

**Final Report, Grant R40MC08957,
“Parent-Newborn Correlates of Health Issues in Infants with Down syndrome”**

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I & II-Introduction & Review of the Literature

Research Problem & Literature Review. Over the past three decades, the life-spans of individuals with Down syndrome have risen from approximately 30 years (Yang et al., 1997) to almost 60 years (Bittles & Glasson, 2002). Yet many health issues continue to plague these children. Particularly during the first years of life, these children find themselves at higher-than-average risks for congenital heart defects (40%-60%), pneumonia, bronchitis, and other respiratory problems (So et al., 2007; Boulet et al., 2008); and other serious health issues (Roizen, 2003). Partly as a result, rates of infant mortality appear much higher among infants with Down syndrome (Rasmussen et al., 2006).

Although such basic information is known, few detailed studies exist about the correlates of mortality and morbidity during the first year of life for infants with Down syndrome. In terms of morbidity, for example, we know that roughly 1/2 (So et al., 2007) to 2/3 (Boulet et al., 2008) of these infants experience in-patient hospitalization during their first 2-3 years, but little is known about characteristics of the mother, newborn, or mother’s prenatal practices that might relate to such hospitalizations.

Similarly, we know that newborns with Down syndrome die at alarmingly high rates, but the correlates of infant mortality remain generally unknown. For example, Rasmussen et al. (2006) identified such predictors of survival as maternal race (with African-American babies dying more often than Caucasian babies), presence of heart defects, low birth weight, and the interaction of maternal race and heart defects. At the same time, however, we know little about when during the first year such deaths occur or the causes of these deaths.

Finally, we suspect—but do not really know—that a host of social factors relate to infant morbidity. In Rasmussen et al.’s (2006) study, for example, newborns with heart defects and born to African-American mothers were most likely to die. But a whole host of other social factors went unexamined in that study. We thus know little about the effects of rural versus non-rural residence, maternal education, or number of prenatal visits. We also do not know if Rasmussen’s racial findings also hold true for hospitalization.

Purpose, Scope, and Methods. To address these issues, we have been examining linked vital statistics and hospital discharge records from the state of Tennessee for periods back to 1990. By linking these records together, we have access to a large amount of information. Such information concerns the mother, the newborn, the mother’s prenatal practices, the child’s hospitalization, and (for those who died) the child’s death. Particularly for analyses employing all of the official Birth records, such information exists on over 1,300 newborns with Down syndrome. As the original reviewers of our grant noted, we have access to incredibly rich datasets, for exceptionally large numbers of infants with Down syndrome, over time spans rarely seen in most studies.

Nature of Findings. Given such a large amount of information over so many years, we have chosen to limit our focus to the first year of life. In contrast to the few prior studies of this issue—which mostly highlighted these children’s first 2 or 5 years (Boulet et al., 2008; Frid et al., 2002), we note that many of the most severe problems occur within the first year. In our initial study, So et al. (2007) followed the hospitalization records of 213 newborns born from 1997-1999 over each child’s first 3 years. Of these infants, 54% (115) had congenital heart defects (CHD’s). Almost fifty percent of all children were hospitalized before age 3; these 106 children were admitted 245 times. But the large majority (88%) of those who were hospitalized underwent hospitalization early on, during their first year. Median time to first hospitalization was 96.0 days (CI: 78 - 114) for CHD infants, 197 days (CI: 46 - 347) for non-CHD infants. Children with Down syndrome and with co-occurring congenital heart defects are at especially high risk for hospitalization within the first year of life.

A second decision concerned linking our many records. Although we have important information from any one of our five datasets, our major contributions occur when we can link both within and across record types. In So et al. (2007), we performed linkage using only the 8,325 inpatient visits (1996-2002) of individuals (all ages) with Down syndrome. First-order linkage (i.e., linking records of a single individual) was performed, resulting in a study that examined for the first 3 years of life only those infants born from 1997 through 1999. In our study of divorce among families of children with Down syndrome, Urbano and Hodapp (2007) used second-order linkage (Tu & Mason, 2004) to identify families of children born to the same mother, utilized all of the state’s families giving birth from 1990-2002 (1.056 million), and then linked the birth-families to Divorce Records. By doing so, we were able to examine the differential effects on divorce of the child with Down syndrome’s being first- vs. latter-born, or of having smaller or larger families. Such linkage techniques are discussed in Urbano (2007).

In this project, we have been faced with—and have now overcome—an unexpected linkage problem. Specifically, the Birth record provides a wealth of identifying information, specifically the newborn’s and the mother’s names and social security numbers. In contrast, the Hospital Discharge records include only the patient’s social security number (no names), along with other, less-than-perfect identifiers (patient’s gender, date of birth, town of residence). Unfortunately, newborns do not yet have social security numbers and, unlike the Birth records, such numbers are not entered retroactively. Thus, until infants received social security numbers (at 6-9 months, on average), we had difficulty in linking across these two record types. Fortunately, however, we have recently discovered another variable (ClaimID) that links mothers to their newborns. We can now make the following connections: Newborn with DS to mother (both within the Birth record); mother’s social security number (from Birth record) to mother’s social security number (Hospital Discharge of the hospitalization when mother delivers); mother’s ClaimID (from delivery hospitalization) to newborn’s ClaimID (at birth hospitalization); and newborn’s birth hospitalization (backward) to the newborn’s Birth record and (forward) to the infant’s subsequent hospitalizations (based on patient ID and hospital record numbers). This difficulty in linking, which initially slowed our progress, has now been resolved, and we are rapidly progressing on many analyses (for discussions of issues using large-scale datasets, see Hodapp & Urbano, in press).

A third decision involves our inclusion of many social factors in our analyses. We have endeavored to go beyond the “obvious”—that is, maternal race and age—to examine potential correlates of the health of children with Down syndrome. By capitalizing on our demographic variables (from the Birth records) and on 2nd-order linkage (Tu & Mason, 2004) to make complete families, we can now examine many additional issues. Such issues transcend those of the infant with Down syndrome per se, to also include issues of divorce (Urbano & Hodapp, 2007), family demographics (Hodapp & Urbano, 2008; Hodapp, Urbano, & Rosenbloom, 2008), and subsequent parental childbearing (Burke, Urbano, & Hodapp, 2009). Throughout these analyses, we employ social factors as both correlates of infant morbidity-mortality and as outcomes in their own right.

III. Study Design and Methods

A-Study design. This project uses large-scale, linked databases to examine, in a retrospective longitudinal design, issues of morbidity-mortality among infants with Down syndrome, as well as to examine correlates of health in the infants and of functioning in the families. Depending on the individual study, the time-frame is from 1990-2006, although studies examining hospitalization go back only to 1997 (when Tennessee’s Hospital Discharge system became fully operational).

B & C. Population studied & sample selection. The main samples of interest are infants with Down syndrome and their families. Many of these studies examine correlates or sub-groups within the Down syndrome group alone—for these studies, infants with Down syndrome (1,305 from 1990-2006) are used. In other studies, we compare various health or family characteristics of infants with Down syndrome (or their families) to infants-families of non-Down syndrome children. For these studies, we use both the Down syndrome and Tennessee population infants (from 1990-2005, N = 1.3 million).

D-Records Employed in These Studies. For these studies, the main records used involve the state of Tennessee’s Birth, Hospital Discharge, and Death records.

1) **Birth Records, 1990-2005.** From 1990 to 2005, approximately 81,000 children were born annually in Tennessee, with numbers rising over the period and totaling 1.3 million births. For each child, local recorders administered a questionnaire to the mother and followed-up by themselves recording information from hospital records. The official Birth records contain approximately 140 variables pertaining to:

---**mother**—age; race; marital status; education; home address, county, and state; prior live births; inter-delivery interval from last birth; maternal smoking; number of prenatal doctor’s visits; month during pregnancy when prenatal visits began;

---**infant**—gender; birthweight; estimated gestational age; APGAR score at 5 minutes; birth complications; abnormal conditions; congenital anomalies; and indicator that death occurred within the infant’s first year of life (placed retrospectively on all birth certificates of deceased infants to discourage identity theft);

---**family**—As the Birth Records of individual mothers are linked to constitute families and because the mother’s exact residential address (street number, name, town, and zip code) is

provided, we can determine the effects on child hospitalization of family structure and of geographic variables (based on the family's residence).

2) Hospital Discharge Records, 1997-2006. The Tennessee Hospital Discharge Data System records any hospitalization (in- or out-patient), for all children and adults within the state. In recent years, these in-patient visits number approximately 900,000 (per year, entire state). In addition to demographic information, the in-patient Hospital Discharge file provides a principal ICD-9 diagnostic code and up to 8 secondary diagnostic codes (where Down syndrome, 758.0, is most often listed); procedures performed; days and units of service required; identifiers of specific hospitals and doctors; patient's home zip code; and insurance/ payer information.

3) Death Records, 1990-2006. Each year approximately 56,000 people die in Tennessee. Official death records include information about the individual's name, age, sex, residence, and cause of death. The underlying cause of death is a single cause that the physician chooses from one of the eight multiple causes of death. Within the Tennessee Death Records, causes of death were indicated using ICD-9 codes if the death occurred during or before 1998, with ICD-10 diagnostic codes if the death occurred between 1999 and 2006.

D-Statistical techniques employed. In addition to basic descriptive statistics to determine means, medians, standard deviations, and quartile scores within the Down syndrome (and, for some studies, the Tennessee population control group), univariate analyses such as chi-squares, t-tests, and one-way ANOVA's are used when comparing across groups. We also use logistic regressions to determine the effects of different predictor variables in one or several groups.

IV. Detailed Findings

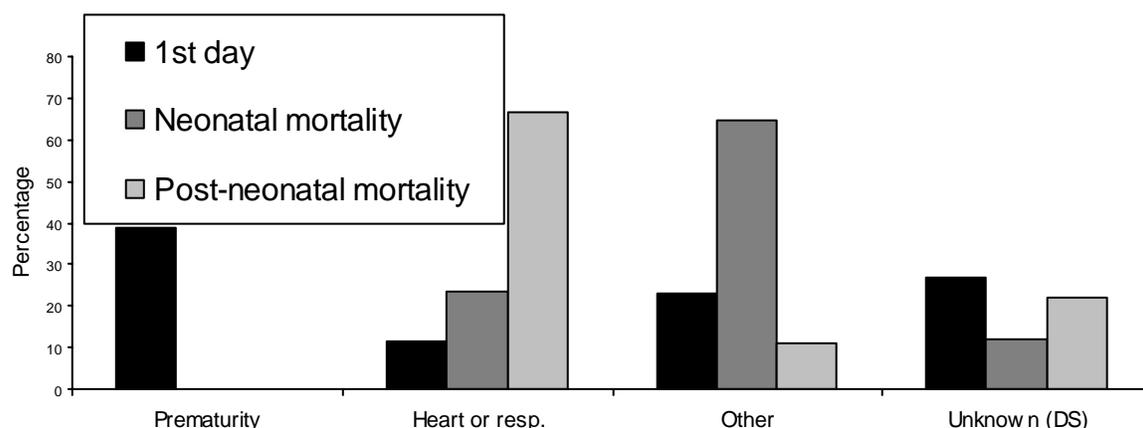
In the pages below, we organize our main findings so far. As noted above, these findings relate to both the mortality and morbidity of infants with Down syndrome, examine in-depth several important social factors, and use family and other variables both as correlates and as outcomes.

1-Infant Mortality. The first main study relates to mortality among infants with Down syndrome. Examining 1,305 Tennessee infants with Down syndrome over the 1990-2006 period, Goldman, Urbano, and Hodapp (2009) have linked these Birth Records to Tennessee's official Death Records to examine the amount, timing, and causes of infant mortality. With 97 deaths occurring within the first year, we found a death rate of 74 per 1,000 deaths (or 7.4%), a rate well within the previously reported range of 4.0% (Weijerman et al., 2008) to 24.5% (Sadetzki et al., 1999) among the seven other existing studies of this issue (the median value of the 7 other studies—7.1%--was found by Rasmussen et al., 2006).

Beyond the mortality rate itself, we were struck by the timing and causes of such deaths. Compared to early deaths within the Tennessee population, greater proportions of deaths occurred from 1 month (28 days) to 1 year than occurred during earlier periods (see similar findings from Sadetzki et al., 1999; Shin et al., 2007). In our sample, 56% of deaths in the first year of life occurred during this post-neonatal period, compared to 36% of deaths in the first year among Tennessee population infants ($p < .001$).

From our data, however, it seems important to differentiate children who died during their first day (N = 26), from 2-27 days (N = 17), and from 28-364 days (N = 54). Compared to infants who died during the two later periods, newborns who died at 1-day of age were much more likely to be born prematurely (<37 weeks) and to have low birthweights (<2500 grams). Prematurity occurred in 91% of newborns who died during their first day, 47% of those who died during the neonatal period (2-27 days) and 26% of those dying beyond 1 month ($p < .001$). Figures for very premature newborns (< 32 weeks), as well as those of extremely low birthweights (< 1,000 grams) and extremely low APGAR scores (<4), all mirrored this overall pattern.

Conversely, those infants who died from 1 month to 1 year were more likely to have causes of death that involved either heart or respiratory problems, whereas the causes of death during the neonatal period (2-27 days) were less clear, with many “other” causes noted (often involving intestinal causes). As in Rasmussen et al. (2006), we too found that “Down syndrome” was listed as the sole cause of death on a substantial proportion of death certificates. The Figure below summarizes this pattern, with every case of death categorized into one of four causes (“Down syndrome” was chosen only when Down syndrome was listed in the Death record but no other cause of death—as either an underlying or multiple cause—could be determined). Deaths during the first year of life in Down syndrome seem to occur at three separate time-periods, with the youngest deaths related to prematurity, later deaths to heart and respiratory conditions.



2) Morbidity Among Newborns with Down syndrome. A second focus involves morbidity among newborns with the syndrome. Given our difficulties in linking from the Birth to newborn Hospital Discharge records, we are only now performing some of these analyses. We have, however, completed and submitted other studies, including one that examines whether newborns with Down syndrome differ from general population rates for low birthweight, prematurity, and small for gestational age. This study also related demographic and prenatal practice variables to these birth outcomes. Using Tennessee Birth Records from 1990-2005, Hodapp, Urbano, and Rosenbloom (2009) compared information concerning 1,043 newborns with Down syndrome to remaining Tennessee resident births. Information was examined about the newborn (birthweight, estimated gestational age, sex, birth order), mother (ethnicity, age, race, education), and maternal prenatal practices (number and beginning month of prenatal doctor’s visits during pregnancy; weight gain during pregnancy; maternal smoking). Compared

to the general population, newborns with Down syndrome had greater risks for being born at low birthweights (OR = 3.1; CI: 2.6–3.5), preterm (OR = 2.2; CI: 1.9–2.6), and small for gestational age (OR = 2.6; CI: 2.3–3.1).

As shown by the Figure below, such findings translate into an enormous public-health burden. Not only do such outcomes occur 2-3 times more often among newborns with versus without Down syndrome, but from 20% to 25% of all newborns with Down syndrome are born with each of these three adverse birth outcomes. Within the Down syndrome group, maternal weight gain of below 25 pounds was the sole predictor for all three outcomes, with <5 prenatal checkups also predicting low birthweight and prematurity. Other predictors, including those usually found within non-DS populations (and also found within our larger Tennessee population), did not independently predict these birth outcomes in newborns with Down syndrome.

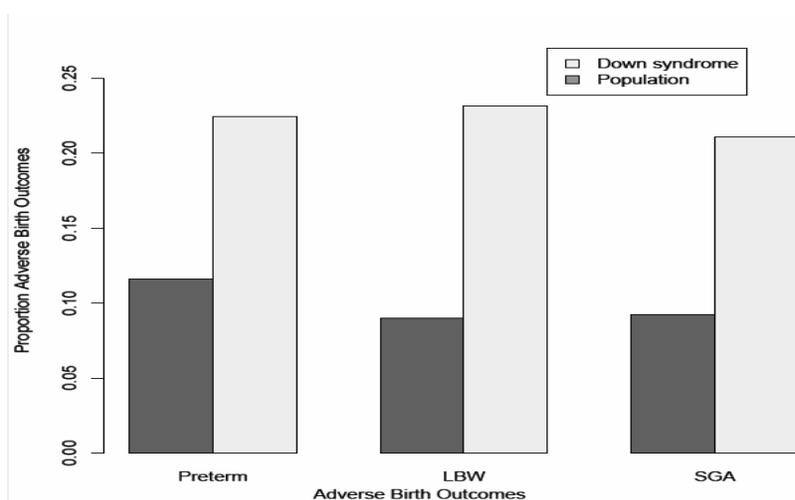


Figure 1. Proportion of Preterm, Low Birthweight and Small for Gestational Age by Group

3) Parent-Family Correlates, Demographics, and Behaviors. As noted above, we feel strongly that many characteristics of the family may be important for the health of infants with Down syndrome. In this particular group, however, major issues exist even in terms of the field's knowledge of basic family demographics. Specifically, both parents and researchers have asked whether Down syndrome is “disappearing.” This concern, found in both the popular press (Newsweek, 2008) and in the scientific literature (Collins, Muggli, Palma, & Halliday, 2008), relates to the recent calls for universal prenatal screening (ACOG, 2007), and the supposed effects that such screening might have on the syndrome's prevalence rates.

In response to these issues, we have also begun examining whether mothers of children with Down syndrome continue to be older than population-group mothers and, if so, whether advancing age relates to other maternal characteristics. Using Tennessee Birth and Hospital Discharge records from 1990-2002, Hodapp, Urbano, and Rosenbloom (2009) compared mothers of 918 infants with Down syndrome to mothers of 1,055,360 population infants. More mothers of children with (vs. without) Down syndrome delivered their infants when aged 35 or older, fewer before 25. In both groups, increasing age was associated with higher education and

SES, and White (vs. African-American) mothers had their babies at older ages. Compared to population mothers, more mothers of children with Down syndrome graduated college and fewer had not graduated high school. Compared to Tennessee-population births, disproportionately more White versus African-American mothers delivered a child with Down syndrome (also found in Boulet et al., 2008; Canfield et al., 2006).

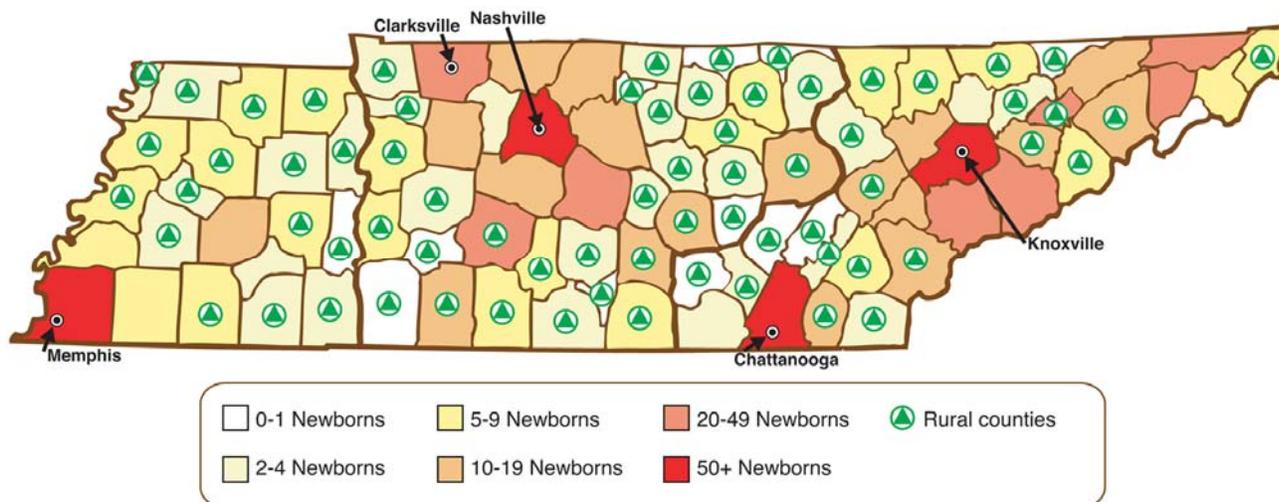
Beyond the general characteristics of these families, we have been interested in those children with the Down syndrome who are from minority families. In a recent article on this issue, Hodapp and Urbano (2008) compared African-American to White mothers of newborns with Down syndrome in terms of education, SES, and marital status, all variables that have previously been related to varying levels of utilization of health services (Elster, Jarosik, VanGeest, & Fleming, 2003; Garland et al., 2005; Thompson & May, 2006). Using Birth Records of infants with Down syndrome across the 1990-2002 period, we compared 759 White (Non-Hispanic) to 145 African-American (Non-Hispanic) mothers of newborns with Down syndrome; outcome measures included maternal age at infant's birth, education level (particularly whether the mother completed high school), marital status, and neighborhood median family income. Compared to White mothers, African-American mothers were of lower SES and were younger, with more African-American (vs. White) mothers giving birth at 23 years or younger (37.2% vs. 22.9%). In both groups, young (vs. older) mothers showed the highest percentages of not graduating high school and of being unmarried. To our knowledge, our article is the first to examine African-American and White mothers of children with Down syndrome on personal characteristics that generally relate to health care access.

In a soon-to-be completed study, we are looking as well at subsequent childbearing in these families (Burke, Urbano, & Hodapp, 2009). From the psychoanalytic literature, families of children who have died are more likely to have subsequent or "replacement" children, and we are using our linked family datasets to determine whether parents of children with disabilities are also more likely to have subsequent children. Using family-wise, linked Birth records, the study examined subsequent children and family size across families of three groups, those of: children without disabilities, children with spina bifida, and children with Down syndrome. The study included 727,563 families of children without disabilities, 1,092 families of children with Down syndrome, and 302 families of children with spina bifida. Compared to families of children without disabilities (40.4%), proportionally more families of children with Down syndrome (57.5%) and spina bifida (52.0%) were multi-child families. Families of children with disabilities were more likely to have subsequent children and larger family sizes regardless of maternal race, age, educational attainment, or marital status. Furthermore, compared to families of children with spina bifida, families of children with Down syndrome were slightly more likely to have subsequent children. This study, one of only a few of its kind, has enormous implications for family planning and counseling.

Finally, we are beginning to examine the effects of where children live on their morbidity and mortality. As shown in the map below, children with Down syndrome come from throughout the state, including its most rural areas. As 67 of Tennessee's 95 counties are designated as "rural" by the federal government (Office of Rural Health Policy, 2001), many children reside in these sparsely populated counties. As the map illustrates, from 5 to 20 children

live in several rural counties, and cumulatively those children who live in rural counties constitute approximately 1/3 of our total Down syndrome group.

Tennessee's Children with Down syndrome, by County (1990-2005)



In analyses of the effects of rural vs. non-rural residence, we have seen major effects of rural living status on certain family variables, but not on health outcomes. In our earlier study of divorce in families of children with Down syndrome, we found exceptionally high rates of divorce among fathers (and mothers) who had not graduated high school and who lived in rural areas (Urbano & Hodapp, 2007). But such findings have yet to translate into health outcomes. Thus, in the Goldman et al. (2009) study of mortality among infants with Down syndrome, rural counties were home to 27.0% of all infants who died and 31.8% of all infants who survived their first year. In these preliminary analyses, few differences have emerged concerning the health of infants with Down syndrome who reside in rural compared to non-rural Tennessee counties.

Clearly, we are early in these analyses and various factors may account for the “non-effects” of residential status. It may simply be that death is an overly gross health outcome and that small-sized effects may not be detectable. To address this problem, we have begun examining whether rural-nonrural differences emerge on prematurity, low birthweight, or hospitalizations. Similarly, our measure of “rural-ness” may be overly gross. To address this issue, we have begun categorizing each of Tennessee’s 95 counties in terms of “Beale codes.” Assigned by the USDA’s Economic Research Service, Beale codes measure a county’s degree of ruralness on a scale ranging from 1 (most rural) to 9 (least rural). These codes have versions based on the status of the county as measured in the 2000 Census (2003 Beale codes) and in the 1990 Census (1993 Beale codes). Finally, the main determinant for health outcomes may not relate to rural-nonrural status per se, but instead to mothers’ easy access to pre- and post-natal healthcare,. To address this issue, we are beginning to geocode a locality’s degree of accessible health care in terms of “rational service areas.” As determined by the state of Tennessee, “Rational service areas are individual counties, groups of counties, or communities that have displayed certain obvious primary, obstetrical, and pediatric care service patterns for residents of that county, community, and/or surrounding areas.” (State of Tennessee, 2005, p. 7). Although

the fate of these analyses remains uncertain, we are working hard to use our geographic data to inform us about the health needs and outcomes of infants with Down syndrome.

V. Discussion and Interpretation of Findings

A & C. Main Study Findings and Comparison to Other Studies. With major help from this grant's funding, we have in a little over a year's time produced several important studies. Such work, which we are continuing beyond the grant period, is providing critical, policy-related specifics to what had previously been a more general outline. We see our work as producing four major findings.

1) Amount, Timing, and Causes of Infant Mortality. Although a few earlier studies had shown that newborns with Down syndrome face extremely high rates of death during the first year, our study is the first to highlight this finding and to examine in more depth when and why such deaths occur. As in earlier studies, we too found that newborns with Down syndrome are over 8 times more likely to die than newborns in the general population. Our overall infant mortality rate in the Down syndrome group, 7.1 per 1000 births, is almost identical to the median death rate from prior studies (7.4/1000), leading us to conclude that our methods were indeed reasonable to examine this question. Similarly, we too found that disproportionately more infant deaths occurred beyond the 1-month period, a finding earlier found for Down syndrome infant deaths (e.g., Shinn et al., 2007).

New to our study, however, is the designation of three distinct “death periods”—at 1 day, within the neonatal (2-27 days) period, and beyond 1 month (28-364 days). In addition, these periods seem tied to causes of death. Indeed, most prematurity, low birthweight, and low Apgar scores occurred in the group who died at 1-day. Conversely, much more of the heart and respiratory causes of death were noted for infants who died at 1-month or beyond. This connection between “death periods” and specific causes of death is new to our study.

2) Amount of Infant Morbidity. Just as infant mortality is rampant among newborns born with Down syndrome, so too is hospitalization and adverse birth outcomes. Our earlier study found that over half of these infants—and even more of those with congenital heart defects—experienced in-patient hospitalization. Most such hospitalization occurred within the first year of life, most often for pneumonia, bronchitis, and other respiratory conditions (So et al., 2007).

In extending this earlier work, we find that newborns with Down syndrome are also at increased risk for prematurity and low birthweight (Hodapp, Urbano, & Rosenbloom, 2009). We also find that maternal low weight gain is the most important correlate of such birth outcomes, with few prenatal visits a correlate for two of the three outcomes. As noted above, we are also finishing up our analyses connecting information from the Birth files to the birth hospitalization (from the Hospital Discharge files). Again, the main take-home message here concerns the sheer amount of health issues experienced by these newborns, a state of affairs that seems rarely acknowledged within various publications of parent-advocacy groups that promote knowledge of this syndrome.

3) Social Correlates. Our third major finding is, in some sense, a non-finding. Across several different areas, we are oftentimes not finding clear social and familial correlates of health among

infants with Down syndrome. In considering infant mortality, for example, we did not find that infants from rural counties died at rates higher than did those living in non-rural areas. Similarly—and in contrast to Rasmussen et al., (2006)—we did not find that a disproportionate number of our infants with Down syndrome died when mothers were African-American. With few exceptions (e.g., low number of prenatal visits as a predictor of prematurity and low birthweight), we have found few “social” predictors of health among infants with Down syndrome.

In response to this somewhat surprising finding, we have two reactions. First, the physical changes related to Down syndrome in many ways “trump” more social factors. Indeed, compare these findings to those in the larger population. In studies of non-Down syndrome newborns, mothers who are from minority groups, unmarried, less educated, or who have received no or inadequate prenatal care most often have low birthweight and/premature newborns. In our studies of infants with Down syndrome, however, such usual potential correlates were only sporadically correlated to adverse health outcomes.

A second (and not mutually exclusive) explanation may relate to the specificity of social correlates. It may, for example, be that such social correlates are operating, but only when examined in a more fine-grained manner. Our new analyses using Beale codes and rational service areas for different types of healthcare may reveal such relationships.

4) Parent-Family Outcomes. Our fourth main finding relates to family functioning. As ultimately families are responsible for knowing about, accessing, and receiving health care for their children, understanding family functioning is vitally important to public health. In the case of Down syndrome, we are finding that these mothers continue to be, on average, more likely to give birth at older ages (and fewer of these mothers are younger; Hodapp, Urbano, & Rosenbloom, 2009). And, as in the general population, more mothers of infants with Down syndrome are more educated; there also appears to be a slight over-representation of White—as opposed to African-American—mothers within the Down syndrome (versus population) group (see also Canfield et al., 2006).

But it is also important not to lose sight of sub-groups within these families. In our study comparing mothers of newborns with Down syndrome who are African-American versus White, we found that higher percentages of African-American mothers were relatively young (23 or younger). For both races, younger mothers were also likely to be less educated (i.e., not high school graduates) and unmarried (Hodapp & Urbano, 2008). Given the scarcity of attention paid to African-American families within the Down syndrome group, our study is critically important for the organization and delivery of early health and early intervention services.

Finally, our finding related to so-called “replacement” children gets at another public health issue related to these families. If families are more often having subsequent children in response to their child having either Down syndrome or spina bifida, then public health workers, counselors, social workers, and school personnel need to consider the issues and concerns faced by these “replacement children.” Until now, this issue has not been considered within those examining siblings of children with disabilities (Hodapp & Urbano, 2007).

B) Main Limitations. There are two main limitations in this project. First, we are using administrative data, which some consider suspect. Fortunately, at least for children with Down syndrome seen at Vanderbilt, we have compared our sample (i.e., those identified through Birth and Hospital Discharge records) to those children seen at Vanderbilt's state-supported Clinical Genetics Clinic. Essentially, almost all children (99%) who we identified as having Down syndrome do indeed have the syndrome, but we do miss some children (Urbano et al., 2007).

A second limitation concerns the lack of psychological data. In this case, we oftentimes know that something happens, but not always why. Consider, for example, our newest study of subsequent childbearing. We know from these data that parents of newborns with either Down syndrome or spina bifida are more likely than other parents to have a subsequent child in the family, but not what parents are thinking and why they have subsequent children.

D & E) Potential Applications to Care and Policy

Our findings from these studies suggest two main care and policy implications, one more specific and the other more general. The specific care recommendations relate to the need to alert Ob/Gyn's and other physicians about the risks for prematurity and low-birthweight, with the goal of alerting physicians so that they increase their attempts to delay delivery. Even a few extra weeks in gestation would help enormously in decreasing rates of low birthweight and premature infants. In addition to helping to avert some percentage of prematurity and low birthweight births, some substantial percentage of 1st-day deaths among newborns with Down syndrome will also be averted.

For those who do survive their first day—and who generally are not born at low birthweights or prematurely—physicians need to take extra care to avoid later infant mortality. Heart and respiratory problems seem the main causes of these later deaths. Although most physicians are alert to the health issues relating to congenital heart defects, respiratory problems have received inadequate attention (as has the increased rates of respiratory problems among those infants with congenital heart defects; So et al., 2007; Boulet et al., 2008). Within Roizen's (2003) scheme for physicians treating young children with Down syndrome, respiratory problems need to become much more vigilant about the prevalence and effects (both hospitalization and death) that may arise from such conditions.

More generally, our studies highlight the need to consider Down syndrome as a “health condition,” particularly during the earliest years. Within the Down syndrome community, the general feeling has been that, having saved the large majority of these children from early death due to heart defects, we need not worry much about the early years for these children. Unfortunately, even now over 8 times as many newborns with Down syndrome die in their first year, the three time-periods feature different causes of death, and prematurity and low birthweight occur in 20-25% of all infants. Simply stated, the field needs to better appreciate—and make provisions to address—the health concerns faced by so many of these infants.

F) Suggestions for Further Research

Given the paucity of research on the health of young children with Down syndrome, many areas of fruitful investigation remain. Beyond sheer amount of health problems, we now need to:

- search harder for familial and social correlates of early health concerns;
- know the degree to which these parents have difficulties in knowing about, accessing, and receiving health care;
- examine how early health services fit within more general early intervention services, and,
- understand how the needs of and services for younger, minority, less educated, or rural parents might differ from services for more advantaged groups.

We thank HRSA for your support and hope to continue on what we consider to be an important area of public health inquiry.

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