HHS Public Access

Author manuscript

Int J Care Coord. Author manuscript; available in PMC 2019 December 01.

Published in final edited form as:

Int J Care Coord. 2018 December ; 21(4): 140–152. doi:10.1177/2053434518818448.

Changes in care coordination and health insurance in the population of US children with muscular dystrophy, 2005–2006 and 2009–2010

Jaimie Z Shing[1], **Tiebin Liu**[2], and **Rodolfo Valdez**[2]

[1]Vanderbilt University, USA

[2]Centers for Disease Control and Prevention, USA

Abstract

Introduction: We aimed to assess changes in care coordination and health insurance coverage among US children with muscular dystrophy.

Methods: We used 2005–2006 and 2009–2010 data from the National Survey of Children with Special Health Care Needs. We examined the distribution of sociodemographic and health characteristics of children with muscular dystrophy by survey cycle. Multivariable regression was used to calculate odds of not receiving effective care coordination, not having adequate health insurance coverage, receiving no help coordinating care, and having problems obtaining referrals in each survey cycle.

Results: In the 2005–2006 and 2009–2010 survey cycles, there were 135 and 117 children with muscular dystrophy (representing 34,672 and 31,169 US children with muscular dystrophy), respectively. The percentage of children with muscular dystrophy who did not receive effective care coordination changed from 59.2% (95% confidence interval (CI), 45.6%–72.7%) in 2005–2006 to 53.4% (95% CI, 38.3%–68.6%) in 2009–2010. The odds of not receiving effective care coordination (adjusted odds ratio (aOR) = 0.77; 95% CI, 0.32–1.89) or having problems obtaining referrals (aOR = 0.52; 95% CI, 0.17–1.59) did not change significantly between the two periods, whereas odds of having inadequate insurance coverage decreased significantly (aOR = 0.41, 95% CI, 0.18–0.93) and odds of not receiving help coordinating care increased significantly (aOR = 4.22, 95% CI, 1.24–14.29) between the two periods.

Corresponding author: Rodolfo Valdez, Division of Human Development and Disability, National Center on Birth Defects and Developmental Disabilities, Centers for Disease Control and Prevention, 1600 Clifton Rd. NE, Mailstop E88, Atlanta, GA 30333, USA. rvaldez@cdc.gov.

Author contributions

All authors contributed to the design and execution of the study, including analysis of the data, presentation and interpretation of the results, and writing of the manuscript.

Ethical approval

This study is exempt from IRB approval because it used existing, publicly available data from unidentified individuals.

Supplemental material

Supplemental material for this article is available online.

Declarations of conflicting interests

The authors declared no potential conflicts of interest related to the productions and publication of this article.

Conclusion: Our results suggest key health care needs for many families with children with muscular dystrophy have remained unmet for a prolonged period. Although there were significant improvements in health insurance coverage, nearly one-third of children with muscular dystrophy still had inadequate health insurance coverage in 2009–2010; it is likely that this situation has not changed much since then.

Keywords

Muscular dystrophies; health insurance; health service needs

Introduction

Care coordination has many definitions[1]; however, it is generally recognized as a multilevel, organized approach to care that links children and their families with necessary health services, health care providers/ professionals, resources, assistance, and communication.[2,3] The US Agency for Healthcare Research and Quality defines care coordination as the deliberate organization of patient care activities between two or more participants (including the patient) involved in a patient's care to facilitate the appropriate delivery of health care services.[4] Coordinated care is recommended for children with special health care needs (CSHCN) because these children require more assistance with daily living activities and coordinated specialty care, as well as use health services more frequently than their peers without such needs.

Coordinated care has been suggested to reduce unmet specialty care needs and limitations in activity/ body functions, particularly among CSHCN who are receiving such care in medical homes.[5–7] Studies have shown that the health care needs of CSHCN are consistently unmet due to multiple barriers that make it difficult for these children to obtain health care referrals, receive coordinated care services, and use medical homes, such as having inadequate health insur-ance.[5,8–10]

One subgroup of CSHCN is symptomatic children with muscular dystrophy (MD), which is a genetic disease characterized by progressive and irreversible skeletal muscle weakness and degeneration.[11] The major types of MD vary in severity, muscle groups affected, gene/ mutation involved, and age of onset.[12,13] In children, Duchenne and Becker are the most commonly inherited types of MD and mostly affect males,[2] with an estimated combined prevalence of 1.4 per 10,000 males aged 5–24 years in the United States (US).[14]

Compared to other CSHCN, children with MD are more likely to report greater difficulties with functionality and self-care, and have family members who spend more time coordinating care, have financial problems, and have reduced or leaves from employ-ment.[15] Among children covered by the same type of health insurance, health care expenditures for children with MD are about 13 times higher than comparable expenditures for children without MD.[16] Thus, effective care coordination could potentially alleviate these financial burdens on families of CSHCN in general[17,18] and of CSHCN with MD in particular.

Currently, there is no cure for any type of MD.[19,20] Therefore, it is important that children with MD have access to care, therapy, and treatments to improve their health and quality of

life. Some studies have examined the effects of care coordination on health care needs, barriers to care, and disabilities among CSHCN[2,3,5–8,21]; however, no study has examined care coordination specifically among US children with MD. This study examines changes in care coordination and health insurance coverage among children with MD in the US. We hypothesized that for these children, care coordination status and health insurance coverage adequacy have not changed, as one study analyzing health care utilization from 2005 to 2011 among CSHCN overall found quality and utilization of health care services did not improve over time.[8] Our paper will also address the major changes that have occurred in the past decade in the US health care system and how this may have had an impact on care coordination, specifically for US children with MD.

Methods

Study design and data source

Our study is based on two cross-sectional, population-based surveys of US children. We used data from the National Survey of Children with Special Health Care Needs (NS-CSHCN), a complex, random digit-dial telephone-based survey conducted by the Centers for Disease Control and Prevention's National Center for Health Statistics. The NS-CSHCN includes information on CSHCN aged 0–17 years and their families from the non-institutionalized population in all 50 US states and the District of Colombia. General topics covered in the NS-CSHCN include children's health/functional status, health insurance (e.g., access to care and adequacy of coverage), care coordination, and family and financial impacts.

We requested and received 2005–2006 and 2009–2010 NS-CSCHN datasets prepared by the Data Resource Center for Child and Adolescent Health as part of the Child and Adolescent Health Measurement Initiative.[22,23] The data included demographic characteristics, variables to determine CSHCN status, and interview data for all CSHCN. The parent or guardian most knowledgeable about the health status and care of the children in the household responded to the survey as a proxy for the participants.

The NS-CSHCN has been conducted three times (2001–2002, 2005–2006, 2009–2010); however, we only examined data from the latter two periods, which included questions about MD status, allowing us to examine children with MD as a stratum of the general CSHCN population using complex survey methodology. Interviews completed from April 2005 to February 2007 were included in the 2005–2006 NS-CSHCN survey cycle, while interviews completed from July 2009 to March 2011 were included in the 2009–2010 NS-CSHCN survey cycle. Both survey cycles used similar questions; however, the 2009–2010 survey included additional topics such as attention-deficit/ hyperactivity disorder, alternative health care, and brain injury. Additionally, a sample of cell phone users was included in the 2009–2010 NS-CSHCN survey cycle to examine the effect of increased usage of cell phones compared with landline phones.

The average time of the interview was 28 min for the 2005–2006 survey and 33 min for the 2009–2010 survey. Each participant was interviewed after verbally consenting to the interview and being assured of confidentiality.[24,25] For the 2005–2006 NS-CSHCN, the

overall national weighted response rate for all children was 56.1%, but the proportion of interviews completed by children with special needs, once selected within a household, was 96.2%. For the 2009–2010 survey cycle, the national weighted response rates were 43.7% for the landline phone sample and 15.2% for the mobile phone sample (25.5% overall), but the combined proportion of interviews completed by children with special needs, once selected within a household, was 80.8%.[24,25] More detailed information about data collection, survey design, and the questionnaires can be found elsewhere.[24,25]

Description of variables

In the 2005–2006 survey, MD status was established by answering "yes" when asked "to the best of your knowledge, does your child currently have MD?" In the 2009–2010 survey, participants were asked whether their child ever had MD and if yes, whether their child currently had MD. For consistency, we defined children with MD in 2009–2010 as a "yes" response to the second question: "does your child currently have muscular dystrophy?" Our methods, as stated below, also considered that some survey participants answered the questionnaire in Spanish.

We assessed four outcomes: did not receive effective care coordination, had inadequate current health insurance coverage, received no help coordinating care, and had problems obtaining referrals. As predictor variables, we included several socio-demographic characteristics (i.e., sex, age, race/ethnicity, family income, and education level) and health-related characteristics (i.e., activity limitations, usual source for sick care, family-centered care, and current insurance type). All variables were used as prepared by the Data Resource Center.[22,23]

Effective care coordination was evaluated using the Child and Adolescent Health Measurement Initiative algorithm, which was part of a larger algorithm for evaluating the status of the care provided by medical homes. Details about this algorithm can be found else-where.[5] Effective care coordination was determined by affirmative responses to usually or always getting sufficient help coordinating care when needed and being very satisfied with communication among doctors and other health care providers, and among doctors and the child's school or other children programs when needed.

Adequacy of current health insurance coverage was determined by answers to the following questions: (1) Does the child's health insurance offer benefits or cover services that meet his/her needs? (2) Are costs not covered by the child's health insurance (out-of-pocket costs) reasonable? and (3) Does the child's health insurance allow him/her to see the health care providers he/ she needs? Health insurance coverage was considered adequate if the respondent answered "usually" or "always" to all three questions.

Revisions and changes between surveys should be noted when comparing survey cycles. First, in 2009–2010, the variable race/ethnicity was missing in more participants than expected during screening. Therefore, the Data Resource Center imputed all missing values for this variable during this period. Second, in 2005–2006, single imputation was used for missing family incomes (median value for the sample), whereas in 2009–2010, such incomes were imputed using multiple imputation. Third, in 2005–2006, participants were

asked for the highest level of education attained by anyone in the household, but 2009–2010 participants were asked for the highest level of education attained by each parent in the household.

Statistical analyses

We determined the weighted percent distributions of the selected socio-demographic and health characteristics among children with MD by survey cycle. Although the analyses were restricted to children with MD, all children in each survey were retained in the sample for the correct estimation of standard errors. We used Rao-Scott chi-square tests to assess the bivariate associations between survey cycle and the selected characteristics. We also determined the weighted percent distributions of the selected characteristics among children with MD who did not receive effective care coordination and those who had inadequate health insurance coverage by survey cycle. We used Rao-Scott chi-square tests to assess the bivariate associations between survey cycle and each characteristic among children with each outcome.

Using multivariable logistic regression, we estimated crude and adjusted odds of not receiving effective care coordination, having inadequate current health insurance coverage, receiving no help coordinating care, and having problems obtaining referrals among children with MD, comparing the 2009–2010 survey cycle to the 2005–2006 survey cycle. The models tested are depicted in Figure 1. Before these estimations, we performed analyses of collinearity among all predictor variables to be included in our models. These variables include socio-demographic variables (sex, age, race/ ethnicity, family income, and education level) and health care-related variables (activity limitations, usual source for sick care, family centered care, and current insurance type). We used variance inflation factors to assess multi-collinearity in each regression model. Variables with high variance inflation factors (five or greater) were deemed redundant in our model. Finally, all non-redundant variables were included in a backward selection model in which only those yielding p values < 0.10 were retained in the final model. We use this conservative p value to account for multiple testing in each model. As a result, in addition to adjusting for sex, age, and race/ ethnicity in each multivariable model, (1) the model for “did not receive effective care coordination” additionally adjusted for usual source for sick care, (2) the model for “had inadequate current health insurance” additionally adjusted for usual source for sick care and current insurance type, (3) the model for “received no help coordinating care” additionally adjusted for activity limitations, family-centered care, and current insurance type, and (4) the model for “had problems obtaining referrals” additionally adjusted for education level, family-centered care, and current insurance type.

Due to the increased number of cell phone users in 2009–2010, both the state location of the participant and the sampling type (landline or cell phone) were considered as strata in the sampling design to obtain unbiased variance estimations. Interview weights were applied to generalize the findings to CSHCN aged 0–17 years to the non-institutionalized US population. P v alues < 0.05 were considered statistically significant except when otherwise noted (e.g., backward selection procedures). All statistical analyses were performed using SAS version 9.4 (SAS Institute Inc., Cary, NC, USA).

Sensitivity analyses

We examined two methodological concerns using subsequent models. First, in previous studies, either the number of CSHCN were under-reported[26] or the number of MD cases were over-reported[15] by Hispanics who answered the survey in Spanish. We confirmed this over-reporting of MD in our data. Thus, we compared all models listed in Figure 1 before and after excluding participants who answered the survey in a language other than English. Second, only for model 2 shown in Figure 1, we conducted a secondary analysis that compared models with and without current insurance type to explore the possibility of collinearity between inadequacy of health insurance and current insurance type.

Results

Characteristics of children with MD

In the 2005–2006 NS-CSHCN cycle, 135 parents or guardians reported that their child currently had MD, representing 34,672 US children with MD; in the 2009–2010 NS-CSHCN cycle, 117 parents or guardians reported that their child currently had MD, representing 31,169 US children with MD. Compared to children with MD in 2005–2006, children with MD in 2009–2010 had similar distributions of characteristics, except that children in the 2005–2006 survey were more likely to live with two parents that were either biological or adoptive (69% vs. 44%), and to have adequate current health insurance coverage (70% vs. 49%) (Table 1, $p < 0.05$).

In both survey cycles, public health insurance was the most common type of insurance and the group of children with MD who did not receive effective care coordination was larger. In the most recent period (2009–2010), 53.4% (95% CI, 38.3%–68.6%) of children with MD did not receive effective care coordination, 30.2% (95% CI, 17.6%–42.8%) had inadequate current health insurance coverage, 44.0% (95% CI, 24.6%–63.4%) had families that received no help coordinating care when needed, and 28.8% (95% CI, 8.2%–49.4%) had families that had problems obtaining referrals when needed (Table 1). Some categories had an unweighted number of children with MD of less than 10 in 2009–2010 (e.g., “non-Hispanic Other” race/ ethnicity, “Other” family structure, “Never Affected” activity limitations, and currently uninsured).

For both periods (2005–2006 and 2009–2010), a larger group of children with MD who did not receive effective care coordination also did not receive family-centered care (77.5% in 2005–2006 and 53.2% in 2009–2010) (Table 2). Among children with MD who did not receive effective care coordination, bivariate analyses showed no statistically significant differences between the two survey cycles for the following characteristics: sex, age group, race/ethnicity, family income, activity limitations, usual source for sick care, whether the child received family-centered care, and current insurance type. However, highest education level and family structure were statistically, significantly associated with survey cycle ($p < 0.05$). Among children with MD who had inadequate current health insurance coverage, many (64.7%) did not receive family-centered care in 2005–2006; however, many (58.0%) did receive family-centered care in 2009–2010 (Table 2). There were no statistically significant differences between the two survey cycles for the following characteristics: sex,

age group, race/ethnicity, activity limitations, usual source for sick care, whether the child received family-centered care, and current insurance type among children with MD who had inadequate current health insurance coverage. Family income and highest education level were statistically, significantly associated with survey cycle ($p < 0.05$).

Changes in care coordination and health insurance coverage

Multivariable regression analyses listed in Figure 1 showed that the unadjusted associations between survey cycle and the four outcomes of interest (not receiving effective care coordination, having inadequate current health insurance coverage, receiving no help coordinating care, and having problems obtaining referrals) were not statistically significant (Table 3). After adjusting for main demographic variables and important confounders, the odds of having inadequate current health insurance coverage among children with MD were statistically, significantly lower in 2009–2010 compared to in 2005–2006 (adjusted odds ratio (aOR) = 0.41; 95% CI, 0.18–0.93). Conversely, the adjusted odds of receiving no help coordinating care when needed in 2009–2010 were statistically, significantly higher than the odds in 2005–2006 (aOR = 4.22; 95% CI, 1.24–14.29).

Sensitivity analyses

In the “Methods” section, we listed two concerns to be addressed in the analyses: the first concern was that, compared to Hispanics who answered in English, Hispanics who answered the survey in Spanish over-reported their cases of MD. After excluding the participants who answered in a language other than English, we found similar results to those when all participants were included (Supplemental Table A). The only notable difference was that the adjusted association between inadequate current health insurance coverage was slightly higher (aOR = 0.45) and lost statistical significance (95% CI, 0.19–1.09). The odds of receiving no help coordinating care were still higher in 2009–2010 than in 2005–2006, but the confidence intervals became much wider after the exclusion. Given the large difference between the unadjusted and adjusted odds for the coordinated care variable, we tested all combinations of variables to find the variable most strongly associated with these odds, which was family-centered care. After excluding this variable from the model for “received no help coordinating care,” the aOR changed from 4.22 (95% CI, 1.24–14.29) (Table 3) to 1.92 (95% CI, 0.74–4.96) (Supplemental Table B). The second concern was that due to the potential correlation between inadequate current health insurance coverage and current insurance type, the adjusted association tested with the model for “had inadequate current health insurance” could be unreliable. We repeated the modeling excluding current insurance type from the predictors, and the result was no longer statistically significant (Supplemental Table B). The new aOR was 0.50 (95% CI, 0.22–1.17) compared to 0.41 (95% CI, 0.18–0.93) from our original model.

Discussion

The results from our study indicate that care coordination status among children with MD aged 0–17 years in the US did not change from one survey to the next. We found no statistically significant changes in the proportions and odds of children with MD who did not receive effective care coordination among children with MD from 2005–2006 to 2009–2010.

However, we found lower proportions and odds of children with MD who had inadequate health insurance coverage from 2005–2006 to 2009–2010 after controlling for key variables.

More than half of children with MD did not receive effective care coordination in both periods, and children with MD had significantly higher adjusted odds of having families that received no help coordinating care when needed in 2009–2010 than in 2005–2006. These findings suggest that there is potential for improvement in the delivery of effective care coordination among children with MD. For example, one tool that can aid in improving this delivery is the use of electronic personal health records.[18,27] According to the American Academy of Pediatrics, electronic personal health records can increase the availability of information and the exchange of data, can improve communication between families and health providers, and can improve quality of pediatric health care, particularly for CSHCN.[28]

Our finding of improved health insurance coverage adequacy among CSHCN with MD is consistent with another study that found minor but statistically significant improvements in health insurance coverage among CSHCN from 2001 to 2009–2010.[29] One possible explanation for the statistically significant improvements in health insurance coverage may be attributed to the Children's Health Insurance Program Reauthorization Act of 2009, which increased funding for states to expand coverage to more children.[30] Because the implementation of this act started shortly before the data for the second period of our study were being collected, a more likely explanation is that the 2009–2010 sample had more educated parents and an increased number of participants with public insurance. As mentioned by a previous study, public insurance is more affordable than private insurance and may offer more benefits for services that meet the needs of CSHCN.[29]

We also found that a large proportion of children with MD in the US who did not receive effective care coordination were also not receiving family-centered care, although there are known benefits of family-centered care. One systematic review found that family-centered care among CSHCN significantly improved functional outcomes, quality of life, care satisfaction, doctor patient partnerships, access to care, and family-provider communication.[31] In 1987, the US Department of Health and Human Services released a Surgeon General's report on CSHCN, urging a family-centered, community-based approach to health care.[32] Thus, the Title V Maternal and Child Health Services Block Grant Program was created in 2010 to provide family-centered, community-based care for CSHCN, among other goals.[33] Our results support these efforts toward coordinated, family-centered care for CSHCN.

Despite the strength of having a nationally representative sample, our study has several limitations, including the low overall response rates for the surveys, which were somewhat compensated by the high response rate among participants with CSHCN. The combined number of children with MD in both survey cycles was only 252. In consequence, the unweighted number of children with MD was less than 10 for many categories after stratification, limiting the interpretation of the results for these cells; however, we included all unweighted numbers for all cells in our tables for transparency. Another limitation is that our sample of children with MD came from a larger sample of noninstitutionalized CSHCN; therefore, our weighted population estimates may differ from other studies of children with

MD sampled from the general population of US children. Additionally, we were unable to examine more recent data regarding CSHCN with MD and coordinated care because the NS-CSHCN was no longer conducted as a separate survey after 2009–2010. The NS-CSHCN was integrated into the National Survey of Children's Health in 2016, and this survey did not include questions about MD.[34] Moreover, phone coverage bias could be present in our study due to the increased use of cell phones over landlines; although, unlike the 2005–2006 survey, the 2009–2010 survey included cell phones in the sampling scheme along with land lines to reduce this bias. Finally, it is possible that the number of cases of MD is misrepresented in our study for two reasons. First, the diagnosis was reported by parents over the phone and, besides the potential for recall bias, the diagnosis was not validated by medical records or clinical examination. Second, MD is a rare disease and education or access to health care may influence the accuracy and timing of the diagnosis reported. For example, the diagnosis of MD is often delayed among children until the age of five years, but informed parents or pediatricians may notice signs of the disease at earlier ages and seek the proper referral for a diagnosis.[35]

Despite the limitations, our study is the first to examine the possibility of changes over time in care coordination and adequacy of health insurance coverage among a representative sample of CSHCN with MD in the US. A recent comprehensive report issued by three professional medical associations in the US, including the American Academy of Pediatrics, claims that data from national surveys of the last 20 years have shown that most youth and young adults with special health care needs do not receive the support they need for a successful transition to adult care.[36] These claims are not specific to MD or coordinated care, but children with MD are part of the group included in the report, and coordinated care is a key part of this transition in care. In 2010, the US government enacted the Affordable Care Act (ACA) expanding insurance coverage, enhancing the range of services covered, and changing the structure of payments and incentives in the country. The effect of this law on the health care of CSHCN could be limited because it was aimed mostly to adult care. However, it may benefit children indirectly—the ACA was based on a 2006 law enacted by the state of Massachusetts and the effects of this local law on the care of CSHCN were examined in a study using data from the NS-CSHCN,[37,38] which found that the effects of the new law on the health care of CSHCN were rather modest. Therefore, we can say that it is likely that our study still has relevance for the current population of US children with MD.

In conclusion, the findings from our study are informative for all stakeholders involved in the care of children with MD, particularly organizations and individuals in charge of implementing coordinated care among these children, their relatives, and their caregivers. In both NS-CSHCN survey cycles (2005–2006 and 2009–2010), more than half of children with MD did not receive effective care coordination, with no statistically significant changes between the two time periods, suggesting that the needs of many families with CSHCN, especially families with children with MD, have not been met for a prolonged period. Care coordination for children with complex needs can be improved by expanding access to community support services, implementing electronic personal health records, and providing more family-centered care. Furthermore, although our study found improvements in health insurance coverage adequacy, nearly a third of children with MD still reported having inadequate health insurance coverage in the most recent survey cycle. Deficits in

coordinated care could negatively affect the health and quality of life of CSHCN, specifically of children with MD.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

Acknowledgements

JZS performed the initial work for this manuscript under a Study Volunteer Service Agreement with the Centers for Disease Control and Prevention as an MPH student at the Department of Epidemiology and Biostatistics, University of Georgia, Athens, GA and carried out the project to completion at Vanderbilt University.

Funding

JZS was supported by a Clinical and Translational Science Award No. TL1TR002244 from the National Center for Advancing Translational Sciences in the National Institutes of Health. The findings and conclusions in this report are those of the authors and do not necessarily represent the official position of the Centers for Disease Control and Prevention nor the National Institutes of Health.

References

1. Schultz EM and McDonald KM. What is care coordination? Int J Care Coord 2014; 17: 5–24.
2. Antonelli RC, Stille CJ and Antonelli DM. Care coordination for children and youth with special health care needs: a descriptive, multisite study of activities, personnel costs, and outcomes. Pediatrics 2008; 122: e209–e216. [PubMed: 18595966]
3. Alexander J, Cartwright J, Desch L, et al. Care coordination in the medical home: integrating health and related systems of care for children with special health care needs. Policy statement. Pediatrics 2005; 116: 1238–1244. [PubMed: 16264016]
4. McDonald KM, Sundaram V, Bravata DM, et al. Closing the quality gap: a critical analysis of quality improvement strategies (Vol. 7: Care Coordination) Technical Review 9, AHRQ Publication No. 04(07)-0051–7. Rockville: Agency for Healthcare Research and Quality, 6 2007.
5. Boudreau AA, Goodman E, Kurowski D, et al. Care coordination and unmet specialty care among children with special health care needs. Pediatrics 2014; 133: 1046–1053. [PubMed: 24864172]
6. Van Cleave J, Boudreau AA, McAllister J, et al. Care coordination over time in medical homes for children with special health care needs. Pediatrics 2015; 135: 1018–1026. [PubMed: 25963012]
7. Litt JS and McCormick MC. Care coordination, the family-centered medical home, and functional disability among children with special health care needs. Acad Pediatr 2015; 15: 185–190. [PubMed: 25311760]
8. Aboneh EA and Chui MA. Care coordination, medical complexity, and unmet need for prescription medications among children with special health care needs. Res Social Adm Pharm 2017; 13: 524–529. [PubMed: 27260830]
9. Bitsko RH, Visser SN, Schieve LA, et al. Unmet health care needs among CSHCN with neurologic conditions. Pediatrics 2009; 124: S343–S351. [PubMed: 19948598]
10. Sannicandro T, Parish S, Son E, et al. Health care changes for children with special health care needs, 2005–2011. Matern Child Health J 2017; 21: 524. [PubMed: 27469109]
11. Mercuri E and Muntoni F. Muscular dystrophies. Lancet 2013; 381: 845–860. [PubMed: 23465426]
12. Emery AE. The muscular dystrophies. Lancet 2002; 359: 687–695. [PubMed: 11879882]
13. Zhu Y, Zhang H, Sun Y, et al. Serum enzyme profiles differentiate five types of muscular dystrophy. Dis Markers 2015:543282. [PubMed: 26063958]
14. Romitti PA, Yong Z, Puzhankara S, et al. Prevalence of Duchenne and Becker muscular dystrophies in the United States. Pediatrics 2015; 135: 513–521. [PubMed: 25687144]

15. Ouyang L, Grosse SD, Fox MH, et al. A national profile of health care and family impacts of children with muscular dystrophy and special health care needs in the United States. J Child Neurol 2012; 27: 569–576. [PubMed: 21954427]
16. Ouyang L, Grosse SD and Kenneson A. Health care utilization and expenditures for children and young adults with muscular dystrophy in a privately insured population. J Child Neurol 2008; 23: 883–888. [PubMed: 18403582]
17. Xing J, Goehring C and Mancuso D. Care coordination program for washington state medicaid enrollees reduced inpatient hospital costs. Health Aff 2015; 34: 653–661.
18. Schneider A, Donnachie E, Tauscher M, et al. Costs of coordinated versus uncoordinated care in Germany: results of a routine data analysis in Bavaria. BMJ Open 2016; 6: e011621–e011621.
19. Guiraud S, Chen H, Burns DT, et al. Advances in genetic therapeutic strategies for Duchenne muscular dystrophy. Exp Physiol 2015; 100: 1458–1467. [PubMed: 26140505]
20. Hashim HZ, Che Abdullah ST, Wan Sulaiman WA, et al. Hunting for a cure: the therapeutic potential of gene therapy in Duchenne muscular dystrophy. Tzu Chi Med J 2014; 26: 5–9.
21. Marti-Morales M and Rohrer JE. Care coordination, family-centered care, and functional ability in children with special health care needs in the United States. Popul Health Manag 2014; 17: 233–238. [PubMed: 25127206]
22. Maternal and Child Health Bureau in collaboration with the National Center for Health Statistics. 2005/06 National Survey of Children with Special Health Care Needs SAS Indicator Data Set. Prepared by the Data Resource Center for Child and Adolescent Health, Child and Adolescent Health Measurement Initiative. www.childhealthdata.org (accessed 13 November 2018).
23. Maternal and Child Health Bureau in collaboration with the National Center for Health Statistics. 2009/10 National Survey of Children with Special Health Care Needs SAS Indicator Data Set. Prepared by the Data Resource Center for Child and Adolescent Health, Child and Adolescent Health Measurement Initiative. www.childhealthdata.org (accessed 13 November 2018).
24. Bramlett M, Blumberg S, Ormson A, et al. Design and operation of the National Survey of Children With Special Health Care Needs, 2009–2010. Vital Health Stat 1 2014; 57: 1–271.
25. Blumberg S, Welch E, Chowdhury S, et al. Design and operation of the National Survey of Children With Special Health Care Needs, 2005–2006. Vital Health Stat 1 2008; 45: 1–188.
26. Read D, Bethell C, Blumberg SJ, et al. An evaluation of the linguistic and cultural validity of the Spanish language version of the children with special health care needs screener. Matern Child Health J 2007; 11: 568–585. [PubMed: 17562154]
27. Sharma N, O'Hare K, O'Connor KG, et al. Care coordination and comprehensive electronic health records are associated with increased transition planning activities. Acad Pediatr 2018; 18: 111–118. [PubMed: 28428097]
28. Council on clinical information technology using personal health records to improve the quality of health care for children. Pediatrics 2009; 124: 403–409. [PubMed: 19564327]
29. Ghandour RM, Comeau M, Tobias C, et al. Assuring adequate health insurance for children with special health care needs: progress from 2001 to 2009–2010. Acad Pediatr 2015; 15: 451–460. [PubMed: 25864809]
30. Children's Health Insurance Program Reauthorization Act of 2009; Public Law 11–3. February 4, 2009.
31. Kuhlthau KA, Bloom S, Van Cleave J, et al. New research on children with special health care needs: evidence for family-centered care for children with special health care needs: a systematic review. Acad Pediatr 2011; 11: 136–143.e8. [PubMed: 21396616]
32. United States Department of Health and Human Services. Surgeon general's report: Children with special health care needs, campaign '87, 1987.
33. Social Secutiry Act, Title V, U.S.C. §§701–710, Subchapter V, Chapter 7, Title 42, 2010.
34. Data Resource Center for Child and Adolescent Health Measurement Initiative. 2016 National Survey of Children's Health (NSCH): Guide to Topics & Questions Asked; 2017 http://childhealthdata.org/docs/default-source/nsch-docs/2016-nsch-guide-to-topics-and-questions_05-15-17.pdf?sfvrsn=2 (accessed on 13 November 2018).

35. Ciafaloni E, Fox DJ, Pandya S, et al. Delayed diagnosis in Duchenne muscular dystrophy: data from the Muscular Dystrophy Surveillance, Tracking, and Research Network (MD STARnet). J Pediatr 2009; 155: 380–385. [PubMed: 19394035]

36. White PH and Cooley WC. Transitions Clinical Report Authoring Group; American Academy of Pediatrics; American Academy of Family Physicians; American College of Physicians. Supporting the health care transition from adolescence to adulthood in the medical home. Pediatrics 2018; 142: e20182587. [PubMed: 30348754]

37. Berman S Children with special health care needs: the canary in the health care reform coal mine. Pediatrics 2014; 134: 373–374. [PubMed: 25002662]

38. Smith AJ and Chien AT. Massachusetts health reform and access for children with special health care needs. Pediatrics 2014; 134: 218–226. [PubMed: 25002660]

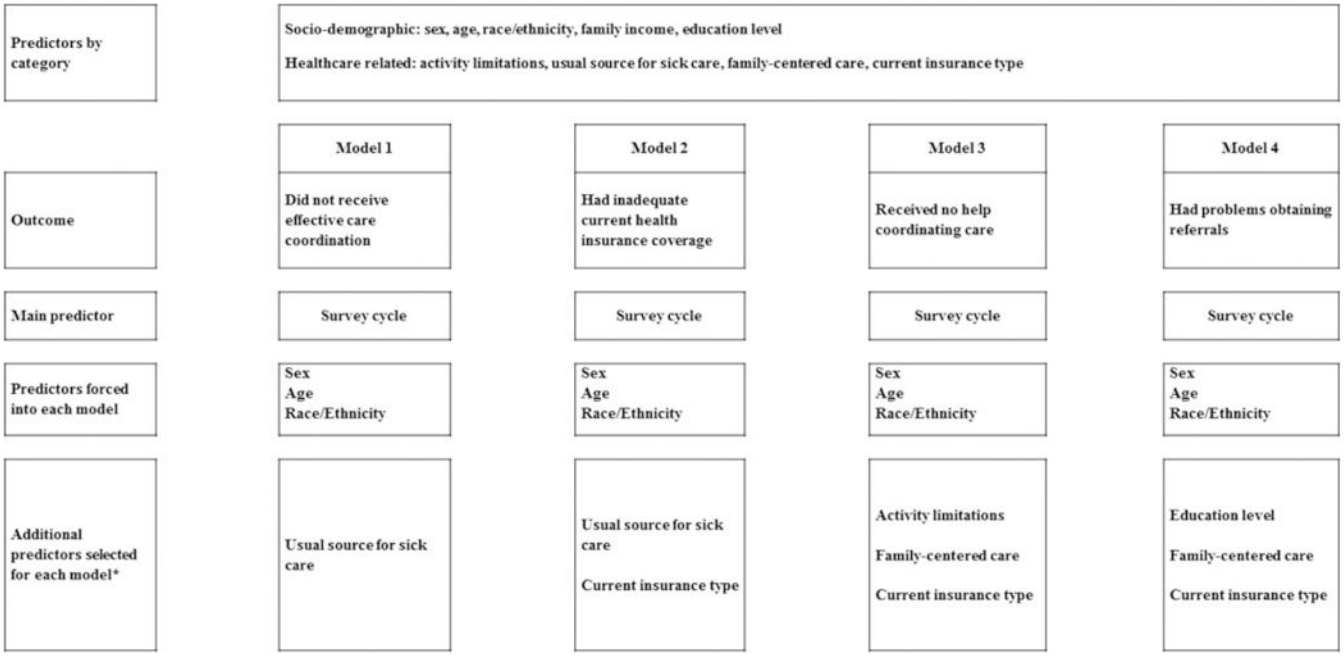

Figure 1.

Multiple logistic models tested among children with muscular dystrophy using data from the National Survey of Children With Special Health Care Needs (2005–2006 and 2009–2010). *Variables were selected with a backward stepwise procedure after assessing for multicollinearity.

Table 1.

Unadjusted comparisons of characteristics among children with muscular dystrophy[a] in the United States from 2005–2006 to 2009–2010, NS-CSHCN 2005–2006 and 2009–2010.

Characteristics	2005–2006 survey cycle			2009–2010 survey cycle			
	Sample n (N = 135)	Weighted n (N = 34,672)	Weighted %(95% CI)	Sample n (N =117)	Weighted n (N = 31,169)	Weighted %(95% CI)	Rao-Scott p value
Sex							
Male	95	24,731	71.3 (58.8–83.8)	78	20,704	66.4 (52.8–80.0)	
Female	40	9941	28.7 (16.2–41.2)	39	10,465	33.6 (20.0–47.2)	0.60
Age group (years)							
0–5	16	5365	15.5 (5.7–25.3)	24	8967	28.8 (15.0–42.6)	
6–11	52	11,094	32.0 (18.3–45.7)	40	9569	30.7 (16.8–44.6)	
12–17	67	18,213	52.5 (36.4–68.6)	53	12,633	40.5 (26.2–54.9)	0.27
Race/ethnicity[b]							
Hispanic	32	9903	28.6 (15.0–42.2)	24	7261	23.3 (10.8–35.8)	
NH White	71	13,023	37.6 (23.4–51.7)	71	14,137	45.4 (30.9–59.8)	
NH Black	20	9801	28.3 (9.3–47.3)	14	4710	15.1 (5.3–24.9)	
NH Other	12	1945	5.6 (0.8–10.4)	8	5061	16.2 (1.8–30.7)	0.17
Family income (% of FPL)[c]							
<100	47	18,357	52.9 (37.1–68.8)	25	11,175	35.9 (20.5–51.3)	
100–199	35	7775	22.4 (10.8–34.1)	31	8803	28.2 (15.0–41.5)	
200–399	33	5092	14.7 (7.2–22.2)	36	5956	19.1 (9.2–29.0)	
≥400	20	3448	9.9 (3.5–16.4)	25	5235	16.8 (7.5–26.1)	0.31
Highest education level[d]							
<High school	18	10,603	30.6 (11.1–50.0)	10	5827	18.7 (5.4–31.9)	
High school	33	7976	23.0 (12.1–33.9)	17	4437	14.2 (4.8–23.6)	
>High school	84	16,093	46.4 (30.6–62.2)	90	20,905	67.1 (52.7–81.5)	0.18
Family structure							
2 parent biological or adoptive	63	13,617	43.7 (27.5–59.9)	81	21,014	68.7 (55.4–82.1)	
2 parent (at least 1 step-parent	14	3691	11.8 (2.3–21.4)	11	1505	4.9 (1.1–8.8)	
Single mother only	38	11,555	37.1 (17.8–56.3)	18	7430	24.3 (11.4–37.2)	

Characteristics	2005–2006 survey cycle			2009–2010 survey cycle			Rao-Scott p value
	Sample n (N = 135)	Weighted n (N = 34,672)	Weighted %(95% CI)	Sample n (N =117)	Weighted n (N = 31,169)	Weighted %(95% CI)	
Other	12	2307	7.4 (0.0–15.0)	4	625	2.0 (0.0–4.4)	0.03 *
Activity limitations							
Consistently affected	99	20,520	59.6 (41.3–77.8)	89	23,834	76.5 (64.5–88.5)	
Moderately affected	25	10,937	31.8 (12.4–51.1)	21	6023	19.3 (8.0–30.7)	
Never affected	10	2983	8.7 (0.1–17.2)	7	1311	4.2 (0.0–9.0)	0.22
Usual source for sick care							
Doctors office	77	19,889	57.4 (41.8–72.9)	75	19,756	63.4 (48.5–78.3)	
Clinic/health center/other	40	9784	28.2 (15.2–41.2)	32	9200	29.5 (14.6–44.5)	
No usual source/ER only	18	5000	14.4 (4.1–24.7)	10	2212	7.1 (1.5–12.7)	0.51
Received family-centered care							
Yes	69	13,373	46.4 (33.3–59.6)	75	19,534	63.0 (48.1–77.9)	
No	58	15,441	53.6 (40.4–66.7)	40	11,481	37.0 (22.1–51.9)	0.11
Current insurance type							
Private only	43	7123	22.6 (12.2–33.0)	41	7178	23.1 (13.4–32.7)	
Public only	46	15,759	50.0 (32.7–67.3)	50	19,011	61.1 (47.7–74.4)	
Both public and private	30	7529	23.9 (11.3–36.5)	21	4143	13.3 (4.0–22.6)	
Uninsured	10	1103	3.5 (0.5–6.5)	3	793	2.5 (0.0–7.0)	0.51
Received effective care coordination[e]							
Yes	47	10,843	40.8 (27.3–54.4)	55	13,363	46.6 (31.4–61.7)	
No	73	15,715	59.2 (45.6–72.7)	58	15,325	53.4 (38.3–68.6)	0.58
Adequacy of current health insurance coverage[f]							
Adequate	67	16,062	48.7 (31.8–65.7)	73	21,197	69.8 (57.2–82.4)	
Inadequate	55	16,896	51.3 (34.3–68.2)	41	9178	30.2 (17.6–42.8)	0.03 *
Received help coordinating care (among those who needed extra help)							
Yes	63	15,324	68.8 (55.9–81.8)	38	11,680	56.0 (36.6–75.4)	
No	34	6941	31.2 (18.2–44.1)	30	9173	44.0 (24.6–63.4)	0.27
Had problems obtaining referrals (among those who needed referrals)							
Yes	24	4672	38.0 (16.6–59.4)	15	5373	28.8 (8.2–49.4)	
No	42	7616	62.0 (40.6–83.4)	46	13,275	71.2 (50.6–91.8)	0.55

ER: emergency room; FPL: federal poverty level; NS-CSHCN: National Survey of Children With Special Health Care Needs; NH: non-Hispanic.

[a] Children with muscular dystrophy were identified by some version of the question “does your child currently have muscular dystrophy?” in both survey cycles.

[b] Race/ethnicity was imputed in 2009-2010 due to having more missing race cases than expected.

[c] Any family income cases expressed as a range were assigned the median value of the range in 2005-2006; cases were imputed in 2009-2010.

[d] The 2005-2006 NS-CSHCN survey asked for the highest education level of anyone in the household while the 2009-2010 NS-CSHCN survey asked for the highest education of each individual parent in the household.

[e] Effective care coordination was determined by having affirmative responses to usually or always getting sufficient help coordinating care when needed, and being very satisfied with communication between doctors and each other, and doctors and other programs when needed.

[f] Adequate current health insurance coverage was determined by having responses of “usually” or “always” to having health insurance that covers services, had reasonable out-of-pocket expenses, and allowed child to see needed health providers.

[*] $p < 0.05$.

Table 2.

Unadjusted weighted percentages of characteristics among children with muscular dystrophy[a] in the United States who did not receive effective care coordination and who had inadequate current health insurance coverage by survey cycle, NS-CSHCN 2005-2006 and 2009-2010.

	Did not receive effective care coordination[b]					Had inadequate current health insurance coverage[c]				
	2005-2006		2009-2010			2005-2006		2009-2010		
Characteristics	**n (N = 73)**	**Weighted% (95% CI)**	**n (N = 58)**	**Weighted% (95% CI)**	**P**	**n (N = 55)**	**Weighted% (95% CI)**	**n (N = 41)**	**Weighted% (95% CI)**	**P**
Sex										
Male	53	69.3 (52.7-85.9)	38	70.3 (52.0-88.6)		41	81.6 (67.2-95.9)	30	67.2 (44.7-89.7)	
Female	20	30.7 (14.1–47.3)	20	29.7 (11.4–48.0)	0.94	14	18.4 (4.1–32.8)	11	32.8 (10.3–55.3)	0.23
Age group (years)										
0–5	7	16.7 (1.8–31.6)	11	19.3 (5.0–33.6)		7	15.6 (0.0–31.6)	5	14.2 (0.3–28.0)	
6–11	28	29.9 (14.4–45.3)	25	41.3 (19.3–63.2)		23	28.8 (7.4–50.1)	19	37.8 (17.3–58.2)	
12–17	38	53.4 (36.0–70.8)	22	39.5 (18.5–60.4)	0.59	25	55.6 (28.8–82.4)	17	48.1 (24.7–71.5)	0.83
Race/ethnicity[d]										
Hispanic	23	48.3 (30.8–65.9)	15	25.2 (8.4–42.0)		12	20.7 (2.8–38.6)	7	15.2 (1.8–28.5)	
NH White	32	24.9 (12.9–37.0)	30	33.8 (16.4–51.2)		31	43.0 (16.9–69.0)	25	44.6 (22.6–66.7)	
NH Black	10	16.8 (4.5–29.1)	8	15.7 (1.0–30.5)		8	32.9 (0.0–66.8)	6	24.7 (2.5–46.8)	
NH Other	8	10.0 (0.4–19.5)	5	25.3 (0.5–50.2)	0.19	4	3.4 (0.0–8.3)	3	15.5 (0.0–37.9)	0.53
Family income (% of FPL)[e]										
<100	29	59.3 (43.0–75.7)	12	31.5 (8.1–54.8)		20	65.4 (43.9–86.8)	6	21.1 (3.4–38.8)	
100–199	17	18.9 (4.8–33.0)	11	18.6 (3.8–33.5)		15	13.4 (2.9–24.0)	13	20.1 (5.1–35.0)	
200–399	17	14.6 (5.5–23.7)	19	24.4 (8.1–40.7)		11	9.9 (1.3–18.4)	12	30.9 (8.6–53.2)	
>400	10	7.1 (1.1–13.2)	16	25.5 (8.5–42.5)	0.09	9	11.3 (0.2–22.4)	10	28.0 (5.6–50.3)	0.001*
Highest education level[f]										
<High school	12	26.8 (9.9–43.7)	4	6.2 (0.0–13.3)		8	48.1 (18.8–77.4)	3	11.9 (0.0–26.4)	
High school	12	19.7 (6.0–33.4)	5	9.5 (0.0–19.6)		12	16.3 (3.3–29.3)	7	14.1 (1.4–26.8)	
>High school	49	53.5 (36.0–71.0)	49	84.3 (72.0–96.5)	0.001*	35	35.6 (13.6–57.6)	31	74.0 (55.6–92.3)	0.002*
Family structure										
2 parent biological or adoptive	35	57.3 (39.4–75.1)	39	72.6 (54.4–90.7)		28	48.7 (17.8–79.6)	27	61.9 (38.0–85.9)	

Characteristics	Did not receive effective care coordination[b] 2005-2006 n (N = 73)	Weighted% (95% CI)	2009-2010 n (N = 58)	Weighted% (95% CI)	P	Had inadequate current health insurance coverage[c] 2005-2006 n (N = 55)	Weighted% (95% CI)	2009-2010 n (N = 41)	Weighted% (95% CI)	P
2 parent (at least 1 step-parent)	9	11.0 (1.8–20.1)	7	6.1 (0.3–12.0)		7	5.6 (0.0–11.9)	7	12.0 (0.2–23.7)	
Mother only	16	16.8 (4.7–29.0)	8	20.5 (3.2–37.8)		12	42.4 (7.7–77.1)	4	24.7 (0.9–48.5)	
Other	9	14.9 (0.0–29.9)		0.8 (0.0–2.3)	0.05*	4	3.2 (0.0–7.5)		1.4 (0.0–4.2)	0.50
Missing	4	-	3	-		4	-	2	-	
Activity limitations										
Consistently affected	57	78.3 (62.6–94.1)	45	84.8 (73.3–96.3)		42	58.9 (27.0–90.8)	32	81.1 (65.9–96.3)	
Moderately affected	13	18.7 (3.1–34.3)	11	13.6 (2.5–24.6)		11	39.2 (6.5–71.8)	7	17.0 (2.2–31.9)	
Never affected	3	3.0 (0.0–7.0)	2	1.6 (0.0–4.2)	0.72	2	2.0 (0.0–5.3)	2	1.9 (0.0–4.7)	0.12
Usual source for sick care										
Doctors office	35	43.6 (26.5–60.7)	38	62.4 (39.5–85.3)		32	61.2 (35.9–86.6)	28	66.6 (43.5–89.7)	
Clinic/health center/other	28	40.0 (22.7–57.4)	16	33.1 (9.6–56.6)		17	22.0 (4.0–39.9)	8	22.9 (0.8–45.0)	
No usual source/ER only	10	16.3 (4.6–28.0)	4	4.5 (0.0–10.3)	0.22	6	16.8 (0.0–35.6)	5	10.5 (0.0–23.9)	0.87
Received family-centered care										
Yes	25	22.5 (10.0–34.9)	27	46.8 (25.4–68.3)		21	35.3 (17.3–53.3)	22	58.0 (35.2–80.8)	
No	46	77.5 (65.1–90.0)	30	53.2 (31.7–74.6)	0.05	32	64.7 (46.7–82.7)	18	42.0 (19.2–64.8)	0.12
Missing	2	-		-		2	-		-	
Current insurance type										
Private only	20	17.9 (7.7–28.2)	18	20.5 (8.0–33.0)		16	13.9 (3.1–24.7)	10	19.6 (5.1–34.1)	
Both public and private	13	22.6 (5.5–39.6)	13	22.9 (5.3–40.4)		15	24.0 (4.7–43.3)	10	24.9 (2.1–47.8)	
Uninsured	6	5.1 (0.0–10.7)	0	-	0.05	-	-	-	-	0.86
Missing	4	-	2	-		0	-	2	-	

ER: emergency room; FPL: federal poverty level; NS-CSHCN: National Survey of Children with Special Health Care Needs; NH: non-Hispanic.

[a]Children with muscular dystrophy were identified by some version of the question “does your child currently have muscular dystrophy?” in both survey cycles.

[b]Not receiving effective care coordination was determined by having affirmative responses to never or sometimes getting sufficient help coordinating care when needed, and not being very satisfied with communication between doctors and each other, and doctors and other programs when needed.

[c]inadequate current health insurance coverage was determined by having responses of “never” or “sometimes” to having health insurance that covers services, had reasonable out-of-pocket expenses, and allowed child to see needed health providers.

[d] Race/ethnicity was imputed in 2009-2010 due to having more missing race cases than expected.

[e] Any family income cases expressed as a range were assigned the median value of the range in 2005-2006; cases were imputed in 2009-2010.

[f] The 2005-2006 NS-CSHCN survey asked for the highest education level of anyone in the household while the 2009-2010 NS-CSHCN survey asked for the highest education of each individual parent in the household.

* $p<0.05$.

Table 3.

Crude and adjusted odds ratios of not receiving effective care coordination, having inadequate health insurance coverage, and related components among children with muscular dystrophy,[a] NS-CSHCN 2005-2006 and 2009-2010: results from multivariable logistic regression.

	Did not receive effective care coordination[b]		Had inadequate current health insurance coverage[c]		Received no help coordinating care (among those who needed extra help)		Had problems obtaining referrals (among those who needed referrals)	
	Crude OR (95% CI)	aOR[d] (95% CI)	Crude OR (95% CI)	aOR[e] (95% CI)	Crude OR (95% CI)	aOR[f] (95% CI)	Crude OR (95% CI)	aOR[g] (95% CI)
Survey cycle								
2005-2006	Ref	Ref	Ref	Ref	Ref	Ref	Ref	Ref
2009-2010	0.79	0.77	0.41	0.41	1.73	4.22	0.66	0.52
	(0.35–1.81)	(0.32–1.89)	(0.17–1.02)	(0.18–0.93) *	(0.64–4.68)	(1.24–14.29) *	(0.17–2.55)	(0.17–1.59)

aOR: adjusted odds ratio; CI: confidence interval; NS-CSHCN: National Survey of Children with Special Health Care Needs; NH: non-Hispanic.

[a] Children with muscular dystrophy were identified by some version of the question "does your child currently have muscular dystrophy?" in both survey cycles.

[b] Not receiving effective care coordination was determined by having affirmative responses to never or sometimes getting sufficient help coordinating care when needed and not being very satisfied with communication between doctors and each other, and doctors and other programs when needed.

[c] inadequate current health insurance coverage was determined by having responses of "never" or "sometimes" to having health insurance that covers services, had reasonable out-of-pocket expenses, and allowed child to see needed health providers.

[d] Adjusted odds ratio for "did not receive effective care coordination" adjusted for sex, age group, race/ethnicity, highest education level, family-centered care, and current insurance type.

[e] Adjusted odds ratio for "had inadequate current insurance coverage" adjusted for sex, age group, race/ethnicity, usual source for sick care, and current insurance type.

[f] Adjusted odds ratio for "received no help coordinating care" adjusted for sex, age group, race/ethnicity, activity limitations, family-centered care, and current insurance type.

[g] Adjusted odds ratio for "had problems getting referrals" adjusted for sex, age group, race/ethnicity, and usual source for sick care.

* $p < 0.05$.