late preterm infants. Each additional year saw an increase of 1.53% and 1.26% for early preterm and late preterm infants respectively (p<0.0001). From 2000 to 2015, SSP improved from 56.3% to 76.1% for early preterm and from 59.4% to 81.7% for late preterm infants.

Figure 3.2 Adjusted Actual and Predicted Supine Sleep Positioning Percents and 95% Confidence Intervals for Early Preterm and Late Preterm Groups
When GA categories were stratified by race/ethnicity, adherence to SSP improved for both NHB and NHW mothers. For NHB mothers, each additional year saw an increase in SSP of 1.72% for early preterm and 1.30%, for late preterm infants. From 2000 to 2015, for NHB mothers, SSP improved from 44.5% to 60.6% for mothers of early preterm infants and from 43.7% to 59.8% for mothers of late preterm infants. For NHW mothers, each additional year saw an increase in SSP of 1.74% for the early preterm and 1.07% for late preterm groups. From 2000-2015, for NHW mothers, SSP improved from 60.8% to 85.9% for mothers of early preterm infants and from 62.9% to 82.9% for mothers of late preterm infants. (Figures 3.3A and 3.3B)

**Supine Sleep Positioning for Mothers of Early Preterm Infants**

Figure 3.3A Adjusted Actual and Predicted Supine Sleep Positioning Percents and 95% Confidence Intervals for Non-Hispanic Black and Non-Hispanic White Mothers of Early Preterm Infants
Figure 3.3B Adjusted Actual and Predicted Supine Sleep Positioning Percents and 95% Confidence Intervals for Non-Hispanic Black and Non-Hispanic White Mothers of Late Preterm Infants

From 2000 to 2015, NHB mothers had lower SSP percentages compared to NHW mothers for early preterm and late preterm groups (p< 0.0001; Figures 3A and 3B). When the adjusted rate of SSP increase was compared between NHB and NHW mothers, for the early preterm group, NHB mothers had a slower rate of improvement compared to NHW mothers (p=0.002). For the late preterm group, there was no difference in the rate of SSP increase between NHB and NHW mothers (p=0.43). (Table 3.2).
TABLE 3.2 Unadjusted and adjusted changes in supine sleep positioning among early preterm and late preterm infants for the non-Hispanic (NHB) group, relative to the non-Hispanic White (NHW) group.

<table>
<thead>
<tr>
<th></th>
<th>UNADJUSTED</th>
<th>ADJUSTED¹</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Effect</td>
<td>Estimate</td>
</tr>
<tr>
<td><strong>EARLY PRETERM</strong></td>
<td>NHB Race</td>
<td>-0.68</td>
</tr>
<tr>
<td></td>
<td>Year</td>
<td>0.12</td>
</tr>
<tr>
<td></td>
<td>Year x NHB Race</td>
<td>-0.03</td>
</tr>
<tr>
<td><strong>LATE PRETERM</strong></td>
<td>NHB Race</td>
<td>-1.00</td>
</tr>
<tr>
<td></td>
<td>Year</td>
<td>0.09</td>
</tr>
<tr>
<td></td>
<td>Year x NHB Race</td>
<td>-0.005</td>
</tr>
</tbody>
</table>

¹ Adjusted for maternal age, education, marital status, previous live birth, insurance status before pregnancy, receipt of prenatal care in the first trimester, and plurality

Discussion

In this population-based study of SSP racial disparity among preterm infants, we found that while improvements were seen for both NHW and NHB mothers of preterm infants, the NHB-NHW disparity persisted during this time-period for early preterm and late preterm infants. In addition, the rate of SSP improvement was lower for NHB mothers of early preterm infants while there was no racial difference in the late preterm group. This ongoing racial disparity seems to be in line with racial/ethnic disparity trends in infant mortality and in SUID rates that have persisted for decades among all infants. In a recently published analysis, CDC epidemiologists analyzed 1995-2013 US period-linked birth-infant death data for all GA infants to assess SUID rates by race/ethnicity. During the study period, SUID rates were consistently highest for American Indian/Alaska Natives and NHBs while lowest for Hispanics and Asian/Pacific Islanders. When stratified by GA categories, NHB infants had the highest rate of SUID for all GA groups.
Moreover, our results are also in line with a prior study of SSP among preterm and term infants which reported that less than half of all NHB infants from 2000-2011 were positioned supine compared to about two-thirds of NHW infants across GA categories. Given that all years of the study period were analyzed in aggregate for this population-based analysis, there has been a persistent lack of data on SSP trends among preterm infants stratified by race/ethnicity. In a more recently published report from the CDC of 32 states and New York City in 2015, 37.6% of NHB mothers placed their infants in the prone position to sleep compared to 16.1% of NHW mothers. This analysis, however, did not stratify infants by GA. Thus, our study findings are an important contribution to the understanding of SSP among NHB preterm infants, who bear the greatest risk for SUID.

**Practice and policy implications**

Additional efforts to understand the barriers and facilitators to SSP adherence among NHB preterm infant caregivers are needed so that novel interventions can then be developed. Hospital-based initiatives have tried to address infant sleep practices among preterm infants but none, to our knowledge, have focused on reducing racial/ethnic disparities. NICU-based quality improvement initiatives were undertaken in one unit in Texas and in all NICUs in Massachusetts to improve safe infant sleep practices among hospitalized infants. Both studies demonstrated significant improvement in compliance with safe infant sleep practices but neither collected nor analyzed results by maternal or infant race/ethnicity. Thus, while compliance for all hospitalized infants improved, it is not known whether racial/ethnic disparities were improved or worsened.

The racial/ethnic disparity in SSP and SUID among term infants has been studied for decades and in response, several public health initiatives and intervention trials were developed. However, efforts focused on racial/ethnic disparities in infant sleep practices among preterm

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babies are lacking despite their higher risk for SUID. In 2000, the National Institute for Child Health Development (NICHD) tailored their infant sleep-related work to the African-American community and developed “Babies Sleep Safest on Their Backs: A Resource Kit for Reducing the Risk for SIDS in African American Communities”. In 2012, the Maternal and Child Health Bureau (MCHB) of the Health Resources and Services Administration (HRSA) partnered with the National Institute for Children’s Health Quality (NICHQ) in developing an infant mortality reduction program utilizing the science of QI and collaborative learning to accelerate improvements in infant mortality called the Collaborative Improvement and Innovation Network to Reduce Infant Mortality (IM CoIIN). This effort was first targeted to the southern U.S. states with the largest NHB infant population, where infant mortality rates were 1.5 to 2 times higher than the national average. In 2013, six other Midwestern states joined the effort and by 2014, nearly all states and U.S. territories were participating. From September 2013 to September 2017, the effort expanded nationally, with its impact still under study. While not focused on SSP, Moon et al sought to test a health messaging intervention among African-American mothers of infants to improve sleep location and decrease bedsharing. In this study, receipt of the intervention did not impact maternal choice of infant sleep location. In fact, during the study period of 6 months, maternal belief that bedsharing increased the risk of SIDS or suffocation declined for control and intervention mothers, and did not differ by treatment assignment.

The lack of public health and intervention efforts focused on racial/ethnic disparities in infant sleep practices among preterm infants and the findings from our study of the persistent NHB-NHW disparity among preterm infants highlights the need to better understand the barriers and facilitators to safe sleep practice adherence among NHB families. To develop effective
interventions to improve safe sleep practices among NHB preterm families, a comprehensive approach to studying the individual, health system, community, and broader socioeconomic factors that influence maternal decision-making will be needed. Tremendous resources are utilized in the care of preterm infants during the first few days and weeks following their birth and rigorous randomized controlled trials and long term longitudinal follow-up studies have been undertaken to study specific medical interventions on short - and long-term health as well as developmental outcomes of preterm infants. However, adherence to health promoting behaviors after hospital discharge among preterm infant caregivers of different racial/ethnic backgrounds requires additional study. There exist modifiable caregiver behaviors that can reduce the risk for adverse outcomes such as SUID. Given the persistent significant racial disparity in SSP among preterm infants despite the prolonged hospitalization in the NICU with greater opportunities for parental education and engagement about safe infant sleep practices, NICU providers should think critically about how to engage more effectively with these families.

Limitations

There are several limitations to the study. First, our primary outcome of SSP was measured through maternal report on the PRAMS survey and mothers may have given the socially acceptable response of adherence to recommended sleep practices despite engaging in prone positioning for their infants. We hypothesize that this would bias our results towards the null with the SSP disparity seen among NHW and NHB mothers being smaller than what we found. Second, as with all retrospective survey studies, there is the potential for recall bias in maternal report of not only our primary outcome but also for the covariates included in our analyses. Finally, given that only 16 of the 41 PRAMS states were included in our study, our results may not be nationally-representative of SSP practices among preterm infants.
Despite these limitations, this study is the first, to our knowledge, to examine the trends in SSP among preterm infants and report the persistent racial/ethnic disparity of SSP in this high-risk group. Future studies are needed to understand the barriers and facilitators to SSP adherence among NHB caregivers so that resources can then be directed to developing effective interventions.

**Aim 2**

**Introduction**

Sudden unexpected infant death (SUID) is defined as “the death of an infant less than 1 year of age that occurs suddenly and unexpectedly, and whose cause is not immediately obvious prior to an investigation” and encompasses three diagnoses: sudden infant death syndrome (SIDS), accidental suffocation and strangulation in bed (ASSB), and unknown cause. SUID is the leading cause of death in infancy beyond the first month of life with approximately 3700 infants dying from SUID each year, at an overall rate reported as 91.4 per 100,000 live births. Significant racial/ethnic disparities in SUID exist, with non-Hispanic Black infants suffering the highest SUID rate in the U.S. when SUID rates are stratified by gestational age categories.

One of the most effective and modifiable parental behaviors to reduce the risk for SUID is adhering to safe infant sleep practices, including supine sleep positioning (SSP). And while the years following the Back to Sleep Campaign in 1994 initially saw a decrease in SUID for infants overall, since 2000, rates have remained stagnant. Thus, several states have identified improving safe infant sleep as a public health priority as part of the Title V Maternal and Child Health Services Block Grant Program. Title V is one of the largest federal block grant programs, providing support for nearly 54 million women, infants, and children in U.S. states and territories. Each year federal dollars are distributed to individual states by taking into account the proportion of low-income children in a state compared to the total number of low-
income children in the entire U.S. In order to receive this federal funding, states must complete comprehensive needs assessments every five years where they identify areas of priority to which resources will be directed and outcomes measured. For states that include safe infant sleep in their list of priorities, data on if and how this state-level priority-setting impacted the racial/ethnic disparity in SSP is lacking.

To address these gaps in the literature, the objective of this study was to investigate whether safe infant sleep prioritization by states differentially impacted maternal SSP report for NHW and NHB U.S.-born infants. We hypothesized that maternal SSP report improved at greater rates for NHW and NHB mothers residing in the intervention states compared to those living in control states.

**Methods**

**Data sources**

The data source for this aim was the Pregnancy Risk Assessment Monitoring System (PRAMS), a surveillance system established by the Centers for Disease Control and Prevention (CDC) and administered by state departments of public health. PRAMS was designed to monitor selected self-reported maternal behaviors and experiences among women who recently delivered a live-born infant in the prior 2-4 months, with a maximum allowable recall of 9 months postpartum. Using standardized data collection methods, monthly stratified samples were selected from recent birth certificates. Surveys were obtained from mothers using a mixed-mode data collection method with mailed questionnaires and telephone-follow-up for non-respondents. Survey data were linked to birth certificate data by the CDC and weighted for sample design, nonresponse, and noncoverage.64,78
The Title V Maternal and Child Health Block Grant Program requires state grantees to complete comprehensive needs assessments every five years. Title V funds are then used to address these priorities in the states. Two states, West Virginia and Oklahoma, specifically listed safe infant sleep, of which SSP is a critical element, in their list of ten priorities in 2010, and also contributed to PRAMS data for the majority of the study period. States that did not either directly or indirectly prioritize safe infant sleep with available PRAMS data served as the comparison group and included Arkansas and Utah. Data on selection of priorities for the 2010 needs assessment for all U.S. states and territories were obtained from Michael Kogan, PhD, Director, MCHB Office of Epidemiology and Research. States that explicitly listed safe infant sleep in their priorities were initially selected. States in the comparison group were selected if there was no explicit mention of infant sleep or the possibility of including infant sleep under broader umbrellas such as infant mortality or childhood injury prevention. In addition, among intervention and control groups, states were only selected for final analysis if PRAMS data from 2005 to 2015 were available, with the exception of missing one year of data.

Response Rates: We analyzed PRAMS state-specific data from 2005-2015 with the following response rates as reported by the CDC: 2005-2006 ≥70%, 2007-2011 ≥ 65%, 2012-2014 ≥ 60%, 2015 ≥55%.

Exclusions
We excluded: 1) surveyed women whose infants were born ≥43 weeks as such extreme post-due date delivery is no longer clinically accepted and thus these infants were likely misclassified; 2) women whose infants’ gestational age was reported as unknown; 3) mothers whose infants were delivered at home, in a clinic, in-route to a hospital, or in free-standing birth centers, in order for groups to be more comparable in their interaction with hospital systems; 4) mother of infants
who died, or were not living with their birth mother at the time of the survey; 5) mothers of infants of unknown race/ethnicity. We excluded mothers of infants who died since mothers are asked about SSP in the present tense, thus this question is not pertinent for mothers of non-survivors.

**Exposure**

Maternal residence in a state that prioritized safe infant sleep; Maternal race/ethnicity was categorized as NHW or NHB as provided by PRAMS

**Outcome**

Data on supine sleep position were obtained from PRAMS. Mothers were asked: “In which position do you usually put your infant to sleep (side, back, and/or stomach)”. Responses were then categorized as supine (back) or non-supine (which included a combination of sleep positions).

**Variables**

Maternal demographic characteristics included data obtained from birth certificates (maternal age, education, and marital status). A history of previous live birth and insurance prior to pregnancy were obtained from the survey. First trimester prenatal care use as well as method of delivery (vaginal or cesarean section), infant sex, and infant birth weight were obtained from birth certificates. Infant gestational age was defined using clinical estimates of gestational age from linked birth certificates.

**Analysis**

Using state-specific sampling weights, we used a two-stage mixed modelling approach to develop adjusted models for NHB and NHW mothers separately. In the first stage, we calculated the adjusted population prevalence of SSP with 95% confidence intervals (95% CI), for the
intervention (Oklahoma and West Virginia) and control states (Arkansas and Utah) for each year of the study period from 2005 to 2015. Given that the needs assessment occurred in 2010, this year was considered the washout year with the pre period defined as 2005-2009 and the post period defined as 2011-2015. We then compared maternal demographic and infant birth characteristics between intervention and control groups.

In the second stage, using generalized linear mixed models, we developed piecewise regression models with year as continuous variable and specified the pre/post intervention periods. We tested the 3-way interaction of year, intervention, and period to assess whether the rate of SSP change differed significantly from the pre to post periods between intervention and control groups for NHW and NHB mothers. We utilized this mixed model approach to account for correlation across years within groups in SSP.

Results

Our weighted study cohort included 499,476 and 605,076 mothers in the intervention and control groups, respectively. Compared to the pre-period, NHW mothers in the post-period for both intervention and control groups were more likely to be older, have more years of education, have a prior live birth, report first trimester prenatal care, and be insured before pregnancy. For NHB mothers, compared to the pre-period, a greater proportion in the post-period had more than 12 years of education, received prenatal care in the first trimester, and were insured before pregnancy. Compared to NHW mothers, NHB mothers in control and intervention groups during the pre and post periods were more likely to be less than 20 years of age, have less than 12 years of education, be unmarried, and lack first trimester prenatal care. (Table 3.3)
Table 3.3: Unadjusted Weighted Prevalence of Maternal and Infant Characteristics in the Pre- and Post-Periods for Non-Hispanic White (NHW) and Non-Hispanic Black (NHB) Mothers in Control and Intervention States

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<tr>
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</thead>
<tbody>
<tr>
<td>Married</td>
<td></td>
<td></td>
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<tr>
<td>Other</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Previous live birth</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Yes</td>
<td>195780 (62.7%)</td>
<td>19292 (60.9%)</td>
</tr>
<tr>
<td>No</td>
<td>116403 (37.3%)</td>
<td>12462 (39.1%)</td>
</tr>
<tr>
<td>First trimester prenatal care</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Yes</td>
<td>251035 (81.4%)</td>
<td>20390 (65.8%)</td>
</tr>
<tr>
<td>No</td>
<td>35011 (18.9%)</td>
<td>9805 (32.0%)</td>
</tr>
<tr>
<td>No PNC</td>
<td>2212 (0.7%)</td>
<td>663 (2.2%)</td>
</tr>
<tr>
<td>Insurance Before Pregnancy</td>
<td></td>
<td></td>
</tr>
<tr>
<td>None</td>
<td>91406 (39.3%)</td>
<td>19160 (60.5%)</td>
</tr>
<tr>
<td>Yes</td>
<td>220780 (70.7%)</td>
<td>12234 (39.5%)</td>
</tr>
<tr>
<td>Gestational Age</td>
<td></td>
<td></td>
</tr>
<tr>
<td>&lt; 34 weeks</td>
<td>5053 (1.6%)</td>
<td>1194 (3.9%)</td>
</tr>
<tr>
<td>34-36 weeks</td>
<td>22413 (7.4%)</td>
<td>3210 (10.1%)</td>
</tr>
<tr>
<td>&gt;36 weeks</td>
<td>284990 (93.1%)</td>
<td>27289 (86.3%)</td>
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<tr>
<td>Birthweight (g)</td>
<td></td>
<td></td>
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<tr>
<td>&lt; 1500</td>
<td>1978 (0.6%)</td>
<td>645 (2.0%)</td>
</tr>
<tr>
<td>1501-2000</td>
<td>3349 (1.1%)</td>
<td>762 (2.4%)</td>
</tr>
<tr>
<td>2001-2500</td>
<td>12627 (4.1%)</td>
<td>2481 (7.7%)</td>
</tr>
<tr>
<td>&gt;2500</td>
<td>50781 (18.1%)</td>
<td>8833 (28.0%)</td>
</tr>
<tr>
<td>Sex of infant</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>160310 (51.4%)</td>
<td>160310 (51.4%)</td>
</tr>
<tr>
<td>Female</td>
<td>151876 (48.6%)</td>
<td>151876 (48.6%)</td>
</tr>
</tbody>
</table>
From 2005 to 2015, for NHW infants, SSP improved from 62.5% and 68.6% to 82.1% and 78.9% for intervention and control states, respectively. After adjustment for maternal characteristics, there was no difference in the rate of SSP change from the pre- to post-intervention periods for both intervention (p=0.26) and control (p=0.48) groups. When the rate of SSP change was compared between the intervention and control groups from the pre- to the post-period, there was no statistically significant difference (p=0.21).

Supine Sleep Positioning (SSP) Among Non-Hispanic White Infants

Adjusted SSP Percent and 95% Confidence Intervals

Figure 3.4: Adjusted Prevalence of Supine Sleep Positioning (SSP) in Intervention and Control Groups Among Non-Hispanic White Infants

For NHB infants, SSP improved from 39.8% and 46.3% to 62.3% and 58.3% for intervention and control states, respectively. When adjusted for maternal characteristics, there was no difference in the rate of SSP improvement from the pre- to the post-period for either the intervention (p=0.80) or control (p=0.18) groups. In the comparison of the SSP rate change from pre- to post-periods between the intervention and control groups, there was no statistically significant difference (p=0.25).
Supine Sleep Positioning (SSP) Among Non-Hispanic Black Infants
Adjusted SSP Percents and 95% Confidence Intervals

Figure 3.5: Adjusted Prevalence of Supine Sleep Positioning in Intervention and Control Groups Among Non-Hispanic Black Infants

Table 3.4: Adjusted changes in supine sleep positioning among non-Hispanic (NHB) and non-Hispanic White (NHW) infants in intervention and control groups

<table>
<thead>
<tr>
<th></th>
<th>Control</th>
<th>Intervention</th>
<th>Difference-in-Difference</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Estimate</td>
<td>SE</td>
<td>P-value</td>
</tr>
<tr>
<td>NHW</td>
<td>Pre</td>
<td>0.7912</td>
<td>0.5369</td>
</tr>
<tr>
<td></td>
<td>Post</td>
<td>0.1859</td>
<td>0.664</td>
</tr>
<tr>
<td></td>
<td>Difference</td>
<td>0.6053</td>
<td>0.8539</td>
</tr>
<tr>
<td>NHB</td>
<td>Pre</td>
<td>-0.8031</td>
<td>2.4516</td>
</tr>
<tr>
<td></td>
<td>Post</td>
<td>4.509</td>
<td>3.0106</td>
</tr>
<tr>
<td></td>
<td>Difference</td>
<td>-5.3121</td>
<td>3.8825</td>
</tr>
</tbody>
</table>

1 Adjusted for maternal age, education, marital status, previous live birth, insurance status before pregnancy, receipt of prenatal care in the first trimester, and plurality
Discussion

In this analysis of the impact of states’ prioritization of safe infant sleep practices in the 2010 Title V Block Grant needs assessment on maternal adherence to SSP, we found that SSP increased both for NHB and NHW infants during the study period. However, the rate of SSP change for NHB and NHW infants in intervention and control groups was not significantly different from the pre- to post- periods. Thus, the states’ prioritization of safe infant sleep did not independently lead to increases in SSP for NHW or NHB infants. These results are in line with recent data from the CDC that reported decreasing rates of SUID among NHB infants but with very little narrowing of the disparity in SUID between NHW and NHB infants. Across the U.S., SUID and preterm-related death are the leading causes of the NHW-NHB racial disparity in infant mortality.

While the lack of impact on both infant groups is concerning given that the Title V block grant priorities drive resource allocation, the lack of additional improvement in SSP rate among NHB infants in the intervention compared to control states is particularly worrisome given their much higher risk for SUID. There are several decades of research that have identified factors associated with maternal adherence to safe infant sleep practices. Moreover, these investigators also investigated barriers and facilitators to safe sleep adherence among Black mothers and discovered that, in fact, predictors of adherence among Black mothers are not the same as for other White mothers. Namely, Black mothers were most concerned about the safety of the infant and the possibility of choking. They also reported greater likelihood of adhering to safe sleep practices if advised by a medical provider. Whether or not the Title V programs in the intervention states specifically addressed the differing factors of maternal adherence to safe sleep practice by race/ethnicity is not known.
Historically, there has been significant attention given to the need to reduce health disparities at state and national levels. Given the stark racial/ethnic disparities in SUID and unsafe sleep practices, the NICHD developed initiatives targeted to the highest risk racial/ethnic groups. In 2000, they tailored their work to the African-American community and developed “Babies Sleep Safest on Their Backs: A Resource Kit for Reducing the Risk for SIDS in African American Communities”. More recently, the National Conference of State Legislatures recently published a list of recommendations for how state policymakers could address health disparities. However, details about how interventions can address the unique needs and concerns of specific populations were lacking in this report. Furthermore, there are no studies, to our knowledge, that assessed the differential impact of these programs on adherence to safe infant sleep practices among different racial/ethnic groups.

The Title V program, the primary exposure of this analysis, recognized the importance of measuring and linking processes to outcomes and holding states accountable to their selected measures and outcomes. In 2015, under the leadership of Michael Lu, the Associate Administrator for Maternal and Child Health at HRSA, a new three-tiered performance measure framework was introduced in order to allow states to better demonstrate the impact of Title V on their selected health priorities, and included national outcomes measures, national performance measures, and evidence-based strategy measures. Prior to this new measurement framework, it was not known if the priority areas selected by states and to which resources were directed actually, demonstrated direct improvement in their process and outcome measures. It is possible that this lack of directly tying interventions to process and outcomes measures during our study period of 2005-2015 may partially explain the lack of impact of safe sleep prioritization by intervention states on increasing the SSP improvement rate compared to control states. In fact,
multiple attempts by investigators to obtain detailed information from intervention states about specific programs and policies related to states’ prioritization of safe infant sleep through the Title V program were unsuccessful. Perhaps with the establishment of the new performance measure framework, causal relationships between programs and outcomes can be better demonstrated.

There are several limitations to this work. First, given SSP was measured by maternal report, there may be recall bias with potentially an over-reporting of SSP as it may be a more socially acceptable response. We hypothesize that this may bias our results towards the null as we would be less likely to find differences in SSP. Conversely, it is also possible that this bias may be randomly distributed across the intervention and control groups and would not affect the magnitude or direction of our outcomes. Second, we lacked more granular information about the specific programs and policies that were implemented after safe infant sleep was prioritized in the Title V program by intervention states. Moreover, the control states may have also undertaken safe infant sleep-related work outside of the Title V program, which may have led to our lack of difference in the SSP rate improvement between intervention and control groups.

Despite these limitations, our study is the first, to our knowledge, that has investigated the impact of states’ prioritization of safe infant sleep on maternal adherence to infant sleep practices, stratified by racial groups. By utilizing the piecewise regression approach, we were able to undertake a natural experiment, creating intervention and control groups to isolate the effect of states’ safe sleep prioritization, despite the limitations of observational data. Overall, our findings demonstrate that while SSP improved over time, the rate of increase did not differ from intervention and control groups for both NHW and NHB infants. NHW term infants comprise the largest percentage of births in the U.S., but they have considerably lower risk for
SUID compared to NHB infants.\textsuperscript{6,22,26} The lack of improvement in SSP rate in WV and OK compared to AR and UT after safe sleep prioritization for the Title V Block Grant indicates that more targeted approaches may be needed, particularly for NHB infants who bear the highest risk.

\textbf{Aim 3}

\textbf{Introduction}

Approximately 4 million infants are born in the United States (U.S.) each year and nearly 24,000 die in their first year of life for an infant mortality rate of about 6 per 1,000 live births.\textsuperscript{1,22} Beyond the first month of life, the leading cause of infant death is due to sudden unexpected infant death (SUID). SUID typically encompasses three diagnoses: sudden infant death syndrome (SIDS), accidental suffocation and strangulation in bed (ASSB), and unknown.\textsuperscript{23} The distinction across these three diagnoses is often not clear\textsuperscript{24,25} and thus most current literature related to post-neonatal infant mortality, use the overarching term of SUID in referring to deaths attributed to suffocation due to unsafe sleep practices or which ultimately are not known even after thorough investigation.\textsuperscript{23} There are approximately 3700 SUIDs in the U.S. each year.\textsuperscript{22} While the 1990’s saw a dramatic decline in SIDS due to the Back-to-Sleep Campaign, there has been little improvement in SUID rates since 2000.\textsuperscript{26} Moreover, preterm infants (< 37 weeks gestational age) are at greater risk for SUID than their full term counterparts. Compared to infants born at 39-41 weeks, preterm infants have a 2 to 4-fold greater incidence of SUID.\textsuperscript{7} The majority of the epidemiologic, prospective cohort, and intervention studies of infant sleep practices have focused on the healthy full term population, overlooking the unique risk profile of preterm infants and the need to better understand the relationship between state programs and policies related to SUID-reduction and adherence to safe sleep practices in the full term and preterm populations.
Starting in 2011, Massachusetts (MA) implemented a multifaceted safe infant sleep program, including the development of a state infant sleep policy by the department of public health, trainings for several public health organizations, and a widespread public health messaging campaign. In contrast, Maine (ME) and VT (VT), neighboring New England states, took a much more limited approach with some education training and public messaging.

Thus, the objective of this study was to assess the impact of the MA safe infant sleep programs on maternal adherence to supine sleep positioning (SSP) compared to ME and VT, which did not undertake a comprehensive safe infant sleep intervention approach. We hypothesized that compared to ME and VT, MA would have greater improvement in SSP for full term and preterm infants in the years following their multi-faceted effort to improve adherence to safe sleep practices.

Methods

Data source

The data source for this aim was the Pregnancy Risk Assessment Monitoring System (PRAMS), a surveillance system established by the Centers for Disease Control and Prevention (CDC) and administered by state departments of public health. PRAMS was designed to monitor selected self-reported maternal behaviors and experiences among women who recently delivered a live-born infant in the prior 2-4 months, with a maximum allowable recall of 9 months postpartum. Using standardized data collection methods, monthly stratified samples were selected from recent birth certificates. Surveys were obtained from mothers using a mixed-mode data collection method with mailed questionnaires and telephone-follow-up for non-respondents. Survey data were linked to birth certificate data and weighted for sample design, nonresponse, and noncoverage.64
PRAMS data for MA, ME, and VT were the focus for this study. While ME has been collecting PRAMS data since the inception of this surveillance system, VT and MA joined the PRAMS effort in 2001 and 2007, respectively. This study included PRAMS data from these states from 2007-2015 with the following response rates, as set by the CDC: 2007-2011 ≥ 65%, 2012-2014 ≥ 60%, and 2015 ≥55%.

Exclusions: We excluded: 1) surveyed women whose infants were born ≥43 weeks as such extreme post-due date delivery is no longer clinically accepted and thus these infants were likely misclassified; 2) women whose infants’ gestational age was reported as unknown; 3) mothers whose infants were delivered at home, in a clinic, in-route to a hospital, or in free-standing birth centers, in order for the preterm and full term groups to be more comparable in their interaction with hospital systems; 4) mother of infants who died, or were not living with their birth mother at the time of the survey; 5) mothers of infants of unknown race/ethnicity. We excluded mothers of infants who died since mothers are asked about SSP in the present tense, thus this question is not pertinent for mothers of non-survivors.

Variables

Using clinical estimates of gestational age from linked birth certificates, gestational age was categorized into preterm (< 37 weeks) or term (≥ 37 weeks).

Maternal demographic characteristics included data obtained from birth certificates (maternal age, education, race/ethnicity, and marital status). Maternal race/ethnicity was categorized as non-Hispanic white, non-Hispanic black, Hispanic, and other. A history of previous live birth and insurance prior to pregnancy were obtained from the survey.

Data on supine sleep position were obtained from PRAMS. Mothers were asked: “In which position do you usually put your infant to sleep (side, back, and/or stomach)”. Responses
were then categorized as supine (back) or non-supine (which included a combination of sleep positions). First trimester prenatal care use as well as method of delivery (vaginal or cesarean section), infant sex, and infant birth weight were obtained from birth certificates.

**Interventions**

MA, ME, and VT undertook different approaches to improving infant sleep practices during the study period, allowing for natural experimental analyses to measure the effect of states’ policies and practices to changes in supine sleep positioning by state. Starting in 2011, MA undertook a multifaceted effort to improve safe infant sleep practices. Some components of the safe sleep work are listed below.

In ME, the safe sleep effort began in March of 2010 with a presentation to the state Perinatal Nurse Managers of ME group, a voluntary collaborative group of the nurse leaders of birthing hospitals in ME. At that time, birthing hospitals were encouraged to use the NICHD safe sleep educational materials. In addition, an educational video created by safe sleep advocates in ME and the First Candle safe sleep video were distributed to the birthing hospitals over the course of 2010. Since 2012, educational lectures on safe infant sleep were given by safe sleep experts at hospitals, community partner agencies, ME Families visiting program, Public Health Nursing, and Office of Child and Family services. The public awareness campaign and legislative focus on infant sleep was not included in Maine’s efforts.

In more recent years, ME has become a Cribs for Kids state, with ME Medical Center being the state host for this chapter. ME’s network of home visiting programs has provided crib distribution and safe sleep education to families upon referral, and expanding to include participants of the Child Abuse Prevention Councils (in all of Maine’s 16 counties), and any
birthing facility that wants to distribute directly to families, as long as safe sleep education is also provided at the time of portable crib delivery.

Table 3.5 Safe sleep program and policies implemented by Massachusetts

<table>
<thead>
<tr>
<th>Safe Sleep Work by Massachusetts</th>
</tr>
</thead>
<tbody>
<tr>
<td>Developed, implemented and disseminated a MA Department of Public Health (MDPH) Safe Sleep policy to align with the 2011 American Academy of Pediatrics (AAP) guidelines for infant safe sleep practices.</td>
</tr>
<tr>
<td>Promoted the adoption of an infant safe sleep policy, consistent with the AAP and the MDPH, among several MA state agencies that are involved in the care of infants such as the Department of Child and Families, Department of Early, Education and Care, Office of Child Advocate, State Child Fatality Review Team, Department of Housing and Community Health Development and The Children Trust.</td>
</tr>
<tr>
<td>Developed and implemented, in collaboration with the Women, Infant, and Children (WIC) Program, new messaging, materials and a train-the-trainer presentation, which were disseminated to all WIC statewide programs.</td>
</tr>
<tr>
<td>Collaborated with the MA Department of Early Education and Care’s Community Family Engagement program to provide education and training on infant safe sleep in November 2012. In 2014, assisted with the development and the dissemination of Request for Qualifications for trainers to become approved safe sleep trainers across the Commonwealth.</td>
</tr>
<tr>
<td>Trained home visiting nurses from the Welcome Family Program, a branch of the state home visiting program, on safe sleep practices in August and September 2014.</td>
</tr>
<tr>
<td>Trained 18 Early Intervention (EI) senior staff on infant sleep practices in November 2014, who in return trained over 1400 Early Intervention staff members, as well as provided information and resources to over 30,000 families enrolled in EI to improve safety and prevention of infant fatalities.</td>
</tr>
<tr>
<td>In 2014, MA Governor’s office and the Executive Office of Health and Human Services convened a task force with membership from multiple state agencies, leading to the following activities:</td>
</tr>
<tr>
<td>1. Public Awareness: posters on public transportation, billboards</td>
</tr>
<tr>
<td>3. Resources for Physicians: Partnering with the MA Chapter of the American Academy of Pediatrics and Massachusetts Hospital Association</td>
</tr>
<tr>
<td>In October 2014, The Governor of the Commonwealth of MA issued a proclamation and October became Infant Sleep Awareness Month. Website.</td>
</tr>
</tbody>
</table>
In contrast, in VT, the safe infant sleep efforts have only recently gotten underway. For instance, in Spring 2018, the VT Department of Public Health began a public messaging effort focused on infant safe sleep for health care providers, community organizations, and parents.

**Analysis**

We used a two-stage mixed modelling approach to develop adjusted models for full term and preterm groups separately. In the first stage, using state-specific sampling weights, we calculated the adjusted population prevalence of SSP with 95% confidence intervals (95% CI), for intervention (MA) and control (ME and VT) groups for each year of the study period of 2007-2015, controlling for maternal sociodemographic factors: maternal age, education, marital status, previous live birth, insurance status before pregnancy, and plurality. Given that MA initiated their multifaceted safe infant sleep work in 2011, this year was considered the washout year. The pre-period was defined as 2007 to 2010 and the post-period was defined as 2012-2015. We compared maternal demographic and infant birth characteristics between intervention and control groups in the pre- and post- periods.

In the second stage, using generalized linear mixed models, we developed piecewise regression models with year as continuous variable and specified the pre/post intervention periods. We tested the 3-way interaction of year, intervention, and period to assess whether the rate of SSP change differed significantly from the pre to post periods between intervention and control groups. We utilized this mixed model approach to account for correlation across years within groups in SSP.

**Results**

Our weighted study cohort included 513,317 mothers from MA and 121,865 mothers from ME and VT. Compared to the pre-period, a greater proportion of MA mothers were older,
non-White, of higher education, married, and with a previous live birth. A greater proportion were also more likely to have first trimester prenatal care, be insured before pregnancy, and deliver a preterm infant. For mothers from ME and VT, compared to the pre-period, a greater proportion in the post-period were older, unmarried, have higher education, and with a previous live birth. In the post-period, ME and VT mothers were more likely to have received prenatal care in the first trimester and be insured before pregnancy, but less likely to deliver a preterm infant. (Table 3.6)

From 2007 to 2015, for full term infants, SSP improved from 77.4% and 81.8% to 86.7% and 84.8% for intervention and control states, respectively. (Figure 3.6) For preterm infants, SSP improved from 58.1% and 78.6% to 85.5% and 91.1% for intervention and control states, respectively. (Figure 3.7) In the adjusted models, there was no difference in the rate of SSP change between intervention and control groups from pre to post periods for either full term or preterm groups (p=0.36 and p=0.10, respectively). (Table 3.7)
Table 3.6: Cohort Characteristics of the Control and Intervention Groups in the Pre- and Post-Intervention Periods

<table>
<thead>
<tr>
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<th></th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>MA Intervention</td>
<td>MA Control</td>
</tr>
<tr>
<td><strong>Age (years)</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>&lt; 20</td>
<td>15619 (5.9%)</td>
<td>5798 (8.1%)</td>
</tr>
<tr>
<td>20-24</td>
<td>44888 (16.9%)</td>
<td>17200 (24.0%)</td>
</tr>
<tr>
<td>25-34</td>
<td>148919 (56.1%)</td>
<td>38279 (53.4%)</td>
</tr>
<tr>
<td>≥ 35</td>
<td>55937 (21.1%)</td>
<td>10345 (14.4%)</td>
</tr>
<tr>
<td><strong>Race/Ethnicity</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Non-Hispanic White</td>
<td>180576 (68.0%)</td>
<td>69194 (96.6%)</td>
</tr>
<tr>
<td>Non-Hispanic Black</td>
<td>22218 (8.4%)</td>
<td>676 (0.9%)</td>
</tr>
<tr>
<td>Hispanic</td>
<td>37525 (14.1%)</td>
<td>615 (0.9%)</td>
</tr>
<tr>
<td>Other</td>
<td>23043 (9.4%)</td>
<td>1136 (1.6%)</td>
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<tr>
<td><strong>Education (years)</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>0-8</td>
<td>7282 (2.7%)</td>
<td>651 (0.9%)</td>
</tr>
<tr>
<td>9-11</td>
<td>19017 (7.2%)</td>
<td>5883 (8.2%)</td>
</tr>
<tr>
<td>12</td>
<td>69537 (26.2%)</td>
<td>22876 (31.9%)</td>
</tr>
<tr>
<td>13-15</td>
<td>49538 (18.7%)</td>
<td>17648 (24.6%)</td>
</tr>
<tr>
<td>≥ 16</td>
<td>119889 (45.2%)</td>
<td>24563 (34.3%)</td>
</tr>
<tr>
<td><strong>Marital status</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Married</td>
<td>172489 (65.0%)</td>
<td>42678 (59.6%)</td>
</tr>
<tr>
<td>Other</td>
<td>92874 (35.0%)</td>
<td>28943 (40.4%)</td>
</tr>
<tr>
<td><strong>Previous live birth</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>No</td>
<td>131115 (49.4%)</td>
<td>33873 (47.3%)</td>
</tr>
<tr>
<td>Yes</td>
<td>134187 (50.6%)</td>
<td>37747 (52.7%)</td>
</tr>
<tr>
<td><strong>First trimester prenatal care</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Yes</td>
<td>233970 (89.5%)</td>
<td>61434 (86.9%)</td>
</tr>
<tr>
<td>No</td>
<td>25827 (9.9%)</td>
<td>8938 (12.6%)</td>
</tr>
<tr>
<td>No PNC</td>
<td>1554 (0.6%)</td>
<td>340 (0.5%)</td>
</tr>
<tr>
<td><strong>Insurance Before Pregnancy</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Yes</td>
<td>225943 (85.1%)</td>
<td>52727 (73.6%)</td>
</tr>
<tr>
<td><strong>Gestational Age</strong></td>
<td></td>
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</tr>
<tr>
<td>Preterm</td>
<td>17357 (6.5%)</td>
<td>5119 (7.3%)</td>
</tr>
<tr>
<td>Full Term</td>
<td>248005 (93.5%)</td>
<td>66421 (92.7%)</td>
</tr>
<tr>
<td><strong>Birthweight (grams)</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>&lt; 1500</td>
<td>2671 (1.0%)</td>
<td>435 (0.6%)</td>
</tr>
<tr>
<td>1501-2000</td>
<td>3208 (1.2%)</td>
<td>784 (1.1%)</td>
</tr>
<tr>
<td>2001-2500</td>
<td>11740 (4.4%)</td>
<td>2656 (3.7%)</td>
</tr>
<tr>
<td>2501-3000</td>
<td>42508 (16.0%)</td>
<td>10561 (14.7%)</td>
</tr>
<tr>
<td>≥ 3000</td>
<td>205236 (77.3%)</td>
<td>57184 (79.8%)</td>
</tr>
<tr>
<td><strong>Gender</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>136863 (51.6%)</td>
<td>37009 (51.7%)</td>
</tr>
<tr>
<td>Female</td>
<td>128500 (48.4%)</td>
<td>34611 (48.3%)</td>
</tr>
</tbody>
</table>
Supine Sleep Positioning (SSP) Among Full Term Infants in Intervention (MA) and Control (ME and VT) States

![Graph showing adjusted prevalence percents and 95% confidence intervals of supine sleep positioning among full term infants in intervention (MA) and control (ME and VT) states.]

Figure 3.6: Adjusted Prevalence Percents and 95% Confidence Intervals of Supine Sleep Positioning Among Full Term Infants in Intervention (MA) and Control (ME and VT) States

Supine Sleep Positioning (SSP) Among Preterm Infants in Intervention (MA) and Control (ME and VT) States

![Graph showing adjusted prevalence percents and 95% confidence intervals of supine sleep positioning among preterm infants in intervention (MA) and control (ME and VT) states.]

Figure 3.7: Adjusted Prevalence Percents and 95% Confidence Intervals of Supine Sleep Positioning Among Preterm Infants in Intervention (MA) and Control (ME and VT) States
Table 3.7: Adjusted changes in supine sleep positioning among full term and preterm infants in intervention (MA) and control (ME and VT) groups

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1 Adjusted for maternal age, education, marital status, previous live birth, insurance status before pregnancy, receipt of prenatal care in the first trimester, and plurality

Discussion

In this population-based difference-in-difference analysis of the impact of MA’s comprehensive approach to improving safe infant sleep practices, we found no difference in the rate of SSP change for full term and preterm infants between intervention and control states. We had initially hypothesized that MA full term and preterm infants would experience greater SSP improvement compared to ME and VT and were surprised to see that with our causal inference approach, neither infant group were directly impacted by the MA safe sleep effort. We speculate that this lack of difference may be due to relatively high SSP percents in both the intervention and control groups, making differences in rate of SSP change more difficult to detect between these groups. Prior CDC analyses demonstrated that SSP compliance ranged from 62.4% to 83.9% for U.S. mothers surveyed in 2015. While this CDC analysis did not stratify infants by preterm status, these published 2015 estimates are generally lower than our SSP percentages.

The lack of difference seen in the preterm intervention was particularly disappointing. Full term infants comprise over 90% of births in MA and represent the majority of infants who
suffer SUID, but preterm infants are over-represented in SUIDs in MA. From 2011-2014, preterm infants comprised 8.9% of all births in MA but represented 26.7% of SUIDs in the state. Similarly for low birth weight infants, while they comprised 7.5% of births in the states, they made up 26.7% of SUID cases. While the number of full term infants affected by SUID is high, the relative contribution by preterm infants to SUID remain significant. Thus the lack of relative improvement in SSP among MA preterm infants when compared to ME and VT requires greater attention. Additional research on the factors that influence caregiver adherence to safe infant sleep practices for preterm infants is needed so that effective interventions can then be developed. For the full term population, several population-based studies such as the National Infant Sleep Position Study and the Study of Attitude and Factors Effecting Infant care Practices have demonstrated that maternal perceptions of safety, fear of choking, and trust in medical providers’ counseling related to safe infant sleep are significantly associated with adherence. Such data for preterm infants is lacking and thus, a recently funded national study will seek to identify factors associated with maternal adherence to recommended infant care practices among preterm infants (Study of Factors Effecting PREterm infant care Practices – SAFE PREP; 1R01HD095060-01).

In MA, starting in 2015, neonatal intensive care units (NICUs) across the state recognized that safe sleep practices were not being modeled during the preterm infants’ prolonged hospital stay. While caregivers may have been given verbal discharge education about safe infant sleep, these practices were not being demonstrated on a regular basis in the NICU. To address this deficit in care, the MA NICU Safe Sleep Collaborative was formed and sought to standardize the sleep care of infants in the NICU. Included in the initiative, were guidelines about which infants would be eligible to engage in safe sleep practices, intervention
toolkits with bedside crib card denoting sleep designation based on medical condition, education of all NICU providers about prematurity and SUID, crib audits with data entry into a centralized database, and regularly scheduled webinars and forums to share best practices.43,44 Every NICU participated in this quality improvement collaborative and compliance with safe sleep practices during hospitalization improved from 47% in 2015 to 75% in 2017. It is not known if these hospital-based efforts led to improved adherence in the home after discharge and thus additional efforts including ongoing PRAMS SSP analyses are needed to assess post-discharge caregiver compliance with recommended infant sleep practices for preterm infants.

Reducing infant mortality has been a priority at state and national levels for decades. Given that two-thirds of infant deaths occur in the first month of life, mainly attributed to prematurity, a significant amount of attention has been given to strategies to reduce preterm birth and low birthweight. However, both broad public health and clinical efforts have only been successful in initially reducing the birth of late preterm infants from 7.51% in 2007, the highest rate in decades, to 6.82% by 20141, and in fact, there was an increase in late preterm births in 2015 and 2016.8,57 The rates of very preterm infants and extremely low birth weight (ELBW; < 1000 grams) and very low birth weight (VLBW; < 1500 grams) births have not improved to the same extent.1 In addition, since 2000, post-neonatal death due to SUID, the leading cause of infant death after the first month of life, has been stagnant.23 Recognizing that reducing SUID rates through maternal behavior change in infant care practices may be a more winnable shorter-term battle compared to preterm birth with its broad array of social and medical risk factors, several national public health efforts have been developed. However, the public health messaging has been targeted to caregivers of healthy full term infants, and this approach may not
resonate with families of preterm infants who have had very different postnatal experiences compared to their full term counterparts.

There are several limitations to this study. First, the PRAMS data is primarily obtained from maternal surveys and thus recall bias and pressure to give socially acceptable answers related to infant care practices, such as adherence to SSP, may have introduced bias. Second, we did not have data related to the presence of other caregivers, interactions with medical providers, and maternal sources of educational information about infant sleep practices. We know from prior studies that there is significant variability in who mothers trust about infant care advice and from where they receive such education. Finally, in our statistical approach of two stage modeling, we may have lacked the power to detect significant differences in SSP rate change over time given that SSP rates were generally high for all groups.

Despite these limitations, to our knowledge, this is the first study to assess the impact of state infant sleep programs and policies on compliance with safe sleep practices stratified by full term and preterm status. This distinction is an important one given the prolonged hospitalization of preterm infants compared to full term infants and the vastly different parental experiences of preterm infant care. These differences should be taken into consideration when interventions are developed.

In conclusion, by utilizing the piecewise regression approach for intervention and control groups, we were able to undertake a natural experiment to isolate the effect of a state’s multifaceted approach to improving safe sleep practices, despite the limitations of observational data. Overall, our findings demonstrate that compared to control states, MA infants did not experience a greater rate of SSP change in the post-intervention period. The lack of improvement in SSP among preterm infants in MA compared to ME and VT was particularly
notable given the higher risk for SUID in this group, indicating that targeted approaches tailored to the needs of preterm infants may be needed.
CHAPTER IV

DISCUSSION AND CONCLUSIONS

Summary of findings

Across the 3 aims of this thesis, we demonstrate that while SSP has increased for preterm and term infants from 2000 to 2015, the rate of improvement varied by gestational age category and by race/ethnicity. Moreover, state-level public health interventions focused on safe infant sleep did not directly lead to improved SSP rates compared to control states. NHB and preterm infants have the highest SUID risk but their SSP rate changes over the study period did not differ from NHW and full term infants. These findings highlight how public health interventions may need to be more targeted to specific high risk groups to ensure optimal impact.

Aims 1 and 3: Prematurity and supine sleep positioning

Among preterm infants, our finding in aim 1 that SSP improved more rapidly for early preterm infants compared to late preterm infants was surprising given that prior studies, albeit few in number, reported the youngest gestational age preterm infants had lower rates of SSP compared to older infants. One of the few studies that has investigated SSP among preterm and term infants analyzed PRAMS data from 2000 to 2011 and found that the prevalence of SSP declined with decreasing gestational age such that 59.7% of infants ≤ 27 weeks were placed in SSP compared to 63.6% of late preterm infants and 66.8% of term infants. This study, however, did not investigate trends over time in SSP by gestational age and thus while the aggregate prevalence of SSP may have been lower in the early preterm group, it may have been possible when analyzed over the study period, as done in our current work, there could have been a differential rise in SSP with higher rates in the more recent years.
We find the higher rate of SSP increase among early preterm infants to be encouraging given that the smallest and youngest newborns have the highest risk for SUID, nearly three to four times the risk compared to 39-40 week gestational age infants. Investigators from the CDC published a groundbreaking manuscript in 2017 that assessed SUIDS from 1995 to 2013, with particular focus on racial/ethnic disparities as well as varying risk profiles of preterm and term infants. They found that preterm SUID rates dropped significantly between 1995-1997 and 2011-2013 for all races/ethnicities except for Asian Pacific Islander. While we cannot explicitly state that this SUID reduction in preterm infants was caused by the improvements in SSP that we saw in our work, we can conclude that SSP improvement is associated with SUID reduction given that non-supine sleep positioning has been shown to be a significant risk factor for SUID.

In terms of understanding the factors associated with this higher rate of SSP increase among early preterm infants, we hypothesize that changes in hospital-based infant sleep practices may have improved parental adherence to SSP after discharge home. Recognizing that safe sleep practices are not routinely practiced in the NICU for preterm infants, several birthing hospitals throughout the U.S. have implemented quality improvement projects focused on modeling appropriate infant sleep behaviors during the prolonged hospital stays of preterm infants, with the most premature infants having the greatest period of exposure given their prolonged birth hospitalization. Gelfer, et al.’s NICU-based quality improvement effort developed a comprehensive safe sleep educational program for their nurses and parents including a post-discharge telephone reminder for families. This NICU team from Houston, Texas improved SSP from 39% to 83% (P < .001) as well as other recommended sleep practices during the study period of May to November 2010. In Massachusetts, clinicians and maternal and child
health public health representatives collaborated to integrate safe sleep practices into the care of preterm infants in every level III NICU in the state. First initiated in two level III NICUs in MA, the effort expanded to all ten MA NICUs and demonstrated significant improvement in overall safe sleep compliance which was defined for this quality improvement effort as SSP, flat head-of-bed positioning, absence of blankets/dolls, and positioning devices. Compliance increased from 47.7% to 81.0% during the study period.

We hypothesize that these NICU-based efforts may have engaged and educated parents about safe infant sleep practices and led to improved adherence after hospital discharge in the preterm population. In the work by Gelfer, et al., the percent of former NICU parents who were full compliant with safe infant sleep practices increased from 23% to 83% in the post-discharge period. The MA investigators did not assess post-discharge adherence. Walcott, et al. from the Division of Health Protection and Safety at the Georgia Department of Public Health surveyed primary caretakers of all infants born in Georgia from August to October 2016 one month after birth hospital discharge. Their adjusted analyses showed that receipt of safe sleep information from hospitals was significantly associated with safe sleep behaviors. While this study did not focus on preterm infants, it may lend support to the idea that hospital-based efforts may influence home care behaviors. Given the retrospective design of this study along with the very low response rate of 420 respondents out of 20,500 mailed surveys, generalizability is extremely limited.

In contrast, a recently published randomized clinical trial investigating the effect of nursing quality improvement and mobile health interventions on infant sleep practices demonstrated that only the mobile health intervention but not the hospital-based nursing quality improvement intervention increased maternal adherence to safe sleep practices when compared
to the control group. As was the case with the study from Georgia, the focus on this clinical trial was not on preterm infants. The nursing quality improvement interventions sought to standardize infant sleep-related messages that mothers received during birth hospitalization through verbal education and demonstration through modeling of sleep practices. The mobile health interventions included maternal receipt of messages and videos related to safe sleep on a daily basis for the first 11 days after discharge and then every 3 to 4 days for 60 days. The efficacy of the mobile health intervention in combination with the nursing quality improvement intervention, but not the nursing quality improvement intervention alone indicates that ongoing support beyond the hospital stay through novel approaches may have greater impact. Indeed, this randomized clinical trial demonstrated much higher rates of improvement in SSP (92.5%) than prior intervention studies. Despite these remarkable results, it is unclear whether this same method would be as effective for the preterm population given the very different medical problems of preterm infants and also the greater social-emotional and financial distress experienced by parents of preterm infants.

The barriers and facilitators to parental adherence to recommended infant sleep practices among preterm infants are likely very different than those for parents of term infants. Numerous quantitative and qualitative observational studies have reported that parents of preterm infants experience greater anxiety and trauma and may feel less engaged with their infants. But whether these adverse experiences impact parental adherence to safe infant sleep practices is not known. The ways in which we engage with parents of preterm infants during and after the birth hospitalization may need to be tailored to meet their specific needs. As outlined in the following section on future research directions, we first need to better understand the barriers and
facilitators to parental adherence to recommended infant sleep practices among preterm infants so that effective targeted interventions for this high risk group can be developed.

**Aims 1 and 2: The NHB-NHW racial disparity in supine sleep positioning**

Our works demonstrated that while SSP improved for all gestational age groups and NHB and NHW infants, there was significant variation across racial groups. Moreover, despite improvements in SSP, the NHB-NHW racial disparity remained significant with little narrowing from 2000 to 2015 with NHB infant having lower rates of SSP compared to NHW infants for early preterm and late preterm categories. This lack of disparity reduction even in the early preterm population who spends months hospitalized in NICUs highlights the unintended consequences of hospital-based quality improvement efforts. Despite what we perceive to be tremendous opportunities for parental engagement about safe infant sleep practices during a prolonged birth hospitalization, the education may not be delivered and/or accepted in equitable ways among NHB and NHW families. Most hospital-level and even state-level perinatal quality improvement efforts focused on improving the health of mothers and infants rarely consider the diversity of their patient population during the development of their key driver diagrams, intervention toolkits and project database. With respect to infant sleep practices among preterm infants, despite the several published quality improvement studies\(^{42-44}\) which improved SSP during birth hospitalization, it is not known if these rates of improvement varied by race/ethnicity or other maternal/infant sociodemographic characteristics. Thus, it is possible that the NHB-NHW racial disparity may have worsened during these study periods.

Most recently, Parker, et al. demonstrated this unintended consequence of continued racial/ethnic disparities in their quality improvement initiative to improve use of mother’s milk among very low birth weight infants.\(^{102}\) These Massachusetts hospitals quality improvement
teams sought to increase use of mother’s own milk among all very low birth weight infants in Massachusetts through participation in their state’s Neonatal Quality Improvement Collaborative. These investigators and clinicians developed a bundle of interventions to promote use of mother’s milk in 9 Massachusetts NICUs, including human milk prenatal education, first milk expression <6 hours after delivery, lactation consultation < 24 hours, and receipt of skin-to-skin care. In their cohort of over 1300 mother-infant pairs, they found that while initiation of mother’s milk was similar across racial/ethnic groups, infants of Hispanic mothers and NHB mothers stopped receiving milk earlier in the hospitalization compared to NHW mothers. In addition, Hispanic mothers were less likely to provide skin-to-skin care at less than one month of hospitalization compared to NHW mothers. Based upon these results, these researchers are now gathering more data on specific hospital-based practices that may have contributed to this racial/ethnic persistent disparity in provision of human milk.

The approach taken by Parker, et al. is also needed to understand the impact of safe sleep work in NICUs among preterm infants. While knowledge about the barriers and facilitators to parental adherence to safe infant sleep practices is lacking, there has been significant work done in the full-term population in both observational and intervention studies. Through several qualitative studies, Moon, et al. demonstrated that African-American parents have a low degree of self-efficacy related to reducing the risk for sudden infant death syndrome (SIDS) in their infants but a high degree of self-efficacy around preventing death due to suffocation. In these studies, African-American parents seemed not to make the connection between suffocation and SIDS. Based upon this observational data, investigators then performed a randomized controlled trial, specifically targeting African-American mothers of infants during birth hospitalization, focusing on suffocation prevention messaging versus the standard approach of
providing education on SIDS risk reduction to reduce parental report of bed sharing. Despite the evidence-informed approach to the development of the intervention, this randomized clinical trial showed no difference in African-American mothers likelihood to bedshare between those who received the enhanced educational messages compared to those who received the standard infant sleep education. Interestingly, the authors discovered that mothers in both the intervention and control groups were more likely to bedshare to prevent SIDS, believing that their vigilance during sleep would offer greater protection against suffocation. These results seem to indicate that we do not yet have a comprehensive understanding of the factors that influence parental decision-making related to infant sleep practices among African-American mothers, and in addition, that hospital-based interventions may not be enough.

The lack of reduction in racial/ethnic disparity in SSP among preterm infants despite prolonged hospitalization with many opportunities for engagement between parents and hospital providers is in line with a growing body of literature focused on racial/ethnic disparities in neonatal outcomes among preterm infants. While the precise mechanisms for these disparities are still being elucidated, studies point to both individual and broader hospital or systems-level factors. The California Perinatal Quality Care Collaborative (CPQCC) is a statewide network for California NICUs that adopt quality improvement methods to improve the health of preterm infants in the state and collect data on process and outcome measures. In a recently published analysis of their data, Profit, et al demonstrated significant racial/ethnic variation in the quality of care between NICUs as well as within NICUs in California. By using a validated measurement tool of neonatal quality, the Baby-MONITOR which aggregates 9 risk-adjusted measures, investigators demonstrated significant racial disparities in key outcomes
such as provision of human milk, hypothermia, receipt of antenatal steroids, and chronic lung
disease.

Elizabeth Howell and colleagues have contributed significantly to the body of literature
related to broader structural factors which may be contributing to racial/ethnic disparities in
health outcomes of preterm infants.\textsuperscript{111-117} In a population-based cohort study of all live births
and deaths of very low birth weight (VLBW; <1500 grams) infants weighing 500 to 1499 grams
born in New York City hospitals between 1996-2001, Howell, et al. found that NHB very low
birthweight infants were more likely to be born in hospitals with higher risk-adjusted neonatal
mortality rates compared to white very low birthweight infants.\textsuperscript{118} This study highlighted two
important points: 1) neonatal outcomes vary by NICU even after adjusting for case mix; and 2)
NHB infants are born in hospitals with worse outcomes. Howell replicated this study with more
recent data from 2010-2014 and demonstrated similar results, with NHB and Hispanic preterm
infants being more likely to be born at hospitals with higher risk-adjusted neonatal morbidity and
mortality rates.\textsuperscript{114}

The persistent racial/ethnic disparity in SSP among preterm infants who have prolonged
birth hospitalizations may be due to unit-level factors such as the unit’s prioritization of safe
sleep education or the degree to which NICU providers engage equitably with parents in
culturally sensitive ways so that consistent safe sleep messages are communicated and
subsequently understood. The impact of parental engagement in the NICU on short and long
term infant and family outcomes is actively being studied with some research indicating racial
differences in parental satisfaction in the NICU. For instance, Martin et al. undertook a
prospective cohort study of families of preterm infants presenting to primary care clinics and
surveyed them about satisfaction with the nursing care they received in the NICU.\textsuperscript{97} Among the
249 parents surveyed, 57% of comments were positive with NHB parents reporting more negative feelings compared to NHW parents. NHB parents were most dissatisfied with the lack of support, compassion, and respectful communication from nurses. In contrast, NHW parents were most dissatisfied with inconsistent nursing care and lack of education about their infants. It is nearly impossible to determine if these aspects of neonatal nursing care were truly deficient or perceived as being inadequate. Regardless, the neonatal community is becoming more aware of the impact that positive parental engagement can have on infant health outcomes and thus the parental perception of satisfaction is a key component of optimizing infant health. In the recently published randomized clinical trial of the Family-Integrated Care model, infants in the intervention arm demonstrated better infant weight gain, exclusive breastfeeding, and lower parental stress and anxiety scores. To date, it is not known if parental engagement in the NICU impacts adherence to health promoting behaviors such as safe infant sleep practices after hospital discharge. It is critical to understand if and how parental engagement influences parental behavior so that targeted interventions can then be developed. Furthermore, the mediating effect of parental engagement on infant health should be studied for specific racial/ethnic groups to address the unique barriers encountered by the highest risk groups.

Implications for practice and policy

State policies and programs related to safe infant sleep

Recognizing that a broader public health approach was needed to reduce SUID by improving adherence to safe sleep practices, several states prioritized infant sleep a priority in their Title V Maternal and Child Health Bureau Block Grant Program (West Virginia and Oklahoma), and also developed new programs aimed at improving safe infant sleep practices (Massachusetts). Our in-depth analyses was the first, to our knowledge, that investigated the
impact of states’ prioritization of safe infant sleep on maternal adherence to infant sleep practices by race/ethnicity and by preterm status of which supine sleep positioning was the focus here. By utilizing the segmented regression approach, we were able to undertake a natural experiment, creating intervention and control groups for aims 2 and 3 to isolate the effect of states’ safe sleep prioritization, despite the limitations of observational data. Overall, our findings demonstrated that intervention states did not experience a greater SSP improvement rate compared to control states. Despite preterm and NHB infants having the highest SUID risk, the interventions states’ safe infant sleep programs and policies did not lead to SSP improvement in these groups. As previously discussed, this again highlights the need for understanding the barriers and facilitators to SSP adherence among high risk groups because these factors may not be the same as for healthier full-term infants. The persistent racial/ethnic disparity indicates that targeted approaches tailored to the needs of NHB infants and their caregivers will be necessary to increase SSP.

As highlighted in “Perinatal legislative policies and health outcomes” journal article, Lorch makes the case that detailed patient-level data is needed to develop and assess the impact of perinatal health interventions across various patient subpopulations. In his review of 3 legislative policies (elective deliveries prior to 39 weeks gestation, perinatal regionalization, and paid maternity leave), Lorch outlines challenges to utilizing the current epidemiologic data systems to study the impact of these policies on maternal and infant health outcomes. Similarly, he argues that a more comprehensive investigation of factors impacting maternal decision making around safe infant sleep is only possible if more granular data at individual, hospital, and community-levels are available. Despite these limitations, Lorch calls for the use of causal
inference methods to study the impact of policies on perinatal health outcomes which is what was done in this dissertation.

**Inconsistent safe sleep messaging**

For mothers of preterm infants, to our knowledge, there are no public health safe infant sleep messaging campaigns focused on their unique experiences. Perhaps the lack of targeted efforts focused on preterm infants may be a factor in the lack of improvement in SSP we found among Massachusetts-born preterm infants. And for mothers of healthy term infants, the messages they receive about infant sleep practices from public health organizations, the lay press, retail stores, family members and friends can be conflicting. While most public health organization such as departments of public health, government-based service providers, national maternal and child health non-profit organizations define safe infant sleep as sleeping alone, in a separate sleep space, on the back without the presence of blankets or other potentially dangerous items, mothers are surrounded by very different messaging in their daily lives. In a recent report on National Public Radio (NPR), a reporter places the risk for sudden infant death due to unsafe sleep practices along a spectrum of risk for death due to other causes. She makes the argument that among low risk infants the likelihood of death is much lower than the chance of being hit by lightning in the U.S. in a person’s lifetime and lower than the chance of being killed in a car accident in the U.S. in a year. Moreover, Goodstein et al evaluated images depicting sleeping infants on stock photography websites, and found that only 50.8% of photos showed the infant in SSP and only 5% of all photos accurately represented the safe sleep environment as outlined by the American Academy of Pediatrics. Thus mothers and other caregivers are surrounded by infant sleep messages that are in sharp contrast to what is recommended by the medical and public health community. Highlighting this disconnect, a recently published
editorial in the Journal of the American Medical Association Pediatrics stated that “no recent breakthrough has occurred for this public health issue (infant sleep related death), which kills a surprisingly large number of society’s most vulnerable – typically healthy infants”. While the medical community continues to raise alarms about the nearly 3500 babies who die from unsafe sleep practices, the broader public may be hearing a very different message. And even advice received by mothers from doctors and nurses about infant care practices, including safe infant sleep, may vary considerably in frequency and content. Eisenberg et al found that 11-75% of mothers of generally healthy term infants in a nationally-representative sample reported receiving no advice from doctors about infant care practices, with percentages varying by the specific care practice such as sleep position, vaccination, and breastfeeding. And when advice was given, it was not consistent with AAP recommendations 2-29% of the time, again depending on the specific care practice.

Furthermore, mothers and other caregivers may have varying degrees of trust in the advice sources about infant care practices, including infant sleep. For the predominantly healthy term population, the previously mentioned nationally-representative prospective study of mothers from 32 U.S. maternity hospitals demonstrated that NHB mothers are less likely to trust doctors about advice on bed sharing and pacifier use compared to NHW mothers. When the analysis was isolated to physicians, investigators found that NHW mothers were more likely to trust physicians about infant care advice if mothers reported the doctor was the same race. For mothers of all races/ethnicities in this study, they were more likely to trust their doctors if they reported that the doctors were qualified or if the doctor had asked their opinion. While we know the prevalence of maternal receipt of advice about infant safe sleep as well as the predictors of maternal trust in advice about infant care practices for the healthy term population,
data for preterm and particularly NHB preterm infants are lacking. In light of our research findings that demonstrate persistent racial/ethnic disparities in SSP among preterm infants, future research should seek to elucidate the content and mode of communication about infant sleep practices that are most impactful for mothers of preterm infants.

**Limitations**

There were several limitations to this work related to the survey method of data collection and the lack of information on certain covariates. First, as with all data collected from self-reported surveys, there was the possibility of recall bias as well as pressure mothers may feel to report socially-acceptable answers about supine sleep positioning. While we hypothesized that this bias would be randomly distributed across the study period, only observed infant sleep positions by investigators would adequately address this issue. Second, while PRAMS contained some clinical data from birth certificates, it lacked information about infants’ severity of illness which may impact the quantity and quality of education provided to families about safe infant sleep. For instance, for an extremely preterm infant with chronic lung disease, a complication of prematurity, both NICU staff and parents may be focused on the acute medical needs of the infant during hospitalization, and less on home care practices like safe infant sleep. Third, we lacked data around the quality and quantity of interactions between hospital providers and parents, and advice received about safe infant sleep practices. Given that in term infants, there is tremendous variability in advice received, these data may be a significant predictor of maternal SSP adherence. Fourth, we did not have data on which other caregivers or child care centers were involved in infant care, including sleep practice and environment, after hospital discharge. In a recently published study of grandmothers who provided care at least weekly for an infant grandchild < 6 months old, only 45% of the 239 respondents reported adherence to SSP at the
grandmothers’ house and 58% reported SSP while in the mothers’ house. The presence of non-parental caregivers may influence maternal decision-making about infant sleep practices in the preterm population.

**Strengths**

Despite these limitations, our body of work was the first, to our knowledge, to focus on the most vulnerable population of infants at greatest risk for SUID, preterm infants and NHB infants. Our cohort of over 50,000 preterm infants (in Aim 1) represents a population of preterm infants born in 16 states who were discharged home to the care of the mother. Our advanced statistical methods allowed for consideration of maternal-level and state-level factors that could impact our primary outcome, SSP. Moreover, by utilizing the segmented regression approach, we were able to undertake a natural experiment to test the impact of states’ prioritization of safe infant sleep on SSP. Given the difficulty in undertaking a population-based randomized clinical trial of state-level policies on infant sleep practices, this causal inference method was a robust approach to address the observable and unobservable confounding present in epidemiologic observational data.

**Future Directions**

For preterm infants and their families, we do not yet have a comprehensive understanding of the barriers and facilitators to adherence to safe infant care practices, including safe sleep practices, after hospital discharge. Moreover, we do not know how these barriers and facilitators vary by race and ethnicity. Until we can conduct population-based observational studies of independent predictors of parental decision-making around infant care practices among preterm infants, stratified by race and ethnicity, we will not be able to develop effective interventions to improve safe sleep practices for preterm infants, and reduce or eliminate the racial disparity.
To address these deficits, we are undertaking two studies to develop the body of evidence which will inform future intervention studies. The first is a mixed methods prospective observational cohort study of 300 maternal-infant dyads from 4 NICUs in Denver and Fort Collins, CO. Entitled “the Impact of Maternal Engagement in the NICU on Infant Health Outcomes”, this study seeks to understand how maternal engagement in the NICU may impact infant short term and longer-term health outcomes, including post-discharge adherence to safe sleep practices, breastfeeding, and minimizing second hand smoke exposure.

Figure 4.1 Conceptual model for impact of maternal engagement in the NICU

The conceptual model demonstrates our hypothesis that maternal engagement may mediate the relationship between barriers to engagement (maternal sociodemographic, mental health, and infant health factors) and infant outcomes. At present, approximately 150 maternal-infant dyads have been recruited from the NICUs at University Hospital and Children’s Hospital Colorado with recent expansion to Poudre Valley Hospital and Denver Health.

Our second study entitled “Study of Attitudes and Factors Effecting PREterm infant care Practices” (SAFE PREP), will use a mixed methods approach to conduct a nationally
representative survey of mothers of preterm infants discharged from U.S. NICUs and special care nurseries with a wide range of gestational ages and medical complexities. We will use qualitative and national surveillance methods to determine prevalence of adherence to AAP recommended infant care practices known to reduce the risk of SUID (safe sleep, breastfeeding, reducing second-hand smoke exposure) and to characterize the barriers and facilitators of adherence among mothers of preterm infants. This will fill a critical knowledge gap, which is needed to inform intervention development to reduce SUID mortality among this vulnerable population. Shown below is our conceptual model.

Figure 4.2 SAFE PREP Conceptual Model
Using the Theory of Planned Behavior (TPB) as our main theoretical framework, we will first perform qualitative analysis to identify TPB domains and the broad array of barriers and facilitators to adherence of SUID reducing behaviors (Aim 1). Following development and administration of a national survey of 1500 mothers of preterm infants, we will determine prevalence of SUID risk-reducing infant care practices (Aim 2), the extent to which TPB domains predict intention and adherence to SUID risk-reducing infant care practices (Aim 3), and the extent to which maternal, infant, medical system, and/or other factors identified in our qualitative analysis are associated with TPB domains (Aim 4A) and adherence to AAP recommended infant care practices (Aim 4B).

For both the local and national studies, we will conduct analyses stratified by race/ethnicity to investigate the differing impact of identified barrier and facilitators on adherence to safe infant care practices in order to investigate racial/ethnic disparities in greater detail.

**Conclusion**

In this population-based analysis of SSP among preterm infants, with particular focus on the NHB-NHB racial disparity in SSP, we found that 1) NHB mothers of preterm infants are significantly less likely to adhere to SSP compared to NHW mothers of preterm infants across the entire study period, 2) West Virginia and Oklahoma’s prioritization of infant sleep practices did not improve SSP adherence among NHW or NHB mothers compared to control states, and 3) Massachusetts’ comprehensive effort to improve safe infant sleep compliance did not lead to improved adherence among full term or preterm infant mothers. These results provide robust epidemiologic SSP data for preterm infants and for the persistent NHB-NHW racial disparity in SSP and highlights the need for observational and interventional studies that will inform local
and national public health initiatives to understand and address the unique needs of specific infant groups.
REFERENCES


