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### Birth Outcomes Among Women with Congenital Neuromuscular Disabilities

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### Abstract

**Background:** Women with disabilities are at an increased risk for adverse birth outcomes; however, research among women with congenital neuromuscular disabilities (cNMD) is limited.

**Objective:** To describe characteristics and compare birth outcomes among women with and without cNMD.

**Methods:** Data were from the Slone Birth Defects Study (case-control, conducted from 1976–2015), which collected information on demographic, reproductive, and lifestyle characteristics. cNMD included spina bifida, cerebral palsy, muscular dystrophy, contractures, or arthrogryposis and were identified by participant report. Those with cNMD were matched to participants without cNMD by interview year and study site. We use modified Poisson regression to estimate relative risks (RR) for low birthweight, macrosomia, preterm birth and small/large for gestational age (SGA/LGA). Given the case-control design and overrepresentation of infants with congenital anomalies, data were weighted to reflect a 3% national prevalence of infants with congenital anomalies.

**Results:** Women with cNMD (n=125) were more likely to be white, nulliparous, have a cesarean section, have an unplanned pregnancy, report a pre-pregnancy BMI 25kg/m<sup>2</sup>, smoke during pregnancy, and report genitourinary infections. Women with cNMD had infants with shorter gestational length (mean difference: -7.44 days, 95% CI: -13.94, -0.95) compared to women without cNMD. cNMD was associated with higher risk of preterm birth (RR=3.98, 95% CI: 1.33, 11.95) and SGA (RR=2.14, 95% CI: 0.74, 6.15).

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**Conclusion:** Women with cNMD were more likely to deliver preterm and have a SGA infant. These findings highlight disparities faced by women with cNMD and stress the need to provide optimal perinatal and reproductive care.

#### Keywords

Physical Disability; Perinatal Health; Birth Outcomes; Pregnancy; Health Disparity

#### INTRODUCTION

An estimated 6–12% of women of childbearing age in the United States have a physical disability.<sup>1</sup> The most common mobility and dexterity disabilities are congenital neuromuscular disabilities (cNMD), including spina bifida, cerebral palsy, muscular dystrophies, contractures, and arthrogryposis, with approximately 12,000 infants born with these conditions annually. Average survival among people with cNMD has increased to beyond childbearing years, yet little is known about pregnancy and birth outcomes in this population.<sup>2,3</sup>

Growing literature suggests that women with disabilities are at an increased risk for adverse pregnancy complications and birth outcomes.<sup>4–9</sup> Severity of disability and other underlying health conditions may contribute to some adverse health outcomes experienced by individuals with cNMD. There are currently no pregnancy care guidelines from the American College of Obstetricians and Gynecologists specific to women with physical disabilities. Thus, factors such as inadequate preconception and prenatal care, physical accessibility barriers during office visits, and lack of proper training among doctors can contribute to negative health care experiences and adverse outcomes among women with physical disabilities.<sup>7,10–12</sup>

To date, only a few studies have documented pregnancy and birth outcomes among women with cNMD. One study reported an increase in the rate of deliveries among women with spina bifida, from 16 per 100,000 deliveries to 25 per 100,000 deliveries from 2003 to 2011 (a 56% increase).<sup>5</sup> Over the course of the study, more than half of women with spina bifida had cesarean deliveries (52%) compared to a third of women without spina bifida (32%). Another study found that pregnancy complications, including preterm delivery, hematologic events, blood transfusion, and urinary tract infections were more common among women with spina bifida.<sup>13</sup> Women with spina bifida who had a caesarean delivery-related complication compared to women without spina bifida who had a caesarean delivery. Additionally, recent studies found that infants born to women with cerebral palsy were more likely to be born preterm and be small for gestational age compared to women with cerebral palsy or muscular dystrophy have reported pregnancy complications (e.g., preterm birth and low birthweight); however these studies lacked control groups.<sup>16–18</sup>

Similarly, studies of women with other disabilities (e.g., intellectual and developmental disabilities, deaf or hard of hearing) have found similar increased risks in adverse birth outcomes such as preterm birth and low birth weight compared to non-disabled

peers.<sup>4,6,14,19–21</sup> However, very few of these studies have examined birth outcomes specific to women with cNMD, with a majority utilizing broad definitions of physical disability or other types of disability.

To this end, we sought to examine birth outcomes among women with cNMD by utilizing data from the Boston University Slone Epidemiology Center Birth Defects Study (U.S. and Canada, years 1976–2015). We describe maternal characteristics and birth outcomes including low birthweight, preterm birth, macrosomia, and small and large for gestational age (SGA/LGA) among women with and without cNMD.

#### METHODS

#### **Study Population**

Study data were retrieved from the Boston University Slone Epidemiology Center Birth Defects Study (BDS), a multi-site, case-control study with the goal of examining risk factors for congenital anomalies. BDS enrolled over 51,000 women during years 1976 to 2015 from the greater metropolitan and surrounding areas of Boston, Toronto, Philadelphia, San Diego, Nashville, and upstate New York. Cases were defined as women with pregnancies affected by at least one major structural congenital anomaly. Each study site ascertained infant cases with congenital anomalies via birth and tertiary care hospitals and vital records within 5 months of delivery. Controls were infants without a congenital anomaly, selected from the same recruitment areas as the cases, also within 5 months of delivery. Trained nurses conducted standardized interviews with the women, in person (1976-mid 1998) or by telephone (1998–2015) within 6 months after delivery. Interviews included standardized questions on sociodemographic information, lifestyle behaviors (e.g., smoking, alcohol use), and reproductive and medical history. All subjects provided informed consent.

As a part of the interview women were asked "Were you or the baby's father or any of your family members born with any of the following birth defects?: Brain/head/eye/ spine/spinal cord/spina bifida; muscles/bones/arms/legs; cleft lip/palate/gum; heart/blood vessels; lungs/throat/windpipe; kidney/ureter/bladder/sex organs; tumor/cysts; food pipe/ stomach/intestines/bowel/rectum; or other defect." Congenital disabilities were recorded and classified with the British Pediatric Association modification of International Classification of Diseases (ICD) diagnoses codes.<sup>22</sup> All reports of "mother/self" as the family member with the congenital disability were reviewed by research staff. Women with cNMD were defined as participants who reported themselves having one of the following: spina bifida, cerebral palsy, muscular dystrophy, contractures, or arthrogryposis, for a total of 132 women with cNMD. Those without cNMD were participants who reported no congenital disabilities for "mother/self" and were selected at a four to one ratio, matched to women with cNMD by interview year and study site, for a total of 528 women without cNMD. We matched women with cNMD to women without cNMD to help control for time and place differences over the course of the study. Matching also aided in cost efficiency for data that needed to be reviewed line-by-line, such as comment fields. We restricted analyses to participants whose infant was liveborn with a final analytic sample of 125 for women with cNMD and 505 for women without cNMD (see Ancillary material, Table 1 for distributions of the specific cNMD phenotypes and matched characteristics).

#### **Outcome Measures**

Low birthweight (<2500 grams), macrosomia (>4000 grams), preterm birth (<37 weeks), and early preterm birth (<32 weeks) were derived using data collected from mother reports. Percentiles for the infants' birthweights were assigned using U.S.-based reference curves for singleton liveborn infants adjusted for gestational age and sex.<sup>23</sup> We categorized infants' birthweight as <10<sup>th</sup> percentile (SGA), 10<sup>th</sup> to 90<sup>th</sup> percentile (reference), and > 90<sup>th</sup> percentile (LGA). We restricted the SGA/LGA variables to singleton only births, as the birthweight reference curves we utilized are only estimated among singleton births.

#### Analytic Methods

The BDS study was a case-control design, where pregnancies affected by congenital anomalies were the infant cases. For this secondary analysis, we were interested in the delivery outcomes among a selected sample of cases and controls in women with and without cNMD. In order to use BDS case-control data to calculate valid estimates for these secondary outcomes, we utilized a reweighting approach; this methodology is explained in further detail elsewhere.<sup>24</sup> In brief, we reweighted the data because women with infants with congenital anomalies are overrepresented in the dataset in comparison to the underlying source population, due to the case-control design of the BDS (72% of our total sample had an infant with a congenital anomaly). Further, women with cNMD were more likely to have an infant affected by a congenital anomaly, and infants with congenital anomalies are more likely to have the delivery outcomes of interest (e.g., low birthweight, preterm birth).<sup>25</sup> Because it is plausible that congenital anomaly occurrence may be in the causal pathway between maternal cNMD and delivery outcomes, if we were not to account for the sampling scheme, observed associations may be biased. Therefore, we utilized an approach that weighted cases (weight=1.0) and upweighted controls (weight=84.7) in the dataset to represent a 3% prevalence of infants with congenital anomalies, which approximates the prevalence of congenital anomalies among livebirths in the US.<sup>26</sup> The reported confidence intervals (CIs) around the weighted estimates represented the variance in the original (unweighted) data, to not make the estimates appear more precise than they were.

First, we examined distributions of maternal characteristics and sociodemographic factors between women with and without cNMD. We assessed infant biological sex, mother's age at delivery (<20, 20–24, 25–29, or 30 years), race/ethnicity (non-Hispanic White, non-Hispanic Black, Hispanic, and Other), mother's education level (<12, 12, >12 years), parity (number of births > 24 weeks gestation), gravidity (number of times participant has been pregnant), plurality (singleton or multiples), whether pregnancy was planned (yes or no), timing of first prenatal visit ( 8 or >8 weeks of pregnancy), multivitamin/folic acid supplementation in the first trimester, delivery type (vaginal or cesarean section), pre-pregnancy body mass index (BMI) (kg/m<sup>2</sup>; <18.5, 18.5–24.9, 25.0–29.9, or 30), smoking during pregnancy (yes or no), alcohol use from the last menstrual period through the first trimester (no drinks, <4 drinks/day, or 4 drinks/day), and genitourinary infections (yes or no) during pregnancy including: sexually transmitted infections (STI), vaginal infections, and kidney/bladder infections.

We present unweighted proportions or means and weighted proportion or mean estimates for all analyses. We examine birthweight and gestational age as both continuous and categorical outcomes (low birthweight, preterm birth, macrosomia, SGA and LGA). Unadjusted and adjusted generalized linear regressions were conducted to estimate mean differences in gestational age and birthweight. Unadjusted and adjusted Poisson regressions were conducted to estimate relative risks (RR) for low birthweight, preterm birth, macrosomia, SGA and LGA. Birthweight between 2500 and 4000 grams was used as the comparison for the low birthweight and macrosomia outcome models. Birthweight in the 10-90<sup>th</sup> percentile for gestational age was used as the comparison for the SGA and LGA outcome models. In the weighted models, we computed robust confidence intervals (CIs), a more conservative approach to account for the sampling weights. Adjusted models included race/ethnicity, as it was the only measured covariate meeting our definition of a potential confounder. Specifically, to be a confounder, a variable had to be related to and antecedent to both cNMD and birth outcomes. Due to the potential for adverse birth outcomes in multifetal pregnancies compared to singleton pregnancies, we conducted a sensitivity analyses only including singleton births (excluding SGA/LGA given these variable definitions only included singleton births). We used a total sample size of 609 women when excluding multiple births (n=19, 3% of the total sample) in the sensitivity analyses. We also conducted a sensitivity analysis where we used a 5% prevalence of infant congenital anomalies for weighting, instead of 3%. All analyses were conducted using SAS version 9.4.

#### RESULTS

Distributions of nearly all characteristics changed after weighting, although the relative, nominal comparisons of women with versus without cNMD were similar for some variables (Table 1). In the unweighted sample, infants with congenital anomalies were overrepresented in women with and without cNMD, 84.8% and 69.3% respectively, while reweighted results to account for the prevalence of infant with congenital anomalies produced a distribution of 6.1% and 2.6% respectively. Weighted results show that compared to women without cNMD, women with cNMD were more likely to self-identify as white, to have 12 years of education, be nulliparous before study pregnancy, have a prenatal visit before 8-weeks of gestation, have a caesarean delivery, and report vitamin use in the first trimester. Women with cNMD were less likely to have a planned pregnancy compared to women without cNMD were more likely to have a BMI 25 kg/m<sup>2</sup> (43.4% vs 30.0%), smoke during pregnancy (31.5% vs 19.5%), drink during the first trimester of pregnancy (52.0% vs 46.3%), report a vaginal infection during pregnancy (25.6% vs 17.8%) and report a kidney/bladder infection during pregnancy (20.6% vs 14.0%) compared to those without cNMD.

Table 2 shows the distributions of birth outcomes before and after weighting by cNMD status. In the weighted analysis, women with cNMD had infants with a lower mean birthweight (3302g vs 3408g) and a shorter mean gestational length (268 days vs 275 days) compared to women without cNMD. In weighted results, women with cNMD were more likely to have a preterm birth (20.8% vs 4.8%) and deliver an SGA infant (16.4% vs 10.0%) compared to women without cNMD. They were also less likely to have an infant with macrosomia (10.0% vs 13.4%) compared to those without cNMD. There were no observed

differences in low birthweight and LGA. Unweighted analyses generally yielded the same trends as weighted results.

Weighted, adjusted generalized linear regressions show that the average birthweight of infants born to women with cNMD was 137.22 grams less than infants delivered to women without cNMD (95% CI: -424.49, 150.05; Table 3). Women with cNMD also had a shorter average gestational length of 7.44 days compared to women without cNMD (95% CI: -13.94, -0.95). After weighting and adjustment, women with cNMD were more likely to have a preterm birth (RR=3.98, 95% CI: 1.33, 11.95) compared to women without cNMD. Women with cNMD were also more likely to deliver a SGA infant (RR=2.14, 95% CI: 0.74, 6.15) and had a moderately decreased risk of having an infant with macrosomia (RR=0.67, 95% CI: 0.17, 2.63), although estimates were imprecise. There were no observed differences for low birthweight and LGA.

Sensitivity analyses showed no appreciable changes to our interpretation of the findings when we restricted to singleton births (Ancillary material, Table 2) or when we used a 5% prevalence of infant congenital anomalies for weighting (Table 3). Proportions and means remained consistent and estimates for RRs and mean differences yielded similar estimates in direction and magnitude.

#### DISCUSSION

In these data, women with cNMD were at increased risk for certain adverse birth outcomes including preterm birth and having a small for gestational age infant, although the precise magnitude is unclear as our estimates were fairly imprecise. Findings from this study are consistent with growing literature that documents adverse birth outcomes for women with congenital neuromuscular disabilities, as well as individuals with other disabilities.

There are several possible explanations for the findings in the current study. First, several studies have documented that women with physical disabilities report disparities in their healthcare utilization before, during and after pregnancy, which may explain the birth outcome disparities found in the present study. In this study, 42% of women with cNMD reported having an unplanned pregnancy. This may be result of poor sexual health education during adolescence and young adulthood. Adolescent health research has documented that female adolescents with physical disabilities report receiving little education from health care providers about sexuality, fertility, and social relationships as they relate to disability, and stress the need for knowledge surrounding sexuality and fertility.<sup>27,28</sup> Despite our data showing that women with cNMD reported receiving earlier prenatal care more often than those without cNMD, studies have documented that women with disabilities receive suboptimal reproductive and prenatal care to meet their specific needs. For example, one study showed that due to lack of equipment accessibility, women reported not being routinely weighed and were examined in their chairs during prenatal appointments.<sup>10</sup> Women with physical disabilities have also reported that their healthcare providers are not adequately trained in providing appropriate reproductive care.<sup>29</sup> Poor communication between patient and providers, as well as inadequate health care can potentially result in various adverse outcomes.<sup>30</sup>

Women with physical disabilities are generally at an increased risk for experiencing secondary health conditions such as chronic pain, diabetes, obesity, high blood pressure, and genitourinary infections. These conditions may predispose women with disabilities to pregnancy complications and adverse birth outcomes.<sup>31,32</sup> We found that women with cNMD were more likely to have a higher BMI and report genitourinary infections compared to women without cNMD. Additionally, women with cNMD were more likely to smoke throughout pregnancy, a well-established risk factor for adverse outcomes and other pregnancy complications.<sup>33</sup> However interactions between cNMD and these other exposures with respect to birth outcomes could not be assessed in the present study due to small sample sizes. Lastly, other studies have found that women with physical disabilities are more likely to experience depression, stress, anxiety and interpersonal violence, all potential risk factors for adverse pregnancy outcomes.<sup>34–37</sup> For example, one study found that women who experience intimate partner violence before and during pregnancy had an increased risk for preterm deliveries and having a low birthweight infant.<sup>38</sup> Similar findings have been documented for women who report depression, stress, anxietys, and anxiety.<sup>39</sup>

There are some limitations to the study. First, the number of women who report having cNMD is small, which affects precision of our study findings. To maximize the number of women affected by cNMD in the analysis, we included data from the entire span of the Slone BDS. To control for possible time trends, we matched by interview year. The broad study period may mean that the covariate distributions, and clinical practices affecting pregnant women with cNMD, are not representative of a more current sample. Given the small sample size, we were not able to stratify birth outcomes by diagnosis type to assess differences in outcomes. Second, there may be interest in understanding medically indicated preterm deliveries due to cNMD status versus those which are naturally occurring. Unfortunately, this distinction was not systematically collected via the BDS interview. We did thoroughly review the free-text notes within each interview to assess whether there was any medical indication for preterm delivery among women with cNMD and found no such relationships. Third, given the original case-control sampling scheme, pregnancies where an infant was born with a congenital anomaly were far more prevalent in both the cNMD and without cNMD groups than would be expected in the general population. To address concern of bias due to overrepresentation of infants with congenital anomalies, we used a method which heavily upweighted pregnancies not affected by a congenital anomaly. We computed weighted estimates based on 3% and 5% prevalence of infant congenital anomalies. Specifically, in the main analysis, each of the 174 infant controls represented 84.7 individuals in the weighted analysis. We computed robust 95% CIs to account for the variability due to the extreme weights. We also presented the unadjusted and adjusted results, all of which demonstrated similar associations in terms of direction. Still, it is a limitation that we relied on U.S. national estimates rather than having metropolitan specific estimates to assign the weights. Controls that are not representative of the underlying source population could bias the associations observed in the study. We reviewed the distribution of the birth outcomes among the infant controls with no congenital anomalies (data not shown), and they followed the expected distribution based on U.S. estimates for livebirths with one exception. Our estimate of preterm birth in the weighted data among women without cNMD was lower than would be expected based on the general North American population (5% in

the current study). This may have led to an overestimate of the relative increase in preterm birth for women with cNMD compared to women without cNMD. However, to rule out the possibility of the observed association solely due to bias, we explored additional analyses and found that the RR remained elevated even under extreme selection bias scenarios (e.g., 50% of women with cNMD and term births are missing). Lastly, there is potential due to bias from other sources of systematic error. We controlled for a few covariates (year, study site, and race/ethnicity). Conceptually, there may not be many common causes of both maternal cNMD and their birth outcomes, given the early occurrence of cNMD in the lifespan. Many covariates that one may wish to adjust for in analysis, such as maternal age, may be a downstream effect of having a cNMD or not, rather than a precursor. Also, information reported by the women on pregnancy outcomes may be subject to recall errors. However, an interval validation study within in the Slone BDS found good agreement with the medical record for gestational age and weight at delivery.<sup>40</sup>

Despite these limitations, we were able to examine birth outcomes among a group of women with congenital physical disabilities, a population that has not been widely studied. Most literature has focused specifically on women with spina bifida or cerebral palsy. Women with muscular dystrophy, arthrogryposis or contractures have not been included in many previous studies and we were able to include them in the current study. Additionally, we utilized the reweighting approach to account for the prevalence of infants with congenital anomalies in the general population, which allowed us to utilize a unique data source to examine birth outcomes among women with cNMD. Given the scarce data on birth outcomes among this population, we urge researchers to use novel approaches and data sources to further understand pregnancy outcomes and health care needs among women with cNMD.

#### CONCLUSION

Our study contributes to growing literature that women with disabilities are more likely to experience chronic conditions and adverse birth outcomes compared to their counterparts, highlighting the need for further research to identify ways to reduce adverse experiences and birth outcomes in this population. Further research is needed to understand the experiences of women with cNMD during the preconception, pregnancy, and post-partum periods to develop interventions and policies that will ensure they receive the appropriate resources and care they need for optimal pregnancy and birth outcomes.

#### Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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#### References

- Okoro CA, Hollis ND, Cyrus AC, Griffin-Blake S. Prevalence of Disabilities and Health Care Access by Disability Status and Type Among Adults — United States, 2016. MMWR Morb Mortal Wkly Rep. 2018;67(32):882–887. doi:10.15585/mmwr.mm6732a3 [PubMed: 30114005]
- Signore C, Spong CY, Krotoski D, Shinowara NL, Blackwell SC. Pregnancy in women with physical disabilities. Obstet Gynecol. 2011;117(4):935–947. doi:10.1097/AOG.0b013e3182118d59 [PubMed: 21422868]
- Glinianaia SV, Morris JK, Best KE, et al. Long-term survival of children born with congenital anomalies: A systematic review and meta-analysis of population-based studies. PLoS Med. 2020;17(9):e1003356. doi:10.1371/journal.pmed.1003356 [PubMed: 32986711]
- Mitra M, Clements KM, Zhang J, Iezzoni LI, Smeltzer SC, Long-Bellil LM. Maternal characteristics, pregnancy complications, and adverse birth outcomes among women with disabilities. Med Care. 2015;53(12):1027–1032. doi:10.1097/MLR.000000000000427 [PubMed: 26492209]
- Shepard CL, Yan PL, Hollingsworth JM, Kraft KH. Pregnancy among mothers with spina bifida. J Pediatr Urol. 2018;14(1):11.e1–11.e6. doi:10.1016/j.jpurol.2017.08.001 [PubMed: 28943353]
- Mitra M, McKee MM, Akobirshoev I, et al. Pregnancy, Birth, and Infant Outcomes Among Women Who Are Deaf or Hard of Hearing. Am J Prev Med. 2020;58(3):418–426. doi:10.1016/ j.amepre.2019.10.012 [PubMed: 31952943]
- 7. Smeltzer SC. Pregnancy in Women With Physical Disabilities. J Obstet Gynecol Neonatal Nurs. 2007;36(1):88–96. doi:10.1111/j.1552-6909.2006.00121.x
- Tarasoff LA, Ravindran S, Malik H, Salaeva D, Brown HK. Maternal disability and risk for pregnancy, delivery, and postpartum complications: a systematic review and meta-analysis. Am J Obstet Gynecol. 2020;222(1):27.e1–27.e32. doi:10.1016/j.ajog.2019.07.015 [PubMed: 31306650]
- Tarasoff LA, Murtaza F, Carty A, Salaeva D, Hamilton AD, Brown HK. Health of Newborns and Infants Born to Women With Disabilities: A Meta-analysis. Pediatrics. November 2020:e20201635. doi:10.1542/peds.2020-1635 [PubMed: 33203648]
- Iezzoni LI, Wint AJ, Smeltzer SC, Ecker JL. Physical accessibility of routine prenatal care for women with mobility disability. J Women's Heal. 2015;24(12):1006–1012. doi:10.1089/ jwh.2015.5385
- Mitra M, Long-Bellil LM, Iezzoni LI, Smeltzer SC, Smith LD. Pregnancy among women with physical disabilities: Unmet needs and recommendations on navigating pregnancy. Disabil Health J. 2016;9(3):457–463. doi:10.1016/j.dhjo.2015.12.007 [PubMed: 26847669]
- Iezzoni LI, Wint AJ, Smeltzer SC, Ecker JL. "how did that happen?" Public responses to women with mobility disability during pregnancy. Disabil Health J. 2015;8(3):380–387. doi:10.1016/ j.dhjo.2015.02.002 [PubMed: 25944504]
- Shepard CL, Yan PL, Kielb SJ, et al. Complications of Delivery Among Mothers With Spina Bifida. Urology. 2019;123:280–286. doi:10.1016/j.urology.2018.04.045 [PubMed: 29908216]
- Rubenstein E, Ehrenthal DB, Mallinson DC, Bishop L, Kuo H, Durkin MS. Birth outcomes affecting infants of mothers with intellectual and developmental disabilities. Paediatr Perinat Epidemiol. May 2021:ppe.12765. doi:10.1111/ppe.12765
- Sundelin HEK, Stephansson O, Johansson S, Ludvigsson JF. Pregnancy outcome in women with cerebral palsy: A nationwide population-based cohort study. Acta Obstet Gynecol Scand. 2020;99(4):518–524. doi:10.1111/aogs.13773 [PubMed: 31738455]
- Johnson NE, Hung M, Nasser E, et al. The Impact of Pregnancy on Myotonic Dystrophy: A Registry-Based Study. J Neuromuscul Dis. 2015;2(4):447–452. doi:10.3233/JND-150095 [PubMed: 27858748]

- Awater C, Zerres K, Rudnik-Schöneborn S. Pregnancy course and outcome in women with hereditary neuromuscular disorders: Comparison of obstetric risks in 178 patients. Eur J Obstet Gynecol Reprod Biol. 2012;162(2):153–159. doi:10.1016/j.ejogrb.2012.02.020 [PubMed: 22459654]
- Hayward K, Chen AY, Forbes E, Byrne R, Greenberg MB, Fowler EG. Reproductive healthcare experiences of women with cerebral palsy. Disabil Health J. 2017;10(3):413–418. doi:10.1016/ j.dhjo.2017.03.015 [PubMed: 28428111]
- McConnell D, Mayes R, Llewellyn G. Women with intellectual disability at risk of adverse pregnancy and birth outcomes. J Intellect Disabil Res. 2008;52(6):529–535. doi:10.1111/ j.1365-2788.2008.01061.x [PubMed: 18422528]
- 20. Rubenstein E, Ehrenthal DB, Mallinson DC, Bishop L, Kuo HH, Durkin M. Pregnancy complications and maternal birth outcomes in women with intellectual and developmental disabilities in Wisconsin Medicaid. PLoS One. 2020;15(10 October). doi:10.1371/ journal.pone.0241298
- Morton C, Le JT, Shahbandar L, Hammond C, Murphy EA, Kirschner KL. Pregnancy Outcomes of Women With Physical Disabilities: A Matched Cohort Study. PM R. 2013;5(2):90–98. doi:10.1016/j.pmrj.2012.10.011 [PubMed: 23200116]
- Correa A, Cragan JD, Kucik ME, et al. Reporting birth defects surveillance data 1968–2003. Birth Defects Res A Clin Mol Teratol. 2007;79(2):65–186. doi:10.1002/bdra.20350 [PubMed: 17278144]
- Oken E, Kleinman KP, Rich-Edwards J, Gillman MW. A nearly continuous measure of birth weight for gestational age using a United States national reference. BMC Pediatr. 2003;3(1):6. doi:10.1186/1471-2431-3-6 [PubMed: 12848901]
- 24. Richardson DB, Rzehak P, Klenk J, Weiland SK. Analyses of case-control data for additional outcomes. Epidemiology. 2007;18(4):441–445. doi:10.1097/EDE.0b013e318060d25c [PubMed: 17473707]
- Dolan SM, Gross SJ, Merkatz IR, et al. The contribution of birth defects to preterm birth and low birth weight. Obstet Gynecol. 2007;110(2 I):318–324. doi:10.1097/01.AOG.0000275264.78506.63 [PubMed: 17666606]
- 26. Hoyert DL, Mathews TJ, Menacker F, Strobino DM, Guyer B. Annual summary of vital statistics: 2004. Pediatrics. 2006;117(1):168–183. doi:10.1542/peds.2005-2587 [PubMed: 16396875]
- 27. Heller MK, Gambino S, Church P, Lindsay S, Kaufman M, McPherson AC. Sexuality and Relationships in Young People With Spina Bifida and Their Partners. J Adolesc Heal. 2016;59(2):182–188. doi:10.1016/j.jadohealth.2016.03.037
- Verhoef M, Barf HA, Vroege JA, et al. Sex education, relationships, and sexuality in young adults with spina bifida. Arch Phys Med Rehabil. 2005;86(5):979–987. doi:10.1016/j.apmr.2004.10.042 [PubMed: 15895345]
- Streur CS, Schafer CL, Garcia VP, Quint EH, Sandberg DE, Wittmann DA. "If Everyone Else Is Having This Talk With Their Doctor, Why Am I Not Having This Talk With Mine?": The Experiences of Sexuality and Sexual Health Education of Young Women With Spina Bifida. J Sex Med. 2019;16(6):853–859. doi:10.1016/j.jsxm.2019.03.012 [PubMed: 31010781]
- Sword W, Heaman MI, Brooks S, et al. Women's and care providers' perspectives of quality prenatal care: A qualitative descriptive study. BMC Pregnancy Childbirth. 2012;12(1):29. doi:10.1186/1471-2393-12-29 [PubMed: 22502640]
- Mitra M, Clements KM, Zhang J, Smith LD. Disparities in Adverse Preconception Risk Factors Between Women with and Without Disabilities. Matern Child Health J. 2016;20(3):507–515. doi:10.1007/s10995-015-1848-1 [PubMed: 26518009]
- 32. Kim M, Kim HJ, Hong S, Fredriksen-Goldsen KI. Health disparities among childrearing women with disabilities. Matern Child Health J. 2013;17(7):1260–1268. doi:10.1007/s10995-012-1118-4 [PubMed: 22918712]
- Ion R, Bernal AL. Smoking and preterm birth. Reprod Sci. 2015;22(8):918–926. doi:10.1177/1933719114556486 [PubMed: 25394641]

- 34. Mitra M, Manning SE, Lu E. Physical abuse around the time of pregnancy among women with disabilities. Matern Child Health J. 2012;16(4):802–806. doi:10.1007/s10995-011-0784-y [PubMed: 21556697]
- Nosek MA, Hughes RB, Robinson-Whelen S. The complex array of antecedents of depression in women with physical disabilities: Implications for clinicians. Disabil Rehabil. 2008;30(3):174– 183. doi:10.1080/09638280701532219 [PubMed: 17852240]
- Smith KJ, Peterson MD, O'Connell NE, et al. Risk of Depression and Anxiety in Adults with Cerebral Palsy. JAMA Neurol. 2019;76(3):294–300. doi:10.1001/jamaneurol.2018.4147 [PubMed: 30592485]
- 37. Sabharwal R The link between stress disorders and autonomic dysfunction in muscular dystrophy. Front Physiol. 2014;5 JAN. doi:10.3389/fphys.2014.00025
- 38. Silverman JG, Decker MR, Reed E, Raj A. Intimate partner violence victimization prior to and during pregnancy among women residing in 26 U.S. states: Associations with maternal and neonatal health. Am J Obstet Gynecol. 2006;195(1):140–148. doi:10.1016/j.ajog.2005.12.052 [PubMed: 16813751]
- Alder J, Fink N, Bitzer J, Hösli I, Holzgreve W. Depression and anxiety during pregnancy: A risk factor for obstetric, fetal and neonatal outcome? A critical review of the literature. J Matern Neonatal Med. 2007;20(3):189–209. doi:10.1080/14767050701209560
- Petersen JM, Mitchell AA, Van Bennekom C, Werler MM. Validity of maternal recall of gestational age and weight at birth: Comparison of structured interview and medical records. Pharmacoepidemiol Drug Saf. 2019;28(2):269–273. doi:10.1002/pds.4699 [PubMed: 30488571]

#### Table 1.

Descriptive characteristics of women in the Slone Birth Defects Study, 1976–2015; by maternal congenital neuromuscular disability status

	Unweig	hted, n (%)	Weig	hted, (%)
	cNMD (n=125)	No cNMD (n=505)	cNMD	No cNMD
Infant with Congenital Anomaly				
Yes	106 (84.80)	350 (69.31)	(6.18)	(2.60)
No	19 (15.20)	155 (30.69)	(93.82)	(97.40)
Infant Sex				
Male	75 (60.00)	279 (55.25)	(58.05)	(44.30)
Female	50 (40.00)	226 (46.00)	(41.95)	(55.70)
Mother Age				
<20	9 (7.20)	36 (7.13)	(0.52)	(5.23)
20–24	30 (24.00)	82 (16.24)	(21.27)	(16.75)
25–29	35 (28.00)	146 (28.91)	(26.44)	(27.79)
30	51 (40.80)	241 (47.72)	(51.77)	(50.23)
Race/Ethnicity				
Non-Hispanic White	104 (83.20)	385 (76.24)	(89.02)	(75.51)
Non-Hispanic Black	10 (8.00)	35 (6.93)	(5.46)	(6.47)
Hispanic	9 (7.20)	60 (11.88)	(5.40)	(12.24)
Other	2 (1.60)	25 (4.95)	(0.12)	(5.77)
Education				
<12 years	18 (14.40)	59 (11.68)	(1.05)	(10.99)
12 years	33 (26.40)	115 (22.77)	(26.32)	(12.65)
>12 years	74 (59.20)	331 (65.54)	(72.63)	(76.35)
Parity				
0	65 (52.00)	206 (40.79)	(47.71)	(39.41)
1	38 (30.40)	194 (38.42)	(26.61)	(44.91)
2	15 (12.00)	76 (15.05)	(10.63)	(12.98)
3	7 (5.60)	29 (5.74)	(15.05)	(2.70)
Gravidity				
1	50 (40.00)	155 (30.75)	(41.95)	(29.72)
2	33 (26.40)	155 (30.75)	(21.44)	(33.44)
3	42 (33.60)	194 (38.52)	(36.61)	(36.84)
Missing	-	1	-	-
Pregnancy Planned				
Yes	61 (53.98)	290 (63.46)	(57.64)	(66.77)
No	52 (46.02)	167 (36.54)	(42.36)	(33.23)
Missing *	12	48	-	-
Plurality				
Singleton	121 (97.58)	488 (96.83)	(99.83)	(99.26)
	() (100)		(,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,	(,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,

	Unweig	hted, n (%)	Weig	nted, (%)
	cNMD (n=125)	No cNMD (n=505)	cNMD	No cNME
Multiple	3 (2.42)	16 (3.17)	(0.17)	(0.74)
Missing	1	1	-	-
First Prenatal Visit				
8 weeks	47 (56.63)	184 (54.72)	(68.79)	(60.15)
> 8 weeks	36 (43.37)	155 (45.72)	(31.21)	(39.85)
Missing $^{\dagger}$	42	166	-	-
Delivery Type				
Vaginal	62 (53.45)	314 (67.09)	(57.59)	(77.53)
Cesarean Section	54 (46.55)	154 (32.91)	(42.41)	(22.47)
Missing <sup>‡</sup>	9	37	-	-
Pre-pregnancy BMI (kg/m <sup>2</sup> )				
Underweight (<18.5)	8 (8.99)	22 (6.25)	(6.42)	(4.90)
Normal (18.5-24.9)	47 (52.81)	206 (58.52)	(50.17)	(65.09)
Overweight (25.0-29.9)	16 (17.98)	79 (22.44)	(30.42)	(18.85)
Obese ( 30.0)	18 (20.22)	45 (12.78)	(12.98)	(11.16)
Missing	36	153	-	-
Smoking during pregnancy				
Yes	38 (30.40)	119 (23.56)	(31.50)	(19.50)
No	87 (69.60)	386 (76.44)	(68.50)	(80.50)
Alcohol (LMP – 1 <sup>st</sup> trimester)				
0 drinks	70 (56.00)	290 (57.43)	(48.00)	(53.69)
<4 drinks	40 (32.00)	165 (32.67)	(46.25)	(36.00)
4 drinks	15 (12.00)	50 (9.90)	(5.75)	(10.31)
Sexually Transmitted Infection				
Yes	2 (1.60)	12 (2.38)	(0.12)	(3.19)
No	123 (98.40)	493 (97.62)	(99.88)	(96.81)
Vaginal Infection				
Yes	21 (16.80)	64 (12.67)	(25.62)	(17.86)
No	104 (83.20)	441 (87.33)	(74.38)	(82.14)
Kidney/Bladder Infection				
Yes	19 (15.20)	46 (9.11)	(20.63)	(14.00)
No	106 (84.80)	459 (90.89)	(79.37)	(86.00)
Vitamin Use in 1st trimester				
Yes	109 (87.20)	415 (82.83)	(99.06)	(91.75)
No	16 (12.80)	86 (17.17)	(0.93)	(8.25)
Missing	0	4	-	-

\*Question not asked 1976–1983

 $^{\dagger}$ Question not asked 1976–1983 and 1993–1998

<sup>‡</sup>Question not asked 1993–1998

<sup>§</sup>Question not asked 1976–1992

BMI body-mass-index, cNMD congenital neuromuscular disability, LMP last menstrual period

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# Table 2.

Unweighted and weighted birth outcomes comparing women with and without congenital neuromuscular disabilities in the Slone Birth Defects Study 1976–2015

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	Unwe	Unweighted	Weig	Weighted
N (%)	cNMD (n=125)	No cNMD (n=505)	cNMD	No cNMD
Birthweight				
< 2500g	19 (15.20)	70 (13.86)	(5.99)	(6.11)
2500g - 4000g	101 (80.80)	385 (76.24)	(83.96)	(80.48)
> 4000g	5 (4.00)	50 (9.90)	(10.05)	(13.41)
Birthweight (g), mean $\pm$ SD	$3068.55 \pm 630.02$	$3245.25 \pm 664.05$	$3302.95 \pm 2326.30$	$3408.11 \pm 2861.28$
Preterm Birth (<37 weeks)				
Yes	22 (17.60)	69 (13.66)	(20.80)	(4.86)
No	103 (82.40)	436 (86.34)	(79.20)	(95.14)
Early Preterm Birth (<32 weeks)				
Yes	3 (2.40)	10 (1.98)	(5.05)	(0.70)
No	122 (97.60)	495 (98.02)	(94.95)	(99.30)
Gestational Age (days), mean ± SD	$269.86 \pm 15.36$	$272.27 \pm 16.55$	$268.04 \pm 54.35$	$275.54 \pm 66.04$
Size for Gestational Age				
< 10 <sup>th</sup> percentile (SGA)	29 (24.17)	81 (16.63)	(16.40)	(10.00)
10 <sup>th</sup> – 90 <sup>th</sup> percentile	86 (71.67)	366 (75.15)	(73.54)	(81.57)
> 90 <sup>th</sup> percentile (LGA)	5 (4.17)	40 (8.21)	(10.06)	(8.43)

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# Table 3.

Weighted birth outcomes comparing women with and without congenital neuromuscular disabilities in the Slone Birth Defects Study 1976–2015

		weigntea, 5%	mrgrau	weighten, 570
	Unadjusted RR or Mean Difference (95% CI)	Adjusted RR or Mean Difference (95% CI)*	Unadjusted RR or Mean Difference (95% CI)	Adjusted RR or Mean Difference (95% CI)*
Birthweight				
< 2500g	1.05 (0.19, 5.65)	1.25 (0.22, 7.03)	1.08 (0.24, 4.92)	1.27 (0.27, 5.93)
$2500\mathrm{g}-4000\mathrm{g}$	ref	ref	ref	ref
> 4000g	0.75 (0.20, 2.86)	$0.67\ (0.17,\ 2.63)$	0.73 (0.19, 2.76)	0.66(0.17, 2.54)
Birthweight (g), mean $\pm$ SD	-105.16 (-380.79, 170.47)	-137.22 (-424.49, 150.05)	-112.78 (-377.59, 152.02)	-143.36(-418.90, 132.19)
Preterm Birth (<37 weeks)				
Yes	4.28 (1.49, 12.30)	3.98 (1.33, 11.95)	4.05 (1.48, 11.09)	3.76 (1.32, 10.70)
No	ref	ref	ref	ref
Gestational Age (days), mean $\pm$ SD	-7.50 (-13.93, -1.06)	-7.44(-13.94, -0.95)	-7.32 (-13.49, -1.14)	-7.25 (-13.48, -1.02)
Size for Gestational Age				
< 10 <sup>th</sup> percentile (SGA)	$1.68\ (0.60,4.71)$	2.14 (0.74, 6.15)	1.69(0.64, 4.44)	2.11 (0.78, 5.68)
$10^{th} - 90^{th}$ percentile	ref	ref	ref	ref
> 90 <sup>th</sup> percentile (LGA)	$1.29\ (0.33, 5.10)$	1.20(0.31, 4.68)	$1.26\ (0.33, 4.88)$	1.18(0.31, 4.49)

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CI confidence interval, g grams, LGA large-for-gestational age, ref reference, RR risk ratio, SD standard deviation, SGA small-for-gestational age