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

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SCHOLARONE™
Manuscripts

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5 2 context: Study protocol
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47 37 **Ethics approval and consent to participate** 48

49
50 38 Ethics approval for this protocol was granted by the institutional research ethics board at the
51

52
53 39 IWK Health Centre (REB # 1026245). A waiver of consent was approved.
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3 **41 Abstract**
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5
6 **42 Introduction:** Children with medical complexity and their families are an important population of
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8 **43** interest within our Canadian healthcare system. Despite representing less than 1% of the
9
10 **44** pediatric population, children with medical complexity require extensive care and account for
11
12 **45** one third of pediatric healthcare expenditures. Opportunities to conduct research to assess
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14 **46** disparities in care and appropriate allocation of health resources relies on our ability to accurately
15
16 **47** identify this heterogeneous group of children.
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20 **48 Objectives:** This study aims to better understand the population of children with medical
21
22 **49** complexity in the Canadian Maritimes, including Nova Scotia (NS), New Brunswick (NB) and
23
24 **50** Prince Edward Island (PEI). This will be achieved through three objectives: (1) Evaluate the
25
26 **51** performance of three algorithms to identify children with medical complexity in the Canadian
27
28 **52** Maritimes in administrative data; then using the “best fit” algorithm we will (2) Estimate the
29
30 **53** prevalence of children with medical complexity in the Canadian Maritimes from 2003-2017; and
31
32 **54** (3) Describe patterns of healthcare utilization for this cohort of children across the Canadian
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34 **55** Maritimes.
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39 **56 Methods:** This research will be conducted in three phases. In Phase 1, an expert panel will co-
40
41 **57** develop a gold-standard definition of pediatric medical complexity relevant to the Canadian
42
43 **58** Maritime population. A two-gate validation process will then be conducted using NS data and
44
45 **59** the gold-standard definition to determine the “best fit” algorithm. During Phase 2 we will apply
46
47 **60** the “best fit” algorithm to estimate the prevalence of children with medical complexity in NS,
48
49 **61** NB and PEI. Finally, in Phase 3 will describe patterns of healthcare utilization across the
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51 **62** Canadian Maritimes.
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63 Discussion: The project outputs will provide critical information that will guide the design and
64 implementation of future population-level research. They will inform the development of
65 policies and interventions to improve the healthcare delivery and health outcomes of children
66 with medical complexity and their families.

68 **Keywords**

69 Medical complexity, administrative data, pediatric, healthcare utilization

71 **Strength and limitations of this study**

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

83 **Introduction**

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3 84 Since 2010, children with medical complexity have gained increasing attention as an important
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5 85 population in critical need of practice and policy reform within our Canadian healthcare
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8 86 system.^{1,2} Medical complexity is generally characterized as having one or more complex chronic
9
10 87 condition(s) associated with significant functional limitations, high health resource use and
11
12 88 family-identified needs.^{1,3} One seminal Canadian study conducted nearly 10 years ago, suggested
13
14 89 that despite representing less than 1% of the pediatric population in Ontario, children with
15
16 90 medical complexity account for one third of pediatric healthcare expenditures.¹ Adding to the
17
18 91 findings stemming from this study, a recent publication from The Canadian Institute for Health
19
20 92 Information reported that in 2015-2016 there were 948 per 100,000 children and youth with
21
22 93 medical complexity.⁴ Recent findings from the United States also suggest that the prevalence of
23
24 94 children living with medical complexity is increasing,^{5,6} likely due to the increased survival rates
25
26 95 of a variety of life-limiting and life-threatening conditions.³ To date, knowledge about children
27
28 96 with medical complexity has relied primarily on data and generalized findings from the United
29
30 97 States with only two main reports being conducted the Canadian health care system.^{7,8} However,
31
32 98 due to important population differences, it is critical that we understand this vulnerable
33
34 99 population in the Canadian context to assist in mapping health outcomes and healthcare
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36 100 utilization for this vulnerable population. Doing so will ensure that more relevant, needs based
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38 101 and sustainable models of service delivery are developed to optimize quality health experiences
39
40 102 and outcomes for children and their families.
41
42 103 Caregivers of children with medical complexity carry a tremendous amount of responsibility,
43
44 104 stress and financial burden to attend to their intensive care needs.^{8,9} Opportunities to assess
45
46 105 disparities in care, and appropriately allocate health resources to better serve children with
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48 106 medical complexity, are dependent on being able to accurately identify them.¹ A standard
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3 107 definition of children with medical complexity does not currently exist in Canada and
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5 108 determining a classification system for these children is a necessary first step to optimize health
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7 109 service delivery for this vulnerable group of children. Given the heterogeneity of this group, with
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9
10 110 variation in the severity and combination of comorbid chronic conditions, there are unique
11
12 111 challenges when attempting to identify a cohort at a population level.¹ Identification begins with
13
14 112 identifying a method to classify and characterize children with medical complexity.
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17 113 The Canadian Maritimes is a unique pediatric care setting composed of three provinces: Nova
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19 114 Scotia (NS), New Brunswick (NB), and Prince-Edward-Island (PEI). The only pediatric tertiary
20
21 115 care facility in the Maritimes is in NS, which results in families crossing provincial jurisdictions
22
23 116 for specialty care. This adds a layer of contextual difference that may intersect with and/or
24
25 117 contribute to medical complexity and health care utilization in the Maritimes.
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29 118 Definitional frameworks

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32 119 A recent scoping review, examining the use of health administrative data in the study of children
33
34 120 with medical complexity, identified three methods that are commonly used to identify this
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36 121 cohort:⁷ (1) Cohen et al.'s³ list of complex chronic conditions, technological assistance and
37
38 122 neurological impairment; (2) Simon et al.'s¹⁰ *Pediatric Medical Complexity Algorithm* (PMCA);
39
40 123 and (3) Feudtner et al.'s¹¹ complex chronic conditions classification system. Cohen et al.'s
41
42 124 algorithm was developed in Ontario using hospital discharge data but has not yet been validated.
43
44 125 Algorithms developed by Simon et al.¹⁰ and Feudtner et al.¹¹ have not undergone validation in
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46 126 Canadian health data.
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51 127 *Algorithm 1.* Cohen et al.³ operationalized a definitional framework in an Ontario administrative
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53 128 dataset by using sets of ICD-10-CA codes relevant to complex chronic conditions, neurological
54
55 129 impairment and technological assistance. This definitional framework aligns with the work of
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3 130 other experts conducting research on this population outside of Canada.^{5,12,13} However, the
4
5 131 sensitivity and specificity of the list of ICD-10-CA codes for identifying children with medical
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8 132 complexity in Canada has not yet been reported.
9

10 133 *Algorithm 2.* Simon et al.¹⁴ used the Chronic Disability Payment System as a guide to develop
11
12 134 the *Pediatric Medical Complexity Algorithm* (PMCA) at the Seattle Children's Hospital in
13
14
15 135 Washington State. The authors employed a systematic process beginning with the development
16
17 136 of consensus definitions for three levels of medical complexity (complex chronic, noncomplex
18
19 137 chronic and no chronic conditions). They classified children with medical complexity as those
20
21 138 who fit their definition for having a complex chronic condition. They proceeded by selecting and
22
23 139 modifying an existing algorithm to conform to the consensus definitions, and selected a gold
24
25
26 140 standard pediatric population through medical chart review to evaluate the sensitivity and
27
28 141 specificity of the algorithm.¹⁴ The PMCA had high sensitivity and specificity (complex chronic:
29
30 142 86% sensitivity, 86% specificity; non-complex chronic: 65% sensitivity, 84% specificity;
31
32 143 children without complex chronic: 77% sensitivity and 93% specificity) for identifying children
33
34 144 with medical complexity and was subsequently updated and validated for ICD-10-CM codes in
35
36 145 2018.¹⁰
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41 146 *Algorithm 3.* Feudtner et al.¹⁵ followed a similar approach as Simon et al.¹⁰ in Washington State,
42
43 147 creating a working definition of complex chronic conditions and subsequently operationalized
44
45 148 the definition using clinical knowledge and existing literature. This resulted in a list of conditions
46
47 149 and their corresponding ICD-9-CM codes that are highly sensitive (87%) for identifying children
48
49 150 with complex chronic conditions. The algorithm was updated in 2014 and the ICD codes were
50
51 151 translated into the 10th edition (ICD-10-CM), including both diagnostic and procedural codes
52
53
54 152 indicative of technology dependence or organ transplantation.¹¹
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153 Aims and objectives

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

162

163 **Methods**

164 Patient and Public Involvement

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

172 Data Sources

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

1
2
3 175 (MASTER), and Physician Billing Databases (MED) from each province. The administrative
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5 176 datasets for NS, NB and PEI are housed by their respective data repository organizations: Health
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8 177 Data Nova Scotia (HDNS), New Brunswick Institute for Research Data and Training (NB-
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10 178 IRDT), and Secure Island Data Repository (SIDR).

13 179 Study Design

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16 180 This study will be conducted in three phases to identify children with medical complexity in the
17
18 181 Canadian Maritimes and their healthcare utilization over a 15-year period (Figure 1).

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21 182 In Phase 1, we will select the “best fit” algorithm for identifying children with medical
22
23 183 complexity in the Maritimes using sensitivity and specificity. Using NS administrative data, we
24
25 184 will establish a cohort for each of the three algorithms. We will develop a gold-standard
26
27 185 definition for children with medical complexity in the Maritime provinces through an expert
28
29 186 consensus meeting. Through the consensus process, this definition will subsequently be
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31 187 translated into a chart audit tool. The “best fit” algorithm will be selected using a two-gate case-
32
33 188 control study design.

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37 189 In Phase 2, we will estimate the prevalence of children with medical complexity in the Maritime
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39 190 provinces. This will be done in all three Maritime provinces using NS, NB and PEI
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41 191 administrative data cohorts identified by the previously selected “best fit” algorithm.

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45 192 In Phase 3, we will describe patterns of healthcare utilization of children with medical
46
47 193 complexity across the Maritime provinces. Ethical approval was received from the primary
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49 194 research institution in NS (REB#: 1026245). A waiver of consent has been granted by our
50
51 195 research ethics board and therefore informed participant consent is not required for this study.

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

198 Phase 1: Identify the “best fit” algorithm for identifying children with medical complexity in the
199 Maritimes

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

217 We will use an internal and external validation process to evaluate the gold-standard definition.²²

218 The internal process will involve the comparison of the definition with the chart diagnosis of 3-4

219 patients identified independently by two NS clinical experts, not involved in the expert panel.

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

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

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

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3 242 are many discrepancies present between reviewers, we will reconvene as a research team to
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5 243 improve the reliability of the extraction tool and an additional 10% sample will be extracted and
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8 244 compared. This process will be repeated until the chart extraction tool is determined to be
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10 245 reliable.

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13 246 In the second step, expert clinicians will identify two clinically derived groups; complex and not
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15 247 complex. These clinicians will be identified by our clinician partners on the research team. They
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17 248 will be composed of number of healthcare providers from across NS of varying specialties and
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19 249 settings (i.e. primary and tertiary care) who provide direct patient and care coordination activities
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21 250 to children with medical complexity and their families. Sixty children with medical complexity
22
23 251 will be identified from relevant clinical areas that fit within our study's gold standard definition.
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26 252 Additionally, a cohort of 400 controls who are children without medical complexity, according
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28 253 to our study's gold standard definition will be identified from a range of clinical areas by these
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30 254 clinicians. All clinical experts participating in this step will undergo training prior to identifying
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32 255 patients for inclusion to ensure they understand the gold-standard definition and the clinical
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34 256 variables that informed the development of the medical chart audit tool.

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39 257 *Sample size and data analysis:* There are 4100 children and youth discharged from our pediatric
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41 258 hospital in NS each year and the majority (3200) reside in NS. Assuming the prevalence rate of
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43 259 children with medical complexity is 0.67%, an estimated 21 children with medical complexity
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45 260 are discharged to a NS residence each year.³ Personal communication with expert clinicians at
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47 261 the pediatric hospital suggests that there are approximately 50 children with medical complexity
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49 262 being discharged at the pediatric hospital per year. This provides a range estimate of the number
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51 263 of children with medical complexity discharged each year. To ensure a large enough cohort to
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53
54 264 power a regression analysis, we are examining prevalence over a 15-year time period.

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3 265 The validation of the algorithms will occur in three parts (Figure 3). The gold-standard sample
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5 266 from step 1 will be evaluated using positive predictive values (PPV), to determine what
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8 267 percentage of algorithm-identified patients truly qualified as medically complex. The gold-
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10 268 standard sample from step 2 will be evaluated using sensitivity and specificity, to evaluate how
11
12 269 well the algorithms can identify both complex and not complex patients. Finally, the two samples
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14 270 will be combined into a single analysis. All statistics will be estimated with 95% confidence
15
16 271 intervals (CIs), and F-statistics will be used to investigate the trade-offs between sensitivity and
17
18 272 specificity.²⁵ As medical complexity is by definition a rare occurrence, we will pay particular
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20 273 attention to the specificity, in order to ensure that the algorithm is not overwhelming the final
21
22 274 cohort with false positives.
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26
27 275 Phase 2: Prevalence of children with medical complexity in all three Maritime provinces
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29 276 The algorithm identified as the “best fit” in phase 1, based on NS data, will be applied to NB’s
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31 277 and PEI’s CIHI DAD through NB-IRDT and SIDR respectively, to identify the cohort of
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33 278 children with medical complexity using the same eligibility parameters as NS. Statistical code to
34
35 279 generate the cohorts developed in NS during the validation step, will be utilized, with minor
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37 280 modification as needed.
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41 281 Prevalence of medical complexity will be calculated using discharge abstract data during the
42
43 282 years 2003-2017 in each Maritime province. Individual level data will use encrypted health card
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45 283 numbers and birthdate in month/year as unique identifiers to link each child’s data across
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47 284 datasets and over time.
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51 285 *Sample size and data analysis:* To estimate the annual prevalence, and corresponding 95% CIs,
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53 286 of children with medical complexity per 100,000 pediatric population in the Maritime provinces
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55 287 during our study period, the number of children identified by our selected algorithm will be
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288 divided by the total number of children (≤ 18 years) in the 2016 pediatric population for each
289 province. Prevalence estimates will be stratified based on relevant variables (e.g. age, sex).

290 Phase 3: Patterns of healthcare utilization

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

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

306

307 Discussion

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

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2
3 310 to comprehensively validate algorithms, that is able to identify children with medical complexity,
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5 311 addresses a significant gap in health research and is essential for better management and
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7 312 improvement of health outcomes. The methods we develop (e.g. the gold-standard definition)
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9
10 313 may be useful to other jurisdictions across Canada that also want to validate the algorithms
11
12 314 specifically for their region. Further, selection of an algorithm to identify children most
13
14 315 accurately with medical complexity will allow us to characterize population-level prevalence and
15
16 316 identify patterns in health service utilization, as well as identify gaps in service, for this
17
18 317 vulnerable population. The formation of this cohort will act as a starting point to conduct future
19
20 318 population-based research that can contribute evidence to support health system planning and
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22 319 policy making and help improve health outcomes. The involvement of key stakeholders
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24 320 throughout this project will ensure knowledge of our research findings are translated to directly
25
26 321 influence clinical practice and health system and policy change by providing data to more
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28 322 appropriately outline health resource gaps.
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323 Limitations and Mitigating Strategies

324 Our team has reflected on potential challenges in the conduct of our study and have planned
325 strategies accordingly to mitigate these difficulties should they arise. A challenge lies in the lack
326 of consensus about how to operationalize a gold-standard definition of children with medical
327 complexity. Our plan to convene an expert panel and employ multiple methods of algorithm
328 validation will address this limitation. Simon et al.⁹ and Feudtner et al.'s¹⁰ administrative data
329 algorithms were developed and validated using ICD-CM codes, while in Canada, we use the
330 ICD-CA iteration of the global ICD codes. The differences between these iterations are expected
331 to be negligible as they relate to administrative needs, not the medical diagnoses. Additional
332 limitations concerning the use of health data are the availability and accuracy of important

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3 333 variables of interest. For example, the order that diagnoses are coded (e.g. primary diagnosis,
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5 334 secondary diagnoses) is not always consistent. To help mitigate this, all diagnoses from a patient
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7 335 encounter coded in the CIHI DAD will be included. Finally, estimating healthcare utilization
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9 336 using health administrative data has limitations as not all services are reflected in existing
10
11 337 databases. Our team of patient partners, researchers, clinicians and administrators recognize the
12
13 338 importance of capturing alternative or relevant non-health service utilization (e.g. massage
14
15 339 therapy, informal respite care) and indirect costs for families (e.g. travel, lost time at work). We
16
17 340 have already begun to explore the use of additional administrative data from other sources as
18
19 341 well as qualitative interviews with patients, families, and providers in a future study.
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343 **Competing interests**

344 The authors declare that they have no competing interests.

345

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348 #452905).

349

350 **Authors' contributions**

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

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

357

358 **References**

- 359 1. Berry JG, Hall M, Cohen E, O'Neill M, Feudtner C. Ways to Identify Children with
360 Medical Complexity and the Importance of Why. *J Pediatr.* 2015 Aug;167(2):229–37.
- 361 2. Cohen E, Berry JG, Sanders L, Schor EL, Wise PH. Status Complexicus? The Emergence
362 of Pediatric Complex Care. *Pediatrics.* 2018 Mar;141(Supplement 3):S202–11.
- 363 3. Cohen E, Berry JG, Camacho X, Anderson G, Wodchis W, Guttman A. Patterns and Costs
364 of Health Care Use of Children With Medical Complexity. *PEDIATRICS.* 2012 Dec
365 1;130(6):e1463–70.
- 366 4. Canadian Institute for Health Information. Children and Youth With Medical Complexity in
367 Canada [Internet]. p. 1–30. Available from:
368 [https://www.cihi.ca/sites/default/files/document/children-youth-with-medical-complexity-](https://www.cihi.ca/sites/default/files/document/children-youth-with-medical-complexity-report-en.pdf)
369 [report-en.pdf](https://www.cihi.ca/sites/default/files/document/children-youth-with-medical-complexity-report-en.pdf)
- 370 5. Simon TD, Berry J, Feudtner C, Stone BL, Sheng X, Bratton SL, et al. Children With
371 Complex Chronic Conditions in Inpatient Hospital Settings in the United States. *Pediatrics.*
372 2010 Oct 1;126(4):647–55.
- 373 6. Burns KH, Casey PH, Lyle RE, Bird TM, Fussell JJ, Robbins JM. Increasing Prevalence of
374 Medically Complex Children in US Hospitals. *Pediatrics.* 2010 Oct 1;126(4):638–46.

- 1
2
3 375 7. Breneol S, Shin D, Carrier J, Macdonald M, Martin-Misener R, Vine J, et al. Improving
4
5 376 Health Care for Children with Medical Complexity Through the Use of Health
6
7 377 Administrative Data: A Scoping Review. Drafted.
8
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10
11 378 8. CAPHC Guideline for the Management of Medically Complex Children and Youth
12
13 379 Through the Continuum of Care [Internet]. Available from:
14
15 380 [https://ken.childrenshhealthcarecanada.ca/xwiki/bin/download/Management+of+Medically+](https://ken.childrenshhealthcarecanada.ca/xwiki/bin/download/Management+of+Medically+Complex+Children+and+Youth+Across+the+Continuum+of+Care/WebHome/CAPHC%20National%20Complex%20Care%20Guideline%202018_final.pdf)
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17 381 [Complex+Children+and+Youth+Across+the+Continuum+of+Care/WebHome/CAPHC%20](https://ken.childrenshhealthcarecanada.ca/xwiki/bin/download/Management+of+Medically+Complex+Children+and+Youth+Across+the+Continuum+of+Care/WebHome/CAPHC%20National%20Complex%20Care%20Guideline%202018_final.pdf)
18
19 382 [National%20Complex%20Care%20Guideline%202018_final.pdf](https://ken.childrenshhealthcarecanada.ca/xwiki/bin/download/Management+of+Medically+Complex+Children+and+Youth+Across+the+Continuum+of+Care/WebHome/CAPHC%20National%20Complex%20Care%20Guideline%202018_final.pdf)
20
21
22
23 383 9. Porepa M, Hoffman A, Fellin M, Kublick L. Children with medical complexities:
24
25 384 Addressing the gaps in respite care during transition from paediatrics to adult health care in
26
27 385 Ontario. *Paediatr Child Health*. 2017 Oct;22(7):369–71.
28
29
30
31 386 10. Simon JL, Daelmans B, Boschi-Pinto C, Aboubaker S, Were W. Child health guidelines in
32
33 387 the era of sustainable development goals. *BMJ*. 2018 Jul 30;362:bmj.k3151.
34
35
36
37 388 11. Feudtner C, Feinstein JA, Zhong W, Hall M, Dai D. Pediatric complex chronic conditions
38
39 389 classification system version 2: updated for ICD-10 and complex medical technology
40
41 390 dependence and transplantation. *BMC Pediatr*. 2014 Dec;14(1):199.
42
43
44
45 391 12. Berry JG. Hospital Utilization and Characteristics of Patients Experiencing Recurrent
46
47 392 Readmissions Within Children’s Hospitals. *JAMA*. 2011 Feb 16;305(7):682.
48
49
50
51 393 13. Srivastava R, Berry JG, Hall M, Downey EC, O’Gorman M, Dean JM, et al. Reflux related
52
53 394 hospital admissions after fundoplication in children with neurological impairment:
54
55 395 retrospective cohort study. *BMJ*. 2009 Nov 18;339:b4411.
56
57
58
59
60

- 1
2
3 396 14. Simon TD, Lawrence M, Stanford S, Lyons D, Woodcox P, Hood M, et al. Pediatric
4
5 397 Medical Complexity Algorithm: A New Method to Stratify Children by Medical
6
7 398 Complexity. *Pediatrics*. 2014;133(6):8.
- 9
10
11 399 15. Feudtner C, Christakis DA, Connell FA. Pediatric Deaths Attributable to Complex Chronic
12
13 400 Conditions: A Population-Based Study of Washington State, 1980–1997. *Pediatrics*. 2000
14
15 401 Jul 1;106(Supplement 1):205–9.
- 16
17
18 402 16. Wilchesky M, Tamblyn RM, Huang A. Validation of diagnostic codes within medical
19
20 403 services claims. *J Clin Epidemiol*. 2004;57(2):131–41.
- 21
22
23 404 17. Canadian Institutes of Health Research. Guide to Knowledge Translation Planning at
24
25 405 CIHR: Integrated and End-of-Grant Approaches - CIHR [Internet]. Canada: Government of
26
27 406 Canada; 2012 Dec [cited 2019 Feb 1] p. 1–34. Available from: [http://www.cihr-](http://www.cihr-irsc.gc.ca/e/45321.html)
28
29 407 [irsc.gc.ca/e/45321.html](http://www.cihr-irsc.gc.ca/e/45321.html)
- 30
31
32 408 18. Vassar M, Holzmann M. The retrospective chart review: important methodological
33
34 409 considerations. *J Educ Eval Health Prof*. 2013;10:12.

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413 **Figure 1.** *Outline of project methods*

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

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2
3 416 **Figure 3.** *Analysis of gold-standard sample of cases with medical complexity (complex) and*
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5 417 *controls (not complex)*
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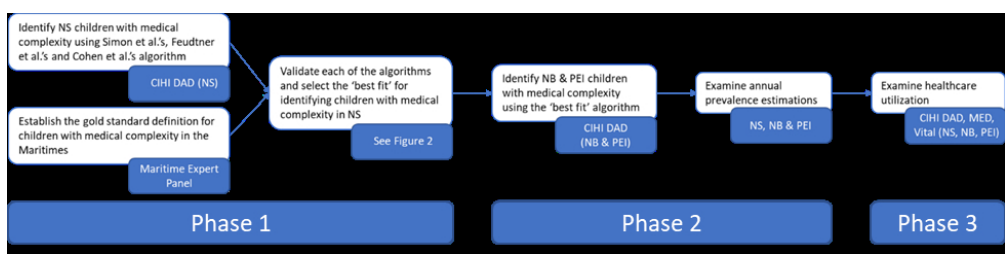


Figure 1. Outline of project methods

419x100mm (59 x 59 DPI)

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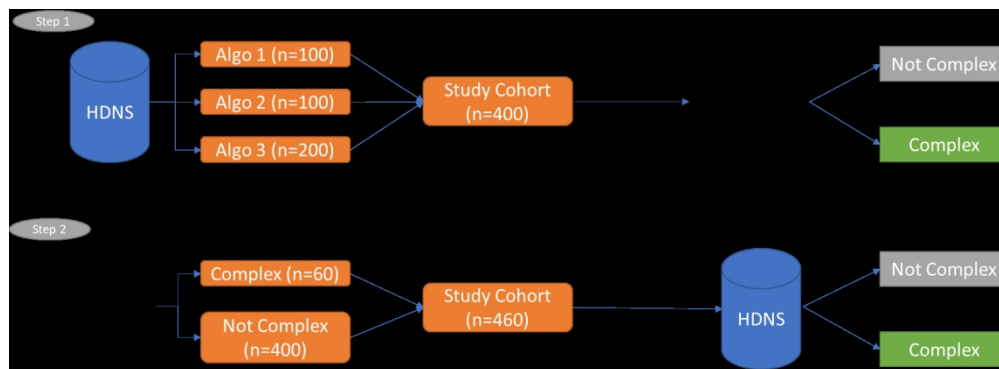


Figure 2. Identification of a gold-standard sample of cases with medical complexity (complex) and controls (not complex)

807x294mm (59 x 59 DPI)

Step 1		WK		
		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

BMJ Open

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

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

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41 Abstract

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

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

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

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

69 **Keywords**

70 Medical complexity, administrative data, pediatric, healthcare utilization

72 **Strength and limitations of this study**

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

81 **Introduction**

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

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3 85 condition(s) associated with significant functional limitations, high health resource use and
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5 86 family-identified needs.^{1,3} One seminal Canadian study conducted nearly 10 years ago, suggested
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7 87 that despite representing less than 1% of the pediatric population in Ontario, children with
8
9 88 medical complexity account for one third of pediatric healthcare expenditures.¹ Adding to the
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11 89 findings stemming from this study, a recent publication from The Canadian Institute for Health
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13 90 Information (CIHI) reported that in 2015-2016 there were 948 per 100,000 children and youth
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15 91 with medical complexity.⁴ Recent findings from the United States also suggest that the
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17 92 prevalence of children living with medical complexity is increasing,^{5,6} likely due to the increased
18
19 93 survival rates of a variety of life-limiting and life-threatening conditions.³ Information regarding
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21 94 children with medical complexity has been primarily derived using reports from the United
22
23 95 States with only a few seminal reports stemming from Canadian-based data.^{4,7} However, due to
24
25 96 important population differences, it is critical to understand this vulnerable population in the
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27 97 Canadian context to assist in mapping health outcomes and healthcare utilization for this
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29 98 vulnerable population. Doing so may ensure that more relevant, needs based and sustainable
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31 99 models of service delivery are developed to optimize quality health experiences and outcomes
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33 100 for children and their families.
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35 101 Caregivers of children with medical complexity carry a tremendous amount of responsibility,
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37 102 stress and financial burden to attend to their intensive care needs.^{7,8} Opportunities to assess
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39 103 disparities in care, and appropriately allocate health resources to better serve children with
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41 104 medical complexity, are dependent on being able to accurately identify them.¹ A standard
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43 105 definition of children with medical complexity does not currently exist in Canada and
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45 106 determining a classification system for these children is a necessary first step to optimize health
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47 107 service delivery for this vulnerable group of children. Given the heterogeneity of this group, with
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108 variation in the severity and combination of comorbid chronic conditions, there are unique
109 challenges when attempting to identify a cohort at a population level.¹ Identification begins with
110 selecting a method to classify and characterize children with medical complexity.

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

116 Health administrative data algorithms

117 A recent scoping review by Breneol *et al.* entitled “Improving Health Care for Children with
118 Medical Complexity Through the Use of Health Administrative Data: A Scoping Review”,
119 examining the use of health administrative data in the study of children with medical complexity,
120 identified three methods that are commonly used to identify this cohort: (1) Cohen *et al.*'s³ list of
121 complex chronic conditions, technological assistance and neurological impairment; (2) Simon *et*
122 *al.*'s⁹ *Pediatric Medical Complexity Algorithm* (PMCA); and (3) Feudtner *et al.*'s¹⁰ complex
123 chronic conditions classification system. Cohen *et al.*'s algorithm was developed in Ontario
124 using hospital discharge data but has not yet been validated. Algorithms developed by Simon *et*
125 *al.*⁹ and Feudtner *et al.*¹⁰ have not undergone validation in Canadian health data.

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

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3 130 sensitivity and specificity of the lists of Canadian Classification of Procedures and Interventions
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5 131 for identifying children with medical complexity in Canada has not yet been reported.
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8 132 *Algorithm 2.* Simon *et al.*¹³ used the Chronic Disability Payment System as a guide to develop
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10 133 the *Pediatric Medical Complexity Algorithm* (PMCA) at the Seattle Children's Hospital in
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12 134 Washington State. The authors employed a systematic process beginning with the development
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14 135 of consensus definitions for three levels of medical complexity (complex chronic, noncomplex
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16 136 chronic and no chronic conditions). They classified children with medical complexity as those
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18 137 who fit their definition for having a complex chronic condition. They proceeded by selecting and
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20 138 modifying an existing algorithm to conform to the consensus definitions, and selected a gold
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22 139 standard pediatric population through medical chart review to evaluate the sensitivity and
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24 140 specificity of the algorithm.¹³ The PMCA had high sensitivity and specificity (complex chronic:
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26 141 86% sensitivity, 86% specificity; non-complex chronic: 65% sensitivity, 84% specificity;
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28 142 children without complex chronic: 77% sensitivity and 93% specificity) for identifying children
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30 143 with medical complexity and was subsequently updated and validated for ICD-10-CM codes in
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32 144 2018.⁹ This algorithm has a least and more conservative version depending on the type of data
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34 145 available to researchers. The least conversative version was shown to perform better in hospital
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36 146 discharge data and the most conservative version was shown to perform better in claims and
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38 147 billing data.¹³
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45 148 *Algorithm 3.* Feudtner *et al.*¹⁴ followed a similar approach as Simon *et al.*⁹ in Washington State,
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47 149 creating a working definition of complex chronic conditions and subsequently operationalized
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49 150 the definition using clinical knowledge and existing literature. This resulted in a list of conditions
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51 151 and their corresponding ICD-9-CM codes that are highly sensitive (87%) for identifying children
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53 152 with complex chronic conditions. The algorithm was updated in 2014 and the ICD codes were
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3 153 translated into the 10th edition (ICD-10-CM), including both diagnostic and procedural codes
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5 154 indicative of technology dependence or organ transplantation.¹⁰ Lindley *et al.* has evaluated the
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8 155 predictive ability of the original and modified versions and determined that the modified version,
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10 156 which will be used in this study, better operationalizes medical complexity.¹⁵
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13 157 Aims and objectives

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16 158 The aim of this study is to better understand the Canadian Maritime's population of children with
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18 159 medical complexity. The ability to accurately identify children with medical complexity is
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20 160 necessary to conduct research that may inform the design and implementation of successful
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22 161 population-level policies and interventions. This study will be conducted in the Canadian
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24 162 Maritime provinces: NS, NB, and PEI. This study will: (1) evaluate the performance of three
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26 163 administrative data algorithms to identify children with medical complexity; then use the "best
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28 164 fit" algorithm to (2) estimate the prevalence; and (3) describe patterns of healthcare utilization
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30 165 for this cohort of children from 2003-2017.
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37 167 **Methods and analysis**

40 168 Data Sources

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43 169 The study cohorts will be constructed using the Canadian Institute for Health Information
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45 170 Discharge Abstract Databases (CIHI DAD) from Nova Scotia and the prevalence and health care
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47 171 utilization estimates will use the CIHI DAD, Vital Statistics (VITAL), Insured Patient Registry
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49 172 (MASTER), and Physician Billing Databases (MED) from each province. The administrative
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51 173 datasets for NS, NB and PEI are housed by their respective data repository organizations: Health
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3 174 Data Nova Scotia (HDNS), New Brunswick Institute for Research Data and Training (NB-
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5 175 IRDT), and Secure Island Data Repository (SIDR).
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8 176 Study Design 9

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11 177 This study will be conducted in three phases to identify children with medical complexity in the
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13 178 Canadian Maritimes and their healthcare utilization over a 15-year period (Figure 1).
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16 179 In Phase 1, the “best fit” algorithm for identifying children with medical complexity in the
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18 180 Maritimes will be selected using sensitivity and specificity. Using NS administrative data, a
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20 181 cohort will be established for each of the three algorithms. A gold-standard definition will be
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22 182 developed for children with medical complexity in the Maritime provinces through an expert
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24 183 consensus meeting. Through the consensus process, this definition will subsequently be
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26 184 translated into a chart audit tool. The “best fit” algorithm will be selected using a two-gate case-
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28 185 control study design.
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33 186 In Phase 2, the prevalence of children with medical complexity in the Maritime provinces will be
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35 187 estimated. This will be done in all three Maritime provinces using NS, NB and PEI
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37 188 administrative data cohorts identified by the previously selected “best fit” algorithm.
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40 189 In Phase 3, patterns of healthcare utilization of children with medical complexity across the
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42 190 Maritime provinces will be described.
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45 191 Phase 1: Identify the “best fit” algorithm for identifying children with medical complexity in the
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50 193 *Developing a gold-standard definition:* A multidisciplinary expert consensus meeting will be
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52 194 convened to co-develop a gold-standard definition of children with medical complexity relevant
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54 195 to the Canadian Maritimes. The consensus meeting will involve relevant stakeholders, including
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3 196 a minimum of two clinicians, two researchers and a parent from each of the three Maritime
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5 197 provinces. Parent participants will be invited to attend a pre-meeting virtual session to introduce
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7 198 them to the consensus meeting agenda and provide them with an opportunity to ask questions
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9 199 related to the project. A parent partner and research coordinator will facilitate the pre-meeting
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11 200 session. An infographic will be developed and distributed prior to the consensus meeting to
12
13 201 introduce participants to existing frameworks and literature that will help guide the
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15 202 conceptualization of medical complexity, functional limitations, and other related concepts.
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17 203 Outputs from relevant exploratory work conducted by members of the research team will be
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19 204 presented during the first session of the consensus meeting.¹⁶ Parent and clinician experts will
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21 205 participate in structured breakout sessions to examine all meeting materials in the context of their
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23 206 clinical or lived experience. Sessions will be audio-recorded, and a note-taker will track
24
25 207 participation. After the breakout sessions are complete, a consensus based decision-making
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27 208 process will be used where participants work together through discussion to reach agreement on
28
29 209 the elements of a clinically meaningful gold-standard definition for this pediatric population in
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31 210 the Maritimes.¹⁷ The definition will be circulated to the expert stakeholder group after the
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33 211 meeting for their review and comments and will be edited through email.
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35 212 To operationalize the definition, a sub-group of clinicians with expertise working with this
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37 213 patient population will use the gold-standard definition to develop a list of clinical variables that
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39 214 will form the basis of a medical chart extraction tool. The expert group will complete this task
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41 215 over a series of meetings, and the final tool will be reviewed by clinicians from both NB and
42
43 216 PEI. A coding manual will be developed to accompany the medical chart extraction tool.
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45 217 *Establishing a Nova Scotia administrative data cohort:* The algorithm validation process will be
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47 218 undertaken using administrative data from the NS provincial data repository organization,
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3 219 HDNS. Children living in NS with medical complexity will be identified and classified with
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5 220 specific ICD-10-CA codes defined within each of the three respective algorithms, resulting in a
6
7 221 distinct cohort being derived from each algorithm. Due to the uncertainty of how the Simon *et al.*
8
9 222 algorithm will perform using Canadian health administrative data, both the least and more
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11 223 conservative versions will be used. All children aged 0 to 18 years of age who have a discharge
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13 224 record in NS CIHI DAD from 2003-2017 will be eligible for inclusion.

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17 225 *Examining the “best fit”*: Given the prevalence of children with medical complexity is rare, a
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19 226 two-gate case-control study design will be used to validate the algorithms (Figure 2).¹⁸ This will
20
21 227 be done in two steps. First, a stratified random sample of 100 children will be selected from each
22
23 228 of the cohorts derived by the algorithms to participate in a chart audit.¹⁹ The chart audit will be
24
25 229 conducted by nurses trained in abstracting records of children with medical complexity using the
26
27 230 medical chart extraction tool developed by the expert panel. The audit will begin with all
28
29 231 abstractors independently extracting the same five charts to calibrate coding. Results will be
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31 232 compared, and the inter-rater reliability will be estimated using a kappa score. If the kappa score
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33 233 is low and there are many discrepancies, the chart audit tool will be refined, and further training
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35 234 provided to the abstractors. An additional five charts will then be extracted and compared. This
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37 235 process will be repeated until the chart extraction is determined to be reliable. A total of 30% of
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39 236 the records in each cohort will be independently abstracted by a second reviewer.

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43 237 In the second step, two clinically derived groups of children (complex and not complex) will be
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45 238 established by healthcare providers from across NS to contribute to the validation procedure.
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47 239 Healthcare providers who contribute to this process will be from varying specialties and settings
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49 240 (i.e., primary and tertiary care) and provide direct patient care or care coordination activities to
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51 241 children with medical complexity and their families. Sixty children with medical complexity will
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242 be identified from relevant clinical areas that fit within the study's gold standard definition.

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

248 *Sample size and data analysis:* There are approximately 3200 Nova Scotian children discharged
249 each year from the pediatric tertiary care facility in the Maritimes. Cohen *et al.* has estimated a
250 0.67% prevalence of children with medical complexity, therefore we estimated approximately 21
251 Nova Scotian children with medical complexity are discharged yearly.³ However, local expert
252 clinicians estimated that there are closer to 50 Nova Scotian children discharged each year with
253 medical complexity. To ensure a large enough cohort to power a regression analysis, prevalence
254 will be examined over a 15-year period.

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

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

266 The algorithm identified as the “best fit” in phase 1, based on NS data, will be applied to NB’s
267 and PEI’s CIHI DAD through NB-IRDT and SIDR respectively, to identify the cohort of
268 children with medical complexity using the same eligibility parameters as NS. Statistical code to
269 generate the cohorts developed in NS during the validation step, will be utilized, with minor
270 modification as needed.
271 Prevalence of medical complexity will be calculated using discharge abstract data during the
272 years 2003-2017 in each Maritime province. Individual level data will use encrypted health card
273 numbers and birthdate in month/year as unique identifiers to link each child’s data across
274 datasets and over time.

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

280 Phase 3: Patterns of healthcare utilization

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

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

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3 287 of acute hospitalizations, unplanned readmissions, day surgeries, hospital-based outpatient visits,
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5 288 community-based primary care visits and specialty care visits. Healthcare utilization follow-up
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8 289 will begin at the index date. The index date will be defined as the first discharge date with a
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10 290 complex chronic condition, by the “best fit” algorithm. Children will be censored at the end of
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12 291 the study period or sooner if they die or turn 18 before 2017. The numerator will be the total
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14 292 number of annual records for each child in the cohort. Each child will contribute one year of
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16 293 person-time to the denominator for every year they are alive and living in the province. Results
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18 294 from each province will be described separately by summarizing findings and comparing
19
20 295 healthcare utilization across provinces.
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24 296 This project is currently in phase 1 in the process of identifying the “best fit” algorithm using the
25
26 297 two-gate case-control study. The project is anticipated to be completed by December 2022.
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29 298 Patient and Public Involvement

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32 299 This study will use an integrated knowledge translation (iKT) approach, where end users are
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34 300 involved in each stage of the project.²¹ The team of researchers, clinicians, administrators and
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36 301 patient partners will work together throughout the research process to develop outputs that are
37
38 302 relevant to knowledge user needs, including development of the research objectives and
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40 303 methods. Opportunities will be created to engage additional parents, clinicians, and
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42 304 administrators to inform different stages of the work as the project unfolds. The proposed iKT
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44 305 method was chosen to increase uptake of the research into policy and practice.²²
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50 51 307 **Ethics and dissemination**

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3 308 Ethical approval was received from the primary research institution in NS (IWK Health Centre;
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5 309 REB#: 1026245). A waiver of consent has been granted by the research ethics board and
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8 310 therefore informed participant consent is not required for this study. Ethical approval will be
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10 311 obtained at the appropriate institutions in NB and PEI before data abstraction commences in
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12 312 these provinces.

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15 313 This study will use an iKT approach, where end users are involved in each stage of the project,
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17 314 which could increase uptake of the research into policy and practice. The findings of this
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19 315 research study will be submitted for publication and dissemination through conference
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21 316 presentations and with our end users.
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26 27 318 **Discussion**

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30 319 To our knowledge, this will be the first time the performance of these three algorithms will be
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32 320 evaluated using Canadian health administrative data. While the recent CIHI report identified
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34 321 children with medical complexity across Canada to provide population-level prevalence rates
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36 322 and an overview of their healthcare utilization, this study will add to the existing knowledge by
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38 323 including a longer to identify any potential trends.⁴
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42 324 Findings from this work may contribute to a priority task, identified by Children's Healthcare
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44 325 Canada, for a national effort to support system change for children with medical complexity.⁷
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47 326 The opportunity to comprehensively identify a "best fit" algorithm, that is able to describe
48
49 327 children with medical complexity, addresses a significant gap in health research and is essential
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51 328 for better management and improvement of health outcomes. The methods developed (e.g., two-
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53 329 gate validation procedure, chart audit tool) may be useful to other jurisdictions across Canada
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3 330 wishing to validate algorithms specifically for their region. Further, identifying a “best fit”
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5 331 algorithm to describe children with medical complexity may allow us to characterize population-
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7 332 level prevalence and identify patterns in health service utilization, as well as identify gaps in
8
9 333 service, for this vulnerable population. The formation of this cohort may act as a starting point to
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11 334 conduct future population-based research that can contribute evidence to support health system
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13 335 planning and policy making and help improve health outcomes. Key stakeholders are being
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15 336 involved throughout this project to help ensure research findings related to service and resource
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17 337 gaps are translated to health system and policy change.
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22 338 Limitations and Mitigating Strategies

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25 339 The team has reflected on potential challenges in the conduct of the study and have planned
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27 340 strategies accordingly to mitigate these difficulties should they arise. A challenge lies in the lack
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29 341 of consensus about how to operationalize a gold-standard definition of children with medical
30
31 342 complexity. The plan to convene an expert panel and employ multiple methods of algorithm
32
33 343 validation was chosen to address this limitation. Simon *et al.*¹³ and Feudtner *et al.*'s¹⁰
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35 344 administrative data algorithms were developed and validated using ICD-CM codes, while
36
37 345 Canada uses the ICD-CA iteration of the global ICD codes. The differences between these
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39 346 iterations are expected to be negligible as they relate to administrative needs, not the medical
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41 347 diagnoses. Additional limitations concerning the use of health data are the availability and
42
43 348 accuracy of important variables of interest. For example, the order that diagnoses are coded (e.g.,
44
45 349 primary diagnosis, secondary diagnoses) is not always consistent. To help mitigate this, all
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47 350 diagnoses from a patient encounter coded in the CIHI DAD will be included. Finally, estimating
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49 351 healthcare utilization using health administrative data has limitations as not all services are
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51 352 reflected in existing databases. The team of patient partners, researchers, clinicians, and
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3 353 administrators recognize the importance of capturing alternative or relevant non-health service
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5 354 utilization (e.g., massage therapy, informal respite care) and indirect costs for families (e.g.,
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7 355 travel, lost time at work). The use of additional administrative data from other sources is being
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10 356 explored as well as qualitative interviews with patients, families, and providers in a future study.
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13 357

15 358 **Competing interests**

17
18 359 The authors declare that they have no competing interests.
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20

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23
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25
26 362 #452905).
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28

29 363 **Contributors**

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32 364 JAC conceived of and designed the study, secured funding for the project, drafted the study
33
34 365 protocol. SB, SS, SM contributed to the study design and the draft of the protocol. MS, JS, SB,
35
36 366 SB, EJ, MAM, SK contributed to the study protocol. JC is the parent knowledge user on this
37
38 367 project and has provided guidance during all phases. HM prepared this manuscript from the
39
40 368 study protocol. All authors critically appraised the intellectual content of the manuscript and
41
42 369 provided input and revisions. All authors read and approved the final manuscript.
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48 371 **References**

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51
52 372 1. Berry JG, Hall M, Cohen E, O'Neill M, Feudtner C. Ways to Identify Children with
53 373 Medical Complexity and the Importance of Why. *J Pediatr*. 2015 Aug;167(2):229–37.
54
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60

- 374 2. Cohen E, Berry JG, Sanders L, Schor EL, Wise PH. Status Complexicus? The Emergence
375 of Pediatric Complex Care. *Pediatrics*. 2018 Mar;141(Supplement 3):S202–11.
- 376 3. Cohen E, Berry JG, Camacho X, Anderson G, Wodchis W, Guttman A. Patterns and Costs
377 of Health Care Use of Children With Medical Complexity. *PEDIATRICS*. 2012 Dec
378 1;130(6):e1463–70.
- 379 4. Canadian Institute for Health Information. Children and Youth With Medical Complexity in
380 Canada [Internet]. p. 1–30. Available from:
381 [https://www.cihi.ca/sites/default/files/document/children-youth-with-medical-complexity-](https://www.cihi.ca/sites/default/files/document/children-youth-with-medical-complexity-report-en.pdf)
382 [report-en.pdf](https://www.cihi.ca/sites/default/files/document/children-youth-with-medical-complexity-report-en.pdf)
- 383 5. Simon TD, Berry J, Feudtner C, Stone BL, Sheng X, Bratton SL, et al. Children With
384 Complex Chronic Conditions in Inpatient Hospital Settings in the United States. *Pediatrics*.
385 2010 Oct 1;126(4):647–55.
- 386 6. Burns KH, Casey PH, Lyle RE, Bird TM, Fussell JJ, Robbins JM. Increasing Prevalence of
387 Medically Complex Children in US Hospitals. *Pediatrics*. 2010 Oct 1;126(4):638–46.
- 388 7. CAPHC Guideline for the Management of Medically Complex Children and Youth
389 Through the Continuum of Care [Internet]. Available from:
390 [https://ken.childrenshealthcarecanada.ca/xwiki/bin/download/Management+of+Medically+](https://ken.childrenshealthcarecanada.ca/xwiki/bin/download/Management+of+Medically+Complex+Children+and+Youth+Across+the+Continuum+of+Care/WebHome/CAPHC%20National%20Complex%20Care%20Guideline%202018_final.pdf)
391 [Complex+Children+and+Youth+Across+the+Continuum+of+Care/WebHome/CAPHC%20](https://ken.childrenshealthcarecanada.ca/xwiki/bin/download/Management+of+Medically+Complex+Children+and+Youth+Across+the+Continuum+of+Care/WebHome/CAPHC%20National%20Complex%20Care%20Guideline%202018_final.pdf)
392 [National%20Complex%20Care%20Guideline%202018_final.pdf](https://ken.childrenshealthcarecanada.ca/xwiki/bin/download/Management+of+Medically+Complex+Children+and+Youth+Across+the+Continuum+of+Care/WebHome/CAPHC%20National%20Complex%20Care%20Guideline%202018_final.pdf)
- 393 8. Porepa M, Hoffman A, Fellin M, Kublick L. Children with medical complexities:
394 Addressing the gaps in respite care during transition from paediatrics to adult health care in
395 Ontario. *Paediatr Child Health*. 2017 Oct;22(7):369–71.
- 396 9. Simon JL, Daelmans B, Boschi-Pinto C, Aboubaker S, Were W. Child health guidelines in
397 the era of sustainable development goals. *BMJ*. 2018 Jul 30;362:bmj.k3151.
- 398 10. Feudtner C, Feinstein JA, Zhong W, Hall M, Dai D. Pediatric complex chronic conditions
399 classification system version 2: updated for ICD-10 and complex medical technology
400 dependence and transplantation. *BMC Pediatr*. 2014 Dec;14(1):199.
- 401 11. Berry JG. Hospital Utilization and Characteristics of Patients Experiencing Recurrent
402 Readmissions Within Children’s Hospitals. *JAMA*. 2011 Feb 16;305(7):682.
- 403 12. Srivastava R, Berry JG, Hall M, Downey EC, O’Gorman M, Dean JM, et al. Reflux related
404 hospital admissions after fundoplication in children with neurological impairment:
405 retrospective cohort study. *BMJ*. 2009 Nov 18;339:b4411.
- 406 13. Simon TD, Lawrence M, Stanford S, Lyons D, Woodcox P, Hood M, et al. Pediatric
407 Medical Complexity Algorithm: A New Method to Stratify Children by Medical
408 Complexity. *Pediatrics*. 2014;133(6):8.

- 1
2
3 409 14. Feudtner C, Christakis DA, Connell FA. Pediatric Deaths Attributable to Complex Chronic
4 410 Conditions: A Population-Based Study of Washington State, 1980–1997. *Pediatrics*. 2000
5 411 Jul 1;106(Supplement 1):205–9.
- 6
7 412 15. Lindley LC, Cozad MJ, Fortney CA. Pediatric Complex Chronic Conditions: Evaluating
8 413 Two Versions of the Classification System. *West J Nurs Res*. 2020 Jun;42(6):454–61.
- 9
10
11 414 16. Curran JA, Breneol S, Vine J. Improving transitions in care for children with complex and
12 415 medically fragile needs: a mixed methods study. *BMC Pediatr*. 2020 May 14;20(1):219.
- 13
14 416 17. Langley J, Wolstenholme D, Cooke J. “Collective making” as knowledge mobilisation: the
15 417 contribution of participatory design in the co-creation of knowledge in healthcare. *BMC*
16 418 *Health Serv Res*. 2018 25;18(1):585.
- 17
18 419 18. Holtman GA, Berger MY, Burger H, Deeks JJ, Donner-Banzhoff N, Fanshawe TR, et al.
19 420 Development of practical recommendations for diagnostic accuracy studies in low-
20 421 prevalence situations. *J Clin Epidemiol*. 2019 Oct 1;114:38–48.
- 21
22 422 19. Vassar M, Holzmann M. The retrospective chart review: important methodological
23 423 considerations. *J Educ Eval Health Prof*. 2013;10:12.
- 24
25 424 20. Lasko TA, Bhagwat JG, Zou KH, Ohno-Machado L. The use of receiver operating
26 425 characteristic curves in biomedical informatics. *J Biomed Inform*. 2005 Oct 1;38(5):404–
27 426 15.
- 28
29 427 21. Wilchesky M, Tamblyn RM, Huang A. Validation of diagnostic codes within medical
30 428 services claims. *J Clin Epidemiol*. 2004;57(2):131–41.
- 31
32 429 22. Canadian Institutes of Health Research. Guide to Knowledge Translation Planning at
33 430 CIHR: Integrated and End-of-Grant Approaches - CIHR [Internet]. Canada: Government of
34 431 Canada; 2012 Dec [cited 2019 Feb 1] p. 1–34. Available from: [http://www.cihr-](http://www.cihr-irsc.gc.ca/e/45321.html)
35 432 [irsc.gc.ca/e/45321.html](http://www.cihr-irsc.gc.ca/e/45321.html)

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435 **Figure 1. Outline of project methods**436 **Figure 2. Identification of a gold-standard sample of cases with medical complexity**437 **(complex) and controls (not complex)**438 **Figure 3. Analysis of gold-standard sample of cases with medical complexity (complex) and**439 **controls (not complex)**

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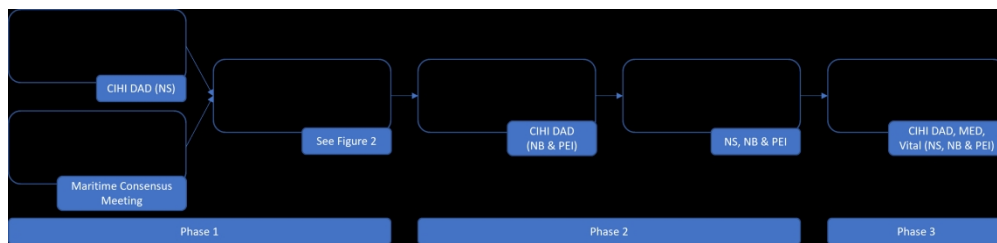


Figure 1. Outline of project methods

346x82mm (300 x 300 DPI)

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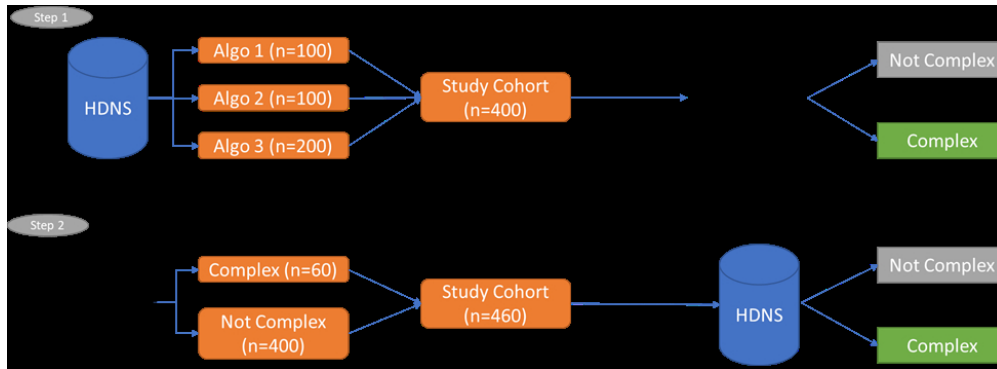


Figure 2. Identification of a gold-standard sample of cases with medical complexity (complex) and controls (not complex)

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)

1
2 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