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Original quantitative research

Cerebral palsy in Canada, 2011–2031: results of a microsimulation modelling study of epidemiological and cost impacts

Nana Amankwah, MSc (1); Maryam Oskoui, MD (2,3,4); Rochelle Garner, PhD (5); Christina Bancej, PhD (1); Douglas G. Manuel, MD (5,6,7,8,9,10,11); Ron Wall, PhD (1); Philippe Finès, PhD (5); Julie Bernier, MA (5); Karen Tu, MD (11,12,13); Kim Reimer, BSc (14)

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Abstract

Introduction: The objective of our study was to present model-based estimates and projections on current and future health and economic impacts of cerebral palsy in Canada over a 20-year time horizon (2011–2031).

Methods: We used Statistics Canada's Population Health Model (POHEM)–Neurological to simulate individuals' disease states, risk factors and health determinants and to describe and project health outcomes, including disease incidence, prevalence, life expectancy, health-adjusted life expectancy, health-related quality of life and health care costs over the life cycle of Canadians. Cerebral palsy cases were identified from British Columbia's health administrative data sources. A population-based cohort was then used to generate the incidence and mortality rates, enabling the projection of future incidence and mortality rates. A utility-based measure (Health Utilities Index Mark 3) was also included in the model to reflect various states of functional health to allow projections of health-related quality of life. Finally, we estimated caregiving parameters and health care costs from Canadian national surveys and health administrative data and included them as model parameters to assess the health and economic impact of cerebral palsy.

Results: Although the overall crude incidence rate of cerebral palsy is projected to remain stable, newly diagnosed cases of cerebral palsy will rise from approximately 1800 in 2011 to nearly 2200 in 2031. In addition, the number of people with the condition is expected to increase from more than 75 000 in 2011 to more than 94 000 in 2031. Direct health care costs in constant 2010 Canadian dollars were about $11 700 for children with cerebral palsy aged 1–4 years versus about $600 for those without the condition. In addition, people with cerebral palsy tend to have longer periods in poorer health-related quality of life.

Conclusion: Individuals with cerebral palsy will continue to face challenges related to an ongoing need for specialized medical care and a rising need for supportive services. Our study offers important insights into future costs and impacts associated with cerebral palsy and provides valuable information that could be used to develop targeted health programs and strategies for Canadians living with this condition.

Keywords: *cerebral palsy, economic burden, Canada, microsimulation modelling, projected incidence and prevalence of cerebral palsy*

Highlights

- A cerebral palsy diagnosis is accompanied by a substantial economic and social burden.
- The number of newly diagnosed cases of cerebral palsy will rise from approximately 1800 in 2011 to nearly 2200 in 2031.
- The number of people living with cerebral palsy is expected to increase from more than 75 000 in 2011 to more than 94 000 in 2031.
- Canadians with cerebral palsy will continue to experience reduced quality of life, increased disabilities and a rising need for supportive services including informal care.

Introduction

Cerebral palsy is the most common cause of motor disability in childhood.[1-3] It is a heterogeneous group of neurological disorders with motor impairments as the core feature. Individuals with cerebral palsy frequently have comorbid conditions that further affect their function.[4] The comorbidities associated with cerebral palsy contribute to individual and family challenges and the financial costs

Author references:

1. Public Health Agency of Canada, Ottawa, Ontario, Canada
2. Departments of Pediatrics and Neurology Neurosurgery, McGill University, Montréal, Quebec, Canada
3. Division of Pediatric Neurology, Montréal Children's Hospital, McGill University Health Centre, Montréal, Quebec, Canada
4. Department of Epidemiology Biostatistics and Occupational Health, McGill University, Montréal, Quebec, Canada
5. Health Analysis Division, Statistics Canada, Ottawa, Ontario, Canada
6. Ottawa Hospital Research Institute, Ottawa, Ontario, Canada
7. University of Ottawa, Ottawa, Ontario, Canada
8. Bruyère Research Institute, Ottawa, Ontario, Canada
9. School of Public and Population Health, University of Ottawa, Ottawa, Ontario, Canada
10. Institute for Clinical Evaluative Sciences, Ottawa, Ontario, Canada
11. North York General Hospital, Toronto, Ontario, Canada
12. Department of Family and Community Medicine, Department of Medicine and Institute of Health Policy, Management and Evaluation, University of Toronto, Toronto, Ontario, Canada
13. Toronto Western Hospital Family Health Team, University Health Network, Toronto, Ontario, Canada
14. Population Health Surveillance and Epidemiology, Office of the Provincial Health Officer, British Columbia Ministry of Health, Victoria, British Columbia, Canada

Correspondence: Nana Amankwah, Applied Research Division, Centre for Surveillance and Applied Research, Health Promotion and Chronic Disease Prevention Branch, Public Health Agency of Canada, 9th floor, Room 903A3, 785 Carling Avenue, Ottawa, ON K1A 0K9; Tel: 613-291-3181; Email: nana.amankwah@canada.ca

related to the medical, rehabilitation and educational needs specific to each child.

Cerebral palsy results from disturbances to the developing brain at any time during pregnancy through early childhood (2 years of age). Congenital cerebral palsy refers to prenatal and perinatal aetiologies before 28 days of life, while postnatal cerebral palsy refers to aetiologies from 28 days of life until 2 years of age. Cerebral palsy has multiple causes, clinical subtypes, associated comorbidities and functional outcomes. The diversity in terms of the onset, neuromotor impairment pattern, severity across functional domains, presence of multiple health conditions or diseases, secondary complications and available interventions makes it nearly impossible to categorize cerebral palsy according to a single classification.[5]

Not surprisingly, multiple risk factors are associated with the condition during the prenatal (prior to labour and delivery), perinatal (during the birthing process) and postnatal (after birth in the first 2 years of life) periods.[3-9] Risk factors include premature birth, intrauterine growth restriction, intrauterine infection/inflammation, male sex, consanguinity, stroke prior to 2 years of age and hypoxic ischemic encephalopathy. Nevertheless, the specific causal mechanism remains elusive for most children with cerebral palsy. Most children are diagnosed in the first 2 years of life,[10] although some diagnoses may not occur until later in childhood.[11,12]

Very little has changed over the past decade in the global prevalence of cerebral palsy.[1-6,8,13-18] A recent meta-analysis reported the birth prevalence of cerebral palsy in developed countries to be 2.11 per 1000 live births.[3] However, population-based studies have reported prevalence estimates ranging from 1.5 to approximately 4 per 1000 live births, and highest in countries with higher incidence of consanguinity.[19-23] The lack of case-validated algorithms for identifying cerebral palsy[24] in administrative health databases may contribute to variability in estimated prevalence as diagnostic code misclassification may be occurring.

The variability in cerebral palsy prevalence estimates across the world and within Canada may also be due to differences in the criteria used for diagnosing the condition,[24,25] as well as differences in pre- and perinatal care in some populations. Studies conducted in Alberta[26], British Columbia[27] and Quebec[28] have reported prevalence estimates similar to the worldwide estimates. Using administrative databases, Alberta reported an estimate of 2.57 per 1000 live births at 8 years,[26] while the figure in British Columbia was 2.68 per 1000 live births.[27] Using data from a Quebec cerebral palsy registry, Oskoui et al.[13] reported an estimate of 2.30 per 1000 children. A more recent Quebec study determined the prevalence of cerebral palsy to be 1.8 per 1000 children (95% confidence interval [CI]: 1.6–2.1), based on data from a cerebral palsy registry versus 2.0 per 1000 children (95% CI: 1.9–2.3) based on data from administrative health databases.[28] Although these estimates are comparable, the concordance in diagnosis between the two databases showed a kappa of 0.62 (95% CI: 0.57–0.67), where a selection bias was seen in the administrative health databases cases.[28] In effect, children with confirmed cerebral palsy who were born preterm were more likely than those born at term to be captured by administrative databases because preterm children tend to receive more specialized developmental care that often leads to correct diagnosis and coding of their condition.

A diagnosis of cerebral palsy comes with a substantial economic and social burden to individuals, families and society in general. The burden can be exacerbated when relevant information for the development of effective health policies, strategies and programs is limited or unavailable. To better understand the incidence, prevalence, impact, health service use and gaps, risk factors and comorbidities of neurological conditions including cerebral palsy, in 2009 the Government of Canada initiated the National Population Health Study of Neurological Conditions (NPHSNC).[29] The NPHSNC included 13 research projects and three national surveys aimed at examining multiple neurological conditions affecting Canadians. One of the projects involved adapting Statistics Canada's Population Health Model (POHEM) framework to create a new population-based longitudinal microsimulation model to project future (up to 2031) health and economic impacts of seven neurological conditions affecting Canadians: this model is called POHEM–Neurological.[29] The NPHSNC set the goal of examining future incidence, prevalence and impact of neurological conditions examined over 5-, 10-, 15- and 20-year horizons. This manuscript focuses on the longest horizon, 20 years, for the model projections.

The objective of our study was to present model-based estimates and projections on current and future health and economic impacts of cerebral palsy in Canada over a 20-year time horizon (2011–2031).

Methods

POHEM–Neurological

The POHEM–Neurological framework is an empirically grounded model that uses Canadian data for demographics and disease incidence and mortality to produce projections over the life cycle of Canadians.[30-34] The model's basic unit of analysis is the individual, or "actor," whose life course is simulated and modelled in continuous time using a Monte Carlo approach.*

The dynamic simulation recreates a synthetic Canadian population at a given point in time (historically and in the future) and ages it, one actor at a time, until each actor's death. Lastly, the model uses status quo assumptions to project future impacts; as such, it assumes that incidence and risk and prognostic factors for the neurological condition being modelled will remain stable throughout the projection period. Further details regarding the methods and procedures involved in POHEM–Neurological can be found elsewhere.[30-34]

The model development process for POHEM–Neurological can be broken into four steps: (1) initialization; (2) yearly updates; (3) model validation; and (4) projection (see Table 1). We used only Canadian, population-based data sources for model initialization and yearly updates.

Step 1: Initialization

Initialization began with historic birth cohorts from 1872 through 2006, which we subjected to the observed historic death rate, as with other Statistics Canada health models.[35,36] Since 1872 represents

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* The Monte Carlo method uses a broad class of computational statistical algorithms that rely on repeated random sampling to obtain numerical results. The essential idea behind the approach is the use of randomness to solve problems that might be deterministic in principle.

TABLE 1
Process for projecting cerebral palsy prevalence, health burden and health care use in Canada using the POHEM–Neurological

	Model-generating step	Model activity	Details of the model step
1	Initialization Generate an initial Canadian population	Initiate a predictive model by creating a synthetic cohort of Canadians.	Historic birth cohorts combined with annual deaths, immigration and emigration. Observed demographic data to 2006 projected to 2031.
2	Yearly Updates Update annual CP incidence and other model parameters	Each year to 2031, update population characteristics, CP incidence, health status and health care costs.	Each year, update the synthetic cohort for demographic changes, CP incidence and deaths. For people with and without CP, estimate Health Utilities Index, health care use (formal and informal) and deaths.
3	Model Validation Validate and calibrate the model	Compare projected CP to observed prevalence in 2009.	Compare projected to observed CP prevalence, by age and sex. Calibrate the model if needed.
4	Projection Generate final projections from 2011–2031	Project CP incidence, prevalence, mortality, health utilities index, HALYs, YLL with CP and health care costs including out-of-pocket expenses and hours of informal caregiving, from 2011–2031.	Generate output tables from projections.

Abbreviations: CP, cerebral palsy; HALY, health-adjusted life year; POHEM, Population Health Model; YLL, years of life lived.

the year of the oldest birth cohort in the LifePaths modelling process,[†] it was used to allow for the simulation and generation of a complete range of ages from newborn to elderly by the year 1971. Hence the model can produce cross-sectional annual tabulations from 1971 that are directly comparable to historical cross-sectional data. For the years 2007 onwards, projected births, deaths and migration (immigration and emigration) used standard Canadian population projections (midgrowth scenario), as estimated by Statistics Canada.[36]

Step 2: Yearly updates of actors' health profiles

An actor's health profile consists of six characteristics: (1) demographics; (2) cerebral palsy status; (3) health status; (4) presence of an informal caregiver; (5) health care costs; and (6) mortality. Each actor's health profile is updated over time, either at an event (e.g. birthday, date of diagnosis of cerebral palsy) or at the change of the calendar year, depending on the profile characteristic. All characteristics were calculated and modelled for people with cerebral palsy and those without neurological conditions. "People with no neurological conditions," as opposed to those without cerebral palsy, was the comparison used because people with cerebral palsy tend to have other comorbid costs that are difficult to separate out (see "Comorbidity, counterfactual population and costs estimation").

Cerebral palsy status: incidence, prevalence and mortality

We applied sex and age-specific cerebral palsy incidence rates to the model's synthetic Canadian population at the beginning of each calendar year. We estimated incidence rates of cerebral palsy using a case identification algorithm that used British Columbia health administrative data from physician billings and hospital discharge abstracts. A case of cerebral palsy was defined as one physician visit or one hospitalization that used the following International Classification of Disease (ICD) codes for cerebral palsy: (1) ICD-10 code G80 (2) ICD-9 code 340; or (3) ICD-9(CM) codes 333.71 or 343. We developed the algorithm with advice from the Canadian Chronic Disease Surveillance System (CCDSS) Neurological Conditions Working Group chaired by the Public Health Agency of Canada.

Based on additional advice from clinical experts in the CCDSS Working Group, we defined incident cases of cerebral palsy as only occurring prior to age 20 years since most children receive a diagnosis in early childhood; after 20 years of age, incidence was assumed to be zero. Incident cerebral palsy cases accumulate over time to generate prevalent cases of cerebral palsy. Relative mortality risks specific to cerebral palsy were applied to actors with cerebral palsy. We estimated sex- and age-specific mortality rates using the same case definition and data used to estimate incidence. The mortality rates specific to cerebral palsy were divided by the mortality rates for the same sex and age group in the general population, thereby generating a relative mortality risk. In POHEM–Neurological, this relative mortality risk for people with cerebral palsy was multiplied by the baseline mortality rate for the Canadian population. In essence, mortality rates for the cerebral palsy population in British Columbia were compared to the mortality rates of the general population (obtained from Statistics Canada), the latter being a combination of life tables and demographic projections.[36] For future projections, the general population's mortality rate is anticipated to gradually decrease over time, reflecting the projected mortality using birth cohorts and the Lee–Carter model as estimated by Statistics Canada.[37] This means that the projected mortality of Canadians with cerebral palsy decreased at the same rate as for those without cerebral palsy. This is an assumption of the POHEM–Neurological model as it incorporates a parameter that allows for an increase or decrease in the relative mortality risk for people with cerebral palsy compared to those without.

During model validation, the model-projected prevalence of cerebral palsy was much higher than that observed in the British Columbia administrative data (see Figure 1). After consulting with clinical experts on the model-projected prevalence, an adjustment parameter was added to POHEM–Neurological that would remove a diagnosis of cerebral palsy from a

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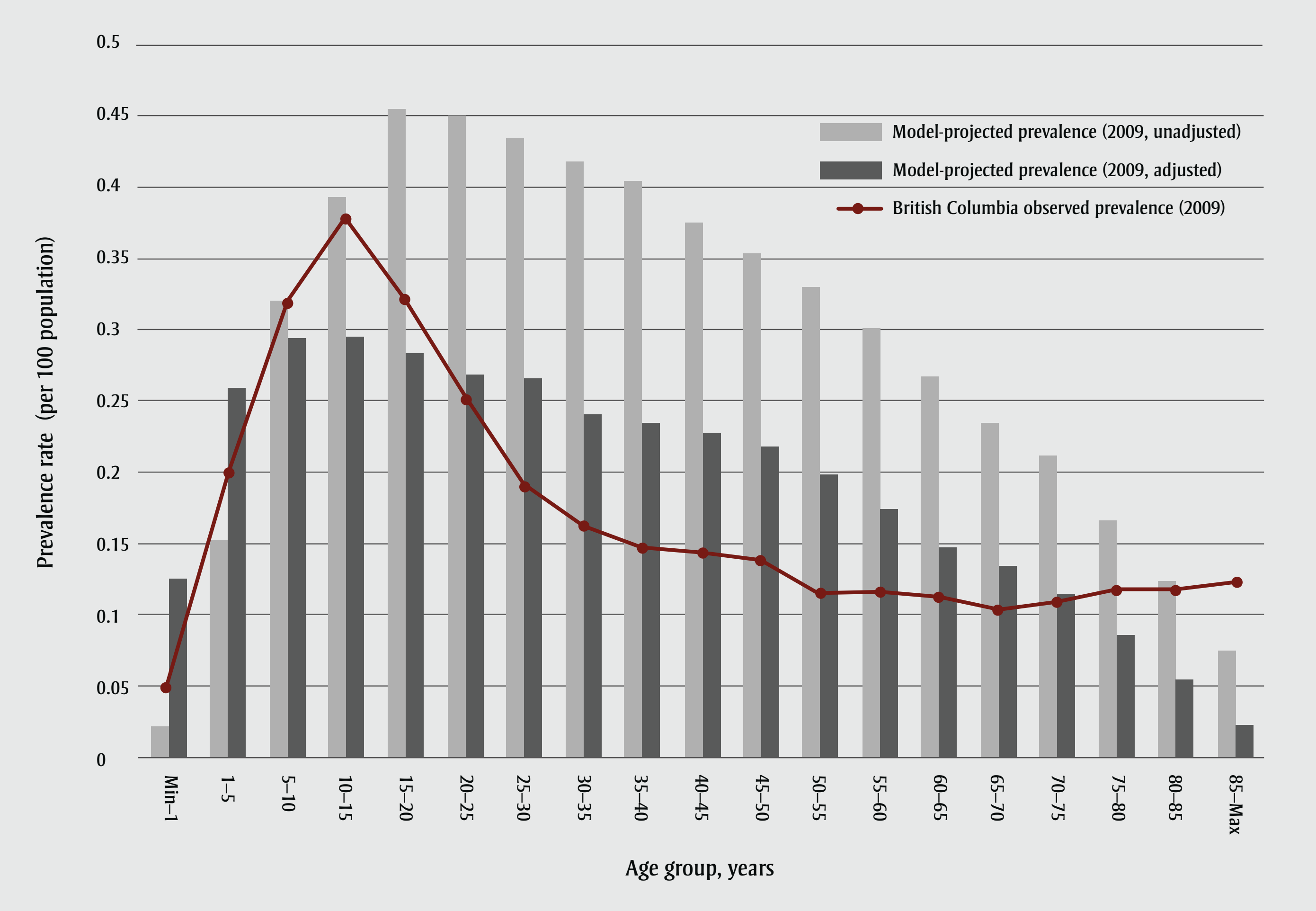
[†] LifePaths is a dynamic longitudinal microsimulation model of Canadian individuals and families that simulates and generates basic individual and family decisions for most discrete events that together constitute an individual's life history. LifePaths creates synthetic life histories from birth to death that are representative of the history of Canada's population.

FIGURE 1
Comparison of 2009 cerebral palsy prevalence rates[a] observed in British Columbia and POHEM–Neurological model-projected, adjusted and unadjusted parameters



Source: POHEM–Neurological (Statistics Canada and Public Health Agency of Canada).

Abbreviations: Max, maximum age; Min, minimum age; POHEM, Population Health Model.

[a] Since POHEM model's basic unit of analysis is the individual person, it creates a population and ages it, one person at a time, until death. The prevalence estimates from POHEM–Neurological can be considered to be rates because the model calculations took into account the person-years of time contributed by an individual over the calendar year and in the disease state in both the numerator and denominator during the modeling process.

proportion of individuals who had previously been prevalent cerebral palsy cases in the model. Although cerebral palsy is a lifelong disorder, the observed incidence rates likely included a high proportion of false-positive diagnoses in the administrative health data source, such as children who were initially given a cerebral palsy code but who were later given a different diagnosis. Although, the standard error and statistical error of the case definition were not validated for this study, the case definition of a single diagnostic code for cerebral palsy within administrative databases has been validated in the Quebec population and found to be highly specific but with lower sensitivity. Aligned with this, the prediction model had a much higher prevalence of cerebral palsy predicted, and it was modified to adjust for this difference in sensitivity. Since the validity of diagnostic codes for cerebral palsy in administrative databases has not yet been established,[28] our study methodology can be considered as the best currently available for predicting cerebral palsy in administrative databases in Canada, being as it is highly specific and providing information that is of value.[28] Evidence of misdiagnoses of neurogenetic disorders as cerebral palsy in early life has been recognized.[38] After adjustment (see Figure 1), the model-generated prevalence "rates"[‡] approached those reported in the literature.[3,19-23]

Health status

The Health Utilities Index Mark 3 (HUI3) is a generic preference-based measure of health-related quality of life that is used to ascertain disease severity.[39] Health-related quality of life is a concept that describes the health of populations and represents a broader health outcome for population health analyses than mortality rates or life expectancy. HUI3 is a utility-based measure that reflects health states with a variety of severity levels, ranging from perfect health

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[‡] Since POHEM's basic unit of analysis is the individual person, it creates a population and ages it, one person at a time, until death. The prevalence estimates from POHEM–Neurological can be considered to be rates because the model calculations took into account the person-years of time contributed by an individual over the calendar year and in the disease state in both the numerator and denominator during the modeling process.

(HUI3 = 1.0) through death (HUI3 = 0). It includes states considered to be worse than death (minimum HUI3 = −0.36).[39] The HUI3 assesses functional health across eight dimensions—cognition, emotion, mobility, dexterity, pain and discomfort, speech, vision and hearing—and provides a valid measure of the functional health status and quality of life experienced by people with cerebral palsy.[39,40] Disability categories have been previously validated for HUI3: none (1.00); mild (0.89 to 0.99); moderate (0.70 to 0.88); and severe (less than 0.70).[41] It should be noted that HUI3 scores can vary depending on the source of valuation, which could either be by proxy or child. A 2019 study[42] reported that HUI3 values can range from −0.013 to +0.84 for children with cerebral palsy. For individuals aged 15 years and older with cerebral palsy, mean HUI3 values (with standard errors) were estimated from the 2011 Survey on Living with Neurological Conditions in Canada (SLNCC).[43] Means were calculated separately for males and females, and for two age groups: 15–34 years and 35 years and older. For children younger than 15 years with a diagnosis of cerebral palsy, a population not covered by the SLNCC, mean HUI3 values were estimated from the 2006 Participation and Activity Limitations Survey (PALS).[44] The estimate was derived for boys and girls combined (i.e. it was not sex specific).

The impact of cerebral palsy can also be assessed by the years of life lost due to the condition, that is, years lost due to premature death from a disease. To calculate years of life lost, we first determined the age of death for all actors with cerebral palsy in each calendar year. Next, we estimated years of life lost for a scenario in which the mortality hazard for cerebral palsy was the same as for actors without cerebral palsy and recalculated the age of death: years of life lost was the difference in age of death between the two calculations. Health-adjusted life years *lost* indicates the years of life lost living in a healthy state, combining morbidity and mortality. Health-adjusted life years (HALYs) were estimated from the product of years of life lived (age at death) multiplied by the annual HUI3 for each actor over their lifetime.

The presence of an informal caregiver

Informal caregiving refers to unpaid care provided by family and friends to Canadians living with a chronic health condition, disability or aging needs.

For each actor in POHEM–Neurological, we assessed the likelihood of having an informal caregiver based on the actor's age, cerebral palsy status and health status (HUI3). If an individual was assigned the presence of an informal caregiver, additional characteristics were assigned based on empirical estimates from the 2011 SLNCC and augmented with the 2012 General Social Survey (GSS)[45]: (1) hours of care received; (2) health status of their caregiver; and (3) out-of-pocket expenses incurred by caregivers.

Health care costs

We estimated formal health care costs in incident and prevalent cohorts with cerebral palsy and ascertained these costs using administrative health data in British Columbia and Ontario. The health care costs were estimated using 2010-dollar values; as such, inflation was not factored into the cost projections. In addition, prevalent and counterfactual cohort costs were obtained for the period 1 April 2009 through 31 March 2010, in both Ontario and British Columbia.

We examined incident cohort costs in Ontario over the same period. We obtained available incident cohort costs in British Columbia for the 12-month period starting on 1 April of the incident year (2006, 2007 or 2008). Formal health care costs were those covered by provincial health plans in health components grouped as follows: (1) acute hospitalization; (2) physician services; (3) prescription drug data;[§] (4) rehabilitation hospitals; (5) Ontario provincially funded home care; (6) Ontario residence and care in a long-term care facility; and (7) Ontario provincially funded assistive devices. Costs included two categories of out-of-pocket costs (expenses paid by an individual but not refunded by insurance or government): those of the patient and those for the caregiver.

Comorbidity, counterfactual population and costs estimation

As noted previously, individuals with cerebral palsy frequently have comorbid conditions. For instance, one in four children with cerebral palsy have epilepsy, one in two have an intellectual disability, and one in four have a behaviour disorder, including Autism Spectrum Disorder, and other musculoskeletal conditions.[4,47] As such, it is sometimes difficult to isolate the contribution of cerebral palsy to health care costs, caregiving or mortality from those due to comorbidities. To address this issue, we identified a counterfactual (nonneurological condition) population cohort to determine the net impact of cerebral palsy. For example, if the prevalence of comorbidity X is greater in individuals with cerebral palsy than those without, the additional health care costs (or receipt of caregiving hours) associated with this greater morbidity should be attributed to individuals with cerebral palsy. Thus, costs over and above those of the age-sex counterparts without a neurological condition were attributed as being the additional direct cost burden of the neurological condition irrespective of comorbid conditions. Conversely, if individuals with cerebral palsy use less of a particular health care resource than those without a neurological condition, our model should also reflect this lower utilization.

The counterfactual nonneurological condition cohort consisted of all individuals in the respective datasets who had not otherwise been classified as having any of the seven neurological conditions of interest for the NPHSNC microsimulation project, namely Alzheimer disease and other dementias, multiple sclerosis, cerebral palsy, epilepsy, Parkinson disease and parkinsonism, traumatic brain injuries and traumatic spinal cord injuries.

Step 3: POHEM–Neurological validation

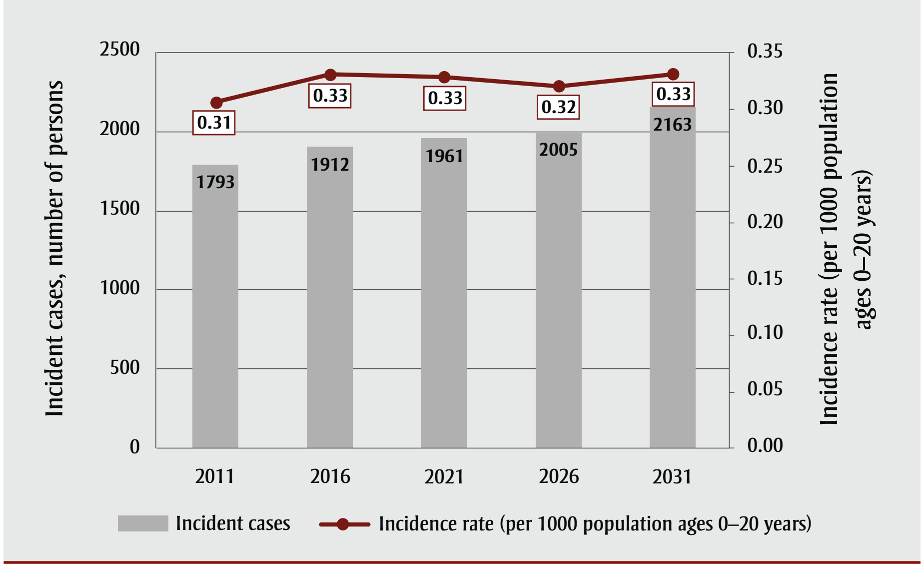
The cerebral palsy prevalence "rates" were derived from an unvalidated case algorithm that was used during feasibility studies as part of the NPHSNC (see Figure 1). As such adjustments were made to calibrate the POHEM–Neurological cerebral palsy prevalence projections to align with the widely accepted prevalence benchmark of 2.5 per 1000 live births cited in systematic reviews and literature.[3,19-23] (See "Step 2: Yearly updates of actors' health profiles".)

[§] British Columbia had comprehensive prescription drug cost data for all prescriptions dispensed in community pharmacies, regardless of age, while Ontario had comprehensive data for persons aged 65 years and older with limited data for lower-income cerebral palsy patients receiving provincial support for their disease-modifying therapies. Where we had costs from both provinces, these were weighted based on an input parameter (75% Ontario, 25% British Columbia). The method used for this analysis was based on Wodchis et al.[46]

Step 4: Projection

We projected multiple main model outputs through to 2031 including incident and prevalent cerebral palsy cases in Canada; years of life lived with cerebral palsy; HALY; health care costs including costs for each of the seven sectors; out-of-pocket expenses; and hours of informal caregiving. In addition, we projected three parameters from the perspective of caregivers: hours of caregiving; out-of-pocket expenses; and health status (HUI3).

FIGURE 2
Projected cerebral palsy incidence cases and rate, ages 0–20 years, both sexes, 2011–2031



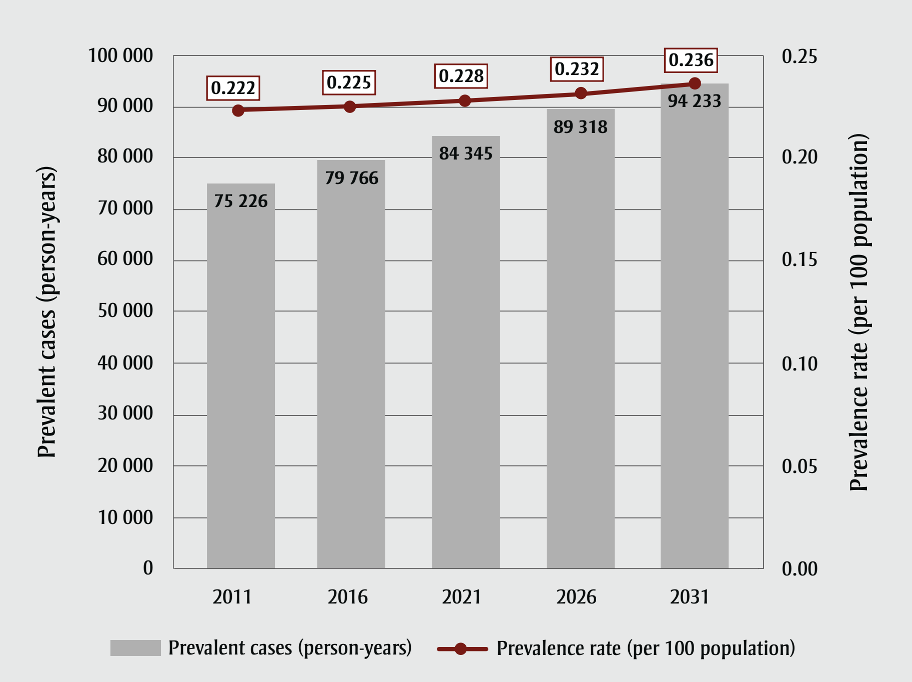
Results

According to POHEM–Neurological projections, the number of children under 20 years of age who will be newly diagnosed with cerebral palsy will increase from approximately 1800 in 2011 to nearly 2200 in 2031 (Figure 2). The rate of cerebral palsy incidence remains stable over the projection period, around 0.3 per 1000 for children less than 20 years old. The majority of the incident cases will occur among children between 1 and 5 years of age, as most cases of cerebral palsy are diagnosed in the first 2 years of a child's life (data not shown).

As with incidence, the number of people with cerebral palsy is expected to increase from more than 75 000 in 2011 to more than 94 000 in 2031, although the prevalence only shows a slight increase over this period, from 2.2 in 2011 to 2.4 per 1000 population in 2031 (Figure 3). The increase can be attributed to the growth of the Canadian population and the increased life expectancy.

In 2011, Canadians with cerebral palsy had reduced functional health (as defined by HUI3 scores) compared with Canadians without neurological conditions. Average HUI3 for Canadians with cerebral palsy was below 0.7, the cut point for "severe disability," at all ages (Figure 4). The average HUI3 for Canadians without a neurological condition was between 1.0 and 0.8; only upon reaching 80 years of age does the average HUI3 score fall below 0.7. The difference in the HUI3 trend remained relatively stable because the model assumes no change in age-specific HUI3 scores.

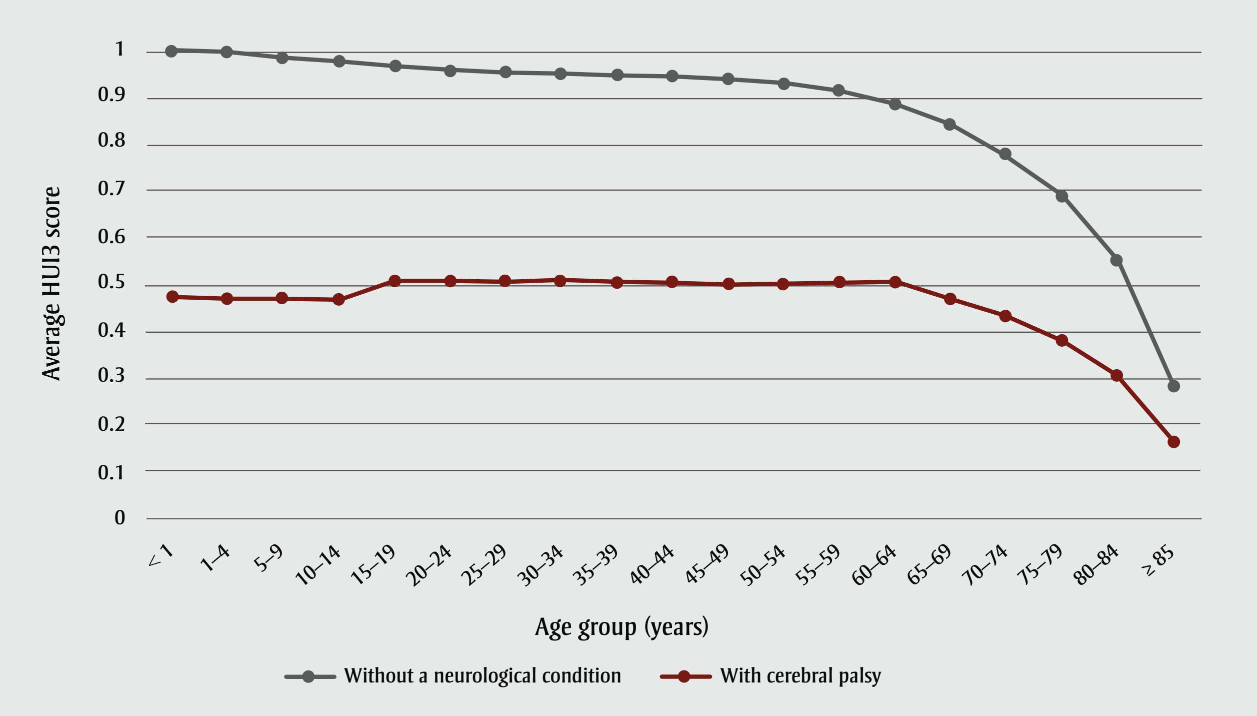
FIGURE 3
Projected cerebral palsy prevalence count (person-years) and rate, Canadian population all ages, both sexes, 2011–2031



Given the lower functional health of Canadians with cerebral palsy, the increased demand for informal care is not surprising. A much larger proportion of individuals with cerebral palsy than those without a neurological condition receive informal caregiving; this holds true at all ages (Figure 5). Overall, individuals with cerebral palsy start to receive informal care at an earlier age, and the proportion receiving informal care rises sharply, remaining consistently higher than among their counterparts without a neurological condition. People without a neurological condition only begin to require greater amounts of informal care as they approach 70 years of age.

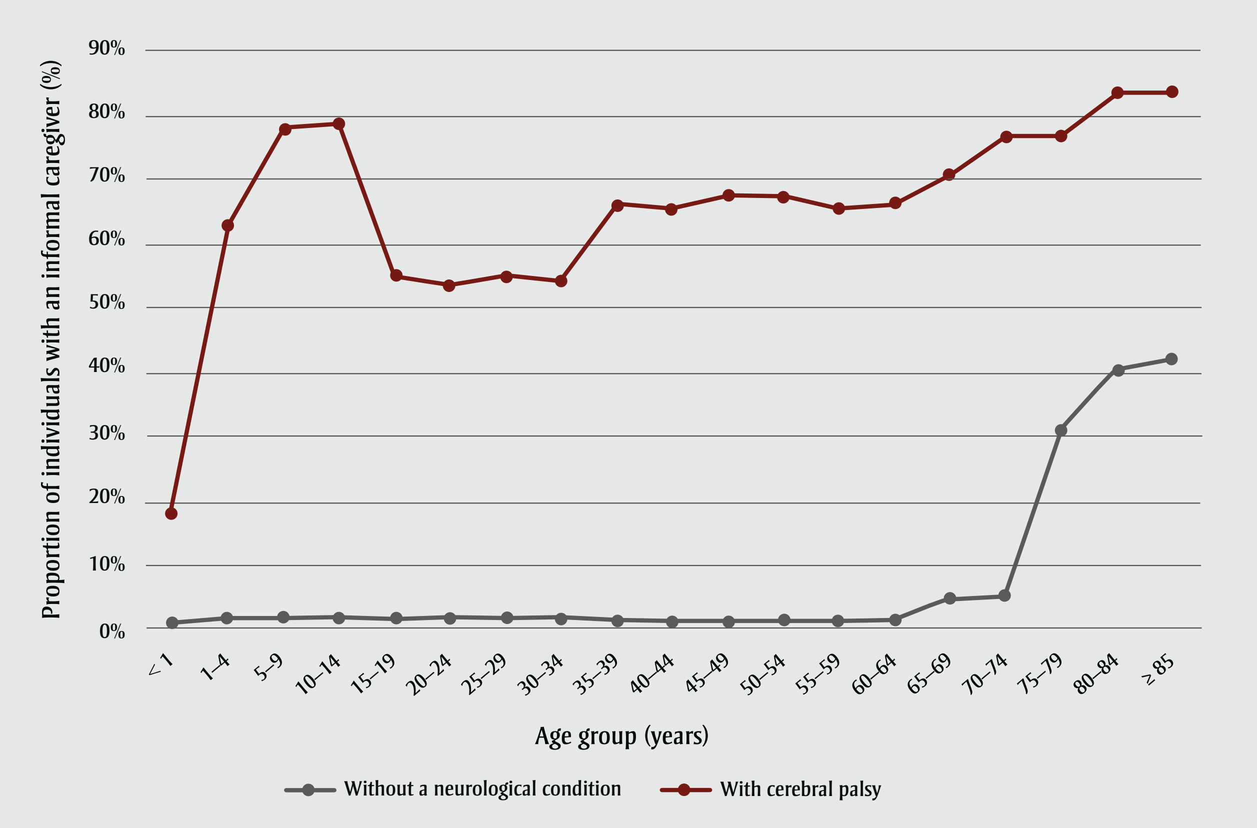
POHEM–Neurological also projects that individuals aged over 35 years will receive considerably more hours of informal care per week than Canadians who do not

FIGURE 4
Average Health Utilities Index score, by cerebral palsy status and age, 2011



Abbreviation: HUI3, Health Utilities Index Mark 3.

FIGURE 5
Proportion of individuals with an informal caregiver, by cerebral palsy status and age, 2011



have cerebral palsy, that is, 30 versus 18 hours per week (Figure 6).

We measured years of life lived with a condition and HALYs to assess the impact of cerebral palsy on the quality of life of Canadians. Figure 7 shows years of life lived and HALYs for Canadians with cerebral palsy and their counterparts without a neurological condition, by decade of birth. The model projects that people born between 2010 and 2020 will have longer periods in poor health than their peers without a neurological condition. Women with cerebral palsy are projected to lose, on average, about 41 years of life in good health, while men with cerebral palsy are expected to lose approximately 33 years. In comparison, Canadian women who do not have a neurological condition are expected to lose about 13 years of life in good health while their male counterparts are expected to lose about 11 years.

FIGURE 6
Average number of hours of care received per week among those receiving informal caregiving, by cerebral palsy status, 2011

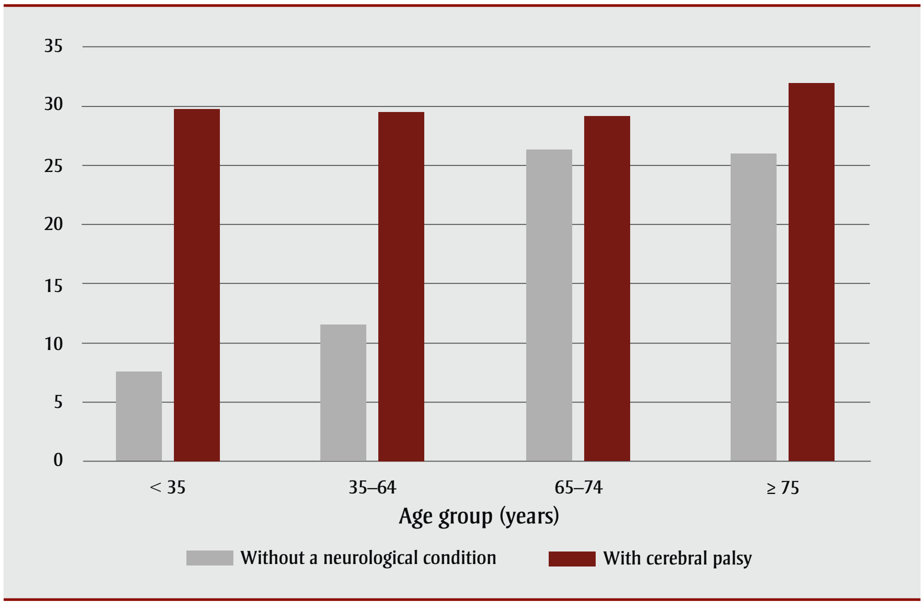
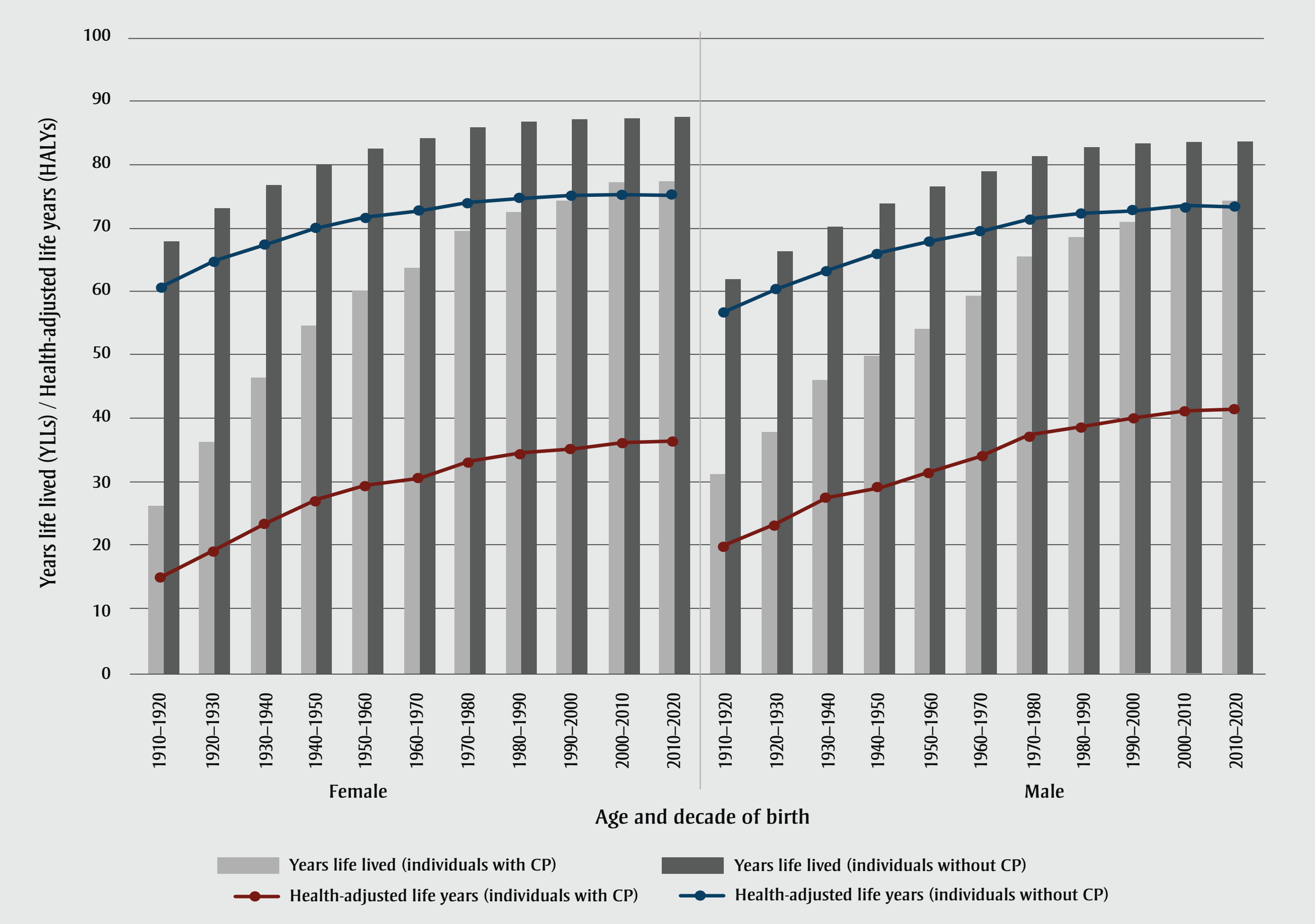


FIGURE 7
Mean years of life lived and health-adjusted life years, individuals with and without cerebral palsy, by sex and decade of birth



Abbreviations: CP, cerebral palsy; HALY, health-adjusted life year; YLL, years of life lived.

Another important difference between Canadians with cerebral palsy and their counterparts who do not have the condition is that their health care expenditures tend to be higher. Table 2 compares mean per capita total health sector costs for Canadians with cerebral palsy and those who do not have a neurological condition.

The comparison shows that the greatest differences in costs occur prior to age 15 years. Around age 20 years, the cost differences begin to decrease (a ratio of 14:1) considerably and level off as people continue to age. By age 30 years, the ratio of total health care costs falls to 7:1. The model projects that the total mean per capita total health care costs will remain stable over the 20-year projection period because no change in costs per year was assumed.

The impact of living with cerebral palsy was also assessed by comparing the out-of-pocket expenses incurred by informal caregivers of Canadians with cerebral palsy and of those without a neurological condition. The difference was highest for informal caregivers of individuals aged under 35 years, with out-of-pocket expenses almost 50 times greater for caregivers of individuals with cerebral palsy than for those caring for individuals without a neurological condition (see Figure 8). A 36-fold difference is seen for informal caregivers of individuals aged 35–65 years. After age 65 years, the differences in out-of-pocket expenses are not as substantial.

As with other POHEM–Neurological parameters, the out-of-pocket expenses trend in 2011 is not expected to change by 2031 due to the status quo assumptions used during the modelling process.

Figure 9 also shows that total health sector costs associated with living with cerebral palsy will rise consistently over the next 20 years while the cost of care not related to cerebral palsy will remain stable over the same period.

Discussion

POHEM–Neurological projects the epidemiological and economic impacts of cerebral palsy over a 20-year period. Although the age- and sex-specific incidence of cerebral palsy was assumed to be constant over the projection period, the model projects an increase in the number of Canadians with cerebral palsy, from more than 75 000 in 2011 to more than 94 000 by 2031. As cerebral palsy is a chronic medical condition, more long-term supportive care services may be required to deal with the increase in the number of people with the condition.

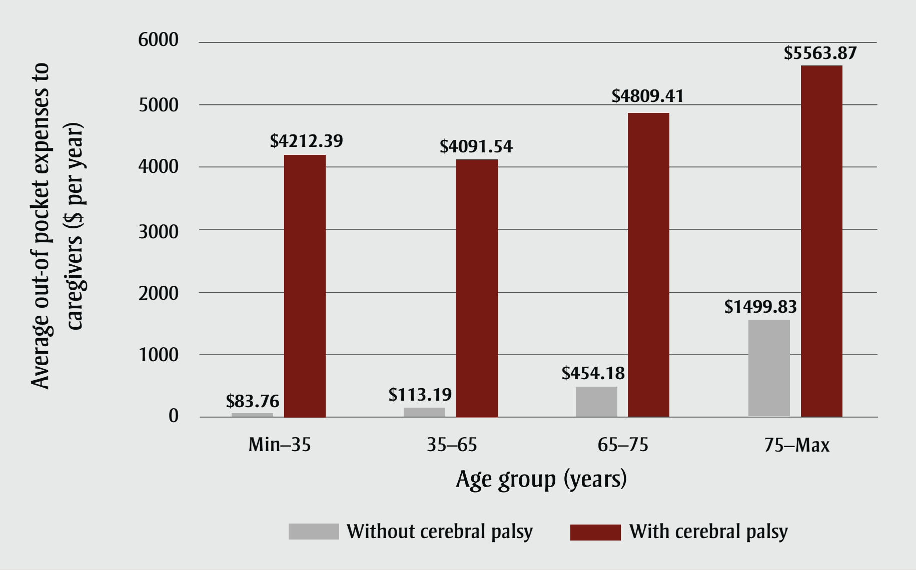
In addition, POHEM–Neurological reflects the fact that people with cerebral palsy tend to have longer periods in poorer health-related quality of life (as reflected by lower scores on the HUI3) than their counterparts who do not have the condition. Although the core feature of cerebral palsy is nonprogressive motor impairment, cognitive impairment, feeding difficulties,

TABLE 2
Projected mean per capita total health care sector costs,[a]
Canadian population with and without the condition, both sexes, by age, 2011 and 2031

Age group, years	Total health care costs, $				Ratio of cerebral palsy to nonneurological health care costs
	With cerebral palsy		Without a neurological condition		
	2011	2031	2011	2031	
< 1	30 497	30 353	1 900	1 899	16:1
1–4	11 666	11 671	564	564	21:1
5–9	10 129	10 140	484	484	21:1
10–14	10 108	10 149	463	463	22:1
15–19	9 765	9 771	702	701	14:1
20–24	8 471	8 489	854	851	10:1
25–29	7 615	7 607	1 066	1 068	7:1
30–34	8 599	8 580	1 215	1 227	7:1
35–39	7 203	7 185	1 216	1 225	6:1
40–44	8 678	8 654	1 305	1 308	7:1
45–49	9 322	9 324	1 575	1 574	6:1
50–54	10 915	10 903	1 998	1 996	5:1
55–59	13 868	13 983	2 514	2 513	6:1
60–64	14 158	14 151	3 204	3 207	4:1
65–69	17 248	17 263	4 129	4 132	4:1
70–74	20 498	20 516	5 168	5 174	4:1
75–79	19 302	19 371	6 343	6 348	3:1
80–84	21 344	21 343	7 546	7 550	3:1
≥ 85	23 555	24 141	9 197	9 177	3:1

[a] Total health care costs include physician, acute and rehabilitation hospital, prescription medication, provincial home care, assistive devices, long-term care and out-of-pocket expenditures for informal care by individuals with the condition and family/friend caregivers. Costs were estimated in 2010 constant (no inflation or discounting) Canadian dollars.

FIGURE 8
Average (per capita) out-of-pocket expenses incurred by informal caregivers, for individuals with and without cerebral palsy, by selected age group, 2011



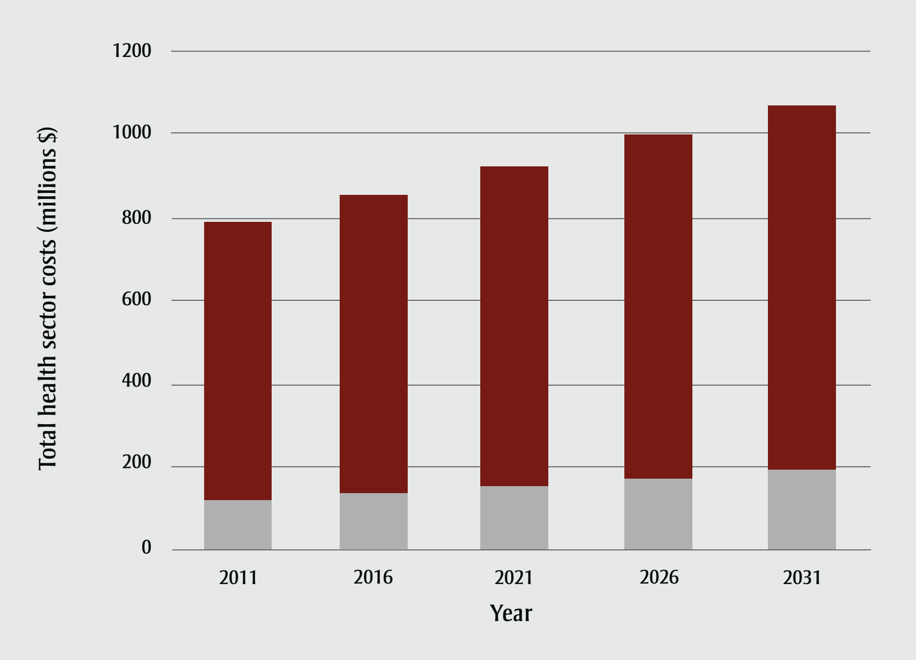
Abbreviations: Max, maximum age; Min, minimum age.

seizure activity, vision and mobility impairment are commonly associated conditions that may progress over time. Our study found that women living with cerebral palsy lose, on average, about 41 years of life in good health compared with Canadian women who do not have the condition: this value was about 33 years for men. The difference in HALYs between men and women is largely due to differences in life expectancy as women live longer in poorer health (see Figure 7).

Long periods living in poor health and functional disability can result in an increased burden on the affected individuals and their families as well as on the health care system.

Total health care costs in all categories for Canadians with cerebral palsy will rise by approximately 33% compared to less than 1% for costs not related to cerebral palsy. In addition, people with cerebral palsy and their families are expected to incur substantial out-of-pocket costs. As medical care and treatments advance, an increasing number of children with cerebral palsy are reaching adulthood. However, the provision of health care services and specialized programs tends not to keep pace with the needs of families and individuals, thereby making the transition from childhood into adulthood a challenge.[14,48,49]

FIGURE 9
Total health sector costs (millions $) for individuals with cerebral palsy, by year



As people with cerebral palsy experience a loss or reduction in the provision of health care services when they transition to adulthood, their demand for informal caregiving increases, and parents and relatives often step in to provide financial support and the required care. As noted in our study, the hours provided of informal care per week and out-of-pocket expenses incurred by the caregivers of people with cerebral palsy are substantially higher than those incurred by the relatives or friends of those who do not have the condition. The economic impact in terms of loss of income for informal caregivers is also an issue that requires more attention over the next 20 years as the impact of this condition is assessed.

Strengths and limitations

Our study offers important insights that strengthen our understanding of a complex neurological condition. POHEM–Neurological projections can help in the planning of future health programs and strategies for Canadians living with cerebral palsy.

The present findings from POHEM–Neurological assumed no future change in age- and sex-specific cerebral palsy incidence, relative mortality or functional health (HUI3). As such, the primary drivers of future prevalence, impact and cost of cerebral palsy derived from POHEM–Neurological are based on projected demographic shifts of the Canadian population. However, future iterations of the model could evaluate alternate scenarios, such as cost discounting, or variations in incidence or relative mortality, for their impact on the future of cerebral palsy in Canada.

Although incidence, mortality and costs were derived from Canadian provincial administrative data, the case definition algorithm for identifying individuals with cerebral palsy had not been validated at the time of implementation. In initial assessments, model-projected prevalence rates were notably higher than observed rates. Therefore, a parameter was included in POHEM–Neurological that would, for a subset of actors, remove the designation of cerebral palsy from some prevalent cases. Such a parameter accounts for potential misdiagnosis, which is known to occur—hence the term "masqueraders of cerebral palsy."[38] A recent Canadian study compared an administrative data-based definition of cerebral palsy to a registry-based approach and found that 41% of cases administratively identified were not in the registry and that the former also missed 34% of registry-captured cases.[28] Therefore, a more sensitive definition of cerebral palsy may be needed if administrative data are to be used to capture incidence and costs associated with this condition. Alternatively, a combination of methods that included combining ICD codes and cerebral palsy diagnoses written in medical or educational records, as described in a 2019 study,[50] can be further analyzed to reduce errors in identifying cerebral palsy cases.

Limitations of POHEM–Neurological microsimulation models are expansively discussed in studies by Finès et al.,[30] Manuel et al.,[31] Amankwah et al.[32] and Hennessy et al.[51] Limitations such as applying incidence and mortality data obtained from one province to the entire nation, health care costs from two provinces, type I and II model error uncertainties and adjustments made to reduce the effects of the limitations are addressed in these studies.

Conclusion

Our study findings suggest that Canadians with cerebral palsy will continue to experience reduced quality of life, increased disabilities and a rising need for supportive services, including informal care. Health care expenditures, particularly out-of-pocket expenses, are projected to remain high over the next 20-year period. As people with cerebral palsy tend to have a normal life expectancy, they face continuous challenges related to an ongoing need for specialized medical care, accommodated education services, developmental assistance and general assisted living support. In addition, the indirect costs of lost productivity and wages to individuals and society, as well as occupational limitations, needs to addressed. More attention may be required for the transitional needs of people living with cerebral palsy as they pass through the different stages of life in situations where appropriate health care services and programs are inadequate or lacking.

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Conflicts of interest

MO is a site investigator for clinical trials in spinal muscular atrophy by Roche, Biogen and Cytokinetics, and a past member of the data safety monitoring board for Avexis.

Authors' contributions and statement

NA, MO and RG contributed to the concept, drafting, analysis interpretation and critical review of the article. MO provided specialized content expertise on cerebral palsy, while RG, CB, DGM, RW, PF, JB, KT and KR contributed to data acquisition, microsimulation modelling and review of the article.

The opinions, results and conclusions reported in this paper are those of the authors, do not necessarily reflect those of the Government of Canada, and are independent from the funding sources. No endorsement by Institute for Clinical Evaluative Sciences or the Ontario MOHLTC is intended or should be inferred. Parts of this material are based on data and information compiled and provided by the Canadian Institute for Health Information. However, the analyses, conclusions, opinions and statements expressed herein are those of the authors, and not necessarily those of the Canadian Institute for Health Information.

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