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Identifying children with medical complexity in administrative datasets in a Canadian context: Study protocol

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Title: Identifying children with medical complexity in administrative datasets in a Canadian context: Study protocol

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Ethics approval and consent to participate

Ethics approval for this protocol was granted by the institutional research ethics board at the IWK Health Centre (REB # 1026245). A waiver of consent was approved.

Abstract

Introduction: Children with medical complexity and their families are an important population of interest within our Canadian healthcare system. Despite representing less than 1% of the pediatric population, children with medical complexity require extensive care and account for one third of pediatric healthcare expenditures. Opportunities to conduct research to assess disparities in care and appropriate allocation of health resources relies on our ability to accurately identify this heterogeneous group of children.

Objectives: This study aims to better understand the population of children with medical complexity in the Canadian Maritimes, including Nova Scotia (NS), New Brunswick (NB) and Prince Edward Island (PEI). This will be achieved through three objectives: (1) Evaluate the performance of three algorithms to identify children with medical complexity in the Canadian Maritimes in administrative data; then using the "best fit" algorithm we will (2) Estimate the prevalence of children with medical complexity in the Canadian Maritimes from 2003-2017; and (3) Describe patterns of healthcare utilization for this cohort of children across the Canadian Maritimes.

Methods: This research will be conducted in three phases. In Phase 1, an expert panel will co-develop a gold-standard definition of pediatric medical complexity relevant to the Canadian Maritime population. A two-gate validation process will then be conducted using NS data and the gold-standard definition to determine the "best fit" algorithm. During Phase 2 we will apply the "best fit" algorithm to estimate the prevalence of children with medical complexity in NS, NB and PEI. Finally, in Phase 3 will describe patterns of healthcare utilization across the Canadian Maritimes.

Discussion: The project outputs will provide critical information that will guide the design and implementation of future population-level research. They will inform the development of policies and interventions to improve the healthcare delivery and health outcomes of children with medical complexity and their families.

Keywords

Medical complexity, administrative data, pediatric, healthcare utilization

Strength and limitations of this study

- the lack of consensus about how to operationalize a gold-standard definition of children with medical complexity
- we will convene an expert panel and employ multiple methods of algorithm validation will address this limitation
- limitations concerning the use of health data are the availability and accuracy of important variables of interest
- to help mitigate this, all diagnoses from a patient encounter coded in the CIHI DAD will be included
- estimating healthcare utilization using health administrative data has limitations as not all services are reflected in existing databases

Introduction

Since 2010, children with medical complexity have gained increasing attention as an important population in critical need of practice and policy reform within our Canadian healthcare system.[1,2] Medical complexity is generally characterized as having one or more complex chronic condition(s) associated with significant functional limitations, high health resource use and family-identified needs.[1,3] One seminal Canadian study conducted nearly 10 years ago, suggested that despite representing less than 1% of the pediatric population in Ontario, children with medical complexity account for one third of pediatric healthcare expenditures.[1] Adding to the findings stemming from this study, a recent publication from The Canadian Institute for Health Information reported that in 2015-2016 there were 948 per 100,000 children and youth with medical complexity.[4] Recent findings from the United States also suggest that the prevalence of children living with medical complexity is increasing,[5,6] likely due to the increased survival rates of a variety of life-limiting and life-threatening conditions.[3] To date, knowledge about children with medical complexity has relied primarily on data and generalized findings from the United States with only two main reports being conducted the Canadian health care system.[7,8] However, due to important population differences, it is critical that we understand this vulnerable population in the Canadian context to assist in mapping health outcomes and healthcare utilization for this vulnerable population. Doing so will ensure that more relevant, needs based and sustainable models of service delivery are developed to optimize quality health experiences and outcomes for children and their families.

Caregivers of children with medical complexity carry a tremendous amount of responsibility, stress and financial burden to attend to their intensive care needs.[8,9] Opportunities to assess disparities in care, and appropriately allocate health resources to better serve children with medical complexity, are dependent on being able to accurately identify them.[1] A standard

definition of children with medical complexity does not currently exist in Canada and determining a classification system for these children is a necessary first step to optimize health service delivery for this vulnerable group of children. Given the heterogeneity of this group, with variation in the severity and combination of comorbid chronic conditions, there are unique challenges when attempting to identify a cohort at a population level.[1] Identification begins with identifying a method to classify and characterize children with medical complexity.

The Canadian Maritimes is a unique pediatric care setting composed of three provinces: Nova Scotia (NS), New Brunswick (NB), and Prince-Edward-Island (PEI). The only pediatric tertiary care facility in the Maritimes is in NS, which results in families crossing provincial jurisdictions for specialty care. This adds a layer of contextual difference that may intersect with and/or contribute to medical complexity and health care utilization in the Maritimes.

Definitional frameworks

A recent scoping review, examining the use of health administrative data in the study of children with medical complexity, identified three methods that are commonly used to identify this cohort:[7] (1) Cohen et al.'s[3] list of complex chronic conditions, technological assistance and neurological impairment; (2) Simon et al.'s[10] *Pediatric Medical Complexity Algorithm* (PMCA); and (3) Feudtner et al.'s[11] complex chronic conditions classification system. Cohen et al.'s algorithm was developed in Ontario using hospital discharge data but has not yet been validated. Algorithms developed by Simon et al.[10] and Feudtner et al.[11] have not undergone validation in Canadian health data.

Algorithm 1. Cohen et al.[3] operationalized a definitional framework in an Ontario administrative dataset by using sets of ICD-10-CA codes relevant to complex chronic conditions, neurological impairment and technological assistance. This definitional framework aligns with the work of

other experts conducting research on this population outside of Canada.[5,12,13] However, the sensitivity and specificity of the list of ICD-10-CA codes for identifying children with medical complexity in Canada has not yet been reported.

Algorithm 2. Simon et al.[14] used the Chronic Disability Payment System as a guide to develop the *Pediatric Medical Complexity Algorithm* (PMCA) at the Seattle Children's Hospital in Washington State. The authors employed a systematic process beginning with the development of consensus definitions for three levels of medical complexity (complex chronic, noncomplex chronic and no chronic conditions). They classified children with medical complexity as those who fit their definition for having a complex chronic condition. They proceeded by selecting and modifying an existing algorithm to conform to the consensus definitions, and selected a gold standard pediatric population through medical chart review to evaluate the sensitivity and specificity of the algorithm.[14] The PMCA had high sensitivity and specificity (complex chronic: 86% sensitivity, 86% specificity; non-complex chronic: 65% sensitivity, 84% specificity; children without complex chronic: 77% sensitivity and 93% specificity) for identifying children with medical complexity and was subsequently updated and validated for ICD-10-CM codes in 2018.[10]

Algorithm 3. Feudtner et al.[15] followed a similar approach as Simon et al.[10] in Washington State, creating a working definition of complex chronic conditions and subsequently operationalized the definition using clinical knowledge and existing literature. This resulted in a list of conditions and their corresponding ICD-9-CM codes that are highly sensitive (87%) for identifying children with complex chronic conditions. The algorithm was updated in 2014 and the ICD codes were translated into the 10th edition (ICD-10-CM), including both diagnostic and procedural codes indicative of technology dependence or organ transplantation.[11]

Aims and objectives

The aim of this study is to better understand the Canadian Maritime's population of children with medical complexity. The ability to accurately identify children with medical complexity is necessary to conduct research that will inform the design and implementation of successful population-level policies and interventions. This study will be conducted in the Canadian Maritime provinces: NS, NB and PEI. This study will: (1) evaluate the performance of three administrative data algorithms to identify children with medical complexity; then use the "best fit" algorithm to (2) estimate the prevalence; and (3) describe patterns of healthcare utilization for this cohort of children from 2003-2017.

Methods

Patient and Public Involvement

This study will use an integrated knowledge translation (iKT) approach, where end users are involved in each stage of the project.[16] Our team of researchers, clinicians, administrators and patient partners will work together throughout the research process to ensure outputs are relevant to knowledge user needs. This includes development of our research objectives and methods. We will also create opportunities to engage additional parents, clinicians, and administrators to inform different stages of our work as the project unfolds. The proposed iKT method will increase uptake of our research into policy and practice.[17]

Data Sources

The study cohorts will be constructed using the Canadian Institute for Health Information Discharge Abstract Databases (CIHI DAD), Vital Statistics (VITAL), Insured Patient Registry

(MASTER), and Physician Billing Databases (MED) from each province. The administrative datasets for NS, NB and PEI are housed by their respective data repository organizations: Health Data Nova Scotia (HDNS), New Brunswick Institute for Research Data and Training (NB-IRDT), and Secure Island Data Repository (SIDR).

Study Design

This study will be conducted in three phases to identify children with medical complexity in the Canadian Maritimes and their healthcare utilization over a 15-year period (Figure 1).

In Phase 1, we will select the "best fit" algorithm for identifying children with medical complexity in the Maritimes using sensitivity and specificity. Using NS administrative data, we will establish a cohort for each of the three algorithms. We will develop a gold-standard definition for children with medical complexity in the Maritime provinces through an expert consensus meeting. Through the consensus process, this definition will subsequently be translated into a chart audit tool. The "best fit" algorithm will be selected using a two-gate case-control study design.

In Phase 2, we will estimate the prevalence of children with medical complexity in the Maritime provinces. This will be done in all three Maritime provinces using NS, NB and PEI administrative data cohorts identified by the previously selected "best fit" algorithm.

In Phase 3, we will describe patterns of healthcare utilization of children with medical complexity across the Maritime provinces. Ethical approval was received from the primary research institution in NS (REB#: 1026245). A waiver of consent has been granted by our research ethics board and therefore informed participant consent is not required for this study.

Ethical approval will be obtained in NB and PEI before data abstraction commences in these provinces.

Phase 1: Identify the "best fit" algorithm for identifying children with medical complexity in the Maritimes

Developing a gold-standard definition: We will convene a multidisciplinary expert panel to participate in a virtual meeting to co-develop a gold-standard definition of children with medical complexity relevant to the Canadian Maritimes that we can use to validate the administrative data algorithms. The expert panel will bring together relevant stakeholders, including a minimum of two clinicians, two researchers and a parent from each of the three Maritime provinces. The parents will be invited to participate in a pre-meeting virtual meet-and-greet to ensure that they know what to expect at the large meeting and provide a collaborative platform to answer any questions that may arise. This pre-meeting will also help facilitate their familiarity with any research and clinical language that may arise in the expert consensus meeting and ensure parents feel empowered and safe to provide their invaluable insights. During the meeting, a consensus process will be used to produce a gold-standard definition that is clinically meaningful for this pediatric population in the Maritimes.[21] The definition will be circulated after the consensus meeting to the expert panel, as well as other relevant stakeholders who may have been unable to attend the meeting, for review and will be edited through email. A sub-group of clinicians at main pediatric hospital in NS will then develop a list of clinical variables that will form the basis of a medical chart extraction tool that aligns with the gold-standard definition. The drafted medical chart extraction tool will be reviewed by clinicians from both NB and PEI.

We will use an internal and external validation process to evaluate the gold-standard definition.[22] The internal process will involve the comparison of the definition with the chart diagnosis of 3-4

patients identified independently by two NS clinical experts, not involved in the expert panel. The external process will involve review of the definition by a clinical expert in NB and PEI, not on the expert panel. During the second virtual meeting, the expert panel will review feedback from the validation process and revise the gold-standard definition as required, until consensus is reached. The final draft of the medical chart extraction tool will also be reviewed and approved by the expert panel.

Establishing a Nova Scotia administrative data cohort: The algorithm validation process will be undertaken in NS using administrative data from our provincial data repository organization, HDNS. Children living in NS with medical complexity will be identified and classified with specific ICD-10-CA codes defined within each of the three respective algorithms, resulting in a distinct cohort being derived from each algorithm. All children aged 0 to 18 years of age who have a discharge record in NS CIHI DAD from 2003-2017 will be eligible for inclusion.

Examining the best fit: Given the prevalence of children with medical complexity is rare, we will use a two-gate case-control study design to validate the algorithms (Figure 2).[23] This will be done in two steps. First, children identified by each algorithm in the administrative data will be selected in a stratified random sample of 400, 100 children from each of the three algorithms (with a 100 children sample pulled from both Simon et al.'s more and less conservative algorithm versions), to have a retrospective chart review completed.[18] A first abstractor, a nurse trained in abstracting records of children with medical complexity and blinded to the ICD-10-CA codes, will perform abstraction on the 400 charts using the medical chart extraction tool developed by our expert panel. A second abstractor will independently abstract a random 30% sample of the medical records for each cohort. Results will be compared and the reliability between the abstractors will be estimated using a kappa score. If the kappa score is low and there

are many discrepancies present between reviewers, we will reconvene as a research team to improve the reliability of the extraction tool and an additional 10% sample will be extracted and compared. This process will be repeated until the chart extraction tool is determined to be reliable.

In the second step, expert clinicians will identify two clinically derived groups; complex and not complex. These clinicians will be identified by our clinician partners on the research team. They will be composed of number of healthcare providers from across NS of varying specialties and settings (i.e. primary and tertiary care) who provide direct patient and care coordination activities to children with medical complexity and their families. Sixty children with medical complexity will be identified from relevant clinical areas that fit within our study's gold standard definition. Additionally, a cohort of 400 controls who are children without medical complexity, according to our study's gold standard definition will be identified from a range of clinical areas by these clinicians. All clinical experts participating in this step will undergo training prior to identifying patients for inclusion to ensure they understand the gold-standard definition and the clinical variables that informed the development of the medical chart audit tool.

Sample size and data analysis: There are 4100 children and youth discharged from our pediatric hospital in NS each year and the majority (3200) reside in NS. Assuming the prevalence rate of children with medical complexity is 0.67%, an estimated 21 children with medical complexity are discharged to a NS residence each year.[3] Personal communication with expert clinicians at the pediatric hospital suggests that there are approximately 50 children with medical complexity being discharged at the pediatric hospital per year. This provides a range estimate of the number of children with medical complexity discharged each year. To ensure a large enough cohort to power a regression analysis, we are examining prevalence over a 15-year time period.

The validation of the algorithms will occur in three parts (Figure 3). The gold-standard sample from step 1 will be evaluated using positive predictive values (PPV), to determine what percentage of algorithm-identified patients truly qualified as medically complex. The gold-standard sample from step 2 will be evaluated using sensitivity and specificity, to evaluate how well the algorithms can identify both complex and not complex patients. Finally, the two samples will be combined into a single analysis. All statistics will be estimated with 95% confidence intervals (CIs), and F-statistics will be used to investigate the trade-offs between sensitivity and specificity.[25] As medical complexity is by definition a rare occurrence, we will pay particular attention to the specificity, in order to ensure that the algorithm is not overwhelming the final cohort with false positives.

Phase 2: Prevalence of children with medical complexity in all three Maritime provinces

The algorithm identified as the "best fit" in phase 1, based on NS data, will be applied to NB's and PEI's CIHI DAD through NB-IRDT and SIDR respectively, to identify the cohort of children with medical complexity using the same eligibility parameters as NS. Statistical code to generate the cohorts developed in NS during the validation step, will be utilized, with minor modification as needed.

Prevalence of medical complexity will be calculated using discharge abstract data during the years 2003-2017 in each Maritime province. Individual level data will use encrypted health card numbers and birthdate in month/year as unique identifiers to link each child's data across datasets and over time.

Sample size and data analysis: To estimate the annual prevalence, and corresponding 95% CIs, of children with medical complexity per 100,000 pediatric population in the Maritime provinces during our study period, the number of children identified by our selected algorithm will be

divided by the total number of children (≤18 years) in the 2016 pediatric population for each province. Prevalence estimates will be stratified based on relevant variables (e.g. age, sex).

Phase 3: Patterns of healthcare utilization

Patterns of healthcare utilization including hospital admissions, outpatient visits, and same-day surgeries during the years 2003-2017, or up to 18 years of age will be described in each Maritime province. Individual level data will use encrypted health card numbers and birthdate in month/year as unique identifiers to link each child's data across datasets and over time.

Sample size and data analysis: We will estimate annual rates, and their corresponding 95% CIs, for several types of healthcare utilization. Specifically, data availability allows describing rates of acute hospitalizations, unplanned readmissions, day surgeries, hospital-based outpatient visits, community-based primary care visits and specialty care visits. Healthcare utilization follow-up will begin at the index date. The index date will be defined as the first discharge date with a chronic condition, by the "best fit" algorithm. Children will be censored at the end of the study period or sooner if they die or turn 18 before 2017. The numerator will be the total number of annual records for each child in the cohort. Each child will contribute one year of person-time to the denominator for every year they are alive and living in the province. We will describe results from each province separately by summarizing findings and comparing healthcare utilization across provinces.

Discussion

This work will contribute to a priority task, identified by Children's Healthcare Canada, for a national effort to support system change for children with medical complexity.[8] The opportunity

to comprehensively validate algorithms, that is able to identify children with medical complexity, addresses a significant gap in health research and is essential for better management and improvement of health outcomes. The methods we develop (e.g. the gold-standard definition) may be useful to other jurisdictions across Canada that also want to validate the algorithms specifically for their region. Further, selection of an algorithm to identify children most accurately with medical complexity will allow us to characterize population-level prevalence and identify patterns in health service utilization, as well as identify gaps in service, for this vulnerable population. The formation of this cohort will act as a starting point to conduct future population-based research that can contribute evidence to support health system planning and policy making and help improve health outcomes. The involvement of key stakeholders throughout this project will ensure knowledge of our research findings are translated to directly influence clinical practice and health system and policy change by providing data to more appropriately outline health resource gaps.

Limitations and Mitigating Strategies

Our team has reflected on potential challenges in the conduct of our study and have planned strategies accordingly to mitigate these difficulties should they arise. A challenge lies in the lack of consensus about how to operationalize a gold-standard definition of children with medical complexity. Our plan to convene an expert panel and employ multiple methods of algorithm validation will address this limitation. Simon et al.[9] and Feudtner et al.'s[10] administrative data algorithms were developed and validated using ICD-CM codes, while in Canada, we use the ICD-CA iteration of the global ICD codes. The differences between these iterations are expected to be negligible as they relate to administrative needs, not the medical diagnoses. Additional limitations concerning the use of health data are the availability and accuracy of important

variables of interest. For example, the order that diagnoses are coded (e.g. primary diagnosis, secondary diagnoses) is not always consistent. To help mitigate this, all diagnoses from a patient encounter coded in the CIHI DAD will be included. Finally, estimating healthcare utilization using health administrative data has limitations as not all services are reflected in existing databases. Our team of patient partners, researchers, clinicians and administrators recognize the importance of capturing alternative or relevant non-health service utilization (e.g. massage therapy, informal respite care) and indirect costs for families (e.g. travel, lost time at work). We have already begun to explore the use of additional administrative data from other sources as well as qualitative interviews with patients, families, and providers in a future study.

Competing interests

The authors declare that they have no competing interests.

Funding

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Authors' contributions

JAC conceived of and designed the study, secured funding for the project, drafted the study protocol. SB, SS, SM contributed to the study design and the draft of the protocol. MS, JS, SB, SB, EJ, MAM, SK contributed to the study protocol. JC is the parent knowledge user on this project and will provide guidance during all phases. HM prepared this manuscript from the study

protocol. All authors critically appraised the intellectual content of the manuscript and provided input and revisions. All authors read and approved the final manuscript.

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Figure 1. *Outline of project methods*

Figure 2. *Identification of a gold-standard sample of cases with medical complexity (complex) and controls (not complex)*

Figure 3. *Analysis of gold-standard sample of cases with medical complexity (complex) and controls (not complex)*

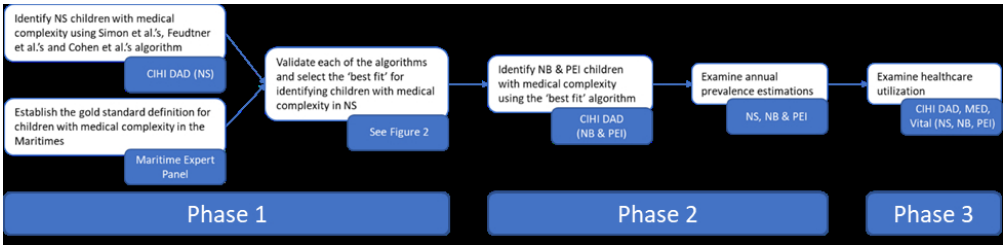

Figure 1. Outline of project methods

419x100mm (59 x 59 DPI)

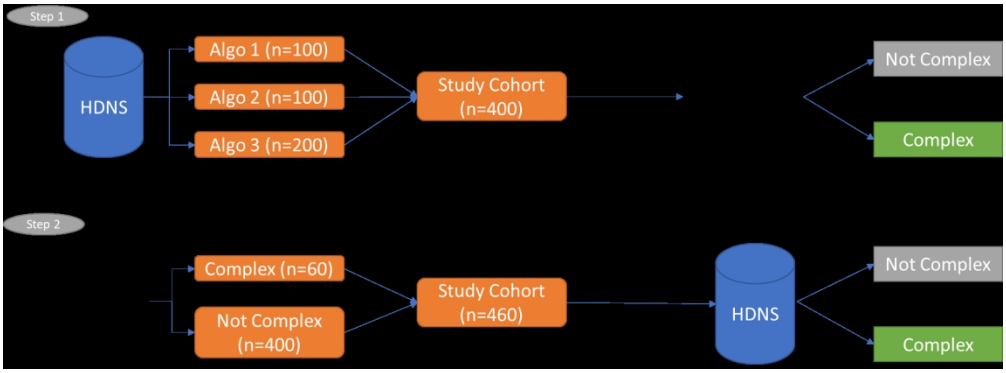

Figure 2. Identification of a gold-standard sample of cases with medical complexity (complex) and controls (not complex)

807x294mm (59 x 59 DPI)

Step 1		IWK		
		Complex	Not Complex	PPV
Algo 1	Complex (n=100)	Y	Z	Y/(Y+Z)
Algo 2	Complex (n=100)	Y	Z	Y/(Y+Z)
Algo 3a	Complex (n=100)	Y	Z	Y/(Y+Z)
Algo 3b	Complex (n=100)	Y	Z	Y/(Y+Z)

Step 2	Algo 1		Algo 2		Algo 3a		Algo 3b	
	Complex	Not Complex	Complex	Not Complex	Complex	Not Complex	Complex	Not Complex
Complex (n=60)	A	B	A	B	A	B	A	B
Not Complex (n=400)	C	D	C	D	C	D	C	D

Step 1+2	Algo 1		Algo 2		Algo 3a		Algo 3b	
	Complex	Not Complex	Complex	Not Complex	Complex	Not Complex	Complex	Not Complex
Complex	A+Y	B	A+Y	B	A+Y	B	A+Y	B
Not Complex	C+Z	D	C+Z	D	C+Z	D	C+Z	D

Figure 3. Analysis of gold-standard sample of cases with medical complexity (complex) and controls (not complex)

442x238mm (59 x 59 DPI)

STROBE Checklist

	Item No	Recommendation	Line number
Title and abstract	1	(*a*) Indicate the study's design with a commonly used term in the title or the abstract	N/A
		(*b*) Provide in the abstract an informative and balanced summary of what was done and what was found	56-81
Introduction			
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	87-155
Objectives	3	State specific objectives, including any prespecified hypotheses	156-164
Methods			
Study design	4	Present key elements of study design early in the paper	181-199
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	230-235, 304-307
Participants	6	(*a*) Give the eligibility criteria, and the sources and methods of case ascertainment and control selection. Give the rationale for the choice of cases and controls	208-209, 230-235
		(*b*) For matched studies, give matching criteria and the number of controls per case	N/A
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	274-283, 298-302, 308-318
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group	174-180
Bias	9	Describe any efforts to address potential sources of bias	241-245, 274
Study size	10	Explain how the study size was arrived at	266-273
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why	273-283, 298-302, 308-318
Statistical methods	12	(*a*) Describe all statistical methods, including those used to control for confounding	273-283, 298-302, 308-318
		(*b*) Describe any methods used to examine subgroups and interactions	273-283, 298-302, 308-318
		(*c*) Explain how missing data were addressed	N/A
		(*d*) If applicable, explain how matching of cases and controls was addressed	N/A
		(*e*) Describe any sensitivity analyses	274-283
Results			
Participants	13*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed	N/A

		(b) Give reasons for non-participation at each stage	N/A
		(c) Consider use of a flow diagram	N/A
Descriptive data	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders	N/A
		(b) Indicate number of participants with missing data for each variable of interest	N/A
Outcome data	15*	Report numbers in each exposure category, or summary measures of exposure	N/A
Main results	16	(*a*) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included	N/A
		(*b*) Report category boundaries when continuous variables were categorized	N/A
		(*c*) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period	N/A
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	N/A
Discussion			
Key results	18	Summarise key results with reference to study objectives	N/A
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias	336-354
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence	N/A
Generalisability	21	Discuss the generalisability (external validity) of the study results	N/A
Other information			
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	41-43

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Identifying children with medical complexity in administrative datasets in a Canadian context: study protocol

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Identifying children with medical complexity in administrative datasets in a Canadian context: study protocol

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Abstract

Introduction: Children with medical complexity and their families are an important population of interest within the Canadian healthcare system. Despite representing less than 1% of the pediatric population, children with medical complexity require extensive care and account for one third of pediatric healthcare expenditures. Opportunities to conduct research to assess disparities in care and appropriate allocation of health resources relies on the ability to accurately identify this heterogeneous group of children. This study aims to better understand the population of children with medical complexity in the Canadian Maritimes, including Nova Scotia (NS), New Brunswick (NB) and Prince Edward Island (PEI). This will be achieved through three objectives: (1) Evaluate the performance of three algorithms to identify children with medical complexity in the Canadian Maritimes in administrative data; then using the "best fit" algorithm (2) Estimate the prevalence of children with medical complexity in the Canadian Maritimes from 2003-2017; and (3) Describe patterns of healthcare utilization for this cohort of children across the Canadian Maritimes.

Methods and analysis: The research will be conducted in three phases. In Phase 1, an expert panel will co-develop a gold-standard definition of pediatric medical complexity relevant to the Canadian Maritime population. A two-gate validation process will then be conducted using NS data and the gold-standard definition to determine the "best fit" algorithm. During Phase 2 the "best fit" algorithm will be applied to estimate the prevalence of children with medical complexity in NS, NB, and PEI. Finally, in Phase 3 will describe patterns of healthcare utilization across the Canadian Maritimes.

Ethics and dissemination: Ethics approval for this protocol was granted by the institutional research ethics board at the IWK Health Centre (REB # 1026245). A waiver of consent was

approved. This study will use an integrated knowledge translation (iKT) approach, where end users are involved in each stage of the project, which could increase uptake of the research into policy and practice. The findings of this research study will be submitted for publication and dissemination through conference presentations and with our end users.

Keywords

Medical complexity, administrative data, pediatric, healthcare utilization

Strength and limitations of this study

- Clinical experts and families with lived experience will develop and operationalize a gold standard definition for children with medical complexity in the Maritimes.
- Multiple methods will be employed to validate the "best fit" algorithm.
- Certain clinical variables relevant to describing children with medical complexity may not be available within health administrative data.
- Health administrative data is limited by type of provider/service and reporting and extraction practices.

Introduction

Since 2010, children with medical complexity have gained increasing attention as an important population in critical need of practice and policy reform within the Canadian healthcare system.[1,2] Medical complexity is generally characterized as having one or more complex chronic

condition(s) associated with significant functional limitations, high health resource use and family-identified needs.[1,3] One seminal Canadian study conducted nearly 10 years ago, suggested that despite representing less than 1% of the pediatric population in Ontario, children with medical complexity account for one third of pediatric healthcare expenditures.[1] Adding to the findings stemming from this study, a recent publication from The Canadian Institute for Health Information (CIHI) reported that in 2015-2016 there were 948 per 100,000 children and youth with medical complexity.[4] Recent findings from the United States also suggest that the prevalence of children living with medical complexity is increasing,[5,6] likely due to the increased survival rates of a variety of life-limiting and life-threatening conditions.[3] Information regarding children with medical complexity has been primarily derived using reports from the United States with only a few seminal reports stemming from Canadian-based data.[4,7] However, due to important population differences, it is critical to understand this vulnerable population in the Canadian context to assist in mapping health outcomes and healthcare utilization for this vulnerable population. Doing so may ensure that more relevant, needs based and sustainable models of service delivery are developed to optimize quality health experiences and outcomes for children and their families.

Caregivers of children with medical complexity carry a tremendous amount of responsibility, stress and financial burden to attend to their intensive care needs.[7,8] Opportunities to assess disparities in care, and appropriately allocate health resources to better serve children with medical complexity, are dependent on being able to accurately identify them.[1] A standard definition of children with medical complexity does not currently exist in Canada and determining a classification system for these children is a necessary first step to optimize health service delivery for this vulnerable group of children. Given the heterogeneity of this group, with

variation in the severity and combination of comorbid chronic conditions, there are unique challenges when attempting to identify a cohort at a population level.[1] Identification begins with selecting a method to classify and characterize children with medical complexity.

The Canadian Maritimes is a unique pediatric care setting composed of three provinces: Nova Scotia (NS), New Brunswick (NB), and Prince-Edward-Island (PEI). The only pediatric tertiary care facility in the Maritimes is in NS, which results in families crossing provincial jurisdictions for specialty care. This adds a layer of contextual difference that may intersect with and/or contribute to medical complexity and health care utilization in the Maritimes.

Health administrative data algorithms

A recent scoping review by Breneol *et al*. entitled "Improving Health Care for Children with Medical Complexity Through the Use of Health Administrative Data: A Scoping Review", examining the use of health administrative data in the study of children with medical complexity, identified three methods that are commonly used to identify this cohort: (1) Cohen *et al*.'s[3] list of complex chronic conditions, technological assistance and neurological impairment; (2) Simon *et al*.'s[9] *Pediatric Medical Complexity Algorithm* (PMCA); and (3) Feudtner *et al*.'s[10] complex chronic conditions classification system. Cohen *et al*.'s algorithm was developed in Ontario using hospital discharge data but has not yet been validated. Algorithms developed by Simon *et al*.[9] and Feudtner *et al*.[10] have not undergone validation in Canadian health data.

Algorithm 1. Cohen *et al*.[3] operationalized a definitional framework in an Ontario administrative dataset by using sets of ICD-10-CA codes relevant to complex chronic conditions, neurological impairment and technological assistance. This definitional framework aligns with the work of other experts conducting research on this population outside of Canada.[5,11,12] However, the

sensitivity and specificity of the lists of Canadian Classification of Procedures and Interventions for identifying children with medical complexity in Canada has not yet been reported.

Algorithm 2. Simon *et al*.[13] used the Chronic Disability Payment System as a guide to develop the *Pediatric Medical Complexity Algorithm* (PMCA) at the Seattle Children's Hospital in Washington State. The authors employed a systematic process beginning with the development of consensus definitions for three levels of medical complexity (complex chronic, noncomplex chronic and no chronic conditions). They classified children with medical complexity as those who fit their definition for having a complex chronic condition. They proceeded by selecting and modifying an existing algorithm to conform to the consensus definitions, and selected a gold standard pediatric population through medical chart review to evaluate the sensitivity and specificity of the algorithm.[13] The PMCA had high sensitivity and specificity (complex chronic: 86% sensitivity, 86% specificity; non-complex chronic: 65% sensitivity, 84% specificity; children without complex chronic: 77% sensitivity and 93% specificity) for identifying children with medical complexity and was subsequently updated and validated for ICD-10-CM codes in 2018.[9] This algorithm has a least and more conservative version depending on the type of data available to researchers. The least conversative version was shown to perform better in hospital discharge data and the most conservative version was shown to perform better in claims and billing data.[13]

Algorithm 3. Feudtner *et al*.[14] followed a similar approach as Simon *et al*.[9] in Washington State, creating a working definition of complex chronic conditions and subsequently operationalized the definition using clinical knowledge and existing literature. This resulted in a list of conditions and their corresponding ICD-9-CM codes that are highly sensitive (87%) for identifying children with complex chronic conditions. The algorithm was updated in 2014 and the ICD codes were

translated into the 10th edition (ICD-10-CM), including both diagnostic and procedural codes indicative of technology dependence or organ transplantation.[10] Lindley *et al.* has evaluated the predictive ability of the original and modified versions and determined that the modified version, which will be used in this study, better operationalizes medical complexity.[15]

Aims and objectives

The aim of this study is to better understand the Canadian Maritime's population of children with medical complexity. The ability to accurately identify children with medical complexity is necessary to conduct research that may inform the design and implementation of successful population-level policies and interventions. This study will be conducted in the Canadian Maritime provinces: NS, NB, and PEI. This study will: (1) evaluate the performance of three administrative data algorithms to identify children with medical complexity; then use the "best fit" algorithm to (2) estimate the prevalence; and (3) describe patterns of healthcare utilization for this cohort of children from 2003-2017.

Methods and analysis

Data Sources

The study cohorts will be constructed using the Canadian Institute for Health Information Discharge Abstract Databases (CIHI DAD) from Nova Scotia and the prevalence and health care utilization estimates will use the CIHI DAD, Vital Statistics (VITAL), Insured Patient Registry (MASTER), and Physician Billing Databases (MED) from each province. The administrative datasets for NS, NB and PEI are housed by their respective data repository organizations: Health

Data Nova Scotia (HDNS), New Brunswick Institute for Research Data and Training (NB-IRDT), and Secure Island Data Repository (SIDR).

Study Design

This study will be conducted in three phases to identify children with medical complexity in the Canadian Maritimes and their healthcare utilization over a 15-year period (Figure 1).

In Phase 1, the "best fit" algorithm for identifying children with medical complexity in the Maritimes will be selected using sensitivity and specificity. Using NS administrative data, a cohort will be established for each of the three algorithms. A gold-standard definition will be developed for children with medical complexity in the Maritime provinces through an expert consensus meeting. Through the consensus process, this definition will subsequently be translated into a chart audit tool. The "best fit" algorithm will be selected using a two-gate case-control study design.

In Phase 2, the prevalence of children with medical complexity in the Maritime provinces will be estimated. This will be done in all three Maritime provinces using NS, NB and PEI administrative data cohorts identified by the previously selected "best fit" algorithm.

In Phase 3, patterns of healthcare utilization of children with medical complexity across the Maritime provinces will be described.

Phase 1: Identify the "best fit" algorithm for identifying children with medical complexity in the Maritimes

Developing a gold-standard definition: A multidisciplinary expert consensus meeting will be convened to co-develop a gold-standard definition of children with medical complexity relevant to the Canadian Maritimes. The consensus meeting will involve relevant stakeholders, including

a minimum of two clinicians, two researchers and a parent from each of the three Maritime provinces. Parent participants will be invited to attend a pre-meeting virtual session to introduce them to the consensus meeting agenda and provide them with an opportunity to ask questions related to the project. A parent partner and research coordinator will facilitate the pre-meeting session. An infographic will be developed and distributed prior to the consensus meeting to introduce participants to existing frameworks and literature that will help guide the conceptualization of medical complexity, functional limitations, and other related concepts. Outputs from relevant exploratory work conducted by members of the research team will be presented during the first session of the consensus meeting.[16] Parent and clinician experts will participate in structured breakout sessions to examine all meeting materials in the context of their clinical or lived experience. Sessions will be audio-recorded, and a note-taker will track participation. After the breakout sessions are complete, a consensus based decision-making process will be used where participants work together through discussion to reach agreement on the elements of a clinically meaningful gold-standard definition for this pediatric population in the Maritimes.[17] The definition will be circulated to the expert stakeholder group after the meeting for their review and comments and will be edited through email.

To operationalize the definition, a sub-group of clinicians with expertise working with this patient population will use the gold-standard definition to develop a list of clinical variables that will form the basis of a medical chart extraction tool. The expert group will complete this task over a series of meetings, and the final tool will be reviewed by clinicians from both NB and PEI. A coding manual will be developed to accompany the medical chart extraction tool.

Establishing a Nova Scotia administrative data cohort: The algorithm validation process will be undertaken using administrative data from the NS provincial data repository organization,

HDNS. Children living in NS with medical complexity will be identified and classified with specific ICD-10-CA codes defined within each of the three respective algorithms, resulting in a distinct cohort being derived from each algorithm. Due to the uncertainty of how the Simon *et al*. algorithm will perform using Canadian health administrative data, both the least and more conservative versions will be used. All children aged 0 to 18 years of age who have a discharge record in NS CIHI DAD from 2003-2017 will be eligible for inclusion.

Examining the "best fit": Given the prevalence of children with medical complexity is rare, a two-gate case-control study design will be used to validate the algorithms (Figure 2).[18] This will be done in two steps. First, a stratified random sample of 100 children will be selected from each of the cohorts derived by the algorithms to participate in a chart audit.[19] The chart audit will be conducted by nurses trained in abstracting records of children with medical complexity using the medical chart extraction tool developed by the expert panel. The audit will begin with all abstractors independently extracting the same five charts to calibrate coding. Results will be compared, and the inter-rater reliability will be estimated using a kappa score. If the kappa score is low and there are many discrepancies, the chart audit tool will be refined, and further training provided to the abstractors. An additional five charts will then be extracted and compared. This process will be repeated until the chart extraction is determined to be reliable. A total of 30% of the records in each cohort will be independently abstracted by a second reviewer.

In the second step, two clinically derived groups of children (complex and not complex) will be established by healthcare providers from across NS to contribute to the validation procedure. Healthcare providers who contribute to this process will be from varying specialties and settings (i.e., primary and tertiary care) and provide direct patient care or care coordination activities to children with medical complexity and their families. Sixty children with medical complexity will

be identified from relevant clinical areas that fit within the study's gold standard definition. Additionally, a cohort of 400 controls who are children without medical complexity, according to the study's gold standard definition will be identified. All clinical experts participating in this step will undergo training prior to identifying patients for inclusion to ensure they understand the gold-standard definition and the clinical variables that informed the development of the medical chart audit tool.

Sample size and data analysis: There are approximately 3200 Nova Scotian children discharged each year from the pediatric tertiary care facility in the Maritimes. Cohen *et al.* has estimated a 0.67% prevalence of children with medical complexity, therefore we estimated approximately 21 Nova Scotian children with medical complexity are discharged yearly.[3] However, local expert clinicians estimated that there are closer to 50 Nova Scotian children discharged each year with medical complexity. To ensure a large enough cohort to power a regression analysis, prevalence will be examined over a 15-year period.

The validation of the algorithms will occur in three parts (Figure 3). The algorithm-identified cohorts from step 1 will be evaluated using positive predictive values (PPV), to determine what percentage of the algorithm-identified patients truly qualified as medically complex. The gold-standard sample from step 2 will be evaluated using sensitivity and specificity, to evaluate how well the algorithms can identify both complex and not complex patients. Finally, the two samples will be combined into a single analysis. All statistics will be estimated with 95% confidence intervals (CIs), and F-statistics will be used to investigate the trade-offs between sensitivity and specificity.[20] As medical complexity is by definition a rare occurrence, particular attention will be paid to the specificity, in order to ensure that the algorithm is not overwhelming the final cohort with false positives.

Phase 2: Prevalence of children with medical complexity in all three Maritime provinces

The algorithm identified as the "best fit" in phase 1, based on NS data, will be applied to NB's and PEI's CIHI DAD through NB-IRDT and SIDR respectively, to identify the cohort of children with medical complexity using the same eligibility parameters as NS. Statistical code to generate the cohorts developed in NS during the validation step, will be utilized, with minor modification as needed.

Prevalence of medical complexity will be calculated using discharge abstract data during the years 2003-2017 in each Maritime province. Individual level data will use encrypted health card numbers and birthdate in month/year as unique identifiers to link each child's data across datasets and over time.

Sample size and data analysis: To estimate the annual prevalence, and corresponding 95% CIs, of children with medical complexity per 100,000 pediatric population in the Maritime provinces during the study period, the number of children identified by the selected algorithm will be divided by the total number of children (≤18 years) in the 2016 pediatric population for each province. Prevalence estimates will be stratified based on relevant variables (e.g., age, sex).

Phase 3: Patterns of healthcare utilization

Patterns of healthcare utilization including hospital admissions, outpatient visits, and same-day surgeries during the years 2003-2017, or up to 18 years of age will be described in each Maritime province. Individual level data will use encrypted health card numbers and birthdate in month/year as unique identifiers to link each child's data across datasets and over time.

Sample size and data analysis: Annual rates, and their corresponding 95% CIs, will be estimated for several types of healthcare utilization. Specifically, data availability allows describing rates

of acute hospitalizations, unplanned readmissions, day surgeries, hospital-based outpatient visits, community-based primary care visits and specialty care visits. Healthcare utilization follow-up will begin at the index date. The index date will be defined as the first discharge date with a complex chronic condition, by the "best fit" algorithm. Children will be censored at the end of the study period or sooner if they die or turn 18 before 2017. The numerator will be the total number of annual records for each child in the cohort. Each child will contribute one year of person-time to the denominator for every year they are alive and living in the province. Results from each province will be described separately by summarizing findings and comparing healthcare utilization across provinces.

This project is currently in phase 1 in the process of identifying the "best fit" algorithm using the two-gate case-control study. The project is anticipated to be completed by December 2022.

Patient and Public Involvement

This study will use an integrated knowledge translation (iKT) approach, where end users are involved in each stage of the project.[21] The team of researchers, clinicians, administrators and patient partners will work together throughout the research process to develop outputs that are relevant to knowledge user needs, including development of the research objectives and methods. Opportunities will be created to engage additional parents, clinicians, and administrators to inform different stages of the work as the project unfolds. The proposed iKT method was chosen to increase uptake of the research into policy and practice.[22]

Ethics and dissemination

Ethical approval was received from the primary research institution in NS (IWK Health Centre; REB#: 1026245). A waiver of consent has been granted by the research ethics board and therefore informed participant consent is not required for this study. Ethical approval will be obtained at the appropriate institutions in NB and PEI before data abstraction commences in these provinces.

This study will use an iKT approach, where end users are involved in each stage of the project, which could increase uptake of the research into policy and practice. The findings of this research study will be submitted for publication and dissemination through conference presentations and with our end users.

Discussion

To our knowledge, this will be the first time the performance of these three algorithms will be evaluated using Canadian health administrative data. While the recent CIHI report identified children with medical complexity across Canada to provide population-level prevalence rates and an overview of their healthcare utilization, this study will add to the existing knowledge by including a longer to identify any potential trends.[4]

Findings from this work may contribute to a priority task, identified by Children's Healthcare Canada, for a national effort to support system change for children with medical complexity.[7] The opportunity to comprehensively identify a "best fit" algorithm, that is able to describe children with medical complexity, addresses a significant gap in health research and is essential for better management and improvement of health outcomes. The methods developed (e.g., two-gate validation procedure, chart audit tool) may be useful to other jurisdictions across Canada

wishing to validate algorithms specifically for their region. Further, identifying a "best fit" algorithm to describe children with medical complexity may allow us to characterize population-level prevalence and identify patterns in health service utilization, as well as identify gaps in service, for this vulnerable population. The formation of this cohort may act as a starting point to conduct future population-based research that can contribute evidence to support health system planning and policy making and help improve health outcomes. Key stakeholders are being involved throughout this project to help ensure research findings related to service and resource gaps are translated to health system and policy change.

Limitations and Mitigating Strategies

The team has reflected on potential challenges in the conduct of the study and have planned strategies accordingly to mitigate these difficulties should they arise. A challenge lies in the lack of consensus about how to operationalize a gold-standard definition of children with medical complexity. The plan to convene an expert panel and employ multiple methods of algorithm validation was chosen to address this limitation. Simon *et al.*[13] and Feudtner *et al.*'s[10] administrative data algorithms were developed and validated using ICD-CM codes, while Canada uses the ICD-CA iteration of the global ICD codes. The differences between these iterations are expected to be negligible as they relate to administrative needs, not the medical diagnoses. Additional limitations concerning the use of health data are the availability and accuracy of important variables of interest. For example, the order that diagnoses are coded (e.g., primary diagnosis, secondary diagnoses) is not always consistent. To help mitigate this, all diagnoses from a patient encounter coded in the CIHI DAD will be included. Finally, estimating healthcare utilization using health administrative data has limitations as not all services are reflected in existing databases. The team of patient partners, researchers, clinicians, and

administrators recognize the importance of capturing alternative or relevant non-health service utilization (e.g., massage therapy, informal respite care) and indirect costs for families (e.g., travel, lost time at work). The use of additional administrative data from other sources is being explored as well as qualitative interviews with patients, families, and providers in a future study.

Competing interests

The authors declare that they have no competing interests.

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Contributors

JAC conceived of and designed the study, secured funding for the project, drafted the study protocol. SB, SS, SM contributed to the study design and the draft of the protocol. MS, JS, SB, SB, EJ, MAM, SK contributed to the study protocol. JC is the parent knowledge user on this project and has provided guidance during all phases. HM prepared this manuscript from the study protocol. All authors critically appraised the intellectual content of the manuscript and provided input and revisions. All authors read and approved the final manuscript.

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Figure 1. Outline of project methods

Figure 2. Identification of a gold-standard sample of cases with medical complexity (complex) and controls (not complex)

Figure 3. Analysis of gold-standard sample of cases with medical complexity (complex) and controls (not complex)

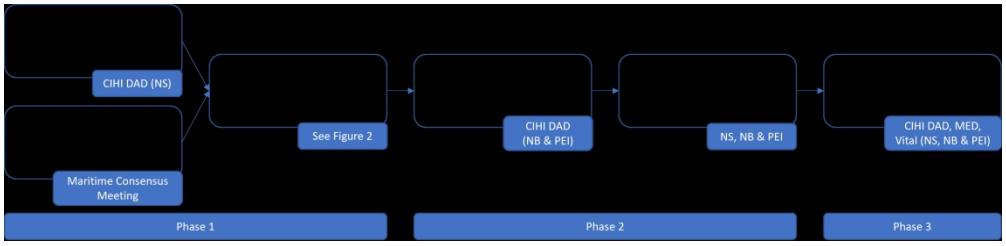


Figure 1. Outline of project methods

346x82mm (300 x 300 DPI)

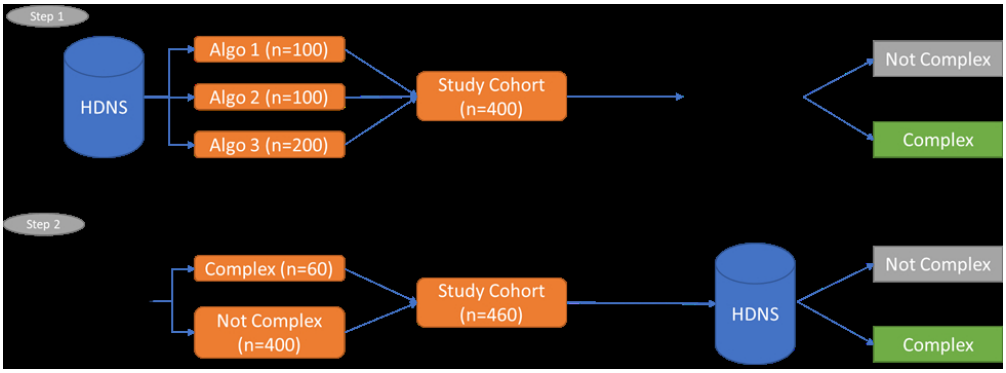
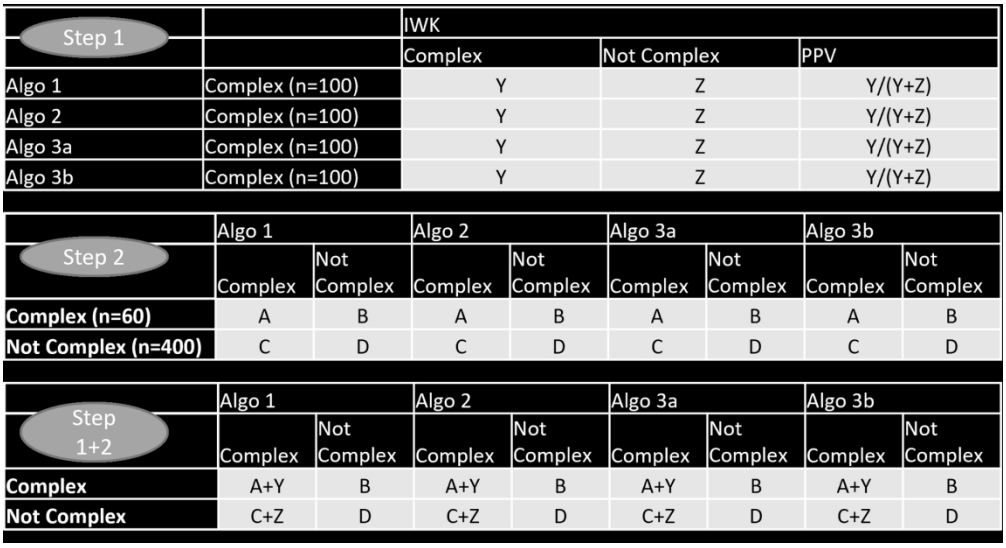


Figure 2. Identification of a gold-standard sample of cases with medical complexity (complex) and controls (not complex)

82x30mm (300 x 300 DPI)



Step 1		IWK		
		Complex	Not Complex	PPV
Algo 1	Complex (n=100)	Y	Z	Y/(Y+Z)
Algo 2	Complex (n=100)	Y	Z	Y/(Y+Z)
Algo 3a	Complex (n=100)	Y	Z	Y/(Y+Z)
Algo 3b	Complex (n=100)	Y	Z	Y/(Y+Z)

Step 2	Algo 1		Algo 2		Algo 3a		Algo 3b	
	Complex	Not Complex	Complex	Not Complex	Complex	Not Complex	Complex	Not Complex
Complex (n=60)	A	B	A	B	A	B	A	B
Not Complex (n=400)	C	D	C	D	C	D	C	D

Step 1+2	Algo 1		Algo 2		Algo 3a		Algo 3b	
	Complex	Not Complex	Complex	Not Complex	Complex	Not Complex	Complex	Not Complex
Complex	A+Y	B	A+Y	B	A+Y	B	A+Y	B
Not Complex	C+Z	D	C+Z	D	C+Z	D	C+Z	D

Figure 3. Analysis of gold-standard sample of cases with medical complexity (complex) and controls (not complex)

174x93mm (300 x 300 DPI)

STROBE Checklist

	Item No	Recommendation	Line number
Title and abstract	1	(*a*) Indicate the study's design with a commonly used term in the title or the abstract	N/A
		(*b*) Provide in the abstract an informative and balanced summary of what was done and what was found	56-81
Introduction			
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	87-155
Objectives	3	State specific objectives, including any prespecified hypotheses	156-164
Methods			
Study design	4	Present key elements of study design early in the paper	181-199
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	230-235, 304-307
Participants	6	(*a*) Give the eligibility criteria, and the sources and methods of case ascertainment and control selection. Give the rationale for the choice of cases and controls	208-209, 230-235
		(*b*) For matched studies, give matching criteria and the number of controls per case	N/A
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	274-283, 298-302, 308-318
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group	174-180
Bias	9	Describe any efforts to address potential sources of bias	241-245, 274
Study size	10	Explain how the study size was arrived at	266-273
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why	273-283, 298-302, 308-318
Statistical methods	12	(*a*) Describe all statistical methods, including those used to control for confounding	273-283, 298-302, 308-318
		(*b*) Describe any methods used to examine subgroups and interactions	273-283, 298-302, 308-318
		(*c*) Explain how missing data were addressed	N/A
		(*d*) If applicable, explain how matching of cases and controls was addressed	N/A
		(*e*) Describe any sensitivity analyses	274-283
Results			
Participants	13*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed	N/A

		(b) Give reasons for non-participation at each stage	N/A
		(c) Consider use of a flow diagram	N/A
Descriptive data	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders	N/A
		(b) Indicate number of participants with missing data for each variable of interest	N/A
Outcome data	15*	Report numbers in each exposure category, or summary measures of exposure	N/A
Main results	16	(*a*) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included	N/A
		(*b*) Report category boundaries when continuous variables were categorized	N/A
		(*c*) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period	N/A
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	N/A
Discussion			
Key results	18	Summarise key results with reference to study objectives	N/A
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias	336-354
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence	N/A
Generalisability	21	Discuss the generalisability (external validity) of the study results	N/A
Other information			
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	41-43