

# A National Profile of Caregiver Challenges Among More Medically Complex Children With Special Health Care Needs

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**Objectives:** To profile the national prevalence of more medically complex children with special health care needs (CSHCN) and the diversity of caregiver challenges that their families confront.

**Design:** Secondary analysis of the 2005-2006 National Survey of Children With Special Health Care Needs (unweighted n=40 723).

**Setting:** United States–based population.

**Participants:** National sample of CSHCN.

**Main Exposure:** More complex CSHCN were defined by incorporating components of child health and family need, including medical technology dependence and care by 2 or more subspecialists.

**Main Outcome Measures:** Caregiver challenges were defined by family-reported care burden (including hours providing care coordination and home care), medical care use (on the basis of health care encounters in the last 12 months), and unmet needs (defined by 15

individual medical care needs and a single nonmedical service need).

**Results:** Among CSHCN, 3.2% (weighted n=324 323) met criteria for more complex children, representing 0.4% of all children in the United States. Caregivers of more complex CSHCN reported a median of 2 (interquartile range, 1-6) hours per week on care coordination and 11 to 20 (interquartile range, 3->21) hours per week on direct home care. More than half (56.8%) reported financial problems, 54.1% reported that a family member stopped working because of the child's health, 48.8% reported at least 1 unmet medical service need, and 33.1% reported difficulty in accessing nonmedical services.

**Conclusions:** Extraordinary and diverse needs are common among family caregivers of more complex CSHCN. Enhanced care coordination support, respite care, and direct home care may begin to address the substantial economic burden and the multiple unmet needs that many of these families face.

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**T**HE SUBSET OF MORE MEDICALLY complex children with special health care needs (CSHCN) is clinically recognized by at least 1 chronic condition resulting in high family-identified service need, medical equipment addressing functional difficulties, multiple subspecialist involvement, and elevated health service use.<sup>1-5</sup> More complex CSHCN are increasing in number,<sup>6</sup> account for growing proportions of hospitalized children,<sup>7,8</sup> and consume a disproportionate amount of health care resources. One study<sup>9</sup> found that 0.5% of a population of children incurred 15% of total health care charges.

Families of CSHCN report that the medical care system is fragmented and difficult to navigate.<sup>9,10</sup> The care requirements of children with any level of special needs create additional financial costs and stress for families.<sup>11-13</sup> As medical diagnoses and care needs rise, families report higher rates of unem-

ployment and unmet needs.<sup>14,15</sup> The challenges of more complex CSHCN may be especially daunting for families because specific care requirements typically include multiple and frequent subspecialist visits and hospitalizations, medical equipment (eg, a tracheostomy or gastrostomy tube), and therapies addressing neurodevelopmental concerns.<sup>16,17</sup>

Little is known on a national level about the effects of more complex CSHCN caregiving on families. Results of prior studies<sup>10,18-21</sup> among small cohorts of families suggest that more complex CSHCN caregiving may contribute to financial, marital, mental, and physical health difficulties. A better understanding of how families spend their time caring for more complex CSHCN may inform more actionable and programmatic strategies to help them overcome these difficulties. The objective of this study was to use a nationally representative sample of CSHCN to describe the di-

versity of caregiver challenges. We hypothesized that family caregivers would report high levels of home care, unemployment, and unmet health care needs.

## METHODS

### STUDY DESIGN

The study is a secondary analysis of the 2005-2006 National Survey of Children With Special Health Care Needs (NS-CSHCN). Funded by the Maternal and Child Health Bureau, the NS-CSHCN provides prevalence estimates of CSHCN and describes aspects of health care use, health status, and family burden.<sup>22</sup> The NS-CSHCN is a module of the National Immunization Survey, conducted annually by the National Center for Health Statistics, with computer-assisted telephone interviews among a national random population sample.<sup>22</sup> Participants for the NS-CSHCN were identified using the CSHCN screener, which asks whether the child had a chronic condition lasting at least 12 months that resulted in any of the following: (1) need for prescription medications; (2) need for more medical care than usual; (3) limited ability to do things; (4) need for occupational, physical, or speech therapy; or (5) emotional or behavioral problems.<sup>23</sup> A positive response to any of the 5 questions enrolls a child in the NS-CSHCN. The sampling design specifies a goal of 750 CSHCN in each state, enabling a maximum standard error of 10% for all point estimates exceeding 15%.<sup>22</sup> Because the data set is publicly available and deidentified, this study received exempt status from the institutional review board at the University of Arkansas for Medical Sciences.

### STUDY POPULATION

The criteria for defining the study population were informed by enrollment criteria from geographically diverse clinical programs at tertiary care centers focused on comprehensive care for more complex CSHCN.<sup>1-5</sup> Such criteria consider family-identified service need, medical equipment use for activities of daily living, the involvement of 2 or more subspecialists on an ongoing basis, and a history of elevated health service use, such as multiple hospitalizations. The criteria at such programs are typically not based on any specific diagnosis.

Study participants in the NS-CSHCN were classified as more complex or less complex. Study participants who were more complex had to meet all 4 of the following criteria: (1) They had to have a positive response to the item on the CSHCN screener indicating "need for more medical care" than usual. This criterion addresses family-identified service need. (2) They had to have positive responses to at least 3 of 4 remaining items (listed in the "Study Design" subsection) on the CSHCN screener. We chose this criterion for several reasons. The number of positive responses on the CSHCN screener correlates with higher health care use.<sup>24</sup> Furthermore, CSHCN with positive responses inclusive of different items have reported higher levels of complexity.<sup>25</sup> We decided against requiring a positive response on all 4 items to avoid falsely excluding more complex CSHCN who were missing 1 component (eg, not all more complex CSHCN have family-identified emotional or behavioral problems). (3) The participants had to demonstrate use of medical equipment, defined as the need for a device to maintain activities of daily living. We operationalized equipment use as a positive response to the need for a mobility aid or device, communication aid or device, medical supplies, or durable medical equipment. This criterion was selected to approximate a comprehensive care program requirement of medical equipment use to address activities of daily living. (4) Finally, the participants had to have seen 2 or more subspecialists in the last 12

months. This criterion was selected to address the requirement of multiple subspecialist involvement.

## OUTCOMES

The primary study outcome was family-reported care burden. This was assessed by weekly hours spent on care coordination and home care, financial burden out-of-pocket costs, and necessity for the family member to stop working, cut back on work to care for their child, or require additional income for medical expenses.

Secondary study outcomes were the following: (1) medical care use, including the use of outpatient, emergency department, early intervention, or special education services, and the stability of the child's health care needs and family report of the number of school days missed and (2) unmet needs, assessed by specific questions relating to 15 identified medical care needs, including preventive, dental, and specialty care, and a single question on nonmedical service needs, such as early intervention services, child care, vocational education, rehabilitation, and related community programs.

We examined the demographic, health services, and clinical characteristics of the study cohort in relation to complexity and the study outcomes. Demographics included age, sex, race/ethnicity, census region, insurance type, poverty level, household educational level, and primary language of household. We examined the presence of a usual source of health care and a usual source of preventive care. The specific chronic medical condition of the child may account for some of the variation in health service use<sup>26</sup>; accordingly, we examined whether the family member identified the child as having any of a list of 16 specific conditions.

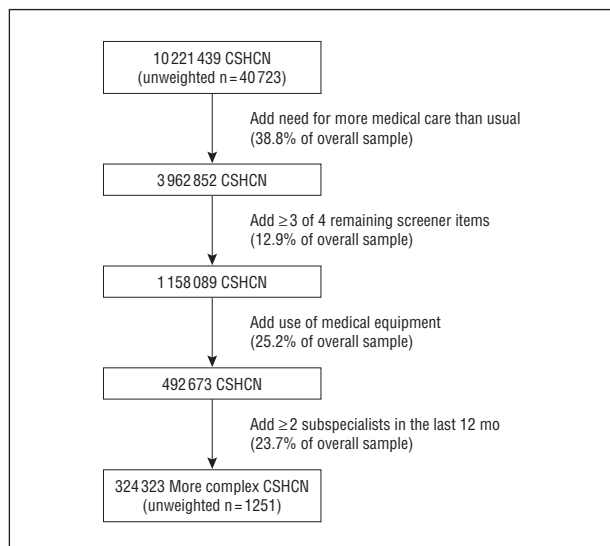
### STATISTICAL ANALYSIS

$\chi^2$  Test was used to compare demographic and diagnosis characteristics between more complex and less complex CSHCN. We identified diagnoses with the largest absolute prevalence differences (defined as being >15.0% between more complex and less complex CSHCN). Mann-Whitney test was used to compare medians or variables with nonnormal distributions (eg, school days missed and hours spent on care coordination). The skewed distribution of these variables led to a categorical large grouping of their highest values; results falling in such categories are reported as the numerical ranges.

The associations of more complex CSHCN with individual diagnoses were verified by logistic regression models that adjusted for significant ( $P < .05$ ) demographics in bivariate analyses. The association of more complex CSHCN with individual diagnoses and unmet health care needs was examined using logistic regression analysis, adjusting for significant ( $P < .05$ ) demographic and diagnosis characteristics in bivariate analyses. Multivariable analysis adjusted for demographic characteristics that were significant at  $P < .05$  and the diagnoses with the largest absolute percentage difference. All the analyses were performed with commercially available software (STATA 10; StataCorp LP, College Station, Texas) using appropriate person-level weights provided in the NS-CSHCN to generate national estimates.<sup>22</sup>

## RESULTS

In 2006, there were 10 221 439 CSHCN aged 0 to 17 years (unweighted NS-CSHCN  $n = 40\,723$ ) in the United States. Of children included in the survey, 38.8% required more medical care than usual according to the CSHCN screener, 12.9% had positive responses to 3 of 4 remaining screener items, 25.2% had use of medical equipment, and 23.7% saw



**Figure 1.** Study sample by criteria. All the values are weighted unless otherwise indicated. CSHCN indicates children with special health care needs.

2 or more subspecialists in the last year. Combining all the study criteria (**Figure 1**) resulted in a study sample of 324 323 (95% CI, 294 064-354 584) more complex CSHCN. This sample represents 3.2% (95% CI, 2.9%-3.5%) of all CSHCN or 0.4% of all children in the United States.

### DEMOGRAPHIC AND DIAGNOSIS CHARACTERISTICS

Compared with less complex CSHCN, more complex CSHCN were more likely to be younger, have public insurance, and have lower family income (all  $P < .001$ ) as well as have a usual source of health care ( $P < .05$ ). No differences were found in sex, race/ethnicity, census region, household educational level, primary language of household, or usual source of preventive care (**Table 1**). Compared with less complex CSHCN, more complex CSHCN had more mean (SD) diagnoses (3.98 [0.13] vs 1.53 [0.01],  $P < .001$ ) per child. Although less likely to have asthma, more complex CSHCN were more likely to have each of the remaining 15 of 16 diagnoses listed. More complex CSHCN had significantly higher prevalences of mental retardation (60.8% vs 9.8%), emotional problems (46.5% vs 20.3%), seizure disorder (26.6% vs 2.7%), autism (24.5% vs 4.0%), cerebral palsy (21.2% vs 1.2%), and joint problems (21.0% vs 3.7%) ( $P < .001$  for all) (**Figure 2**).

### CARE BURDEN

Caregivers of more complex CSHCN reported a median of 2 (interquartile range [IQR], 1-6) hours per week providing care coordination and 11 to 20 (IQR, 3->21) hours providing direct home care per week. Almost half (46.3% [weighted n=150 021]) of families reported paying more than \$1000 in out-of-pocket health care costs in the prior year. Similar proportions reported that they had health care-related financial problems (56.8% [weighted n=184 043]), that a family member stopped working be-

**Table 1. Demographics of Children With Special Health Care Needs by Complexity<sup>a</sup>**

Variable	Less Complex, % (n=9 897 116)	More Complex, % <sup>b</sup> (n=324 323)	P Value
Age, y			
0-5	20.6	30.5	<.001
6-11	37.1	39.4	
12-17	42.2	30.1	
Sex			
Male	59.4	58.8	.81
Female	40.6	41.2	
Race/ethnicity			
White non-Hispanic	65.3	66.2	.14
Black non-Hispanic	16.3	12.4	
Hispanic or Latino	11.7	14.2	
Multiple or other	6.6	7.2	
Census region			
Northeast	17.8	20.2	.50
Midwest	24.1	22.4	
South	38.6	39.2	
West	19.6	18.2	
Insurance type			
Private	68.8	57.6	<.001
Public	27.6	41.0	
Uninsured	3.6	1.4	
Poverty level, %			
≤100	18.8	24.7	<.001
>100-200	21.8	27.0	
>200-300	16.0	14.7	
>300-400	14.2	13.9	
>400	29.2	19.9	
Household educational level			
<High school	6.8	6.4	.94
High school graduate	23.1	23.5	
>High school	70.1	70.1	
Primary language of household			
English	95.4	93.5	.13
Other	4.6	6.5	
Usual source of health care			
No	4.6	2.3	<.05
Yes	95.4	97.7	
Usual source of preventive care			
No	2.5	1.3	.08
Yes	97.5	98.7	

<sup>a</sup>All data are weighted; analyses are by  $\chi^2$  test.

<sup>b</sup>More complex is defined as positive response to "need for more medical care" than usual item and 3 of the remaining 4 items on the children with special health care needs screener; medical equipment use; and seeing 2 or more specialists in the last 12 months.

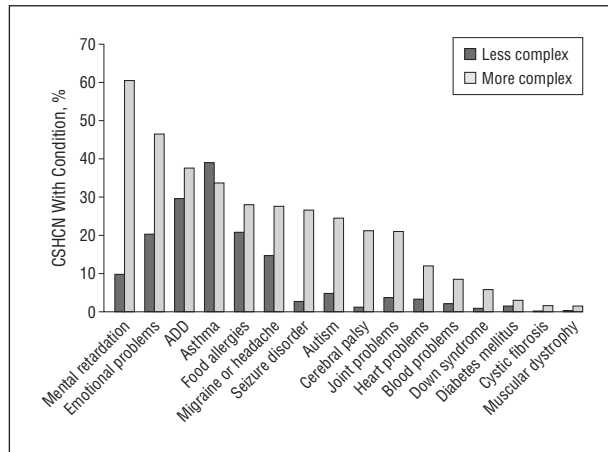
cause of the child's health (54.1% [weighted n=175 294]), that a family member cut back on working hours to care for the child (45.6% [weighted n=147 753]), and that the family needed additional income for medical expenses (48.7% [weighted n=157 798]) (**Table 2**).

### MEDICAL CARE USE AND UNMET NEEDS

Families of more complex CSHCN reported medians of 11 to 15 (IQR, 6 to >21) physician visits and 10 (IQR, 5 to 16-20) school days missed in the last 12 months. Almost two-thirds (65.0% [weighted n=210 810]) of families with more complex CSHCN reported variable health

care service needs of the child (ie, needs change all the time or sometimes) (**Table 3**).

For individual unmet medical needs, families of more complex CSHCN reported an increased likelihood of receiving needed specialty care (adjusted odds ratio [aOR],



**Figure 2.** Parent-reported diagnoses by complexity (weighted n=10 221 439). ADD indicates attention-deficit disorder; CSHCN, children with special health care needs.

2.0; 95% CI, 1.3-3.1) compared with families of less complex CSHCN. Families of more complex CSHCN reported less likelihood of receiving needed prescription medications (weighted n=14 826; aOR, 0.5; 95% CI, 0.3-1.0). No differences were seen in the likelihood of receiving 13 other medical services (**Table 4**). Almost half (48.8% [weighted n=158 270]; aOR, 1.9; 95% CI, 1.4-2.5) of families with more complex CSHCN reported at least 1 unmet medical service need, and 5.4% (weighted n=17 513; aOR, 2.4; 95% CI, 1.3-4.3) reported 5 or more unmet medical service needs in the last 12 months. One-third (33.1% [weighted n=107 350]; aOR, 2.1; 95% CI, 1.6-2.8) of families with more complex CSHCN reported difficulties in accessing nonmedical services. Significantly fewer families (39.4% [weighted n=127 459]; aOR, 0.7; 95% CI, 0.5-0.9) of more complex CSHCN reported being very satisfied with medical services.

### COMMENT

To our knowledge, this is the first study that describes demographics, family care burden, and service needs for the subset of more medically complex CSHCN in a large

**Table 2. Family-Reported Care Burden of Children With Special Health Care Needs by Complexity<sup>a</sup>**

Variable	Less Complex (n=9 897 116)	More Complex <sup>b</sup> (n=324 323)	Adjusted Odds Ratio (95% CI)
Provision of, median (interquartile range), h/wk			
Care coordination	0 (0-2)	2 (1-6)	<.001 <sup>c</sup>
Home care	1 (0-4)	11-20 (3->21)	<.001
In the last 12 mo, %			
Family paid >\$1000 in out-of-pocket health care costs	19.1	46.3	3.0 (2.3-3.9)
Child's health care caused financial problems	16.8	56.8	3.6 (2.7-4.7)
Family member stopped working because of child's health	12.0	54.1	2.9 (2.2-3.9)
Family member cut work to care for the child	15.4	45.6	2.2 (1.7-2.8)
Needed additional income for medical expense	15.2	48.7	3.0 (2.3-3.9)

<sup>a</sup>All data are weighted.  $P < .001$  for all variables ( $\chi^2$  test for proportions and Mann-Whitney test for nonparametric variables). Logistic regression analyses are adjusted for age, insurance type, family income, usual source of health care, mental retardation, emotional problems, seizure disorder, autism, cerebral palsy, and joint problems.

<sup>b</sup>More complex is defined as positive response to "need for more medical care" than usual item and 3 of the remaining 4 items on the complex children with special health care needs screener; medical equipment use; and seeing 2 or more specialists in the last 12 months.

<sup>c</sup> $P < .001$  for comparison of medians by Mann-Whitney test.

**Table 3. Medical Care Use of Children With Special Health Care Needs by Complexity<sup>a</sup>**

Variable	Less Complex (n=9 897 116)	More Complex <sup>b</sup> (n=324 323)
Child's health care needs, %		
Change all the time	5.4	32.0
Change sometimes	27.9	33.0
Are usually stable	66.9	35.0
No. in the last 12 mo, median (interquartile range)		
School days missed	3 (1-8)	10 (5 to 16-20)
Physician visits	4 (2-7)	11-15 (6->21)
Emergency department visits	0 (0-1)	1 (0-3)
Receipt of, %		
Early intervention services at age <3 y	19.0	82.2
Special education services at age range of 3-17 y	27.0	76.9

<sup>a</sup>All data are weighted.  $P < .001$  for all variables ( $\chi^2$  test for proportions and Mann-Whitney test for nonparametric variables).

<sup>b</sup>More complex is defined as positive response to "need for more medical care" than usual item and 3 of the remaining 4 items on the complex children with special health care needs screener; medical equipment use; and seeing 2 or more specialists in the last 12 months.

**Table 4. Families of Children With Special Health Care Needs by Complexity Reporting Difficulty in Accessing Services in the Last 12 Months<sup>a</sup>**

Service Needed	Total Weighted, No.	Less Complex		More Complex <sup>b</sup>		Adjusted Odds Ratio (95% CI)
		No.	Received Service, %	No.	Received Service, %	
Preventive care	7 938 113	7 647 578	97.6	290 535	97.5	1.5 (0.7-3.3)
Specialty care	5 281 140	4 956 878	94.6	324 262	94.4	2.0 (1.3-3.1)
Preventive dental care	8 273 973	8 009 206	92.4	264 767	86.6	0.8 (0.6-1.2)
Other dental care	2 464 893	2 396 862	97.7	68 031	95.5	1.0 (0.4-2.7)
Prescriptions	8 811 327	8 501 168	98.3	310 159	95.2	0.5 (0.3-1.0)
Therapies	2 330 249	2 045 493	87.3	284 756	79.9	0.8 (0.6-1.2)
Mental health care	2 550 503	2 397 218	85.1	153 285	84.3	1.1 (0.7-1.8)
Substance abuse counseling	196 311	190 559	79.2	5752	82.8	0.5 (0.1-2.2)
Home health services	457 942	365 896	90.2	92 046	85.9	1.0 (0.4-2.5)
Eyeglasses or vision care	3 404 342	3 239 572	95.9	164 770	93.2	0.7 (0.4-1.5)
Hearing aid or care	482 090	407 511	91.8	74 579	92.9	1.6 (0.6-4.2)
Respite care	464 223	363 301	52.5	100 922	50.0	1.1 (0.7-1.7)
Genetic counseling	577 468	506 555	75.8	70 913	78.3	1.3 (0.7-2.7)
Family mental health counseling	1 253 180	1 140 519	80.6	112 661	80.1	1.4 (0.9-2.1)
Referrals	3 355 014	3 151 700	79.4	203 314	70.3	0.8 (0.6-1.1)
≥1 Unmet medical service needs in the last 12 mo	10 221 439	9 897 419	19.7	324 020	48.8	1.9 (1.4-2.5)
≥5 Unmet medical service needs in the last 12 mo	10 221 439	9 897 419	0.8	324 020	5.4	2.4 (1.3-4.3)
Very satisfied with medical services	9 978 472	9 656 167	60.5	322 305	39.4	0.7 (0.5-0.9)
Difficulties in accessing nonmedical services	10 128 214	9 806 137	10.2	322 077	33.1	2.1 (1.6-2.8)

<sup>a</sup>All data are weighted. Total size of the subsample indicates weighted subjects for which the question was applicable (eg, preventive care needed). Less complex and more complex numbers are the total weighted study subjects for each category. Analyses are adjusted for age, insurance type, family income, usual source of health care, mental retardation, emotional problems, seizure disorder, autism, cerebral palsy, and joint problems.

<sup>b</sup>More complex is defined as positive response to “need for more medical care” than usual item and 3 of the remaining 4 items on the complex children with special health care needs screener; medical equipment use; and seeing 2 or more specialists in the last 12 months.

nationally representative population. Our findings suggest that families of more complex CSHCN face exceptionally high care demands, increased unemployment and underemployment, and large financial burden and that many more complex CSHCN have unmet medical needs. Our study criteria were specifically designed to select children with complex medical care needs, and it is not surprising to find a high care burden. However, quantifying this burden is revealing: most families with more complex CSHCN in our study reported at least 10 physician visits in the last 12 months, more than 11 missed school days in the past year, and out-of-pocket health care costs exceeding \$1000 annually. More than 175 000 parents terminated employment to stay at home and care for a child with high medical complexity.

Our method is novel in describing a population that is of low prevalence but is nevertheless important. Using criteria adapted from clinical services dedicated to more complex CSHCN and ascertaining such children via family responses as opposed to administrative data focused on diagnostic lists, we focus on attributes that coexist in a distinct population of children within population-level surveys.<sup>26</sup> These attributes are similar to a recently developed definitional framework of more complex CSHCN, termed “children with medical complexity.”<sup>27</sup> The absence of a definitional gold standard<sup>27</sup> makes it difficult to judge the accuracy of our methods to study these children. However, our population estimate (0.4%) is similar to prior prevalence estimates of 0.22% to 0.41% of more complex CSHCN in the United States using case ascertainment from health administrative data.<sup>28,29</sup>

Our criteria captured children with a high prevalence of neurodevelopmental conditions, particularly mental retardation, emotional problems, seizure disorder, autism, and cerebral palsy. This is similar to findings among patients typically enrolled in structured clinical programs that care for more complex CSHCN, although a neurodevelopmental disorder is not generally used as an explicit inclusion criterion.<sup>2,17</sup> Researchers using another identification method via administrative data, known as clinical risk groups, found a high prevalence of neurodevelopmental disorders among patients with the most medical complexity.<sup>30</sup> Children with neurodevelopmental disorders tend to have attributes consistent with more complex CSHCN, such as medical technology assistance, functional status limitations, and multisystem comorbidities, including gastrointestinal (eg, feeding or swallowing), musculoskeletal (eg, spasticity), and respiratory (eg, difficulty in handling secretions).<sup>31-33</sup>

We found that almost 15 000 more complex CSHCN had a low likelihood of receiving prescription medications when needed. The clinical attributes of more complex CSHCN suggest that many of them may need consistent compliance with important medications to maintain their health status. For example, children with cerebral palsy who have epilepsy require daily antiepileptic drug administration to prevent breakthrough seizures. Difficulty in receiving prescription medications may be a factor that is contributing to the high resource use (eg, successive admissions for seizures) observed among some more complex CSHCN.<sup>16</sup>

Aside from prescription medications, we did not find that families of less complex CSHCN had a lower like-

likelihood of any individual unmet medical service need. Access to medical services generally may not be an issue for more complex CSHCN, possibly because their fragile health results in ongoing frequent contact with the medical system. More notable is our finding that families of more complex CSHCN were more likely to have multiple unmet medical service needs; 5.4% reported at least 5 separate unmet medical service needs. This illustrates the difficulty in navigating services and the essential role of care coordination among this subset of CSHCN. We also found that community-based nonmedical services for more complex CSHCN were lacking. Care models that incorporate improved access to and coordination of community-based services are critical for optimal medical, developmental, and service outcomes, yet these are the services that families of more complex CSHCN frequently regard as deficient.<sup>34,35</sup>

More than half of the families among more complex CSHCN in our study reported financial difficulties and employment loss. There is a critical need to help such families overcome these life-disruptive problems. Existing policies can help families sustain employment (eg, the Family Medical Leave Act), but evidence shows that these policies remain underused.<sup>36</sup> Enhanced home caregiving support may help families maintain their employment status and financial stability.<sup>15</sup> There is a perceived home nursing shortage throughout the United States for more complex CSHCN. Other jurisdictions have adopted cost-effective alternative strategies to mitigate this shortage (eg, in Victoria, Australia, a workforce of trained regulated personal care attendants has been created to care for children with tracheostomy who require home mechanical ventilation).<sup>37</sup>

Our study has several limitations. We recognize that there is tautology between the methods used to extract more complex CSHCN (ie, having multiple special needs) and our outcomes (eg, medical care use and unmet needs). Our main intent was to quantify these outcomes rather than highlight the relationship between them and our selected cohort of patients. We had no a priori hypothesis about the magnitude or proportion of health service use in multiple domains that families of more complex CSHCN would report. Specific inclusion criteria, such as having an emotional problem on the CSHCN screener, may be associated with a specific study outcome, such as a parent reporting the presence of an emotional diagnosis or condition. However, of the more complex CSHCN who did not report an emotional problem on the CSHCN screener, 41.6% identified an emotional condition or problem on the survey, suggesting that tautology does not explain all outcomes.

The NS-CSHCN was not originally designed to focus on high medical complexity. It lacks related clinical information (eg, illness severity and duration of functional impairment) and health care use information (eg, hospitalizations) that may help capture more complex CSHCN. Some study criteria, such as the need for supplies, may have increased sensitivity and decreased specificity; not all medical supplies have direct relationships to activities of daily living. However, the prevalence of more complex CSHCN in our study is similar to that in prior studies examining the highest resource users.<sup>28,29</sup>

The less complex CSHCN group in our study likely varies in the level of complexity, and some children in the comparison group likely require a high level of services. However, inclusion of higher levels of complexity in the comparison group would likely lead to a bias toward the null hypothesis, which may strengthen our findings. The survey is cross-sectional and does not permit the determination of causality. Nonmedical service need was characterized as a single question, which did not enable us to specify specific community-based services that may be particularly lacking. Finally, there is no external validation of study responses, although our findings are consistent with clinical experience and prior qualitative literature documenting the pervasive effects of caregiving on the family unit.<sup>20,38</sup>

Given the rising number of more complex CSHCN and their substantial influence on health care costs, an important target for the health care system is to maximize health, development, and function among this subset of CSHCN. Hospital-based programmatic models of care for more complex CSHCN may deliver enhanced care coordination and technical expertise that is necessary to improve care, health outcomes, and family satisfaction.<sup>39</sup> Comprehensive medical care and care coordination for more complex CSHCN in a hospital-based clinic may reduce hospitalizations and overall costs.<sup>40</sup> Family training during transition from hospital to home may decrease morbidity and mortality for children who are dependent on respiratory support.<sup>37</sup> A challenge is to bring these clinical initiatives to the primary care setting, including the training of personnel to manage chronic illnesses and changes to reimbursement patterns that deinstitutionalize comprehensive care.<sup>6</sup>

In conclusion, more complex CSHCN can be distinguished by exceptionally high demands placed on their families and the health care system. Families of more complex CSHCN report multiple unmet needs and many hours devoted to care coordination and home care. Enhanced care coordination support, respite care, and direct home care may begin to address the substantial economic burden and the multiple unmet needs that many of these families face.

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**Author Contributions:** Dr Kuo had full access to all the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis. *Study concept and design:* Kuo, Agrawal, Berry, and Casey. *Acquisition of data:* Kuo. *Analysis and interpretation of data:* Kuo, Cohen, and Berry. *Drafting of the manuscript:* Kuo, Berry, and Casey. *Critical revision of the manuscript for important intellectual content:* Kuo, Cohen, Agrawal, Berry, and Casey. *Statistical analysis:* Kuo, Cohen, and Berry. *Obtained funding:* Kuo.

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## REFERENCES

1. Tanios AT, Lyle RE, Casey PH. ACH medical home program for special needs children: a new medical era. . . . *J Ark Med Soc.* 2009;105(7):163-165.
2. Gordon JB, Colby HH, Bartelt T, Jablonski D, Krauthoefer ML, Havens P. A tertiary care-primary care partnership model for medically complex and fragile children and youth with special health care needs. *Arch Pediatr Adolesc Med.* 2007;161(10):937-944.
3. Kelly A, Golnik A, Cady R. A medical home center: specializing in the care of children with special health care needs of high intensity. *Matern Child Health J.* 2008;12(5):633-640.
4. Cohen E, Friedman JN, Mahant S, Adams S, Jovcevska V, Rosenbaum P. The impact of a complex care clinic in a children's hospital. *Child Care Health Dev.* 2010;36(4):574-582.
5. Berry JG, Agrawal R, Kuo DZ, et al. Characteristics of hospitalizations for patients who use a structured clinical care program for children with medical complexity. *J Pediatr.* 2011;159(2):284-290.
6. Wise PH. The transformation of child health in the United States. *Health Aff (Millwood).* 2004;23(5):9-25.
7. Burns KH, Casey PH, Lyle RE, Bird TM, Fussell JJ, Robbins JM. Increasing prevalence of medically complex children in US hospitals. *Pediatrics.* 2010;126(4):638-646.
8. Simon TD, Berry J, Feudtner C, et al. Children with complex chronic conditions in inpatient hospital settings in the United States. *Pediatrics.* 2010;126(4):647-655.
9. Ghose R. Complications of a medically complicated child. *Ann Intern Med.* 2003;139(4):301-302.
10. Ray LD. Parenting and Childhood Chronicity: making visible the invisible work. *J Pediatr Nurs.* 2002;17(6):424-438.
11. Baker BL, McIntyre LL, Blacher J, Crnic K, Edelbrock C, Low C. Pre-school children with and without developmental delay: behaviour problems and parenting stress over time. *J Intellect Disabil Res.* 2003;47(pt 4-5):217-230.
12. Macias MM, Roberts KM, Saylor CF, Fussell JJ. Toileting concerns, parenting stress, and behavior problems in children with special health care needs. *Clin Pediatr (Phila).* 2006;45(5):415-422.
13. Chevarley FM. *Utilization and Expenditures for Children With Special Health Care Needs.* Rockville, MD: Agency for Healthcare Research and Quality; January 2006.
14. Bitsko RH, Visser SN, Schieve LA, Ross DS, Thurman DJ, Perou R. Unmet health care needs among CSHCN with neurologic conditions. *Pediatrics.* 2009;124(suppl 4):S343-S351.
15. Okumura MJ, Van Cleave J, Gnanasekaran S, Houtrow A. Understanding factors associated with work loss for families caring for CSHCN. *Pediatrics.* 2009;124(suppl 4):S392-S398.
16. Berry JG, Hall DE, Kuo DZ, et al. Hospital utilization and characteristics of patients experiencing recurrent readmissions within children's hospitals. *JAMA.* 2011;305(7):682-690.
17. Srivastava R, Stone BL, Murphy NA. Hospitalist care of the medically complex child. *Pediatr Clin North Am.* 2005;52(4):1165-1187, x.
18. Kratz L, Uding N, Trahms CM, Villareale N, Kieckhefer GM. Managing childhood chronic illness: parent perspectives and implications for parent-provider relationships. *Fam Syst Health.* 2009;27(4):303-313.
19. MacKean GL, Thurston WE, Scott CM. Bridging the divide between families and health professionals' perspectives on family-centred care. *Health Expect.* 2005;8(1):74-85.
20. Macdonald H, Callery P. Parenting children requiring complex care: a journey through time. *Child Care Health Dev.* 2008;34(2):207-213.
21. Thyen U, Kuhlthau K, Perrin JM. Employment, child care, and mental health of mothers caring for children assisted by technology. *Pediatrics.* 1999;103(6, pt 1):1235-1242.
22. Blumberg SJ, Welch EM, Chowdhury SR, Upchurch HL, Parker EK, Skalland BJ. *Design and Operation of the National Survey of Children With Special Health Care Needs, 2005-06* [published online ahead of print November 28, 2007]. [http://www.cdc.gov/nchs/data/series/sr\\_01/sr01\\_045.pdf](http://www.cdc.gov/nchs/data/series/sr_01/sr01_045.pdf). Accessed September 19, 2011.
23. Bethell CD, Read D, Stein RE, Blumberg SJ, Wells N, Newacheck PW. Identifying children with special health care needs: development and evaluation of a short screening instrument. *Ambul Pediatr.* 2002;2(1):38-48.
24. Bramlett MD, Read D, Bethell C, Blumberg SJ. Differentiating subgroups of children with special health care needs by health status and complexity of health care needs. *Matern Child Health J.* 2009;13(2):151-163.
25. Kohen DE, Brehaut JC, Garner RE, et al. Conceptualizing childhood health problems using survey data: a comparison of key indicators. *BMC Pediatr.* 2007;7:40.
26. Andersen RM. Revisiting the behavioral model and access to medical care: does it matter? *J Health Soc Behav.* 1995;36(1):1-10.
27. Cohen E, Kuo DZ, Agrawal R, et al. Children with medical complexity: an emerging population for clinical and research initiatives. *Pediatrics.* 2011;127(3):529-538.
28. Buescher PA, Whitmire JT, Brunssen S, Klutz-Hile CE. Children who are medically fragile in North Carolina: using Medicaid data to estimate prevalence and medical care costs in 2004. *Matern Child Health J.* 2006;10(5):461-466.
29. Neff JM, Sharp VL, Muldoon J, Graham J, Myers K. Profile of medical charges for children by health status group and severity level in a Washington State Health Plan. *Health Serv Res.* 2004;39(1):73-89.
30. Neff JM, Sharp VL, Popalisky J, Fitzgibbon T. Using medical billing data to evaluate chronically ill children over time. *J Ambul Care Manage.* 2006;29(4):283-290.
31. Sullivan PB, Lambert B, Rose M, Ford-Adams M, Johnson A, Griffiths P. Prevalence and severity of feeding and nutritional problems in children with neurological impairment: Oxford Feeding Study. *Dev Med Child Neurol.* 2000;42(10):674-680.
32. Sondheimer JM, Morris BA. Gastroesophageal reflux among severely retarded children. *J Pediatr.* 1979;94(5):710-714.
33. Murphy N, Such-Neibar T. Cerebral palsy diagnosis and management: the state of the art. *Curr Probl Pediatr Adolesc Health Care.* 2003;33(5):146-169.
34. Bodenheimer T, Wagner EH, Grumbach K. Improving primary care for patients with chronic illness. *JAMA.* 2002;288(14):1775-1779.
35. Perrin JM, Romm D, Bloom SR, et al. A family-centered, community-based system of services for children and youth with special health care needs. *Arch Pediatr Adolesc Med.* 2007;161(10):933-936.
36. Chung PJ, Garfield CF, Elliott MN, Carey C, Eriksson C, Schuster MA. Need for and use of family leave among parents of children with special health care needs. *Pediatrics.* 2007;119(5):e1047-e1055.
37. Tibballs J, Henning R, Robertson CF, et al. A home respiratory support programme for children by parents and layperson carers. *J Paediatr Child Health.* 2010;46(1-2):57-62.
38. Rehm RS, Bradley JF. Normalization in families raising a child who is medically fragile/technology dependent and developmentally delayed. *Qual Health Res.* 2005;15(6):807-820.
39. Cohen E, Jovcevska V, Kuo DZ, Mahant S. Hospital-based comprehensive care programs for children with special health care needs: a systematic review. *Arch Pediatr Adolesc Med.* 2011;165(6):554-561.
40. Casey PH, Lyle RE, Bird TM, et al. Effect of hospital-based comprehensive care clinic on health costs for Medicaid-insured medically complex children. *Arch Pediatr Adolesc Med.* 2011;165(5):392-398.