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REVIEW PAPER

Key Population Health Outcomes for Children with Medical Complexity: A Systematic Review

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Abstract

Introduction Despite the significant healthcare policy and program implications, a summary measure of health for children with medical complexity (CMC) has not been identified. It is unclear whether existing population health approaches apply to CMC. We conducted a systematic review of the existing peer-reviewed research literature on CMC to describe the health outcomes currently measured for CMC.

Methods We searched MEDLINE and PsycINFO by linking combinations of key words from three groups of concepts: (1) pediatric, (2) medical complexity, and (3) chronicity or severity. Study eligibility criteria were research studies including CMC with any outcome reported. Data on the outcomes were systematically extracted. Iterative content analysis organized outcomes into conceptual domains and sub-domains.

Results Our search yielded 3853 articles. After exclusion criteria were applied, 517 articles remained for data extraction. Five distinct outcome domains and twenty-four sub-domains emerged. Specifically, 50% of the articles studied healthcare access and use; 43% family well-being; 39% child health and well-being; 38% healthcare quality; and 25% adaptive functioning. Notably lacking were articles examining routine child health promotion as well as child mental health and outcomes related to family functioning.

Conclusions Key health domains for CMC exist. Adaptations of existing sets of metrics and additional tools are needed to fully represent and measure population health for CMC. This approach may guide policies and programs to improve care for CMC.

Keywords Children with medical complexity · Children with special health care needs · Complex chronic conditions · Health outcomes · Population health

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Significance

Despite significant research on children with medical complexity (CMC), a group that comprises roughly 3% of children but generates 40% of child Medicaid expenditures, there is no consensus on how population health for CMC should be defined or measured. Findings from this literature review suggest that common health outcomes for CMC and numerous relevant measurement tools exist. These outcomes may help define population health and guide policies and programs to improve care for CMC.

Introduction

Improving the health of populations is one of the Triple Aims underlying the Affordable Care Act (Berwick et al. 2008). Children with medical complexity (CMC) constitute an important population. According to a definition for CMC developed by Cohen et al. (2011) medical complexity can be characterized by four domains: significant family-identified service needs; chronic, conditions often leading to medical fragility; functional limitations, which are typically severe and associated with technology assistance; and high healthcare use. CMC account for approximately 1–3% of United States (US) children and generate significant costs, including 40% of child Medicaid expenditures (Berry et al. 2013; Cohen et al. 2011). However, valid metrics of population health for CMC are largely unknown, in part due to the substantial heterogeneity and wide-ranging underlying illnesses CMC experience.

Population health provides a conceptual scaffolding for understanding the health outcomes of a group, the determinants of those outcomes, and the distributions of those outcomes across subsets of the population (i.e., disparities) (Kindig and Stoddart 2003). Existing population health frameworks include Healthy People 2020 (Koh et al. 2011), which is based on a frequently cited model developed by Kindig et al. (2008), and models from the Institute for Healthcare Improvement (Steifel and Nolan 2012), Institute of Medicine (Committee on the State of the USA Health Indicators 2009), Robert Wood Johnson Foundation (Parrish 2010), and the World Health Organization (Sadana et al. 2002). While these models include relevant outcomes (e.g., mortality and health-related quality of life) in Kindig's model (Kindig et al. 2008) and important determinants of health, it is unclear whether they fully capture the breadth of health outcomes relevant to CMC.

Attempts to inventory the previously studied outcomes in a way that creates a concise, comprehensive representation of CMC health have not been undertaken. The objective of this study was therefore to systematically review the peer-reviewed research literature on CMC in order to describe the health outcomes being measured, with the downstream goal of contributing to the development of summary measures to describe the health of CMC.

Methods

Data Sources and Article Selection

We identified peer-reviewed articles in the MEDLINE and PsycINFO databases. The search strategy was conducted by using the AND operator to link combinations of Medical Subject Headings (MeSH) terms and key words from three groups of concepts: (1) children, (2) medical complexity, and (3) chronicity or severity (including synonyms for these concepts). The search strategy terms are included (Online Appendix 1).

After running the search, we used the definition for CMC developed by Cohen et al. (2011) to determine inclusion of articles on CMC. The Cohen definition allowed us to select studies involving CMC and simultaneously operationalize a system for sorting articles into conceptually meaningful groups. The approach allowed for a pragmatic categorization of studies resulting from the search, with sub-domains allowed to emerge within the definition in an iterative fashion during extraction and synthesis.

A screening protocol was developed and refined through weekly meetings with the research team, after pilot testing 30 articles. The specific screening procedure was as follows. Per the definition for CMC (Cohen et al. 2011), we included articles with patients meeting the four medical complexity definitional domains. In order to align with the aim of this review, studies that looked exclusively at non-pediatric populations, exclusively at neonatal populations, or patient populations defined solely by mental health diagnoses (i.e., no medical diagnoses, which means not CMC) were excluded. Meta-analyses and non-research articles, such as clinical overviews and guidelines, were excluded as our intent was to capture the breadth of outcomes available in the primary literature. Studies in which no outcomes were reported were similarly excluded. Because the organization and approach of the healthcare system of CMC in developed countries is so different from that of developing countries, studies conducted in non-OECD countries were also excluded. For reasons of feasibility, we additionally excluded articles that were not in the English language or not available through our university's library. Two reviewers independently screened the titles, abstracts, and full text of the article from the search. A third reviewer resolved any disagreements about inclusion or exclusion. The level of agreement between

reviewers evaluating articles for inclusion after full text review was assessed using kappa statistics.

The MEDLINE and PsycINFO databases were initially searched from database inception (1964 and 1985, respectively) through July 20, 2015; quantified percentages in this review report data through this time period. To ensure accuracy of the results, we re-ran the search to examine articles available through December 2017 and qualitatively examined the new articles to identify any trends in the more recent literature.

Data Extraction and Methodological Quality

For included articles, two reviewers independently extracted descriptive data from each article, including study design type, location, CMC diagnoses, authors' medical complexity definitional approach, outcomes measured in the study, and specific measurement tools utilized.

Methodological quality for each article was assessed by two reviewers using Hawker et al.'s critical appraisal tool, designed to assess the quality of heterogeneous studies across nine aspects of an article: abstract and title, introduction and aims, method and data, sampling, data analysis, ethics and bias, findings, transferability, and implications and usefulness (Hawker et al. 2002). Articles were scored for these nine domains on a Likert scale of good to very poor; total scores ranged from nine (i.e., lowest quality) to 36 (i.e., highest quality).

Data Synthesis and Literature Trends

Study outcomes were grouped into conceptual domains and sub-domains created through thematic analysis and iterative team discussions (Braun and Clarke 2006). Two reviewers then categorized outcomes into domains and sub-domains, and kappa statistics were calculated to assess the level of agreement between reviewers. To create a descriptive characterization of the current literature, the frequencies of each domain/subdomain were summarized. Finally, trends in the literature were examined chronologically.

Results

The initial search yielded 3853 articles. After title and abstract screening, 2960 articles did not meet our pre-defined article inclusion criteria, leaving 893 articles for full-text review. After full-text review, an additional 376 articles were excluded based on the pre-defined criteria, leaving 517 articles for data extraction (kappa = 0.79; substantial agreement) (Online Appendix 2, Summary of Articles). The most common reasons for excluding articles at the full-text review stage were study design (e.g., not a research study), followed by no outcome measured (Fig. 1).

The most common study design types among included studies were: observational cross-sectional (219 articles), qualitative (83), and retrospective cohort (80) studies (Table 1). Most of the studies were conducted in the United States (315 articles), United Kingdom (63), and Canada (53) (Table 2). Approximately one-third of the articles included study populations that spanned infancy to 18 years old. Others focused solely on infants, elementary-school aged children, or adolescents.

Medical Complexity Definitions in Included Studies

Medical complexity was defined in study cohorts in a number of ways. Most articles contained a group of diagnostically heterogeneous CMC (367), while other articles (159) identified in the search defined their study population based on specific diagnoses (e.g., childhood cancer, cystic fibrosis). A moderate number of articles (90) utilized technology assistance to define their included study population. Finally, a small number of articles utilized past healthcare utilization (20); a parent questionnaire, such as the Children with Special Health Care Needs (CSHCN) screener (12); or an ICD-9 based algorithm (22) to define the study population. In total, 175 of the studies utilized more than one of these definitions. For the majority of articles that examined a heterogeneous group of CMC, the authors typically did not overtly state the definitional framework they used to characterize their study population.

Outcome Domains

For the full text review, five domain categories and 24 subcategories for types of outcomes on CMC emerged from the iterative coding process through close discussion among the reviewers and research team. Kappa for the domains ranged from 0.43 (moderate agreement) to 0.93 (near perfect agreement). Each of the five domains are elaborated below; domains and sub-domains are summarized in Table 3.

Domain 1: Family Well-Being

This domain referred to outcomes related to the well-being of family members of CMC. Example sub-domains included family service needs, family quality of life, and parent self-efficacy. Outcomes described in these articles tended to focus on parents or caregivers of CMC, with less emphasis given to other family members. Family well-being/stress was the most frequently observed sub-domain within the family well-being domain; 21% of the articles included in the search addressed family stress. In contrast to the family stress aspect of quality of life, measures covering logistical

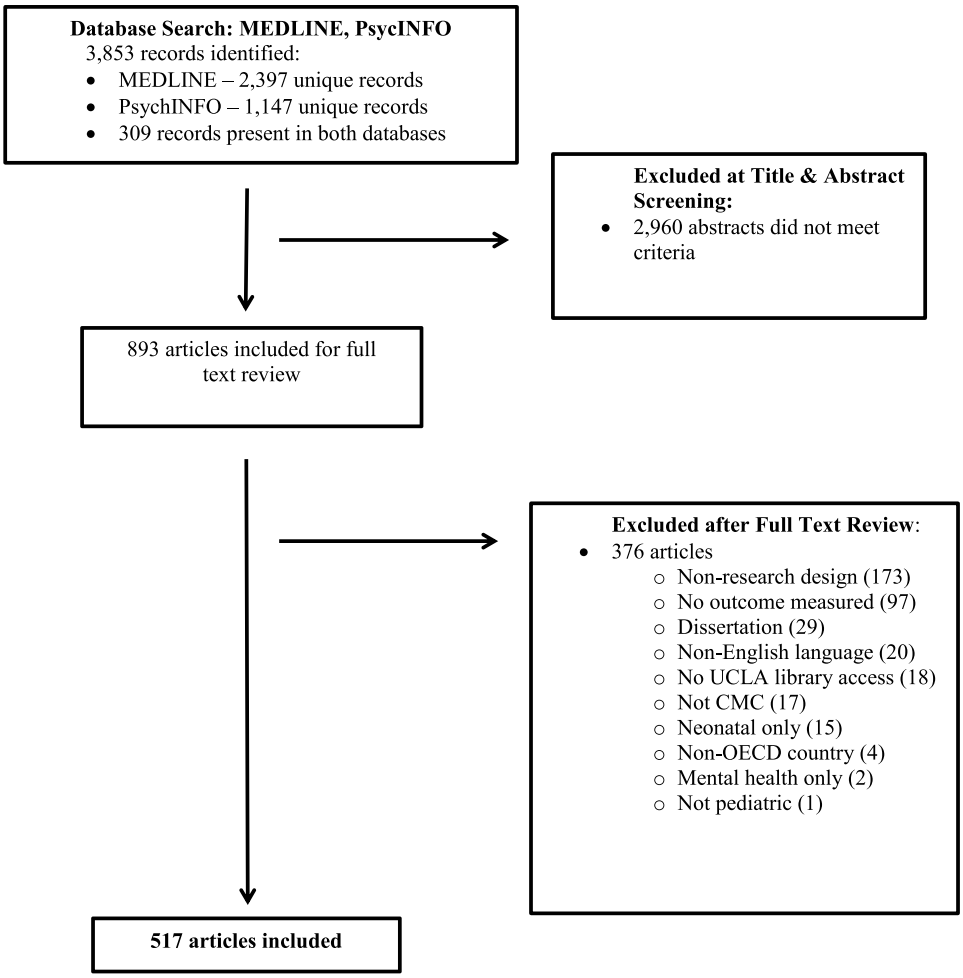

Fig. 1 Flow diagram for article selection

Table 1 Study designs of articles retained after full-text review

Type	Number of articles (n = 517)
Cross-sectional observation	219
Qualitative	83
Retrospective cohort observation	80
Prospective cohort observation	61
Case report	21
Pre-post trial without control	15
Randomized controlled trial	14
Retrospective case–control observation	8
Quasi-experiment	4
Non-equivalent controlled trial	2
Other/multiple	10

aspects of family quality of life, such as family time burden or family financial burden, were included less often in the literature (included in 6.8% and 6.4% of the total articles, respectively). Another outcome relevant to family functioning in the context of caring for a child with medical complexity was family activation/self-efficacy (included in 5% of the articles). A total of 224 articles from the search (43% of the included articles) had at least one outcome within the family well-being domain included.

Domain 2: Child Health and Well-Being

This domain measured health outcomes related to medical complexity. Sub-domains included patient quality of life, progression and complications of disease, and mortality. The majority of articles within this domain measured progression and complications of disease. Common outcome measures capturing disease progression and complications included: health status and measures of disease control, some of which were disease-specific, such as frequency and severity of seizures, pain severity, weight status, hemoglobin A1C levels, pulmonary function tests; number and severity of comorbidities; need for medications; and disease complications. A total of 204 articles (39.5% of the included articles) had at least one outcome within the Child Health and Well-being domain included.

Table 2 Articles by country

Country/continent	Number of articles (n = 517)
United States	315
United Kingdom	63
Canada	53
Europe—non-United Kingdom (Finland, France, Germany, Greece, Ireland, Italy, Netherlands, Norway, Sweden, Switzerland)	50
Oceana (Australia)	22
Asia (Israel, Japan, Turkey)	5
South America (Chile)	1
Multiple countries	8

Table 3 Outcome domains and sub-domains in the research literature on children with medical complexity

Domain	Sub-domain	Articles including domain and sub-domain outcome n (% of all articles)
Family well-being	–	Domain total: 224 (43.3%)
Family well-being	Family quality of life: well-being/stress or burden	108 (20.9%)
Family well-being	Patient/family satisfaction with care	80 (15.5%)
Family well-being	Family service needs	47 (9.1%)
Family well-being	Family quality of life: time burden	35 (6.8%)
Family well-being	Patient–family dynamic	49 (9.5%)
Family well-being	Family quality of life: financial burden	33 (6.4%)
Family well-being	Patient/family activation or self-efficacy	26 (5.0%)
Family well-being	Patient/family-provider dynamic	37 (7.2%)
Child health and well-being	–	Domain Total: 204 (39.5%)
Child health and well-being	Progression/complications of disease	132 (25.5%)
Child health and well-being	Patient quality of life	102 (19.7%)
Child health and well-being	Mortality	37 (7.2%)
Adaptive functioning	–	Domain total: 131 (25.3%)
Adaptive functioning	Activities of daily living	57 (11.0%)
Adaptive functioning	Technology dependence	41 (7.9%)
Adaptive functioning	Work/school attendance	43 (8.3%)
Adaptive functioning	Social engagement	51 (9.9%)
Healthcare access and use	–	Domain total: 261 (50.4%)
Healthcare access and use	Use of medical goods and services	220 (42.6%)
Healthcare access and use	Costs of medical goods and services	98 (19.0%)
Healthcare access and use	Accessibility/geographic distribution	35 (6.8%)
Healthcare access and use	Insurance coverage	14 (2.7%)
Healthcare quality	–	Domain total: 199 (38.5%)
Healthcare quality	Care continuity or coordination	90 (17.4%)
Healthcare quality	Quality of care	78 (15.1%)
Healthcare quality	Adequate medical training	39 (7.5%)
Healthcare quality	Adherence (behavior)	44 (8.5%)
Healthcare quality	Transition care	41 (7.9%)

Domain 3: Adaptive Functioning

This domain measured outcomes related to functional limitations experienced by CMC. Sub-domains included limitations with activities of daily living, challenges with social encounters, technology dependence, and missed school due to illness. A total of 131 articles (25% of the analyzed articles) had at least one outcome within the Functional Limitations domain included.

Domain 4: Healthcare Access & Use

This domain measured outcomes related to healthcare use by CMC. Sub-domains included access to care, utilization, care costs, and insurance coverage. This was the most prevalent category represented in the literature, with 261 articles (51% of the included articles) having at least one outcome within the Healthcare Access & Use domain included. Utilization and costs were the two most common sub-domains (43% and 19%, respectively) identified in the study overall.

Domain 5: Healthcare Quality

This domain measured outcomes related to healthcare quality experienced by CMC. Sub-domains included care coordination, quality of care, transition care, adherence to care, and adequacy of training of medical providers regarding care of CMC. A total of 199 articles (39% of the included articles) had at least one outcome within the Healthcare Quality domain. The two most common sub-domains were care coordination and quality of care.

Measurement Tools

The 517 included studies utilized over 200 specific measures, surveys, and administrative datasets to capture health-related outcomes for children with medical complexity. Surveys were commonly used, e.g., the National Survey of Children with Special Health Care Needs. Types of outcomes captured by the various measurement tools varied widely and included specific measures of disability (e.g., Functional Disability Index Inventory), quality of life (e.g.,

Table 4 Most commonly used measures, surveys, and administrative data sources for health outcomes in children with medical complexity by outcome domain

Measures	Domain
Battelle Developmental Inventory (BDI) (Newborg et al. 1984)	Child health and well-being; adaptive functioning
Center for Epidemiologic Studies Depression (CESD) Scale (Radloff 2016)	Family well-being; child health and well-being
Children with Special Health Care Needs (CSHCN) Screener (Bethell et al. 2002)	Child health and well-being; healthcare access and use
Children's Depression Inventory (Kovacs 1980/1981)	Child health and well-being
Functional Disability Index (FDI) Inventory (Walker and Greene 1991)	Child health and well-being; adaptive functioning
Home Observation for Measurement of the Environment (HOME) Inventory (Caldwell and Bradley 1984)	Family well-being; child health and well-being
Impact on Family (IOF) Scale (Stein and Riessman 1980)	Family well-being
Measure of Processes of Care (MPOC) (King et al. 2004)	Healthcare quality; family well-being
Parenting Stress Index (PSI) (Abidin 1995)	Family well-being
Pediatric Quality of Life Inventory (PedsQL) (Varni et al. 1999)	Family well-being; child health and well-being; adaptive functioning
Questionnaire for Identifying Children with Chronic Conditions (QuICCC) (Stein et al. 1997)	Child health and well-being; adaptive functioning; healthcare access and use
Short Form 36 Health Survey (SF-36) (McHorney et al. 1993)	Child health and well-being; adaptive functioning
Surveys and administrative data sources	
Childhood Cancer Survivor Study (CCSS) (Robison et al. 2002, 2009)	Child health and well-being; adaptive functioning; healthcare access and use
Kids' Inpatient Database (KID) (KID & HCUP 2006/2009)	Healthcare access and use
Medical Expenditure Panel Survey (MEPS) (Cohen et al. 1997)	Healthcare access and use
National Health Interview Survey-Disability (NHIS-D) (United States Department of Health and Human Services et al. 1999)	Multiple domains
National Longitudinal Survey of Children and Youth (NLSCY) (Human Resources Development Canada and Statistics Canada 1997)	Multiple domains
National Survey of Children With Special Health Care Needs (NS-CSHCN) (Blumberg et al. 2008)	Multiple domains

Measures, surveys, and databases included in the table were utilized three of more times in the articles analyzed in the literature review

Pediatric Quality of Life Inventory, or PedsQL), and patient satisfaction and quality of care (e.g., Quality of Discharge Teaching Scale). Measurement tools that were used in three or more studies are presented in Table 4.

Methodological Quality

Most of the included studies were of reasonable quality. Specifically, 161 articles had a rating of very good (Hawker score of 32–36) and 334 articles had scores corresponding to fair quality (Hawker scores of 22–31). The reviewers judged 23 articles to be of poor quality (Hawker score < 22); these articles were included as our intent was to catalogue the breadth of outcomes reported in the research literature on CMC. Average Hawker score by domain ranged from 27 to 29, indicating little variance in study quality by domain.

Trends in the Literature

The literature was reviewed chronologically and shifts in the literature observed. As expected, earlier studies were more likely to be exploratory, with a higher proportion of qualitative or smaller-scale quantitative studies. Outcome measurement for CMC has been a growing field. For the time period before 1990, 19 articles were identified. Sixty-nine articles were identified between 1990 and 1999, 182 between 2000 and 2009, and 247 articles between 2010 and 2015. Across each of these time periods, the distribution of domain outcome types was relatively constant, with the Healthcare Access & Use outcome domain applied to 46 to 68% of the articles reviewed within each of these time periods (Online Appendix 3). In the updated search, encompassing articles indexed between 2015 and 2017, we observed overall highly consistent trends in the literature with the initial search. The exception was a small uptick in articles on caregiver health, with caregiver health reported both as a health outcome in studies on CMC and as a factor influencing the health of CMC.

Discussion

In our review of studies on CMC, five distinct outcome domains and 24 sub-domains emerged. The five emergent domains were healthcare access and use (covered in 50% of articles), followed by family well-being, child health and well-being, healthcare quality, and adaptive functioning. To our knowledge, this is the first attempt to comprehensively review the outcomes studied in the research literature on CMC. This inventory of outcomes represents the concepts that CMC researchers have prioritized to study, and therefore provides a reflection of how researchers might conceptualize health for CMC.

What are Known Measures for CMC that can be Evaluated on a Population Level?

Although the Maternal and Child Health Bureau has tracked population-based measures for CSHCN using six outcomes—partnership in decision making, care within a medical home, insurance coverage, appropriate screening, community-based services, and transitions to adulthood (Administration 1997)—all of these outcomes reflect important measures of system performance that influence the health of children. While recent work has started an important dialogue about what population health might mean for CSHCN, less work has focused specifically on CMC.

Kindig's population health framework, which led to the model for Healthy People 2020 (Koh et al. 2011), has substantial relevance for CMC. In Kindig's model, determinants of population health, such as medical care and social and physical environment, lead to two distinct population health outcomes: mortality and health-related quality of life. Both of these population health outcomes are well-represented by two domains from our study: child health and well-being, and adaptive functioning. Our review's findings, however, suggest a potential need to expand the model to more completely capture health outcomes for the CMC population, and perhaps more broadly for children.

Population Health Gaps

What gaps exist in the literature that need to be filled with further research in order to inform a population health approach for CMC? Most notably, the concepts of child health and well-being and family well-being are underemphasized in existing research, compared to healthcare measures. In particular, child mental health and health promotion seemed under-studied, as were family quality of life measures related to family functioning. Work by Kuo et al. (2011) highlights the broad challenges families face when caring for children with medical complexity, including pragmatic aspects that influence family functioning, such as substantial hours coordinating or directly delivering care, missed work, financial strain, and difficulty accessing services. Often these duties fall most heavily on parents. A substantial challenge on family mental health can ensue, both because of the care attention required and the pain of witnessing one's child suffer; these factors can feedback in a loop (Kuo et al. 2011; Desai et al. 2016). Further, family factors directly impact the care received and the health of CMC (Nelson et al. 2016). The family impact module of the PedsQL scale, a reliable and valid measure in children with complex chronic health conditions (Varni et al. 2004), highlights the relationship between the child's condition, the impact of the condition on the family, and the child's health-related quality of life.

In Kindig's framework, both mortality and health-related quality of life are conceptually sound individual-level outcomes, and both can be aggregated in ways meaningful for populations. More collective outcomes (such as family well-being), however, may be more difficult to measure individually, reflect complex social ties, and yet are critical indicators of population health in their own right. CMC—a group of children whose lives are obviously and inextricably enmeshed with the lives of those who live with and care for them—may help bring these complex ties into sharper focus. Family well-being may likely be an essential population health outcome domain for CMC (and perhaps for all children) that should be explicitly measured. The small increase in articles on CMC caregiver health beginning to emerge in the most recent literature suggests a shifting focus in the CMC field that acknowledges the interdependent nature of caregiver and child health.

Research to Close the CMC Population Health Gaps

To achieve a consensus definition of population health for CMC, more work is needed. The majority of articles to date examine outcomes related to healthcare use and cost, likely reflecting current health policy and health services research priorities. Use and cost outcomes may also be easier to measure, or at least have a longer tradition of measurement, than other outcomes. In contrast, compared to their importance in maintaining health, there remains a dearth of research on preventive health and health promotion outcomes for CMC. Topics for further research on CMC include immunizations, oral health, reproductive health, and broader issues including development and educational attainment, social functioning, and self-management. Research on mental health outcomes for CMC is also strikingly lacking in the medical literature, despite clear evidence that mental health is highly variable among CMC and related to physical health (Bakaniene et al. 2016; Inkelas et al. 2007).

Although our search did not systematically map measures intending to capture health disparities, it is our sense from our review of the literature that disparities among CMC warrant deeper study. Kindig's framework addresses not only mortality and QOL (Kindig et al. 2008), but also disparities within these outcome domains. In the same way, mapping underlying outcome domains for CMC can allow greater examination of the disparities (e.g., racial/ethnic, socioeconomic) within them. In addition, heterogeneity in the CMC literature reflects the underlying heterogeneity of sub-populations being considered. Although the current literature on CMC includes many studies that approach CMC as a heterogeneous group, it also creates relative overrepresentation of certain subgroups, including NICU graduates, children with cerebral palsy, and children with pediatric cancer. The literature on CMC could become more balanced if greater attention was directed to supporting research for lesser-examined conditions, creating an overall body of work that better reflects the CMC population.

If continued research suggests that current population health approaches do not fully represent health for CMC, a refined approach would likely have important programmatic and policy implications. Such an approach could guide the creation of new content for nationally-representative evaluations of children's health across the US, perhaps through surveys such as the National Survey of Children's Health (Blumberg et al. 2008). In addition, the complex care field is rapidly growing with widespread investment through the Health Care Innovation Award Program within the Centers for Medicare and Medicaid Innovation as well as the rapid expansion of complex care programs at US children's hospitals. As these programs grow within accountable care organizations, financial pressures will increasingly require a focus on validated population health outcomes. Policies such as the ACE Kids Act, which propose to improve health of CMC insured by Medicaid across the US by creating a national model for delivery system reform ("The Advancing Care for Exceptional Kids Act of 2015 (ACE Kids Act)," 2015), could also benefit from integrating concepts from a robust population health approach for CMC and their families. Findings suggest that incorporating more metrics related to child and family mental health and family functioning, as well as routine health promotion for children, could prove a worthwhile focus for accountable care organizations and other risk-based payment arrangements. Results also suggest a focus for Medicaid and other health insurance programs on these under-appreciated aspects of quality. The move towards a "whole child, whole family focus" may more accurately and more comprehensively reflect the wellness of the child—which should be the target of all health systems to strive towards—when measuring and rewarding care quality. Ultimately, aligning payment incentives with metrics that capture a more comprehensive view of health for CMC can potentially promote care quality while reducing spending.

Lastly, our work highlights that a variety of measurement tools are available to begin capturing a reasonably diverse set of outcomes; however, important gaps in measurement and in measurement tools remain. Similarly, the feasibility of measuring these domains and sub-domains at the population level is unclear and highly variable. Many measures are caregiver-reported, making them labor-intensive and expensive to implement. Even with a refined approach, additional work will be needed to identify measures that combine reliability and validity with priority and feasibility in order to operationalize measurement of any model of population health for CMC.

Limitations

Our review has some limitations. Given the varying definitions of CMC, some of which are diagnosis specific, it is possible that articles were missed by the search strategy or the databases used. The search may have been biased towards CMC articles reporting physical health rather than mental health outcomes. The search resulted in relatively few articles on developmental outcomes for CMC, yet we are aware of condition-specific studies on development in patients, for example, with low birth weight (Parmelee and Schulte 1970) and congenital heart disease (Wernovsky and Newburger 2003). Outcomes most relevant to CMC in developing countries, which was outside of the scope of this review, may be different than for the populations we examined. Additionally, 367 articles reported on a diagnostically heterogeneous sample of CMC. The extent to which outcomes for a population of children sharing the same disease condition align across studies compared to studies examining heterogeneous groups of CMC merits further examination. Although we made our search as broad as possible, articles that may have only appeared with disease specific searches may have been lost. Finally, bridging a synthesized list of key health-related outcome domains for CMC and domain sub-domains to real-world application will likely require further investigation and inquiry.

Conclusion

Because CMC have very distinct healthcare needs, use, experience, and outcomes compared to the general population and those with less complex chronic conditions, these findings have broad relevance. From a healthcare cost perspective, CMC may be the most important population in pediatrics. From a broader societal perspective, the impact of CMC on the economic and social productivity of families may be prolonged and profound. Furthermore, since healthcare likely contributes only modestly to the health and well-being of most children (Koh et al. 2011), having an agreed upon model of health that extends beyond traditional healthcare boundaries may advance our ability to measure and influence the social and personal factors that contribute to children's health. Results from the existing literature suggest that identifying key common health outcomes for CMC may be possible. Five outcome domains emerge from the literature that appear to capture a variety of the measures—child health and well-being, adaptive functioning, family well-being, healthcare access and use, and healthcare quality. These outcomes suggest the need to adapt current population health approaches to more fully reflect population health for CMC. While the medical complexity field is growing, a broader focus on health promotion as well as child and family mental health is largely absent. Subsequent research building on these findings could ultimately lead to the development of a core set of feasible, impactful population health outcome measures for CMC.

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