Neurodevelopmental and Mental Health Conditions in Children With Medical Complexity

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abstract

BACKGROUND AND OBJECTIVES: Children with medical complexity (CMC) may be at a high risk of neurodevelopmental and mental health conditions given disease comorbidities and lived experiences. Little is known about the prevalence of these conditions at a population level. In this study, we estimated the prevalence of neurodevelopmental and mental health diagnoses in CMC relative to children without medical complexity and measured associations between these diagnoses in CMC and subsequent health care utilization and in-hospital mortality.

METHODS: We applied the Child and Adolescent Mental Health Disorders Classification System to identify neurodevelopmental and mental health diagnoses using all-payer claims data from three states (2012–2017). Poisson regression was used to compare outcomes in CMC with neurodevelopmental and mental health diagnoses to CMC without these diagnoses, adjusting for sociodemographic and clinical characteristics.

RESULTS: Among 85 581 CMC, 39 065 (45.6%) had ≥1 neurodevelopmental diagnoses, and 31 703 (37.0%) had ≥1 mental health diagnoses, reflecting adjusted relative risks of 3.46 (3.42–3.50) for neurodevelopmental diagnoses and 2.22 (2.19–2.24) for mental health diagnoses compared with children without medical complexity. CMC with both neurodevelopmental and mental health diagnoses had 3.00 (95% confidence interval [CI]: 2.98–3.01) times the number of ambulatory visits, 69% more emergency department visits (rate ratio = 1.69, 95% CI: 1.66–1.72), 58% greater risk of hospitalization (rate ratio = 1.58, 95% CI: 1.50–1.67), and 2.32 times (95% CI: 2.28–2.36) the number of hospital days than CMC without these diagnoses.

CONCLUSIONS: Neurodevelopmental and mental health diagnoses are prevalent among CMC and associated with increased health care utilization across the continuum of care. These findings illustrate the importance of recognizing and treating neurodevelopmental and mental health conditions in this population.

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WHAT'S KNOWN ON THIS SUBJECT: Children with medical complexity may have increased risks of neurodevelopmental and other mental health conditions given their complex health care needs and lived experiences; however, the prevalence of these conditions at a population level is not well understood.

WHAT THIS STUDY ADDS: Forty-six percent of children with medical complexity had neurodevelopmental diagnoses and 37% had mental health diagnoses, more than twice the prevalence observed in children without medical complexity. These diagnoses were associated with significantly increased health care use across care settings.

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Children with medical complexity (CMC) comprise a growing population of children in the United States with complex chronic medical conditions, high levels of health system use, and increased risks of care fragmentation, hospitalization, and childhood mortality.[1–5] Chronic disease comorbidities, as well as disease symptoms, treatments, and other lived experiences, may bring substantial stress to CMC and their families.[6–8] Correspondingly, CMC may be at significantly increased risk of neurodevelopmental and mental health conditions.

Approximately 1 in 5 children in the United States have a neurodevelopmental or mental health condition.[9,10] However, few analyses have focused specifically on CMC. Most studies conducted to date have examined individual chronic disease diagnoses or have relied on survey data, which may be limited by self-report bias and small sample sizes. These studies suggest significantly increased rates of autism, attention-deficit hyperactivity disorder, depression, and anxiety in several CMC subgroups compared with children without chronic conditions.[11–17] However, the authors of a 2019 systematic review noted that research on mental health outcomes in CMC was "strikingly lacking."[18]

The analysis of all-payer claims data (APCD), which links health care claims across payers and settings, presents a unique opportunity to study neurodevelopmental and mental health diagnoses in a large cohort of CMC with diverse conditions. As such, in this study, we used APCD to estimate the prevalence of neurodevelopmental and mental health diagnoses in CMC compared with children without medical complexity and investigate associations between these diagnoses in CMC and subsequent health system utilization and in-hospital mortality. Understanding the prevalence of neurodevelopmental and mental health conditions and differences in health service use can inform policies and programs to support CMC and promote equitable care for this population.

METHODS

Study Design and Data Sources

In this retrospective cohort study, we used APCD from Colorado, New Hampshire, and Massachusetts, which included data from Medicaid and employee-sponsored commercial plans and linked individuals within states across payers. These states were chosen to represent geographic diversity, with a particular focus on varied rural-urban composition. Each of the 3 data sets spanned a 5-year period (January 1, 2013–December 31, 2017 for New Hampshire and Massachusetts and October 1, 2012–September 30, 2017 for Colorado), with files indicating the duration of enrollment in a participating health care plan, as well as professional and facility claims. The acquisition, storage, and analysis of the APCD was guided by data use agreements with the Center for Improving Value in Health Care in Colorado, the New Hampshire Comprehensive Health Care Information System and Department of Health and Human Services, and the Massachusetts Center for Health Information and Analysis. The Dartmouth Hitchcock Institutional Review Board deemed this study exempt from further review and informed consent.

Study Population

In this study, we evaluated neurodevelopmental and mental health conditions in CMC and in children not meeting CMC diagnostic criteria as defined below (hereafter called the reference population). We used the first 3 years of each APCD data set to identify CMC and estimate the prevalence of neurodevelopmental and mental health conditions and the subsequent 2 years to examine health care utilization and in-hospital mortality. Eligible children were limited to residents of participating states and those who were 3 to 17 years of age during the 3-year diagnosis ascertainment period (n = 1 984 200 beneficiaries). We excluded children with zip codes that could not be linked to a rural-urban commuting area[19] code (n = 420), children with <12 months of visibility in the diagnosis ascertainment period (n = 776 196), and those with <12 observation months in the 2-year outcome period unless death occurred (n = 257 552). From this cohort of study-eligible children, we identified CMC by applying 2 open-source algorithms that use *International Classification of Diseases, Ninth or 10th revision* (ICD-9/10) codes: the Complex Chronic Conditions Classification System[20] and the pediatric medical complexity algorithm (PMCA).[21] These algorithms were applied concurrently by using a previously published conservative approach for claims data.[1]

We applied the Child and Adolescent Mental Health Disorders Classification System (CAMHD-CS)[22] to identify and group neurodevelopmental and mental health diagnoses in CMC and the reference population. The CAMHD-CS maps ICD-9/10 codes to 30 diagnosis groups consistent with the *Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition*. Children were classified as having these diagnoses if they had eligible CAMDH-CS diagnostic codes on ≥2 separate dates. We categorized eligible diagnoses as neurodevelopmental conditions or mental health conditions (Supplemental Table 4), given differences in age of onset and health services needs associated with these 2 groups of conditions. It was therefore possible for children to have diagnoses in 0, 1, or both condition groups.

Sociodemographic and Clinical Characteristics

Sociodemographic characteristics included age in years at the beginning of the outcome period, sex (female, male, other/unknown), state of residence, rurality determined by linking residential zip codes to rural-urban

commuting area codes,[19] and primary payer. For CMC, additional characteristics derived from the PMCA and Complex Chronic Condition Classification System included the number of body systems with chronic disease diagnoses,[23] a binary variable indicating technology assistance,[1,20] and a binary indicator of a progressive complex disease diagnosis (eg, muscular dystrophy or cystic fibrosis).[21]

CMC Health Care Utilization and In-Hospital Mortality

During the 2-year outcome period, we examined differences in ambulatory clinic visits, emergency department (ED) visits, acute care hospitalizations and hospital days, and in-hospital mortality between CMC with and without neurodevelopmental and mental health conditions. Ambulatory clinic visits were defined as visits, procedures, or treatments in office or clinic settings with physicians, nurse practitioners, physician assistants, and behavioral health and social services providers, as well as at clinics with current procedural terminology codes for office or other outpatient evaluation and management services. ED visits were defined by using ED-specific revenue codes, current procedural terminology codes, and place of service codes limited to those that did not result in hospitalization on the same or subsequent day. Acute care hospitalizations included inpatient and observation stays. In-hospital mortality per 10 000 CMC was determined by using mortality discharge disposition codes and also required the termination of health care claims for in-person encounters subsequent to the hospitalization.

Statistical Analysis

We first examined differences in sociodemographic characteristics between children with and without neurodevelopmental conditions and with and without mental health conditions using descriptive statistics. We then calculated the prevalence of neurodevelopmental and mental health conditions in CMC and the reference population and estimated the relative risk of these diagnoses in CMC compared with the reference population using Poisson regression with robust error variance.[24] Models were adjusted for age, sex, rurality, payer, state, and observation months during the 3-year diagnosis ascertainment period.

To investigate the differences between CMC with and without neurodevelopmental or mental health conditions in health care utilization and in-hospital mortality, we used Student's t tests to examine unadjusted differences of means and χ^2 tests for differences in proportions. Recognizing that health system use and mortality risk may differ for CMC with neurodevelopmental conditions alone, mental health conditions alone, or these conditions in combination, in our regression analyses, we estimated models using a 4-level primary predictor: (1) CMC without neurodevelopmental or other mental health conditions (reference), (2) CMC with neurodevelopmental conditions only, (3) CMC with mental health conditions only, and (4) CMC with both neurodevelopmental and mental health conditions. Models were adjusted for potential confounding on the basis of age, sex, payer, state of residence, rurality, body systems with chronic disease diagnoses, technology assistance, and progressive complex disease diagnosis. Health care utilization models were also adjusted for observation months during the outcome period. Counts of ambulatory care visits, ED visits, and acute care hospital days were modeled by using Poisson regression with a log-link function. The dichotomous outcomes, acute care hospitalization, and in-hospital mortality were modeled by using Poisson regression with robust error variance to account for model misspecification.[24]

Given that some CMC may have met PMCA diagnostic criteria on the basis of having a chronic mental health diagnosis, we performed a sensitivity analysis in which these individuals were excluded. All analyses described above were repeated by using the restricted cohort. Analyses were performed by using SAS software, Version 9.4; all statistical testing was 2-sided with an α of 0.05.

RESULTS

Neurodevelopmental and Mental Health Diagnoses

Among 85 581 CMC meeting eligibility criteria, 39 065 (45.6%) had 1 or more neurodevelopmental diagnoses, and 31 703 (37.0%) had 1 or more mental health diagnoses. CMC with neurodevelopmental diagnoses were most frequently male, insured only by Medicaid, and urban residing (Table 1). They were also more likely to be assisted by technology and have ≥2 body systems affected by chronic diseases than CMC without neurodevelopmental diagnoses. Similar patterns were observed in CMC with mental health diagnoses, although CMC with mental health diagnoses were more likely to be 12 to 17 years of age and less likely to be assisted by technology. Demographic and clinical characteristics of the sample based on 4 mutually exclusive groups (CMC with neurodevelopmental conditions only, mental health conditions only, both neurodevelopmental and mental health conditions, and neither) are provided in Supplemental Table 5.

Among children without medical complexity, 100 849 (11.7%) had 1 or more neurodevelopmental conditions, and 126 046 (14.6%) had 1 or more mental health conditions. In this reference population, 71 162 (70.6%) of children with neurodevelopmental conditions were male (compared with 370 154 [48.4%] of children without these conditions), and 60 187 (59.7%) were Medicaid-insured (compared with 390 760 [51.1%] of children without these conditions, Supplemental Table 6). Children with mental health conditions were most often 12 to 17 years of age (n = 63 338, 50.3%), male (n = 67 076,

TABLE 1 Characteristics of CMC With and Without Diagnoses of Neurodevelopmental Conditions and Other Mental Health Conditions

CMC Characteristics	Full Cohort		Neurodevelopmental Conditions				Other Mental Health Conditions			
			+ Diagnosis		No Diagnosis		+ Diagnosis		No Diagnosis	
	(n = 85 581)		(n = 39 065, 45.6%)		(n = 46 516, 54.4%)		(n = 31 703, 37.0%)		(n = 53 878, 63.0%)	
	n	%	n	%	n	%	n	%	n	%
Age										
3–7 y	21 215	24.8%	11 024	28.2%	10 191	21.9%	5259	16.6%	15 956	29.6%
8–11 y	24 008	28.1%	12 808	32.8%	11 200	24.1%	9420	29.7%	14 588	27.1%
12–17 y	40 358	47.2%	15 233	39.0%	25 125	54.0%	17 024	53.7%	23 334	43.3%
Sex[a]										
Female	38 664	45.2%	12 907	33.0%	25 757	55.4%	13 819	43.6%	24 845	46.1%
Male	46 829	54.7%	26 134	66.9%	20 695	44.5%	17 864	56.3%	28 965	53.8%
No. body systems affected by chronic diseases										
1[b]	23 828	27.8%	1892	4.8%	21 936	47.2%	3745	11.8%	20 083	37.3%
2	43 666	51.0%	24 855	63.6%	18 811	40.4%	19 573	61.7%	24 093	44.7%
3	11 068	12.9%	7073	18.1%	3995	8.6%	5167	16.3%	5901	11.0%
≥4	7019	8.2%	5245	13.4%	1774	3.8%	3218	10.2%	3801	7.1%
Technology assistance[c]	4196	4.9%	2544	6.5%	1652	3.6%	1407	4.4%	2789	5.2%
Progressive complex condition[d]	24 901	29.1%	10 125	25.9%	14 776	31.8%	8245	26.0%	16 656	30.9%
Primary payer										
Medicaid only	48 391	56.5%	25 620	65.6%	22 771	49.0%	20 053	63.3%	28 338	52.6%
Commercial only	27 888	32.6%	8966	23.0%	18 922	40.7%	8136	25.7%	19 752	36.7%
Both Medicaid and Commercial	9302	10.9%	4479	11.5%	4823	10.4%	3514	11.1%	5788	10.7%
Rurality										
Urban-residing	80 181	93.7%	36 809	94.2%	43 372	93.2%	29 789	94.0%	50 392	93.5%
Rural-residing	5400	6.3%	2256	5.8%	3144	6.8%	1914	6.0%	3486	6.5%
State										
Colorado	17 524	20.5%	7839	20.1%	9685	20.8%	5464	17.2%	12 060	22.4%
Massachusetts	61 737	72.1%	28 837	73.8%	32 900	70.7%	23 884	75.3%	37 853	70.3%
New Hampshire	6320	7.4%	2389	6.1%	3931	8.4%	2355	7.4%	3965	7.4%
Observation mo, cohort creation period, mean (SD)	33.5 (5.1)		34.0 (4.4)		33.0 (5.5)		33.9 (4.5)		33.2 (5.3)	
Observation mo, outcome period, mean (SD)	21.9 (3.5)		22.4 (3.1)		21.5 (3.7)		22.1 (3.6)		21.8 (3.5)	

[a] Sex missing or other for n = 88 (<1%) CMC.
[b] Includes 1008 CMC with only technology assistance codes who do not have another chronic disease diagnosis.
[c] Determined using the Complex Chronic Conditions Classification System.
[d] Determined using the PMCA.

53.2%), and insured exclusively by Medicaid (n = 75 482, 59.9%).

In Table 2, we summarize the frequency and prevalence of neurodevelopmental and mental health conditions in CMC and the reference population. Overall, 21.0% (n = 181 709) of children in the reference population had 1 or more neurodevelopmental or mental health conditions, compared with 59.4% (n = 50 848) of CMC. Adjusting for baseline sociodemographic characteristics, CMC had 2.51 times the risk of these conditions overall (rate ratio [RR] = 2.51, 95% confidence interval [CI]: 2.48–2.53). The adjusted risk of neurodevelopmental conditions specifically was 3.46 times greater in CMC than children in the reference population (95% CI: 3.42–3.50), and attention-deficit hyperactivity disorder and developmental delay or unspecified neurodevelopmental disorder were the most common diagnoses. The most substantive differences in risk between CMC and the reference population were observed for developmental delay or unspecified neurodevelopmental disorders (RR = 8.42, 95% CI: 8.22–8.62) and intellectual disability (RR = 7.25, 95% CI: 7.01–7.50).

In adjusted regression analysis, CMC were 2.22 (95% CI: 2.19–2.24) times more likely to be diagnosed with a mental health condition than children in the reference population. The most common mental health conditions for both CMC and children without medical complexity were trauma and stressor-related disorders (n = 59 915, 6.9% in the reference population and n = 12 297, 14.4% of CMC) and anxiety disorders (n = 41 238, 4.8% in the reference population and n = 11 763, 13.7% of CMC).

TABLE 2 Frequency of Neurodevelopmental and Other Mental Health Diagnoses in CMC and in the Reference Population of Children Without Medical Complexity

	Non-CMC, n = 865 451	CMC, n = 85 581	Risk Ratio[a] (95% CI)
At least 1 neurodevelopmental or other mental health condition	181 709 (21.0%)	50 848 (59.4%)	2.51 (2.48–2.53)
≥1 Neurodevelopmental conditions	100 849 (11.7%)	39 065 (45.6%)	3.46 (3.42–3.50)
Attention-deficit hyperactivity disorder	66 973 (7.0%)	20 171 (23.6%)	2.63 (2.58–2.67)
Developmental delay or unspecified neurodevelopmental disorder	14 132 (1.6%)	13 534 (15.8%)	8.42 (8.22–8.62)
Specific learning disorders	12 697 (1.5%)	8793 (10.3%)	6.61 (6.43–6.80)
Autism spectrum disorder	13 389 (1.5%)	8045 (9.4%)	5.46 (5.31–5.61)
Communication disorders	9325 (1.1%)	4957 (5.8%)	5.36 (5.17–5.55)
Intellectual disability	7061 (0.8%)	6281 (7.3%)	7.25 (7.01–7.50)
Motor disorders	4062 (0.5%)	2624 (3.1%)	6.38 (6.07–6.71)
Neurocognitive disorders	2929 (0.3%)	2206 (2.6%)	6.46 (6.11–6.83)
≥1 Mental health conditions	126 046 (14.6%)	31 703 (37.0%)	2.22 (2.19–2.24)
Trauma and stressor-related disorders	59 915 (6.9%)	12 297 (14.4%)	1.80 (1.77–1.84)
Anxiety disorders	41 238 (4.8%)	11 763 (13.7%)	2.52 (2.47–2.57)
Disruptive, impulse control, and conduct disorders	27 051 (3.1%)	9334 (10.9%)	2.95 (2.88–3.02)
Depressive disorders	23 808 (2.8%)	7825 (9.1%)	2.54 (2.47–2.60)
Miscellaneous[b]	18 744 (2.2%)	7417 (8.7%)	3.19 (3.11–3.28)
Mental health symptoms[c]	9557 (1.1%)	4396 (5.1%)	4.04 (3.90–4.19)
Elimination disorders	4240 (0.5%)	2161 (2.5%)	4.94 (4.69–5.20)
Bipolar and related disorders	2856 (0.3%)	1445 (1.7%)	3.85 (3.61–4.10)
Suicide or self-injury	2376 (0.3%)	1153 (1.3%)	3.63 (3.39–3.90)
Obsessive–compulsive and related disorders	2277 (0.3%)	969 (1.1%)	3.78 (3.51–4.08)
Schizophrenia spectrum and other psychotic disorders	2031 (0.2%)	1356 (1.6%)	5.06 (4.72–5.42)
Feeding and eating disorders	1023 (0.1%)	1280 (1.5%)	11.12 (10.23–12.09)
Somatic symptom and related disorders	660 (0.1%)	819 (1.0%)	10.83 (9.76–12.01)
Other[d]	2665 (0.3%)	1351 (1.6%)	4.44 (4.15–4.75)
No. unique neurodevelopmental disorders and/or mental health conditions			
0	683 726 (79.0%)	34 701 (40.6%)	0.50 (0.49–0.50)
1	104 097 (12.0%)	18 075 (21.1%)	1.59 (1.57–1.62)
2	41 398 (4.8%)	12 803 (15.0%)	2.76 (2.70–2.81)
3	18 846 (2.2%)	8310 (9.7%)	3.84 (3.74–3.94)
≥4	17 384 (2.0%)	11 692 (13.7%)	5.64 (5.50–5.77)

All groups of neurodevelopmental and mental health diagnoses are based on the CAMHD-CS. A child can have zero or multiple diagnoses.

[a] Adjusted for age, sex, payer, rurality, state, and months of health care enrollment or claims in the diagnosis ascertainment period.

[b] Most common ICD-9/10 codes included within the miscellaneous group are unspecified episodic mood disorder (5224 [46.4%] CMC; 12 750 [68.1%] in the reference population), unspecified nonpsychotic mental disorder (1889 [25.5%] CMC; 3523, 18.8% in the reference population), and other emotional disturbances of childhood and adolescence (970 [8.6%] CMC; 2601 [13.9%] in the reference population), accounting for 6069 (81.8%) of CMC and 15 187, 81.0% of children in the reference population included in this category.

[c] Most common ICD-9/10 codes included within the mental health symptoms are unspecified mental or behavioral problem (1635 [37.2%] CMC; 4559 [47.7%] in the reference population), other and unspecified special symptoms or syndromes, not elsewhere classified (1497 [34.1%] CMC; 2719 [28.5%] in the reference population), and attention or concentration deficit (549 [12.5%] CMC; 1444 [15.1%] in the reference population), accounting for 3289 (74.8%) of CMC and 7939 (83.1%) of children in the reference population.

[d] Other includes the following groups, combined because of small cell sizes: fetal or newborn damage related to maternal substance abuse, maternal mental illness or substance abuse during pregnancy, delivery, or postpartum, personality disorders, sexuality and gender identity disorders, sleep–wake disorders, substance abuse-related medical illness, substance-related and addictive disorders, and accidental or undetermined poisoning.

The largest risk ratios observed in CMC compared with the reference population were for feeding and eating disorders (RR = 11.12, 95% CI: 10.23–12.09) and somatic symptoms and related disorders (RR = 10.83, 95% CI: 9.76–12.01). In total, 13.7% (n = 11 692) of CMC had 4 or more neurodevelopmental or mental health conditions, compared with 2.0% (n = 17 384) of children in the reference population, reflecting a 5.64 times (95% CI = 5.50–5.77) greater adjusted risk.

Figure 1 reveals prevalence estimates for neurodevelopmental and mental health conditions in CMC, alone and in combination, based on demographic and clinical characteristics. The lowest prevalence of neurodevelopmental and mental health conditions was observed in CMC with a chronic disease affecting just 1 body system; 79.3% (18 897) of CMC in this group did not have any neurodevelopmental or mental health diagnoses. In contrast, only 15.2% (n = 1067) of CMC with chronic diseases affecting 4 or more body systems did not have any neurodevelopmental or mental health diagnoses. In this group, 35.8% (n = 2511) had both neurodevelopmental and mental health conditions, 10.1% (n = 707) had mental health diagnoses alone, and 39.0% (n = 2734) had neurodevelopmental diagnoses only.

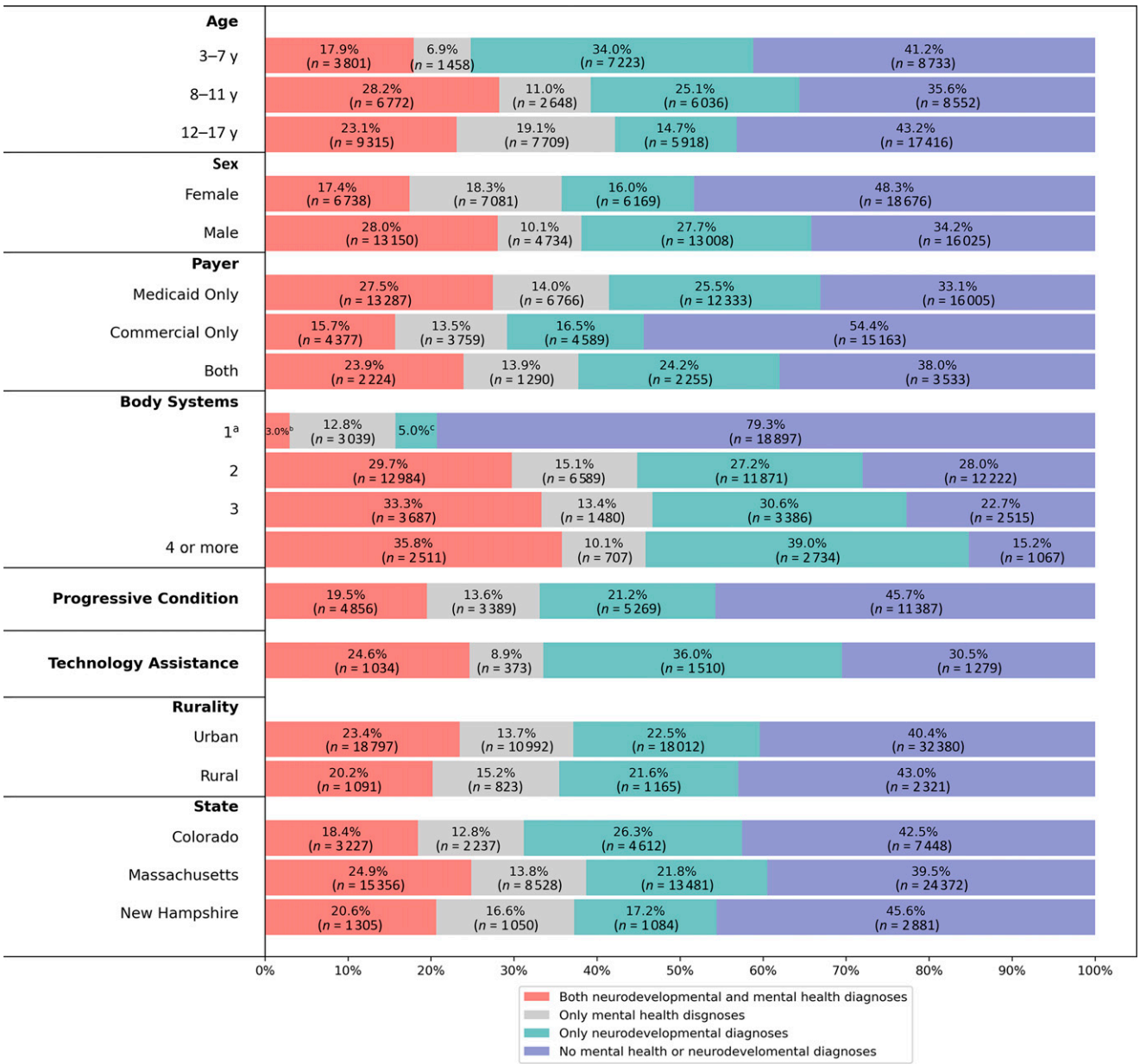

FIGURE 1

Percentage of CMC with cooccurring diagnoses of neurodevelopmental conditions and other mental health conditions by demographic and clinical characteristics. [a] Includes 1008 CMC with only technology assistance codes; [b] $n = 706$; [c] $n = 1186$.

CMC Health Care Utilization and In-Hospital Mortality

On average, CMC with neurodevelopmental diagnoses had more ambulatory clinic visits, more ED visits, more hospitalizations, and more days in hospital during the 2-year outcome period than CMC without these diagnoses (Table 3). One of the largest differences was in the average number of clinic visits; CMC with neurodevelopmental diagnoses had a mean of 28.7 (95% CI: 28.2–29.2) clinic visits over 2 years, compared with 15.4 (95% CI: 15.2–15.6) in CMC without these diagnoses. The in-hospital mortality rate in CMC with neurodevelopmental conditions was also significantly higher than that observed in CMC without these diagnoses: 69 deaths per 10 000 CMC versus 41 deaths per 10 000 CMC ($P < .001$). Similar patterns were observed when comparing health care utilization in CMC with and without mental health diagnoses, although there were no observable differences in mortality risk. CMC with mental health diagnoses had an average of 34.3 (95% CI: 33.7–34.9) clinic visits over 2 years compared with 13.9 (95% CI: 13.7–14.9) visits in those without these diagnoses.

In adjusted regression analyses, CMC with neurodevelopmental conditions or mental health conditions alone

TABLE 3 Health System Utilization and In-Hospital Mortality During the 2-Year Outcome Period in CMC With and Without Cooccurring Diagnoses of Neurodevelopmental Condition and Other Mental Health Conditions

	Full Cohort	Neurodevelopmental Conditions			Other Mental Health Conditions		
	(*n* = 85 581)	+ Diagnosis (*n* = 39 065)	No Diagnosis (*n* = 46 516)	*P*[a]	+ Diagnosis (*n* = 31 703)	No Diagnosis (*n* = 53 878)	*P*[a]
Ambulatory clinic visits, mean (95% CI)							
Total no. clinic visits per CMC	21.4 (21.1–21.7)	28.7 (28.2–29.2)	15.4 (15.2–15.6)	<.001	34.3 (33.7–34.9)	13.9 (13.7–14.1)	<.001
No. clinic visits with a principal diagnosis of a neurodevelopmental condition	4.3 (4.1–4.5)	8.8 (8.5–9.1)	0.6 (0.5–0.7)	<.001	8.0 (7.7–8.3)	2.1 (2.0–2.2)	<.001
No. clinic visits with a principal diagnosis of a mental health condition	6.7 (6.5–6.9)	8.9 (8.6–9.2)	4.9 (4.7–5.1)	<.001	15.0 (14.5–15.5)	1.9 (1.8–2.0)	<.001
No. clinic visits without a principal diagnosis of a neurodevelopmental or mental health condition	10.4 (10.3–10.5)	11.0 (10.9–11.1)	9.9 (9.8–10)	<.001	11.3 (11.2–11.4)	9.9 (9.8–10.0)	<.001
ED visits							
CMC with ≥1 ED visit, *n* (%)	36 883 (43.1%)	18 718 (47.9%)	18 165 (39.1%)	<.001	16 219 (51.2%)	20 664 (38.4%)	<.001
No. ED visits per CMC, mean (95% CI)	2.4 (2.4–2.4)	2.5 (2.5–2.5)	2.2 (2.2–2.3)	<.001	2.7 (2.7–2.8)	2.1 (2.1–2.1)	<.001
ED visits with a principal diagnosis of a neurodevelopmental condition, *n* (%)	1638 (1.9%)	1517 (3.2%)	121 (0.3%)	<.001	1081 (2.4%)	557 (1.3%)	<.001
ED visits with a principal diagnosis of a mental health condition, *n* (%)	6588 (7.5%)	4353 (9.3%)	2235 (5.5%)		5521 (12.4%)	1067 (2.5%)	
ED visits without a principal diagnosis of a neurodevelopmental or mental health condition, *n* (%)	79 417 (90.6%)	40 959 (87.5%)	38 458 (94.2%)		37 826 (85.1%)	41 591 (96.2%)	
Acute care hospitalizations							
CMC with ≥1 acute care hospitalization, *n* (%)	9416 (11.0%)	5012 (12.8%)	4404 (9.5%)	<.001	4500 (14.2%)	4916 (9.1%)	<.001
No. d in hospital per CMC, mean (95% CI)	14.5 (13.6–15.4)	18.1 (16.5–19.7)	10.5 (9.6–11.4)	<.001	19.2 (17.6–20.8)	10.2 (9.2–11.2)	<.001
Hospitalizations with a principal diagnosis of a neurodevelopmental condition, *n* (%)	1130 (2.7%)	1078 (4.6%)	52 (0.3%)	<.001	702 (3.5%)	428 (1.9%)	<.001
Hospitalizations with a principal diagnosis of a mental health condition, *n* (%)	4064 (9.5%)	2700 (11.6%)	1364 (7.0%)		3724 (18.3%)	340 (1.5%)	
Hospitalizations without a principal diagnosis of a neurodevelopmental or mental health condition, *n* (%)	37 503 (87.8%)	19 434 (83.7%)	18 069 (92.7%)		15 849 (78.2%)	21 609 (96.6%)	
In-hospital mortality, *n* (deaths/10 000 CMC)	110 (12.9)	69 (17.7)	41 (8.8)	<.001	42 (13.2)	68 (12.6)	.80

[a] Derived from *t* tests to examine unadjusted differences of means and χ^2 tests for differences in proportions.

or in combination experienced significantly higher health care utilization across all measures (Fig 2). Compared with CMC without neurodevelopmental or mental health conditions, CMC with both condition types had 3.00 (95% CI: 2.98–3.01) times the number of ambulatory visits, 69% more ED visits (RR = 1.69, 95% CI: 1.66–1.72), 1.58 times the risk of being hospitalized (95% CI: 1.50–1.67), and 2.32 times the number of days in hospital (95% CI: 2.28–2.36). In adjusted analyses, the risk of in-hospital mortality in CMC with both neurodevelopmental and mental health conditions was not significantly different from the CMC without these conditions (RR = 1.09, 95% CI: 0.60–1.98).

Sensitivity Analysis

In the sensitivity analysis restricted to children who qualified as CMC when chronic mental health diagnoses were excluded from the PMCA, 22 038 (25.8%) CMC were excluded (Supplemental Table 7). In this restricted cohort of 63 543 CMC, 46.6% (*n* = 29 582) had 1 or more neurodevelopmental or mental health diagnoses and the most common diagnoses were similar to those of the original cohort (Supplemental Table 7). The distribution of sociodemographic characteristics was also similar to the original cohort; however, there were differences in the number of body systems impacted by complexity,

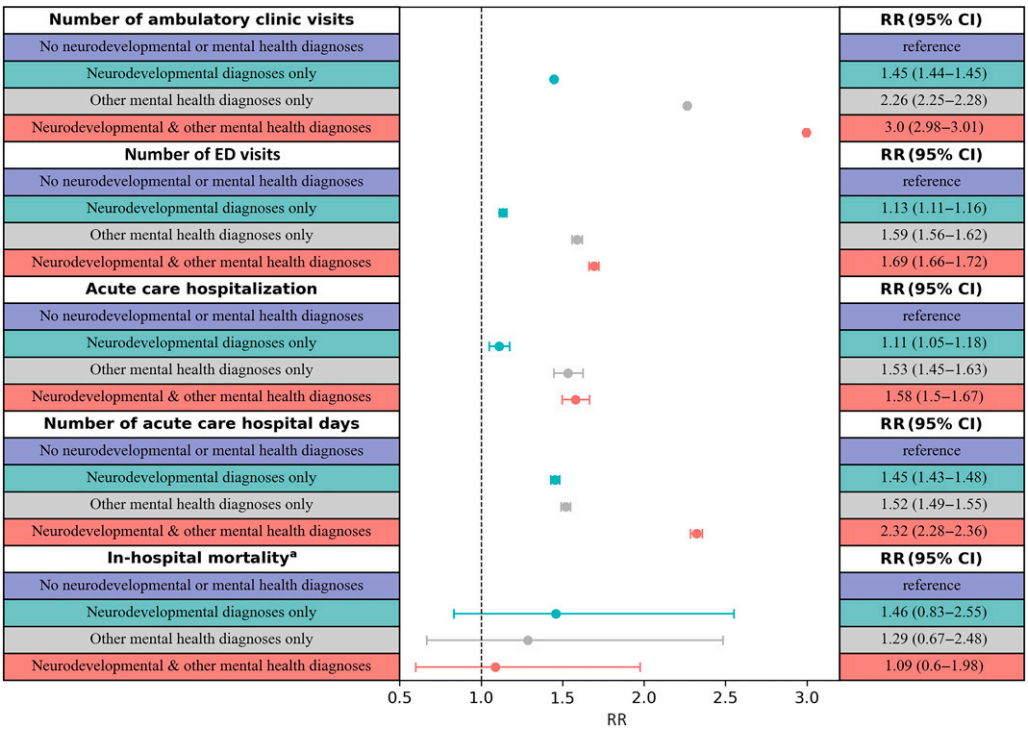

FIGURE 2

Regression analyses demonstrating the likelihood of health care utilization and in-hospital mortality in CMC with cooccurring diagnoses of neurodevelopmental and other mental health conditions, adjusted for demographic and clinical characteristics. Counts of ambulatory care visits, ED visits, and acute care hospital days were modeled by using Poisson regression with a log-link function. The dichotomous outcomes (acute care hospitalization and in-hospital mortality) were modeled by using Poisson regression with robust error variance to account for model misspecification. Models were adjusted for age, sex, payer, state of residence, rurality, body systems with chronic disease diagnoses, technology assistance, and progressive complex disease diagnosis. Health care utilization models were also adjusted for observation months during the outcome period. [a] In the model for in-hospital mortality, diagnoses pertaining to the following body systems were combined into a single group to support stability in modeling a rare outcome: dermatologic, craniofacial, ophthalmologic, and ear, nose, and throat.

technology dependence, and the presence of a progressive complex condition (Supplemental Table 8). Likewise, the patterns of health care utilization remained consistent with the original cohort, with the mean volume of ambulatory clinic visits by CMC with neurodevelopmental (27.8, 95% CI: 27.2–28.4) or other mental health conditions (32.3, 95% CI: 31.6–33.0) being significantly higher compared with CMC without these conditions (Supplemental Table 9). Supplemental Fig 3 reveals adjusted regression analyses that are consistent with the main findings in the original cohort.

DISCUSSION

In this cohort study representing children from 3 states, 45.6% of CMC had 1 or more neurodevelopmental conditions, and 37.0% had 1 or more mental health conditions, reflecting 3 times greater risk of neurodevelopmental diagnoses and 2 times greater risk of mental health diagnoses than observed in children without medical complexity. CMC with neurodevelopmental and mental health conditions used significantly more health care services across the continuum of care from ambulatory to inpatient care than CMC without these diagnoses, and CMC with both neurodevelopmental and mental health conditions experienced the highest utilization. These findings speak to the tremendous importance of the screening, diagnosis, and treatment of these conditions in CMC and can be used to advocate for resources to support their unique health care needs.

Limited research has been conducted to examine the prevalence of neurodevelopmental and mental health conditions in CMC to date. However, studies have reported an increased prevalence of these diagnoses in children with specific complex conditions, such as congenital heart disease, dystonia, and cancer,[11–17] as well as in children with special health care needs or chronic physical conditions.[25,26] In this study, we build on this evidence base, demonstrating a substantially increased risk of these conditions overall and also illustrating specific diagnoses disproportionately experienced by this population. The significantly increased risks of developmental delay (experienced with 8 times greater frequency in CMC than the reference population) and intellectual disability (experienced with 7 times greater frequency) may result from genetic, metabolic, or other conditions that

include both physical and neurodevelopmental manifestations or may be secondary to medical treatments and disrupted routines and learning opportunities due to complex health care needs.[27,28] The results of this study justify focused efforts to support consistent and timely neurodevelopmental screening for CMC, as well as efforts to integrate neurodevelopmental disorder prevention and treatment in their care plans.[29]

Among both CMC and children without medical complexity, trauma and stressor-related disorders and anxiety disorders were the most prevalent mental health conditions, with CMC experiencing 1.8 and 2.5 times the risk of these conditions compared with their peers without medical complexity. These findings are consistent with previous work by Zima et al, who found that trauma and stressor-related disorders were the most common mental health conditions experienced by a Medicaid-insured national sample.[22] The twofold greater risk of these conditions in CMC is likely multifactorial; systematic reviews reveal that the diagnosis of a severe chronic condition may be traumatizing in and of itself, and pediatric medical traumatic stress may result from pain, injury, disease symptoms, and their treatments.[30,31] The substantially increased risk of all mental health conditions observed in this analysis is particularly notable given established challenges in screening, diagnosing, and accessing mental health treatment for CMC.[32–35]

Several previous studies reveal patterns of increased health care use in children with neurodevelopmental or mental health conditions. For example, comorbid mental health conditions have been associated with longer lengths of stay for pediatric medical and surgical hospitalizations,[36] a higher likelihood of recurrent hospitalizations for diabetic ketoacidosis,[37] and higher overall health care costs for children with chronic physical health conditions.[38] Building on this literature, we observed that CMC with neurodevelopmental or mental health conditions, either alone or in combination, had substantially increased health care utilization across care settings. Taken together with past research revealing that the most common reasons for outpatient visits among CMC are mental health-related,[39] our findings speak to the importance of supporting research and public policy to develop and implement effective neurodevelopmental and mental health treatments for CMC.

The results of this study should be interpreted considering its strengths and limitations. Although the study cohort is large, it may not be nationally representative. For example, neurodevelopmental and mental health condition prevalence rates were highest in Massachusetts, which is a state with above-average mental health resources, as well as an established behavioral health screening policy.[40,41] Only neurodevelopmental and mental health conditions that had been coded in health care claims were identifiable, perhaps underestimating disease prevalence. Relatedly, APCD only includes encounters processed by participating insurance plans (excluding encounters paid for out-of-pocket), and as a result, both disease prevalence and the number of health care encounters may have been underestimated. However, we are reassured that the overall prevalence of neurodevelopmental and mental health conditions in the reference population is consistent with previous national analyses.[9,10] APCD lacks details that may be associated with the diagnosis of neurodevelopmental and mental health conditions, such as parental education and socioeconomic status, limiting our ability to identify factors contributing to these diagnoses. Finally, we acknowledge that the study years preceded the coronavirus disease 2019 pandemic and therefore do not reflect more recent potential changes in the prevalence of mental health conditions.[42–45]

CONCLUSIONS

More than 40% of CMC in our cohort had neurodevelopmental conditions, and more than one-third had other mental health conditions. CMC with these diagnoses experienced higher health care utilization across the continuum of care. These findings illustrate the importance of recognizing and treating neurodevelopmental and mental health conditions in this population. Investment in evidence-based strategies to prevent, diagnose, and treat mental health conditions in CMC is needed.

ABBREVIATIONS

APCD: all-payer claims data
CAMHD-CS: Child and Adolescent Mental Health Disorders Classification System
CI: confidence interval
CMC: children with medical complexity
ED: emergency department
ICD-9/10: *International Classification of Diseases, Ninth or 10th revision*
PMCA: pediatric medical complexity algorithm
RR: rate ratio

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